KCE REPORT 241

Federaal Kenniscentrum voor de Gezondheidszorg
Centre Fédéral d'Expertise des Soins de Santé
Belgian Health Care Knowledge Centre

CARDIOVASCULAR PRE-PARTICIPATION SCREENING IN YOUNG ATHLETES

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HEALTH TECHNOLOGY ASSESSMENT

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COLOPHON

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LIST OF ABBREVIATIONS

ABBREVIATION	DEFINITION
ACC	American College of Cardiology
AHA	American Heart Association
ARVC	arrhythmogenic right ventricular cardiomyopathy
CHD	Coronary Heart Disease
CVD	Cardiovascular Disease
ECG	Electrocardiogram
EPS	Electrophysiologic Study
ESC	European Society of Cardiology
H&P	history-taking and physical examination
HCM	hypertrophic cardiomyopathy
HR	Hazard Rate
HR-QoI	Health-Related Quality of Life
HTA	Health Technology Assessment
LQTS	long-QT syndrome
P PS	pre-participation screening
QoL	Quality of Life
RCT	Randomized Controlled Trial
SCA	sudden cardiac arrest
SCA/D	sudden cardiac arrest or death
SCD	Sudden Cardiac Death
SR	Systematic Review
WPW	Wolff-Parkinson-White syndrome

SCIENTIFIC REPORT

1 INTRODUCTION AND SCOPE

A young athlete collapsing and dying during a sports event is a devastating incident with substantial impact on the lay community. In more than half of non-traumatic deaths on the sports field, an underlying unsuspected cardiovascular disease appears to be the responsible underlying condition.[1] In an attempt to try to prevent such tragedies, pre-participation screening for cardiovascular disease has been advocated. The reasoning is that a medical examination can identify or raise suspicion for an increased risk for sudden cardiac death (SCD) in an asymptomatic person, and that appropriate action can eliminate or at least reduce this risk.

This report has been commissioned by the Flemish government (Vlaams Agentschap Zorg en Gezondheid). During the first stakeholders consultations it appeared that this project was also of high interest for the French Community that published a Decree in April 2014 regarding health risk prevention in sports.

Aim and scope of the report

The aim of this report is to assess the effectiveness of cardiovascular pre-participation screening to prevent SCD in non-professional athletes, aged 14-34 years. The target population are young people who plan to become a member of a sports club/federation, or who want to participate in a mass sports event.

The report addresses the following research questions:

1. What is the clinical effectiveness of cardiovascular pre-participation screening of asymptomatic young, non-professional athletes (14-34y)? Both the benefit in terms of SCD prevention as potential harms (overdiagnosis and overtreatment) are to be considered.
2. What is the cost-effectiveness of cardiovascular pre-participation screening of young, non-professional athletes?
3. What is the current practice concerning legal regulations, legal liability of physicians and sports clubs involved in pre-participation screening and existing Belgian initiatives? What are the implications from the point of view of insurers?
4. What ethical issues must be considered in pre-participation screening?

What is "screening"?

This report adopts the definition of "screening" as presented in the seminal paper by Wilson and Jungner[2]: "the presumptive identification of unrecognized disease or defect, by the application of tests, examinations, or other procedures which can be applied rapidly. Screening tests sort out apparently well persons who probably have a disease from those who probably do not". Individuals consulting their doctor because of cardiac or other symptoms - including serious anxiety with respect to sports participation – do not qualify for screening, but are considered "patients". They are not included in the scope of this study.

In its most basic format, cardiovascular pre-participation screening consists of taking a personal and family history with a physical examination. To increase the sensitivity of such basic examination, it has been advocated to include an electrocardiogram (ECG) in the screenings protocol.[3]

Special focus in the report will be on the impact of adding a rest-ECG on the effectiveness of screening. Furthermore, economic, legal and ethical implications will be discussed.

Interventions out of the scope of this study

The immediate management of sudden cardiac arrest (SCA) in athletes with the aim to prevent death, i.e. bystander cardiopulmonary resuscitation, the implementation of emergency medical services and the availability and use of automatic external defibrillators are beyond the scope of the present report.

The benefit/harm of tests other than the rest-ECG, either performed at the initial cardiovascular screening, or later on after a first positive testing will not be discussed in this report. These tests include e.g. exercise-ECG, echocardiography and genetic testing.

The detection of other disorders (e.g. musculoskeletal) that may be a point of interest in a pre-participation screening program, is also out of scope of this report. It will be addressed in a guideline that will be produced by the Flemish association of General Practitioners (Domus Medica).

2 SUDDEN CARDIAC DEATH

The primary objective of cardiovascular pre-participation screening is to prevent sudden cardiac death associated with sports activities. Therefore, in this preliminary section we provide background information on the sudden death phenomenon.

2.1 Definitions

2.1.1 Sudden death

Sudden cardiac death (SCD) is defined as a natural death due to a cardiac cause, heralded by an abrupt loss of consciousness within one hour of the onset of acute symptoms.[4] Pre-existing heart disease may have been known to be present, but SCD may as well be the first (and only) symptom of cardiac disease. Most of these events are caused by a cardiac arrhythmia that is characterised by an extremely fast and uncoordinated activation of the heart (ventricular fibrillation), resulting in a mechanical standstill or sudden cardiac arrest (SCA). Because SCA mostly occurs out-of-hospital and given the very short time interval (minutes) that is available to intervene, SCA in the general population most often leads to death. The only way of restoring normal heart rhythm in these patients is by means of defibrillation, the application of an electrical shock to the chest which depolarises the heart and enables normal heart rhythm to resume. Only in rare instances, victims are lucky enough to develop a SCA in an environment where immediate advanced life support is available. This typically occurs in a hospital or in a public place where bystanders, trained in advanced life support can start resuscitation until the life-saving external defibrillation shock restores heart rhythm. If patients with ventricular fibrillation are defibrillated immediately, survival rate is almost 100%. After delays of 4 to 5 minutes, the survival rate decreases to 15 to 40%, and after 10 minutes or longer, 95% of the victims die.[5] Since most cases of SCA lead to SCD, both expressions are often used interchangeably.

Sports-and exercise-related SCD is defined as SCD occurring during or within 1 hour of moderate- to high-intensity exercise.[6]

In the lay press, the terms "heart failure" and "heart infarction" are often used to denote SCD. However, in scientific language, these terms refer to specific disorders that may lead to SCD but not necessarily do so.

2.1.2 Competitive athletics

Reports on cardiovascular screening have predominantly involved populations of adolescents and young adults participating in competitive athletics, which is defined as "participation in an organised team or individual sport that requires regular competition against others as a central component that places a high premium on excellence and achievement, and requires some form of systematic training".[4]

In this report, we define an athlete as someone who participates in competitive athletics. Professional athletics is beyond the scope.

In practice, differentiating competition from a non-competitive exercise may be rather hypothetical since one can assume a competitive spirit in many participants of any sports related encounter.

2.1.3 Age group of interest

In the present report we have chosen to consider the same age group (14-34 years) as the Belgian Superior Health Council (Hoge Gezondheidsraad - Conseil Supérieur de la Santé) in its 2013 report on the same subject.[7]

The lower age limit is introduced because defining an abnormal rest-ECG is particularly difficult in children and adolescents since the rest-ECG continues to mature until adulthood.[8] Furthermore, most of the genetic cardiac diseases that may lead to SCD do not manifest until puberty.

The upper age limit is chosen because from the age of 35 years on, SCD is most often due to ischemic heart disease, a condition that is almost never encountered in younger individuals. Some authors put this upper age limit at 25 years,[9] others at 39 years.[10]

2.1.4 Electrocardiography

An electrocardiogram (ECG) is a recording of the electrical activity of the heart. It allows to detect abnormalities of the rhythm of the heart. Waveform alterations may be indicative for structural heart disease. A standard ECG is performed at rest and involves the analysis of the electrical activity of the heart from different reference points on the human body resulting in 12 vectors; hence a standard ECG sometimes is also referred to as a 12-lead-ECG or rest-ECG.

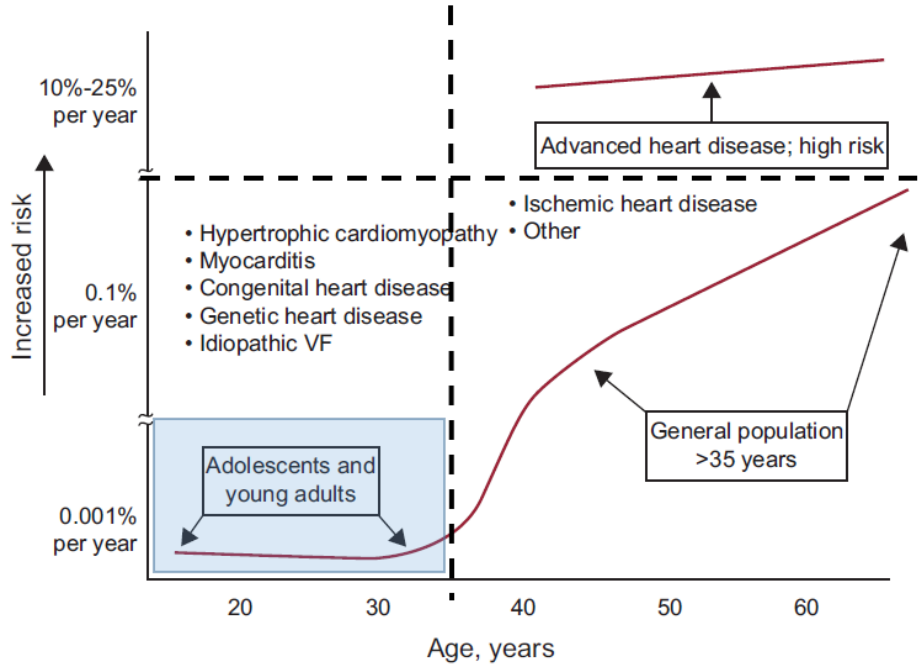
Cardiac disease in the general population may manifest during exercise only, and therefore ECG recording in clinical practice may also be obtained during exercise. However, the exercise ECG will not be further considered in this report.

2.2 Sudden cardiac death in the general population

SCD is among the most common causes of death in developed countries. It is estimated that yearly about 0.1 to 0.2% of the population dies suddenly.[5] Most of them are older than 34 years of age, and the incidence increases sharply with advancing age as shown in Figure 1 (right lower quadrant).

This means that in Belgium presumably more than 10,000 middle-aged or elderly people die each year (i.e. more than one SCD victim each hour) as a consequence of a sudden cardiac arrest. Virtually any cardiac disease can lead to a fatal arrhythmia. The most common (75%) underlying disease is acquired coronary heart disease (ischaemic heart disease), where SCD may be triggered by an acute myocardial infarction (heart attack) or may result from an arrhythmia originating in myocardial scar tissue resulting from a previous, sometimes subclinical, infarction.[5] Apart from ischaemic heart disease, SCD may be caused by cardiomyopathies (heart muscle anomalies) or end-stage valvular or hypertensive heart disease. In very rare instances, SCD is caused by a genetic arrhythmogenic heart disease. However, in children, adolescents and young adults, in whom SCD is an extremely rare event (Figure 1) this group of diseases becomes relatively important since acquired heart disease is very rare in them.

Figure 1 – Age dependency of the incidence and causes of sudden cardiac death

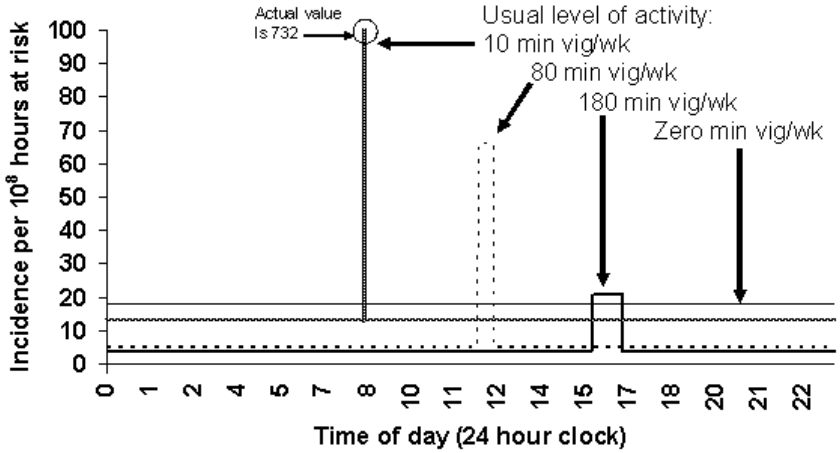


Source: Adapted from Myerburg et al.[11, 12] The shaded area indicates the target population of the present report.

People who regularly engage in physical activity are at lower risk of cardiovascular disease than others, and the more one exercises, the better the outcome.[13] Habitual exercise is associated with an overall reduction in the risk of SCD and diminishes the risk of sudden death during vigorous exertion.[14] It has however been demonstrated that the risk of SCD transiently increases during exercise, but the overall benefit of exercise on the risk of cardiovascular disease and SCD remains convincing.[15]

This apparent paradox becomes clear when considering its graphic representation in Figure 2.

Figure 2 – Risk of SCD during exercise and at rest, by usual level of activity



Source: Haskell et al.[16] The y-axis shows the SCD risk. For example, the graph should be read as follows: the risk for SCD in an individual with an average of 80 minutes of vigorous activity per week is 5 per 10^8 hours while at rest, and 67 per 10^8 hours (0.67 per million sports-hours) during this 80-minute exercise session.

Based on these findings, which are mostly retrieved from observational studies that included people older than 40 years of age, it has been argued that "*To speak of sudden death as a risk of exercise is misleading. Sudden death is, more accurately, a risk of inactivity*".[16]

Key point

In middle-aged and elderly people, sudden cardiac death most often occurs in the context of coronary heart disease. It represents the most common mode of death, affecting more than 10 000 Belgians per year, i.e. more than one per hour.

2.3 Sudden cardiac death in young sporty individuals

There is strong evidence demonstrating that the physical fitness and health status of children and youth are substantially enhanced by frequent physical activity.[16] The US National Heart, Lung and Blood Institute (NHLBI) strongly recommends at least one hour of moderate-to-vigorous physical activity every day of the week for children over 5 years of age and vigorous intense physical activity on 3 days per week. Jogging or playing baseball are given as examples of moderate-to-vigorous physical activities whereas running, playing singles tennis and soccer as examples of vigorous physical activities. The NHLBI also concluded that there is no evidence of harm associated with increased physical activity in children and adolescents.[17]

Nonetheless, SCD in young sporty individuals unfortunately happens.

It remains unknown if SCD in young people follows the same pattern as what has been observed in middle-aged and elderly people as shown in Figure 2. A sudden death of a young person occurring during a sports activity, and especially during a mass competition, attracts more public and media attention than if it occurs in everyday life at home or at school. This may lead to the perception that SCD occurs more often during sports activities (cf. the peaks in the curves in Figure 2).

In an ideal scientific world, incidence rates of SCD in the young would be known in sedentary and sporty individuals, with data on whether sudden death occurred at rest or during exercise, and whether exercise-related SCD occurred during everyday activities, recreational or competitive sports activities. In real world however, our knowledge is limited.

In the absence of strictly standardised registries, it remains unclear whether SCD risk in young athletes is higher than in young non-athletes. The only comparative data supporting a higher risk of SCD in athletes comes from Padua (Italy). During the years 1979-1981 the risk of SCD in athletes was 4.19 per 100 000 person-years versus 0.77 in non-athletes.[18] This information however is strongly influenced by a small number of 14 SCD events in athletes. From other sources, it appears that the SCD risk is similar or may even be higher in non-athletes than in athletes.[19, 20] Whether the incidence rate of sports-related SCD differs between competitive and non-competitive athletes is not clear. In one study,[6] it was similar, in another[21] the incidence rate was higher in competitive than in non-competitive sports.

There is a large variability in the reported incidence rates of SCD, at least partly due to methodological issues. Data are mostly derived from observations of sudden deaths occurring during competitive sports activities. Comparisons between studies can be hampered because of differences in population characteristics (age, gender, and ethnicity), differing definitions of “sports”, and uncertain estimates of the population at risk (denominator). Some authors limit themselves to consider lethal cardiac arrests (SCD) only, while others also include survivors of such an event (SCA). Still others argue that necropsy studies are needed to ascertain the reason of death. The importance of ascertaining the exact cause of SCD has recently been stressed in a report from Israel where researchers found that sudden death in endurance runners is more often the result of heat stroke than from a primary cardiac event.[22, 23]

Estimates of the incidence rate of sports-related SCD in a recent worldwide systematic review varied from 0.11 to 2.66 per 100 000 person-years, i.e. from 1 per 37 593 to 1 per 917 000 person-years.[24] From the available literature, it seems acceptable to conclude that the overall average risk of SCD in apparently normal Europeans, aged 14 to 34 years, is probably not higher than 1 per 100 000 person-years.[10, 25] The rate in females is less than a tenth of that in men, i.e. less than 1 per million person-years.[21, 26] SCD occurs in a wide variety of sports, most commonly soccer, tennis, basketball, running and cycling.[6, 8, 26]

In most - if not all - studies in which the incidence rate of sports-related SCD are reported, the exact number of the individuals at risk (denominator) is not exactly known, rendering the incidence rates imperfect. Therefore, an estimation of the absolute number of young SCD cases per number of inhabitants in a given country might be used as an alternative to quantify the overall magnitude of the public health problem of SCD in the young. Data from the US and from neighbouring countries have recently been reported (Table 1). A nationwide survey in France over the years 2005 to 2010, in people aged 10-35, yielded an estimated absolute yearly number of 15 sports-related SCAs in competitive and 39 in non-competitive athletes.[21] Extrapolating those numbers to Belgium under the assumption of a comparable population composition and similar sports participation, results in an estimation of 9 exercise-related and 2-3 competition-related cases of SCA in young people per year. As mentioned earlier, it may be rather hypothetical to differentiate competitive from non-competitive exercise.

Table 1 – Reported yearly number of exercise-related SCAs/SCDs in young people in different countries

	Total population (mio)	Time window	Age range (years)	Any exercise-related SCA/D (average absolute # per year)	Competition-related SCA/D (average absolute # per year)	Any exercise-related SCA/D per year, per million population	Competition-related SCA/D per year, per million population	Extrapolated to Belgium*: estimated average absolute number per year	
								Any exercise-related	Competition-related
France	66,1	2005 - 2010	10-35	54	15	0,82	0,23	9,15	2,54
N. Holland	2,4	2006 - 2009	10-35	2,3	NA	0,96	NA	10,73	NA
Denmark	5,65	2007-2009	12-35	3	1	0,18	0,06	1,98	0,66
US	320	2001-2006	8-39	NA	66§	0,21§		2,31§	
*Veneto***	*4,4*	*1979-2004*	*12-35*	*2,12§*		*0,48§*		*5,40§*	

** population 11.2 million; extrapolation assumes a comparable population composition and similar proportional sports participation across countries; ** all athletes previously underwent mandatory pre-participation screening; § any SCD. References: France: Marijon et al.[21], North-Holland: Berdowski et al.[26], Denmark: Risgaard et al.[6], US: Maron et al.[1]; data from France and North-Holland include both SCDs and succesfully resuscitated SCAs; data from Denmark, US and Veneto all refer to fatal cases;*

In a recent study from The Netherlands, data of all out-of-hospital cardiac arrests occurring in the province of North-Holland over the years 2006-2009 were collected.[26] Over three years, 7 cases of exercise-related SCA were identified among subjects aged from 10 and 35 years. In Denmark, forensic examination of SCD victims is routinely performed. Risgaard et al. studied 881 SCD certificates for decedents in the period 2007 to 2009 and identified 9 cases of sports-related SCD in the age group 12 to 35 years, of whom 3 occurred during competition.[6] In the US, the average number of SCD per year in competitive athletes aged 8-39 years is 66 (range 50 to 76).[1] In the Italian Veneto region, after the introduction of a mandatory pre-participation screening, 55 cases of SCD occurred over a 26 year period (1979-2004) in the athletic population aged 12 to 35 years.[18] The corresponding estimated number of SCD cases per year in Belgium are shown in Table 1. From these data one may expect that in Belgium, each year no more than 10 exercise-related SCD occur in young people, only a few of those occurring during competitive sports.

The US[1] and Veneto[18] study, any SCD occurring in the age group of interest were counted. No further data are provided in the US study, but in Veneto 50 out of the 55 cases occurred during or immediately after a sports activity.

Under the assumption that one third of Belgians between 14 and 35 years are actively engaged in sports (i.e. 1/3 of 2 873 265 people aged 14-34 years in 2013), 1 million individuals would be at risk of sports-related SCD. Taking into account an incidence rate of 1 SCD per 100 000 per year as discussed earlier, would result in 10 cases per year in Belgium, a number that broadly corresponds to the upper limit of cases reported in neighbouring countries.

2.4 Cardiac disease underlying SCD in the young

In young people, SCD is most often due to one of several genetic heart disorders (Table 2). Some are associated with structural heart disease, such as hypertrophic cardiomyopathy and arrhythmogenic right ventricular cardiomyopathy. Others do not produce structural heart disease but in fact represent aberrations in the electrical activity of the heart. They are due to abnormalities of cardiac ion channels and are also called “channelopathies” or “inherited primary arrhythmia syndromes” such as the long QT syndrome, familial catecholaminergic polymorphic ventricular tachycardia and Brugada syndrome. Other congenital or acquired non-genetic heart disorders may also lead to SCD: Wolff-Parkinson-White syndrome, aortic stenosis, premature coronary atheromatosis. In addition, there are also non-hereditary congenital coronary artery anomalies that may induce a fatal arrhythmia in young people. Structural abnormalities of the aorta, such as in Marfan syndrome and other disorders associated with aortic dilatation, may lead to sudden death because of aortic dissection or rupture.[9]

Very often these conditions are not (or never) diagnosed in affected individuals. The one and only symptom they experience may be a fatal arrhythmia. Some affected individuals are diagnosed with the disease because of the occurrence of self-limiting arrhythmias, presenting as palpitations or syncope. Others are identified when a family member dies suddenly and the possibility of a genetic abnormality is considered, leading to screening of the victim’s relatives.[27]

Over a 27-year period (1980-2006), the US National Registry of Sudden Death in Athletes identified a total of 1866 athletes less than 40 years of age presenting with sudden death. Of those, 1049 (56%) were judged to be due to cardiovascular causes. Other causes included heat stroke, drugs, suicide, lightning, epilepsy. Hypertrophic cardiomyopathy was the most commonly observed underlying cardiovascular disease, occurring in 251 cases (36%). Congenital coronary artery anomalies were next in frequency (17%). Several other cardiovascular diseases each accounted for <7% of the total, with the most common of these being myocarditis (6%), arrhythmogenic right ventricular cardiomyopathy (4%), and channelopathies (4%), including 23 with long-QT syndrome and 2 with Brugada syndrome. Acquired (premature) ischemic heart disease accounted for 23 cases (3.3%) and Wolff-Parkinson-White syndrome (WPW) for 14 cases (2.0%).[1]

The overall prevalence of latent heart disease in apparently normal children and teenagers in this review is estimated to be 3/1000 (0.3%).[25, 28] Overall, those diseases have a low adverse event rate, estimated to be 0.2-0.8% per year.[29]

Below, we briefly describe the most prevalent cardiac diseases that may lead to SCD in young people.

2.4.1 Hypertrophic cardiomyopathy

2.4.1.1 Clinical picture

Hypertrophic cardiomyopathy (HCM) is characterised by a pathological increased thickness of the left ventricular wall. Most cases have a genetic origin but do not manifest until puberty.[30, 31] The proportion of phenotypically affected asymptomatic individuals who actually develop SCD is unknown. The diagnosis of HCM is made by echocardiography and requires a left ventricular wall thickness more than two standard deviations greater than the predicted mean.[30]

A recent systematic review, that included both screening and general population studies, calculated for HCM a summary phenotypic prevalence of 45 (95%CI: 10-79) per 100 000 asymptomatic individuals.[32]

The clinical course of HCM is relatively benign. Most people with HCM are asymptomatic and have a normal lifespan. Some develop symptoms, such as angina, dyspnoea, palpitations and syncope. In rare instances it provokes SCD.[30, 31] The risk for a person with HCM to develop sudden death is dependent on a number of disease characteristics such as left ventricular wall thickness, the presence of an intraventricular pressure gradient, symptoms, or a family history of SCD.[30] In asymptomatic individuals, HCM very rarely leads to death (around 6 per 10 000 per year).[33] The incidence rate of SCD due to HCM in England was calculated to lie between of 0.5-1 per million per year (Table 2).[25, 33]

Table 2 – Estimated prevalence of cardiac disease that may induce SCD in the young and corresponding estimated SCD incidence rate

	Prevalence per million	SCDs per million per year
Genetic disorders		
• Structural cardiovascular disease:		
o Hypertrophic cardiomyopathy	450 (100 à 790)	≤1
o Arrhythmogenic right ventricular cardiomyopathy	200 à 500	<1
o Marfan syndrome	200	<1
o …		
• Primary electric disorders:		
o Long QT syndrome	70 (0 à 140)	<1
o Brugada syndrome		<<1
o Short QT syndrome		<<1
o Catecholaminergic polymorphic ventricular tachycardia		
o …		
Non-genetic congenital or acquired disorders		
• Wolff-Parkinson-White syndrome	1360 (550 à 2180)	<<1
• Congenital anomalies of the coronary arteries	1000	0.5
• Congenital heart disease		<<1
• Aortic stenosis		<<1
• Myocarditis		<1
• Dilated cardiomyopathy		<<1
• Premature coronary atheromatosis		<<1
• Mitral valve prolapse		<<1
• …		
TOTAL	3000	<10

See text for references. "Estimated SCDs per million per year" refers to the yearly number of events in the overall population of young athletes. More than 40 distinct cardiovascular conditions have been identified as a cause of SCD in athletes. For some of those extremely rare diseases, generally accepted data on prevalence and/or SCD risk are lacking.

A recent systematic review analysed the diagnostic performance of rest-ECG and echocardiography in the 3 most common ECG-detectable disorders associated with SCD (HCM, LQTS, WPW) in asymptomatic young people.[32] Most of the data were extracted from studies in relatives of HCM patients. Summary estimates are shown in Table 3. The diagnostic performance of the rest-ECG in HCM (and LQTS) is poor: to detect one case of HCM, more than 2600 individuals need to be screened. For each case, 399 others will be needlessly alarmed.

Table 3 – Diagnostic performance of the rest-ECG in selected conditions

	Prevalence per 10^5 screenees	Sens.	Spec.	PPV	NPV	NNS to detect 1 case	Number of false positives per detected case	Number of false negatives per 100,000 screened
HCM	45 (95%CI 10-79)	0.847	0.848	0.0025 (1/400)	0.9999	2624	399	7
LQTS	7 (95%CI 0-14)	0.861	0.860	0.0004 (1/2324)	0.9999	16592	2323	1
WPW	136 (95%CI 55-218)	1.000	1.000	1.000	1.0000	735	0	0

Adapted from Rodday et al.[32] Sens: Sensitivity; Spec: specificity; HCM: hypertrophic cardiomyopathy; LQTS: long QT syndrome; WPW: Wolff-Parkinson-White. Perfect scores for diagnosing WPW are due to the fact that the Rodday et al. took the rest-ECG as the gold standard for diagnosing WPW.

Patients with HCM are discouraged from participation in competitive sports or engaging in intense physical activity, especially when they have risk factors for SCD.[30] There are no data to support the use of invasive procedures to reduce LV outflow obstruction in asymptomatic patients, regardless of its severity.[30, 31] There is no consensus on the indication for implantation of an implantable defibrillator (ICD), even not in symptomatic patients.[31]

2.4.1.2 Hypertrophic cardiomyopathy in athletes

HCM represents the most common cause of SCD in athletes. It has an estimated prevalence that is 10- to 50-fold greater than other familial diseases affecting the heart and great vessels, such as long QT, Brugada, and Marfan syndromes.[34] In the US National Registry of Sudden Death in Athletes it represents the final diagnosis in 36% of SCD.[1] In this registry, 44 distinct cardiovascular conditions were diagnosed causing SCD.

A recent literature review identified 17 different screening programmes involving nearly 90 000 healthy trained athletes in whom 6 definitive and 38 possible diagnoses of HCM were made (0.05%), a number that is lower than what one would anticipate from published prevalence figures.[34]

As explained in the methodology section of this report, we identified 3 recent (i.e. published during the first six months of 2014) studies on pre-participation screening in the young. One US study involved 2017 high school athletes and detected 1 case (0.05%) of HCM.[35] In a cohort of 22,205 competitive Greek athletes, 7 cases (0.03%) of HCM were identified.[36] Another US study involving 1339 students detected no individuals with HCM.[37]

If the prevalence of HCM in athletes is 0.05% and applying the mortality observed in the asymptomatic general population (6 SCD per 10 000 affected individuals per year), the yearly incidence of SCD due to HCM is 1 per 3 million athletes.

Although the final diagnosis of HCM requires echocardiography, suspicion can be raised by the presence of non-specific abnormalities on the rest-ECG.[30] In trained athletes, the diagnosis may be hampered since hypertrophy (thickening) of the left ventricular wall can result from a normal physiologic adaptation that may induce changes in both the rest-ECG and echocardiogram.

Although it is generally accepted that patients with HCM who are considered at high risk for SCD should be discouraged from engaging in intense physical activity, only 16% of SCDs in HCM occur during moderate-severe exertion, indicating that disqualification from sports may not much affect the occurrence of SCD.[25]

2.4.2 Congenital anomalies of the coronary arteries

2.4.2.1 Clinical picture

Congenital anomalies of the coronary arteries are characterised by an anomalous origin of the right or left coronary artery from the aorta. Since most individuals are asymptomatic and have a normal rest-ECG, diagnosis is mostly made after SCD. Therefore, the prevalence is difficult to ascertain and is probably less than 0.1%.[25, 38]

In a recent review, the risk of sudden death from these conditions in apparently normal children and teenagers is estimated to be 5 per 10 million person-years (Table 2).[25]

A detection of coronary anomalies sometimes occurs accidentally through echocardiography. These individuals are excluded from all participation in competitive sports.[39] Treatment options include coronary stenting and surgery.[40]

2.4.2.2 Congenital anomalies of the coronary arteries in athletes

Congenital anomalies of the coronary arteries were the cause of death in 119 (17.2%) cases in the US National Registry of Sudden Death in Athletes, representing the second most frequent disease responsible for athletic field deaths.[1, 38, 39]

No cases were identified in any of the 3 recently published studies mentioned above.[35-37]

Since affected individuals are asymptomatic, have no particular clinical signs, and there are no rest-ECG signs indicating congenital anomalies of the coronary arteries, these conditions may only be detected during a screening examination if it includes an echocardiogram.

2.4.3 Wolff-Parkinson-White syndrome

2.4.3.1 Clinical picture

The Wolff-Parkinson-White syndrome (WPW) is a cardiac disease characterised by the presence of an extra connection ("accessory pathway") between the atria and the ventricles of the heart. This may lead to an early electrical stimulation of the ventricles ("pre-excitation"), characterised by the presence of a delta-wave on the rest-ECG. In some cases, these ECG abnormalities appear only intermittently, which is prognostically favourable. Affected individuals often remain asymptomatic while others develop palpitations due to arrhythmias. Very rarely, sudden death occurs in a WPW patient.

The prevalence of WPW in the general population is estimated to lie between 0.06 and 0.22%.[25, 32, 41] The incidence of SCD in patients with WPW has been estimated to range from 0.15 to 0.39% over 3 to 10-year follow-up, but cardiac arrest very rarely is the first manifestation in asymptomatic individuals.[41]

In patients with permanent pre-excitation, it is considered reasonable to do an invasive risk stratification by means of an electrophysiologic study. In individuals at risk for SCD, a catheter ablation of the extra connection can be executed.[42] However, since even symptomatic patients considered at risk for SCD have a very low mortality rate, some authors argue that it remains a reasonable option to start no therapy at all.[43] Death as a consequence of accessory pathway ablation has been reported to occur in 0.07% up to 0.19% of cases.[44] Hence, the decision to ablate has to be based on the preference of a well-informed patient who balances a very small immediate ablation risk with a very small longer-term risk without ablation.[43]

2.4.3.2 WPW in athletes

Although the WPW syndrome is the most prevalent disease that may lead to SCD in the young, it is a very rare cause of SCD in athletes (Table 2). WPW accounted for 14 cases (2.0%) of SCD cases in the US National Registry of Sudden Death in Athletes.[1] It is however not known whether these individuals were symptomatic.

In a literature review involving nearly 90 000 healthy trained athletes, WPW was diagnosed in 29 (0.03%) of them.[34]

In recently published studies on pre-participation screening, WPW is the most prevalent diagnosis. In one US study involving 2017 high school athletes, 4 cases of WPW (0.2%) were identified.[35] Another US study involving 1339 students detected 5 (0.4%) cases.[37] In a Greek cohort of athletes (n=22 205) WPW was detected in 8 (0.04%) individuals.[36]

Since SCD is almost never the presenting symptom of a WPW syndrome, and the optimum management of these individuals is not known, its detection in an asymptomatic individual seems futile. Although successful catheter ablation is capable of eliminating the risk of SCD, uniform referral of every individual with WPW for an ablation could also result in serious and potentially life-threatening complications, possibly greater in number than the deaths averted from untreated disease.[42]

2.4.4 Channelopathies

2.4.4.1 Clinical picture

Channelopathies or "inherited primary arrhythmia syndromes" represent a series of rare genetic diseases, characterised by their predisposition to induce SCA but without clear structural abnormalities of the heart. They include long QT syndrome, Brugada syndrome, cathecholaminergic polymorphic ventricular tachycardia, and short QT-syndrome.

Among the channelopathies, the long QT syndrome (LQTS) is the most prevalent. Its reported prevalence varies across authors and ranges from 7[32] to 50[45] per 100 000.

Individuals affected with LQTS may suffer from recurrent syncope and seizures that are induced by a cardiac arrhythmia. Most often, this arrhythmia is self-terminating presenting as a syncopal episode, but it may also lead to SCA.[46] Among 3015 LQTS children that were followed from ages 1 to 12 in the US International LQTS Registry, mortality was low, with only 53 events (not all fatal), yielding an annual serious event rate of 0.15%.[46, 47]

Diagnosis is based on the measurement of the QTc-interval on the rest-ECG. From a 2012 systematic review, it appears that the number needed to screen to detect one case of LQTS is almost 17 000 (Table 3).[32] Recent data have shown that up to 25% of patients with LQTS confirmed by the presence of the LQTS gene mutation have a normal range of the QTc-interval suggesting that the number of false-negative ECGs may be higher than presumed.[45]

The management of LQTS which is essentially based on experts' opinions consists of lifestyle modifications, beta-blockers and implantable defibrillator (ICD) in high risk individuals, e.g. those that survived a SCA. On one hand, avoidance of strenuous exercise is routinely advised in all LQTS patients but on the other hand, participation in competitive sports seems to be still a matter of debate among experts.[45] In the International LQTS Registry, treatment with a beta-blocker reduced the risk of SCD by approximately half. Invasive treatment during childhood was applied in 3.5% of individuals: 1% received an ICD, 1% underwent left cardiac sympathetic denervation and <2% received a pacemaker.[46, 47]

Brugada syndrome is a very rare hereditary channelopathy. Individuals present with syncope or SCD. Brugada Syndrome is not typically associated with exercise-related SCD. The only established treatment is ICD insertion.[48]

2.4.4.2 Channelopathies in athletes

Channelopathies accounted for 25 cases (3.6%) of SCD cases in the US National Registry of Sudden Death in Athletes, They included 23 (3.3%) cases of LQTS and 2 (0.3%) with Brugada syndrome.[1]

In a literature review involving nearly 90 000 healthy trained athletes, LQTS was diagnosed in 11 (0.01%) of them.[34] In recently published studies on pre-participation screening,[35-37] LQTS was diagnosed in one individual out of 22 205 Greek athletes.[36]

Although deaths from Brugada Syndrome characteristically occur at rest, intensive exercise is generally not advised because it may be associated with profound bradycardia and core temperatures exceeding 40°C, both of which may precipitate fatal arrhythmias in affected individuals.[48]

2.4.5 Arrhythmogenic right ventricular cardiomyopathy (ARVC)

2.4.5.1 Clinical picture

Arrhythmogenic right ventricular cardiomyopathy (ARVC) is an inherited disorder characterised by fibro-fatty inflammation affecting both ventricles of the heart. Its true prevalence is unknown, with estimates lying between 1 in 2000 and 1 in 5000.[49]

ARVC cannot be diagnosed by a single test and relies on the demonstration of structural, functional and electrophysiological abnormalities.[49, 50]

Affected individuals are strongly recommended to avoid strenuous exercise. Further treatment consists of beta-blockers, anti-arrhythmics and implantation of an implantable defibrillator (ICD) in selected cases.[49] In a recent review, the risk of sudden death from ARVC in apparently normal children and teenagers is estimated to be 8 per 10 million person-years (Table 2).[25] In a French study on 200 cases of ARVC aged 1-65 years, only 7 cases (3.5%) occurred during sports activity.[51]

2.4.5.2 ARVC in athletes

ARVC accounted for 30 cases (4.3%) of SCD cases in the US National Registry of Sudden Death in Athletes.[1] Remarkably, ARVC is a most prevalent cause of SCD in young athletes in Italy where it is reported to account for 24% SCD cases. In contrast, HCM is a rare (2%) cause of sudden death in athletes.[52]

2.4.6 Myocarditis

Myocarditis, typically caused by viral infections, accounts for up to 7% of SCD in athletes.[1, 48] Acute illness is associated with ventricular arrhythmia. In some cases, myocarditis can lead to a dilated cardiomyopathy and increased risk of SCD.

Athletes diagnosed with myocarditis are recommended to temporary (6 months) refrain from sports activity to reduce the risk of SCD.[48]

Key points

- **In young people, exercise-related sudden cardiac death most often occurs in the context of hereditary or congenital heart disease. It is an extremely rare event, but exact numbers are not known. Extrapolating data from neighbouring countries allows to estimate for Belgium a yearly number of up to 10 cases of which two or three would occur during competitive sports.**
- **Hypertrophic cardiomyopathy (HCM) represents the most common cause of SCD in athletes. The yearly incidence of exercise-related SCD due to HCM is estimated to be less than 1 per million athletes.**
- **Wolff-Parkinson-White syndrome is the most prevalent disease detected at pre-participation screening, but SCD as a presenting symptom in asymptomatic individuals is extremely rare.**
- **There is no hard data on the best management of asymptomatic individuals (i.e. those typically detected at pre-participation screening) affected by one of those diseases. Imperfect data suggest that asymptomatic individuals will almost never (suddenly) die from it.**

3 CLINICAL EFFECTIVENESS OF CARDIOVASCULAR PRE-PARTICIPATION SCREENING

3.1 Methods

In the scoping review of this project we found that the UK National Screening Committee was preparing a report on the subject (expected publication April 2015).[10] Upon our request, we received a pre-final draft in August 2014 including a systematic review of the literature. Shortly thereafter this draft report became available on-line for public consultation. It became clear, both from this document and the 2013 report from the Belgian Superior Health Council (Hoge Gezondheidsraad - Conseil Supérieur de la Santé),[7] that there was no solid evidence in support of pre-participation cardiovascular screening. Not a single randomised trial has been published on the subject. All available data are limited by their observational and anecdotic nature. In September 2014, this was confirmed in an updated scientific statement from the American Heart Association and the American College of Cardiology “on the detection of cardiovascular disease in healthy general populations of young people”.[9]

Based on the fact that recent reviews were available and for the sake of efficiency, we decided not to repeat a systematic literature review on the clinical effectiveness of pre-participation screening. Instead, we performed an update of the literature search that was used by the UK National Screening Committee, using the same search strategy but with date limits of November 2013 until the 11th of August 2014. This search revealed 496 references. During a first selection based on title and abstract, it appeared that the majority of the references did not correspond to our research questions. We retrieved 21 references for full text assessment. Selection of relevant papers was executed by 2 researchers (ADS, HVB) and consensus was reached in case of disagreement. Reasons for exclusion were: already mentioned in the UK report, intervention or outcome not in accordance with our research questions, specific population (children with attention deficit disorder only), design (strictly narrative overview). The flow of the references through the search process is presented in the appendix to this report. At last we retained 3 references. Each reported the results of a previously unpublished pre-participation screening study.[35-37]

Accordingly, the scientific data reported in the present report are predominantly extracted from recently published scientific statements, originating from the UK, the US and the Belgian Superior Health Council,[7, 9, 10] updated with data from more recent primary studies.[35-37]

We also performed a search for existing guidelines and systematic reviews on the subject. This search was carried out in several databases (the National Guideline Clearinghouse, NICE, SIGN, G-I-N and EBMPracticeNet), websites of scientific organisations and Google. Guidelines published in Dutch, English, French or German were selected. After exclusion of duplicates, 20 documents were retained. The quality appraisal of the guidelines was assessed by using the AGREE instrument (http://www.agreetrust.org/resource-centre/the-original-agree-instrument/checklist72). Systematic reviews were assessed using the AMSTAR instrument (http://amstar.ca/Amstar_Checklist.php). Details are provided in the appendix to this document.

As in every KCE report, the methodology and the results were extensively discussed with experts and stakeholders: cardiologists, epidemiologists, economists, an ethicist, sports physicians, general practitioners, representatives from both Communities, insurance companies, mutualities, NIHDI, consumer representatives, and lawyers (for details see colophon). Final decisions were made by KCE experts who take the full responsibility of the contents of the report.

3.2 Diagnostic performance of history-taking and physical examination

Although international scientific associations of cardiologists often recommend personal and family medical history-taking and physical examination in the context of pre-participation screening, hard data supporting the efficacy of such strategy are lacking.[4, 53]

The UK National Screening Committee, whose report we used as the starting point for the present analysis, performed an extensive literature search, but could not identify reliable sensitivity or specificity figures on any type of pre-participation screening testing.[10]

In discussing the diagnostic performance of history-taking and physical examination, the Belgian Superior Health Council distinguishes between diagnosis in the general 14 to 34 years population and diagnosis in those engaged in competitive sports participation. It quotes a sensitivity of history-taking and physical examination of 0.03 in the general young population and no data on specificity.[7, 25] In young people engaging in competitive sports participation, it refers to a US study reporting a sensitivity of 0.44 and a specificity of 0.76 for screening with history and physical examination alone.[54]

Here, we report data from studies published in 2014 that we identified through our literature search as explained earlier. One US study involved a cohort of 2017 high school athletes that underwent screening with a standardised history, physical examination, rest-ECG and an echocardiogram.[35] Five cardiac disorders associated with SCD were identified: 1 HCM and 4 WPW. 14.7% of the participants had an abnormal history or physical examination. History and physical examination detected 2 out of the 5 disorders. Sensitivity and specificity of combined history and physical examination for detecting potentially lethal disease was 0.40 (95%CI 0.12-0.77) and 0.85 (95%CI 0.84-0.87) respectively. Limitations of this study were the narrow age limit of participants (14-18 years), the low proportion of Caucasian participants (34%) and the relatively small sample size in relation to the low prevalence of cardiac diseases that may lead to SCD.

In a large cohort of competitive Greek athletes (n=22 205) studied over an 18-year period (1992-2010), history-taking revealed palpitations at rest in 7% of the athletes and thoracic pain in 4%. On physical examination, 14% had a heart murmur and 4.3% an elevated blood pressure.[36] The study did not provide sufficient data to make sensitivity/specificity calculations.

Another US study involving 1339 students assessed a questionnaire, a physician interview, a physical examination and a rest-ECG.[37] The questionnaire included medical and family history and the heart health questions of the Pre-participation Physical Evaluation Monograph-4 (PPE4) (Figure 3). This questionnaire represents the current standard for conducting cardiovascular screening in the US.[9]

Figure 3 – Heart health questions in the Pre-participation Physical Evaluation Monograph

HEART HEALTH QUESTIONS ABOUT YOU	Yes	No
5. Have you ever passed out or nearly passed out DURING or AFTER exercise?		
6. Have you ever had discomfort, pain, tightness, or pressure in your chest during exercise?		
7. Does your heart ever race or skip beats (irregular beats) during exercise?		
8. Has a doctor ever told you that you have any heart problems? If so, check all that apply: ☐ High blood pressure ☐ A heart murmur ☐ High cholesterol ☐ A heart infection ☐ Kawasaki disease Other: _____		
9. Has a doctor ever ordered a test for your heart? (For example, ECG/EKG, echocardiogram)		
10. Do you get lightheaded or feel more short of breath than expected during exercise?		
11. Have you ever had an unexplained seizure?		
12. Do you get more tired or short of breath more quickly than your friends during exercise?		
HEART HEALTH QUESTIONS ABOUT YOUR FAMILY	**Yes**	**No**
13. Has any family member or relative died of heart problems or had an unexpected or unexplained sudden death before age 50 (including drowning, unexplained car accident, or sudden infant death syndrome)?		
14. Does anyone in your family have hypertrophic cardiomyopathy, Marfan syndrome, arrhythmogenic right ventricular cardiomyopathy, long QT syndrome, short QT syndrome, Brugada syndrome, or catecholaminergic polymorphic ventricular tachycardia?		
15. Does anyone in your family have a heart problem, pacemaker, or implanted defibrillator?		
16. Has anyone in your family had unexplained fainting, unexplained seizures, or near drowning?		

Source: The Pre-participation Physical Evaluation Monograph-4 (PPE4). http://www.aap.org/en-us/about-the-aap/Committees-Councils-Sections/Council-on-sports-medicine-and-fitness/Pages/PPE.aspx

Nine-hundred and sixteen (68%) of the participants reported at least one positive response on a questionnaire. After review by a physician, 421/1339 (31.4%) had at least one relevant response on the questionnaire that required additional cardiac evaluation. Approximately 15% of participants reported a family history of heart problems, unexpected sudden death or a genetic cardiac condition that could not be judged as benign after physician interview. 124 (9.3%) participants had an abnormal physical examination of which 114 (8.5%) had a cardiac murmur and 22 (1.6%) an elevated blood pressure. This study did not provide sufficient data to make sensitivity/specificity calculations. It makes however obvious that the introduction of a standardised questionnaire may induce a massive number of positive screening results. Given the very low prevalence of cardiac disease that may induce SCD, the large majority of these positives are false-positives.

Key points

The diagnostic performance of medical history-taking and physical examination as screening tools for detecting potentially lethal cardiovascular disease in individuals aged 14-34 years is characterised by a low sensitivity (0.03 to 0.44) and specificity (0.69 to 0.85).

3.3 Diagnostic performance of the rest-ECG in pre-participation screening

In order to overcome the very low diagnostic performance of history-taking and physical examination, some experts propose to include a rest-ECG in the pre-participation screening protocol.[55, 56]

The assessment of the diagnostic performance of the rest-ECG in the population of interest in the present report is hampered by a number of particularities of the population involved:

1. Not all diseases that may cause SCD in young athletes can be identified with an ECG since some of them do not directly interfere with the electrical activity of the heart (e.g. congenital coronary anomalies) whilst others can be acquired after the screening examination (e.g. myocarditis, premature coronary heart disease). The exact proportion of those diseases that can be detected by ECG remains unknown and

in case series, numbers varying from 50% to 100% have been observed.[1, 7, 25, 35, 56]

2. The configuration of the ECG in a given individual matures until adulthood, and normal tracings in the young may mimic pathologic tracings in the adult.
3. Intensive training induces physiologic adaptations of the heart that are reflected in changes on the ECG that may mimic pathological conditions. Cardiac alterations can be discerned in those performing ≥3 hours of sports per week but it is unknown in what proportion of the non-professional young athletes and recreational sporty individuals such physiologic adaptations are present.[57]
4. It cannot be taken for granted that health care professionals that are familiar with the interpretation of the ECG in medical practice (cardiologists, paediatricians and selected general physicians) are also skilled to differentiate pathologic from physiologic tracings in healthy young sporty individuals.[29] In Italy, athletes are screened in a national program by physicians who attended a full-time 4 years postgraduate training program.[58]
5. In individuals that are genetically affected with a cardiac disease that may induce SCD, it may take years and even decades before the disease to become phenotypically expressed.[7, 45]

High-quality data on the diagnostic performance of the rest-ECG in the context of pre-participation screening are lacking. The observed proportion of abnormal ECGs varies widely over pre-participation screening studies, depending on methodological issues such as the population characteristics, the ECG reference standard, and the investigators' skills and familiarity with sports cardiology.[59, 60] A given clinician may also incorporate a safety threshold to avoid false-negative ECG readings.

Since up to 50% of the cardiovascular diseases that may induce SCD cannot be detected by rest-ECG,[1, 7, 25, 35, 56] the sensitivity of the ECG can be expected to be situated between 0.50 and 1.00. Unless specified otherwise, in the present text we therefore accept the sensitivity of the ECG to be 0.75.

There is also uncertainty about the specificity of the screening-ECG. This is illustrated by repetitive adaptations of standard ECG criteria, aimed at limiting the number of false-positive results. In 2010, the ESC issued *modern* recommendations for the interpretation of the rest-ECG in athletes.[61] Two years later, an update of the ECG criteria was jointly agreed by US and EU experts ("Seattle criteria").[62] Shortly thereafter, a new revision was called upon.[63] These adaptations largely result from non-peer-reviewed panel proposals.[9] It remains to be established whether and to what extent the increased specificity of new ECG criteria decreases its sensitivity.[63, 64]

Recent studies in which rest-ECG recordings of non-professional athletes were analysed by expert cardiologists using so-called *modern* criteria reported a specificity of the ECG of 0.92,[36] 0.95,[37] and 0.97[35] respectively. But even if a specificity of 0.95 could be obtained by expert physicians, the absolute number of false-positive ECGs would remain enormous. Screening 100 000 individuals with an ECG, taking into account a disease prevalence of 0.3%, and considering a sensitivity of 0.75 and a specificity of 0.95 would lead to 5210 abnormal ECGs of which 4985 (95.7%) would be false-positives.

Although a rest-ECG is an attractive and cheap diagnostic tool, its usefulness in pre-participation screening is limited by its poor diagnostic performance in detecting the extremely rare diseases that are looked for in pre-participation screening. Furthermore, even effective identification of diseases that may induce SCD is not equivalent to identifying individuals who will actually experience SCD.

Key points

- **The diagnostic performance of a rest-ECG as a pre-participation screening tool for detecting potentially lethal cardiovascular disease has not been thoroughly studied in large populations.**
- **Imperfect data suggest a sensitivity of the rest-ECG for detecting cardiac disease that may induce SCD of 0.75 and a specificity of 0.95 provided the test is performed by expert sports electrocardiographists.**

3.4 Diagnostic performance of history, physical examination and ECG combined

The Belgian Superior Health Council adopts ECG performance data from a 2011 US study involving 964 collegiate athletes.[7, 54] Screening with a combination of history-taking, physical examination and ECG was associated with a sensitivity of 0.89 and a specificity of 0.70. The positive predictive value of the combined protocol was 2.7% and negative predictive value is 99.9%. The latter number may seem spectacular, but it is not very informative since even without any testing we know that the large majority of individuals is not affected by one of those (very rare) diseases. The reported sensitivity of 0.89 is rather unexpected if one considers the previously reported very low sensitivity of history-taking and physical examination and previous reports suggesting that up to 50% of cardiac disease that may induce SCD in young people cannot be detected by means of a rest-ECG.

In another study on 510 collegiate athletes, cardiac abnormalities with relevance to sports participation risk were observed in 11 participants (prevalence of 2.2%). Screening with history and examination alone detected abnormalities in 5 of these 11 athletes (sensitivity of 0.46; specificity of 0.94). ECG detected 5 additional participants with cardiac abnormalities, thereby improving the overall sensitivity of screening to 0.91. The addition of an ECG to history and physical examination increased sensitivity from 0.46 to 0.91 and decreased specificity from 0.94 to 0.83.[65]

The data reported in the recently published papers that we discussed in earlier paragraphs do not allow to calculate the sensitivity and specificity of the combined (history, physical examination, ECG) screening protocol.[35-37]

All studies published so far have serious limitations and especially suffer from a small sample size. They aim for detecting diseases with a prevalence often less than 1/5000 in series including only a few thousand individuals. A further limitation is that they most often only use an echocardiogram to exclude structural heart disease and a number of diseases may be left undetected, thus overestimating the sensitivity of pre-participation screening in those studies.[66]

Recent studies stress the impact of improved ECG criteria on the performance of pre-participation screening. However, a lower number of false-positive ECGs does not necessarily annihilate the large number of false-positives induced by history and physical examination. Indeed, it is not clear if a normal ECG will be considered a sufficient reason to exempt for further testing an individual reporting at history-taking exercise-related palpitations, chest pain, dizziness or syncope, or in whom a heart murmur or a high blood pressure is found at physical examination. In the studies mentioned at the start of this paragraph, the addition of an ECG on top of history-taking and physical examination improved sensitivity but decreased the specificity.[54, 65]

Based on the abovementioned reasoning, we feel that for the combined screening protocol a sensitivity of 0.75 (we leave the “accepted” sensitivity of ECG screening unchanged) and a specificity of 0.70 might be a sensible guess. Under this assumption, the positive predictive value (PPV) would be 0.8% and the negative predictive value (NPV) 99.9%. Accordingly, screening 100,000 individuals with a disease prevalence of 0.3% would lead to 30,135 “suspect” screenees of which 29,910 (99.2%) would be falsely suspected.

If the specificity of the combined protocol were – more optimistically - 0.95, the PPV would be 4.3% and the NPV 99.9%. Following screening, 5210 screenees would be “suspect”, 4985 (95.7%) being falsely so.

Key points

- **The diagnostic performance of a screening protocol that combines history-taking, physical examination and rest-ECG for diagnosing cardiac disease that may induce SCD is unknown.**
- **It is not known to what extent the rest-ECG can offset the potentially very high number of false positives induced by history-taking and physical examination.**
- **In the presence of a very rare event, mass screening would result in a huge number of false positive screenees, even if the specificity of the combined screening protocol were 0.95.**

3.5 International practice guidelines on pre-participation screening

A multitude of guidelines, consensus reports and expert opinions claiming how pre-participation screening should be implemented have been published. From those documents it appears that there is no consensus regarding the most effective method for cardiovascular pre-participation screening (Appendix 1). Only a minority of the documents stands up to scientific scrutiny. In most cases, the guideline development group is composed of mono-disciplinary sports physicians, offering a narrative review of the scientific literature in which a critical appraisal of the quality of the evidence is often lacking.

Guidelines produced by the US National Heart, Lung and Blood Institute (NHLBI) and the US Guidelines Advisory Committee rigorously adhere to the highest methodological standards for guideline development. The European and US scientific associations of cardiologists have also adopted strict methodological approaches for their guideline production and dissemination. Here, we briefly summarise the more recent recommendations from those agencies.

In 2006, the European and US scientific associations of cardiologists (ACC/AHA/ESC) jointly issued guidelines for management of patients with ventricular arrhythmias and the prevention of SCD.[4] It includes a chapter on SCD prevention in athletes. In asymptomatic athletes, pre-participation screening with history-taking, including family history and physical examination is recommended. It further stipulates that rest-ECG and possibly echocardiography "may be considered" (class IIb) in screening for heart disorders in athletes. Of note, one year before the publication of this joint European and US guideline, a report was published by the working groups of sport cardiology and cardiac rehabilitation of the European Society of Cardiology (ESC).[56] It recommends "the implementation of a common European screening protocol essentially based on 12-lead ECG". Although this document is only a "proposal for a common European protocol", written by like-minded European experts, most publications on pre-participation screening refer to this document as the official position of the ESC, in this way ignoring the restricted class IIb recommendation from the 2006 joint European and US guideline. In 2006, an additional document known as the "Lausanne recommendations" was published on behalf of the ESC and the International Olympic Committee.[55] It recommends for every athlete a screening protocol that includes history-taking, physical examination, and an ECG for which it provides a detailed questionnaire (http://www.olympic.org/documents/reports/en/en_report_886.PDF).

In 2007, the AHA concluded that personal and family history and physical examination represent the most practical screening strategy in athletes, with echocardiography and/or ECG remaining optional.[53]

In 2008, the US Guidelines Advisory Committee published Physical Activity Guidelines.[16] It found that the protective value of a medical consultation for persons with or without chronic diseases who are interested in increasing their physical activity level is not established.

In 2011, the US National Heart, Lung and Blood Institute (NHLBI) did not support mass screening of young athletes with an ECG.[9, 67]

In 2014, the AHA concluded that there is insufficient information available to support the view that universal screening ECGs in asymptomatic young people for cardiovascular disease is appropriate in competitive athletes or in the general youthful population.[9] In a somewhat contradictory vein, it adds that "Screening, including ECG, may be considered in relatively small cohorts of young healthy people, not necessarily limited to athletes".

Key points

- **There is an ongoing polarised debate on the appropriateness of pre-participation screening.**
- **Most guidelines, consensus reports or expert opinions recommend to include history-taking and physical examination in the screening protocol, although data supporting its effectiveness are limited.**
- **Opinions on the inclusion of a rest-ECG vary from a strong-yes to a strong-no.**

3.6 International experience with pre-participation screening

Protagonists of pre-participation screening consistently refer to observations from Italy where the incidence of SCD in young athletes substantially declined after screening became mandatory in 1971. With an annual pre-participation screening effort involving several millions of athletes each year, the Italian experience is gigantic.[58] Hence, it doesn't come as a surprise that the worldwide debate and scientific literature on pre-participation screening is dominated by Italian investigators. Therefore, we discuss the Italian experience in detail.

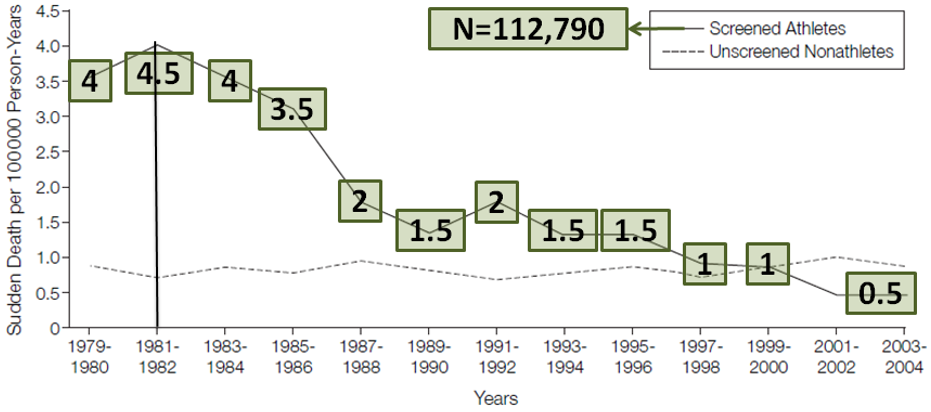
3.6.1 The Italian experience

3.6.1.1 The Veneto study

Since 1982, all Italian competitive athletes underwent an annual pre-participation screening examination, at least including a family and personal history, a physical examination, and a rest-ECG. Italian sport physicians attend postgraduate training programs full-time for 4 years.[58] The Veneto study describes the experience from the Italian region of Veneto (population: 4 379 900) and provides the trends in SCD in athletes aged 12 to 35 years (n~190 000).[18] Between 1979 and 2004, 55 cases (50 males and 5 females) of SCD occurred with an overall incidence rate of 1.9 deaths per 100,000 person-years. In 50 cases (91%), SCD occurred during sports activity (44 cases) or immediately afterward (6 cases). All of the 55 athletes who died suddenly previously had obtained eligibility for competitive sports.

Absolute numbers and incidence rates of SCD during the 26-year period are shown in Figure 4. During the study period, the annual incidence of SCD in screened athletes decreased by 89%. The average incidence of SCD in the pre-screening period was 4.19 (95%CI 1.78-7.59) per 100 000 person-years. The average incidence decreased to 2.35 (95%CI 1.94-2.75) per 100 000 person-years in the early screening period and to 0.87 (95%CI 0.46-1.28) per 100 000 person-years in the late screening period, with the lowest observed rate of 0.43 per 100 000 person-years occurring between 2001 and 2004.

Figure 4 – Trends of SCD in screened athletes and unscreened non-athletes (12 to 35 yrs.) in Veneto



Adapted from Corrado et al.[18] Numbers in squares represent the average absolute yearly number of SCDs per given time period.

The study also reports the trends in reasons for disqualification from competitive sports in a subgroup of 42 386 athletes that underwent pre-participation screening in Padua. Of 42 386 athletes 3914 (9%) were referred for further examination because of positive findings and 879 (2%) were ultimately disqualified from participation in competitive sports.

Since subjects that screened negative in the first screening round (91% of the initial population) did not undergo additional testing such as echocardiography, the proportion of false-negatives in the general population remains unknown. The 2006 paper[18] does not provide data on screenees that were disqualified. The number of false-positives among them is unknown. A previous paper on the same population study published in 1998 mentions that 4 of 621 athletes (out of 33 735) who were disqualified for cardiovascular causes died during a mean follow-up period of 8.2±5 years, one of them because of complex ventricular arrhythmias, the others of non-natural causes.[68]

3.6.1.2 *Critical appraisal of the Veneto study*

In the 2006 paper, the authors conclude: "*The incidence of sudden cardiovascular death in young competitive athletes has substantially declined in the Veneto region of Italy since the introduction of a nationwide systematic screening*."[18] It is clear that by using this wording, they avoid to fall pray of the "post hoc ergo propter hoc" logical fallacy. A causal relationship between screening and a declining SCD rate cannot be assumed here because of a number of methodological weaknesses of the study, more particular its mere observational nature and the lack of an unscreened control athlete population. In fact, the authors use historic data retrieved during a 2 years pre-screening period (1979-1980) as a reference for assessing the impact of screening over the ensuing 22 years (1982–2004). They observed an impressive decline in the incidence rate of SCD from 3.6 per 100,000 person-years in 1979-1980 to 0.4 per 100,000 person-years in the final years of the study (2001-2004).

The yearly event rate was very low as indicated in Figure 4, ranging from 4 cases per year at the start of the study to 1 case per year during the final years. Such low absolute numbers are very sensitive to minor variations, and may be affected by e.g. the adjudication of the cause of death, administrative mistakes, or whether or not a resuscitation of a given SCA victim succeeded. Furthermore, over a time period of more than 20 years, there may have been a substantial change in the composition of the study population, e.g. the proportion of females (of whom it is known that they have a much lower SCD risk) may have increased as time went by. The paper does not provide data in this respect. Observational data suggest that the gradual implementation of emergency action plans and the widespread use of automatic external defibrillators has improved the survival of athletes who developed a SCA.[24]

Experience in Israel, where a mandatory pre-participation screening program was introduced in 1997, indicated a substantial variation in the yearly incidence of SCD. Israel researchers compared their findings that were based on reports of SCD in the general media, with the Italian data as shown in Figure 5. They stress that, if one compares SCD rates during the 2 years preceding the enforcement of screening in Israel with the mortality at the end of the study (as was done in the Italian study), one could conclude that the Israel screening strategy is extremely effective (D to G in Figure 5). It is only when one reviews the entire study period and compares the 12- year period before screening with the 12-year period after screening (from point C to G in Figure 5) that it becomes obvious that this apparent mortality reduction is most likely related to a year-to-year variation and may illustrate a regression-to-the-mean of the SCD incidence rate.[9, 69]

Figure 5 – Variation in reported SCD rates in athletes across studies.

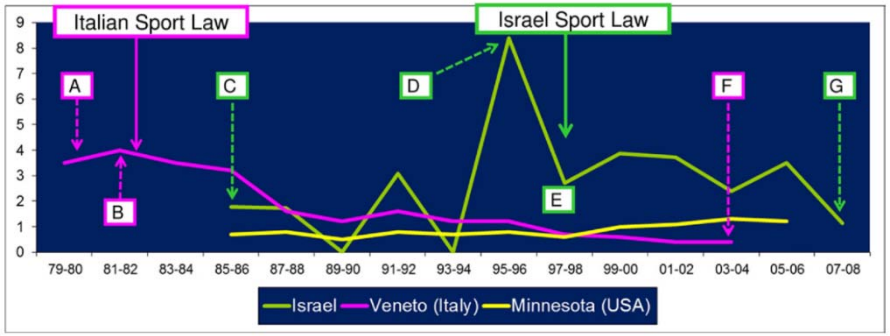


Figure extracted from Steinvil et al.[69] Data refer to the annual incidence of SCD per 100,000 person-years reported in Italy,[18] Israel,[69] and the US[70]. The Italian data are exactly those displayed in Figure 4.

Of note, the incidence rate in the late screening period in Italy does not significantly differ from SCD rates observed in the same era in countries that did not have a mandatory screening program. The mortality rate of athletes in Minnesota (1993 to 2004) was close to that in Italy (Veneto 0.87 per 100 000 person-years vs 0.93 in Minnesota (Figure 5).[1] In France, a nationwide survey from 2005-2010 revealed a similar number, 0.98 cases per 100 000 person-years.[21]

Based on the abovementioned reflections we think that the decreased incidence rate of SCD in Italy during the study was not causally related to the pre-participation screening. Several other authors have also expressed such doubts.[8, 10, 69, 71]

3.6.1.3 Ethical considerations related to the Veneto study

The scientific literature on pre-participation screening is dominated by a relatively small group of researchers. Most prominent among them is Domenico Corrado (University of Padua Medical School, Division of Cardiology, Padua, Italy) who is the principal investigator of the Veneto study. Through the reporting of the 1979-2004 Veneto data in numerous papers and his participation in guideline development groups and consensus meetings, he obviously played an important role in convincing sport physicians and sport federations around the world to promote pre-participation screening.

Corrado et al. prudently concluded in their seminal paper: "*The incidence of sudden cardiovascular death in young competitive athletes has substantially declined in the Veneto region of Italy **since** the introduction of a nationwide systematic screening"*.[18] However, over the years and with no new confirmative evidence, they apparently became convinced on a causal relationship between SCD rate and pre-participation screening in Veneto. This becomes clear if one considers the papers they published in consecutive years in leading medical journals. In a 2008 paper they conclude "*Pre-participation cardiovascular evaluation of competitive athletes essentially based on ECG **seems** according to the long-term Italian experience, to be a lifesaving strategy that adequately meets the criteria for a good screening program*".[60] A few years later, they write that "*The available evidence, based on the long-running Italian experience, **indicates** that ECG screening has to be considered an efficient health strategy for prevention of SCD of young competitive athletes. It meets the most important Wilson and Jungner's criteria*".[72, 73] Always referring to the same data, in 2013 it sounds as follows: "*Pre-participation screening based on a 12-lead electrocardiogram (ECG) **is** effective in identifying athletes with potentially lethal cardiovascular disease and saves lives by disqualifying them from competitive sports activity*."[63]

A number of the objections we summarised in our critical appraisal might be clarified by the Italian investigators if they would provide access to other unpublished Italian data. Firstly, although the Italian screening program is a truly national initiative, published data come only from one small region of this country (Veneto). It would be informative to know to what extent the Veneto data are representative of Italy overall.[8] Secondly, published data about the (remarkably high) pre-screening SCD incidence in Veneto are limited to the years 1979-1980 and are based on no more than 14 cases. Other sources report that annual pre-participation screening in Italy became mandatory by law in 1971.[7, 58] Therefore, one would expect that mortality data during those early years would be available. Thirdly, so far no screening or mortality data from the years after 2004 have been published. These would allow to better appreciate the random variation in SCD incidence.

Researchers from the UK tried to obtain additional data from the Italian screening program via a formal request from Mr. Jeremy Hunt, the UK Secretary of State for Health, to the Minister of Health in Italy, to no avail.[10] Upon our request in June 2014, professor Corrado responded that "we are analysing the SCD rates in the last decade (period 2005-2014)". So far, we did not yet receive more data.

Given the facts that both the scientific and lay communities are largely influenced by the published data from Italy, we think that the Italian investigators have the moral duty to make their data publicly available with no further delay.

Key points

- **In Italy, pre-participation screening of competitive atheletes, including ECG, is mandatory by law since 1971. Data from the Veneto region collected from 1979 until 2004 in athletes aged 12-35 years have been published. The annual incidence of SCD in Veneto decreased by 90% from 3.6/100 000 person-years in 1979-1980 to 0.4/100 000 person-years in 2003-2004.**
- **A critical appraisal of the study indicates that it does not provide sufficient evidence to conclude that the decline in the SCD rate in Veneto was due to the pre-participation screening.**
- **All relevant and up-to-date data collected in the Italian pre-participation screening program should be made available to the international scientific community as they may contain critical information on the impact of a pre-participation screening program.**

3.6.2 Experience with pre-participation screening outside Italy

A recent review identified 17 studies including 89,697 healthy subjects reporting the results of large screening initiatives.[9] The majority of reports were from the United States, but others came from Italy, the United Kingdom, Spain, Germany, and China. The percentage of athletes with abnormal findings on rest-ECG in those studies varies widely from 2.5% in one study of high school athletes to 35% in professional athletes, with the average being 12%. The vast majority (probably >90%) of abnormal ECGs in these populations were false-positives. The most commonly detected clinically relevant diseases reported were bicuspid aortic valve (0.09%), HCM (definitive and possible diagnoses combined: 0.05%), (0.06%), Wolff-Parkinson-White syndrome (0.03%). LQTS was less common, with 11 (0.01%) detected cases. The reported disqualification rate ranged from 0.2% to 4%. None of the studies provide data regarding the effect that such disqualifications have on mortality rate.

In Israel, a mandatory pre-participation screening program was implemented in 1997. There is no centralised national registry. A study that was based on newspaper reports identified 24 SCDs among competitive athletes during the years 1985 through 2009. Eleven of those occurred before the 1997 legislation and 13 occurred after it. Mandatory ECG screening of athletes had no apparent effect on their risk for SCD. The yearly SCD incidence during the decade before and the decade after the 1997 legislation was 2.54 and 2.66 events per 100 000 person-years respectively.[69]

In the Netherlands, a previously mandatory pre-participation screening was abandoned in 1984, except for professional and Olympic athletes.[71] In 2006, the Health Council of the Netherlands formally recommended against pre-participation screening because of lack of evidence that it prevents SCD, the high number of false-positives and the inherent risk for overdiagnosis and stigmatisation.[74] In 2007, an project named “Sportcor” was launched in order to prospectively register sudden death victims and collect cardiovascular abnormalities and electrocardiographic variations in athletes.[75] This initiative however has been halted because of a lack of funding. A Dutch governmental report in 2009 investigated the requirements for conducting a randomised pilot study that would study the clinical and cost-effectiveness of compulsory pre-participation screening. It concluded that performing such a study is infeasible because of the rarity of the underlying disorders and the unrealistically large number of individuals that would needed to be included.[71]

From our own literature search described earlier, we retrieved 3 additional studies of which two originated from the US and 1 large series from Greece. One US study involved a cohort of 2017 high school athletes.[35] One case of HCM (0.05%), 4 WPW (0.2%) and 1 bicuspid aortic valve (0.05%) were identified. Two athletes (0.10%) were disqualified for athletic competition. Another US study involving 1339 students detected 5 (0.4%) cases of WPW, 5 bicuspid aortic valves and 2 (0.15%) with mitral valve prolapse.[37] In a large cohort of competitive Greek athletes (n=22 205) WPW was detected in 8 (0.04%), HCM in 7 (0.03%) and LQTS in 1 (0.005%) individual.[36] In this cohort, 39 athletes (0.18%) were excluded from competitive sports.

3.7 Discussion

Sudden cardiac death in a healthy young individual is a devastating event. Appropriate measures to prevent such tragedies would be more than welcome. Pre-participation cardiovascular screening in this respect seems to be a sensible approach, but there is an ongoing polarised debate on its appropriateness. Solid scientific data on the effectiveness of pre-participation screening are lacking. Protagonists of screening argue that at least some SCDs can be prevented by an early diagnosis and subsequent treatment of a cardiac disease that may induce SCD. Antagonists claim that the diagnostic performance of the investigations presently used in pre-participation screening is insufficient, and that the benefit of detecting one rare true-positive case is annihilated by the harm induced by numerous false-positives.

Sports-related SCD in young people is a rare event with less than 10 incidents per year in Belgium. If a nationwide pre-participation screening program would be initiated, optimistic estimates indicate that 50,000 out of 1 million athletic 14-34-years-old Belgians would be submitted to further medical testing. In many of them additional non-invasive testing would be sufficient to exclude disease, but in others invasive testing with its own inherent risks would be needed.[48] These additional tests would not suffice to completely eliminate false-positives.

All screenees considered at risk for SCD after full investigation, will have to be advised on how to proceed further in life. Some will be temporarily or permanently disqualified from sports participation. Others will be proposed

a lifelong cardiologic follow-up or will be treated with drugs, catheter ablation, or implantation of a defibrillator. Since the absolute risk of SCD in these thoroughly investigated individuals (comprising both true- and false-positives) remains very low, most of them will not benefit from the recommendations they have been proposed, yet incur the harms induced.

Temporary or permanent disqualification from sports participation obviously may directly induce psychological harm both in the screenee and his family,[48] whereas the generally accepted beneficial effects of physical activity on mental and physical health may be lost.

The anxiety mongering resulting from overemphasising extremely rare events, and the resulting medicalisation of the life of the healthiest individuals of our society, raise ethical questions.

Taking into consideration the very small risk of SCD in true-positives and the very small mortality risk associated with invasive testing and/or treatment of all positives, the harm induced by screening might - even in terms of mortality - be larger than the benefit.

Key points

- **It is uncertain whether cardiovascular pre-participation screening is beneficial in terms of the prevention of SCD.**
- **There is less uncertainty on the potential harms induced by cardiovascular screening. In the absence of a perfect diagnostic test, and given the low prevalence of disease, at least 50 000 individuals from a target population of 1 million screenees would test positive. They would be subjected to additional medical investigations and/or treatments. Since more than 95% of them are false-positives, these interventions would most often be futile.**

4 ECONOMIC EVALUATION: A LITERATURE REVIEW AND BELGIAN COST CONSIDERATIONS

4.1 Methods

4.1.1 Research question

The aim of this chapter is to respond to the following question:

- What is the cost-effectiveness of cardiovascular pre-participation screening of young, non-professional atheletes?

A systematic search for economic literature was performed and cost considerations from the Belgian setting were discussed.

4.1.2 Search strategy

The systematic search was performed in two stages:

- First, Health Technology Assessments (HTA) on this topic were searched by consulting the HTA database of the Cochrane Library, websites of HTA institutes members of the International Network of Agencies for Health Technology Assessment (INAHTA) and websites of ex- or non-member HTA institutes such as NICE (National Institute for Health and Care Excellence) (see Appendix 2.1).
- Second, EMBASE, Medline (OVID), Psychinfo (OVID), EconLit (OVID), and NHS EED (National Health Service Economic Evaluation Database of the Cochrane Library) databases were searched to retrieve both full economic evaluations and reviews of full economic evaluations.

No language restrictions were imposed. Because a recent HTA including a review of full economic evaluations was identified in the first stage (i.e. Wiffen 2014)[10], the search strategy in the second stage performed on 17 September 2014 and updated on 24 December 2014 was restricted to the period not covered by this study, i.e. January 2013 up to December 2014. An overview of the search strategy and results is provided in Appendix 2.1.

4.1.3 Selection criteria

All retrieved references were assessed against pre-defined selection criteria, in terms of population, intervention, comparator, and design (Table 4). The population was restricted to athletes aged 14-34 years old and the intervention was a specific cardiovascular screening program. The comparators include other cardiovascular screening approaches or no screening. The design is restricted to full economic evaluations, i.e. studies comparing at least two alternative treatments in terms of costs and outcomes. Cost-minimization, cost-effectiveness, cost-utility, cost-benefit and cost-consequence analyses were eligible.

Table 4 – Economic evaluation selection criteria

	Inclusion criteria	Exclusion criteria
Population	Athletes between 14 and 34 years old	Other populations, such as the general population or athletes < 14 years old or > 35 years old
Intervention	A specific cardiovascular pre-participation screening program	Other interventions
Comparator	Other cardiovascular pre-participation screening approaches or no screening	Other comparators
Design	Full economic evaluations	Other designs such as cost calculations

The selection of relevant articles was performed in a two-step procedure: initial assessment of the title, abstract, and keywords, followed by a full-text assessment of the selected references. When no abstract was available and the citation was unclear or ambiguous, consideration of the citation was directly made on the basis of a full-text assessment. Reference lists of the selected studies were checked for additional relevant citations.

4.1.4 Quantity of research available

The first stage allowed us to identify 34 potential HTAs. After assessment of title and abstract, only one HTA was retained, i.e. the study of Wiffen 2014.[10]

In the second stage, after excluding 230 duplicates, 3144 unique citations were identified from the databases. Because the first stage allowed us to identify a recent HTA including a review of full economic evaluations (i.e. Wiffen 2014)[10], the search strategy in the second stage was restricted to the period not covered by this study, i.e. January 2013 up to December 2014. In total, 404 citations were found in this period. The seven economic evaluations identified in the review of Wiffen et al. were included in the flow chart as “references identified in recent HTA reports”.

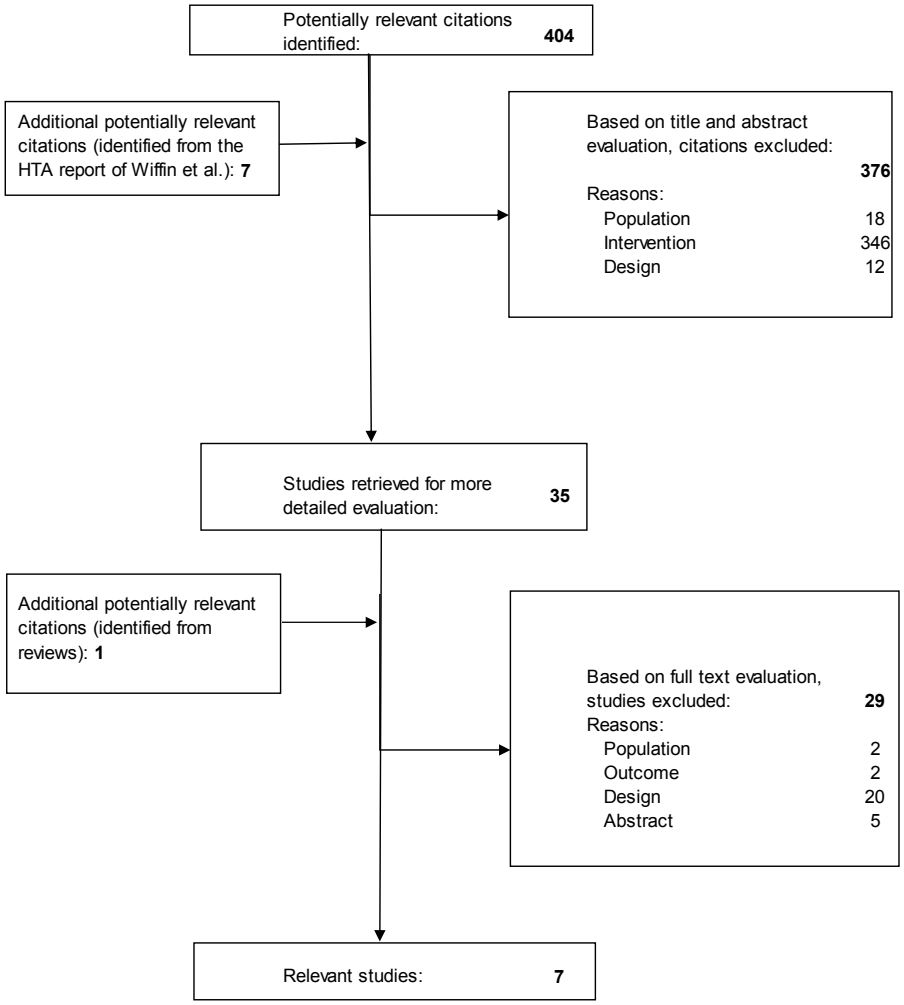
Of this total of 411 references, 376 did not meet the inclusion criteria based on title and abstract evaluation and 35 studies were retained for full text assessment. Based on full text assessment, one of the studies identified by Wiffen et al.[10] (i.e. Maron et al.)[53] was rejected on the design criteria because this study was a guideline that only included a short paragraph description of the potential cost and potential effect of the screening, with incoherence in the results presented.

At this stage, 18 narrative literature reviews were also excluded because these were less recent and had a lower quality[a] than the review of Wiffen et al.[10] Nevertheless, their references were checked to identify potential citations, which allowed us to identify one potential citation that was then excluded based on the population criteria (general population).

Figure 6 provides the flow chart of the selection process, inclusive the reasons for exclusion. In the end, 7 economic evaluations were selected.[3, 76-81]

[a] e.g. no literature search description or search performed in only one database.

Figure 6 – Flow Chart of the second stage (2013-2014)



4.1.5 Data extraction and quality assessment strategies

The identified full economic evaluations were summarized in an in-house data extraction sheet (see Appendix 2.1.3). These data extraction sheets are working documents that provide the basis to make summary tables which are provided in section 4.2. A description of the selected studies, a critical appraisal of their quality and the results is described in section 4.2.

4.2 Overview of economic evaluations

In the following parts, we provide an overview of the retrieved economic evaluations. General characteristics, the target population, interventions, input variables (costs, test accuracy, etc.), results and conclusions are described.

4.2.1 General characteristics

4.2.1.1 Design and analytical technique

Table 5 gives an overview of the characteristics of the 7 economic evaluations identified.[3, 76-81] Two studies were cost-effectiveness analyses,[3, 76] one study was a cost-utility analysis,[80] one study performed both a cost-effectiveness and a cost-utility analysis,[81] and three studies were cost-consequences analyses.[77-79]

4.2.1.2 Countries

All studies assessed the screening of athletes in the United States (US), except the study of Menafoglio et al. that was performed in Switzerland.[79]

4.2.1.3 Conflict of interest

Only the study of Leslie et al. mentioned they had a conflict of interest, reporting that one author was a consultant for Biosense Webster.[3]

4.2.1.4 Perspective

Five studies seemed to adopt a health care payer's perspective (not always clearly mentioned, see Table 5).[3, 76-79] Two studies reported they adopted a societal perspective and included travel time and cost or patient time.[80, 81] Productivity losses were never considered.

Remarks

- **The majority of studies have taken the perspective of a third-party payer. The studies of Schoenbaum et al. and Wheeler et al. have included some indirect costs[80, 81] but have not reported their impact separately, which reduces comparability across studies.**

4.2.1.5 Time horizon and discount rate

The time horizon spanned from 1 year to a lifetime. The studies of Schoenbaum et al. and Wheeler et al. discounted both their costs and outcomes at 3%[80, 81] and said they had analysed the impact of different discount rates in the sensitivity analysis. This impact was nevertheless not clearly reported.

The study of Leslie et al. seemed to have only discounted outcomes (3%) and not costs.[3] In the other studies, no discount rate was applied.

Remarks

- **Given the long term impact of the screening, the two studies based on a time horizon of 15 months (i.e. Menafoglio et al.)[79] and 5 years (i.e. Malhotra et al.)[78] could not be long enough to capture significant clinical endpoints. Nevertheless, the aim of these studies was only to assess the cost of screening per heart disease case detected. The time horizon was therefore sufficient for this objective.**
- **Other studies had a longer time horizon (20 years or lifelong) but had difficulties to find valid long-term data to populate the model. All of them were based on the Veneto study (except the study of Fuller[76]).**
- **Not all evaluations with a long-term time horizon applied a discount rate for costs and outcomes. In other studies, the impact of changing the discount rate was not reported.**

Table 5 – General characteristics of retrieved economic evaluations

Reference - Year (country); CoI	Analytic technique - Design				Time horizon (Discount rate)	Perspective
	CEA	CUA	CCA	Design		
Fuller 2000[76] (US) CoI: not specified.	x			No model, assumptions for long term effect.	Lifetime (not discounted)	Not specified (health care payer)
Wheeler et al. 2010[81] (US) CoI: No	x	x		Decision-tree + Markov model	Lifetime (3%)	Societal
Malhotra et al. 2011[78] (US) CoI: not specified.			x*	Observational study	5 years (not discounted)	Not specified
Leslie et al. 2012[3] (US) CoI: Yes	x			Decision-tree + Markov model – discrete event simulations	Lifetime (Until 75 years) (3%, only for outcomes)	Not specified (health care payer)
Schoenbaum et al. 2012[80] (US) CoI: No		x		Decision-tree + Markov model	Lifetime (3%)	Societal
Halkin et al. 2012[77] (US) CoI: No			x*	Projections (as stated by the authors)	20 years (not discounted)	Not specified (health care payer)
Menafoglio et al. 2014[79] (Switzerland) CoI: No			x**	Observational study	15 months	Not specified

**Cost per cardiac abnormality detected; **Cost per life saved; CCA: Cost-consequences analysis; CEA: Cost-effectiveness analysis; CoI: Conflict of interest; CUA: Cost-utility analysis; US: United States.*

4.2.2 Population

The population characteristics are described in Table 6.

These were (competitive) young athletes in all studies but the way to define them was often unclear. The study of Malhotra et al.[78] focused on elite athletes. Professional athletes are considered as out-of-scope for our study but, because elite does not especially means that these athletes were professionals, we decided to keep this study.

Only the study of Halkin et al.[77] included people younger than 14 years old in their population (i.e. between 12 and 35 years old).

In the CUA and CEA analyses,[3, 76, 80, 81] the assumed prevalence of heart disease in the population varied from 0.01% to 1.2%, while the overall prevalence of cardiac diseases that may induce SCD reported in section 2.4 was 0.3%. Variations in prevalence assumed in economic evaluations could in part be explained by the fact that studies did not take into account the same heart diseases in their model.

Schoenbaum et al. and of Fuller[76, 80] only took into account heart diseases considered at risk of SCD[b], the study of Leslie et al. took most frequent heart diseases at risk (i.e. HCM, WPW, LQTS)[3] into account while the study of Wheeler et al.[81] seemed to take every heart disease detected by the screening into account.

Remarks

- **Definition of (competitive) athletes was often unclear.**
- **The study of Malhotra et al. focused on elite athletes.**
- **Variation in heart disease prevalence was found between studies.**

Table 6 – Population

Reference	Population	Age	% males	Prevalence of heart disease
Fuller 2000[76] (US)	High school athlete participating in sports at risk of SCD (sports at risk not specified)	-	-	0.01%
Wheeler et al. 2010[81] (US)	High school and college competitive athletes	16 years	-	1.2%
Malhotra et al. 2011[78] (US)	Elite (division 1) athletes at university	19+/-2 years (SD)	49%	-
Leslie et al. 2012[3] (US)	High school freshmen participating in organized sports (i.e. 45% of all freshmen)	14 years	-	HCM: 0.05% WPW: 0.14% LQTS: 0.01%
Schoenbaum et al. 2012[80] (US)	Young athlete	14 years	59%	0.1%
Halkin et al. 2012[77] (US)	Registered participants in organized high school, college, and professional sporting activities	Between 12 and 35 years (population of the Veneto study)	-	-
Menafoglio et al. 2014[79] (CH)	Competitive athletes (doing regular exercise training and participating in official athletic competitions)	Between 14 and 35 years old (19.7 +/- 6.3, SD)	75.2%	-

(-): not specified; CH: Switzerland; HCM: Hypertrophic cardiomyopathy, LQTS: Long QT Syndrome; SCD: sudden cardiac death; SD: standard deviation; US: United States; WPW: Wolf-Parkinson-White.

[b] Not specified in the study of Fuller and not clear in the study of Schoenbaum et al: HCM (50%), arrhythmias (25%) and other (25%).

4.2.3 Intervention and comparator

Screening strategies are described in Table 7. Fuller[76] was the only study that seemed not to assess a combination of history and physical examination (H&P) and rest-electrocardiogram (ECG[c]) examination. Comparators were nevertheless unclear in this study. They seemed to compare a general pre-participation examination (not defined) to three other strategies, i.e. H&P according to recommendations of the American Heart Association, an ECG alone, or an echography alone.

Schoenbaum et al.[80] also compared different screening strategies together without comparison with no screening because history and physical examination (H&P) was considered in this study as the current standard practice.

ECG alone was a screening strategy investigated in three studies.[76, 80, 81] In the study of Schoenbaum et al.,[80], nevertheless, the strategy of ECG alone assessed the fact that cardiology referral was based on ECG solely but the cost of a H&P was still taken into account because they considered that in practice, H&P should always be done (likely to provide benefits beyond screening for cardiac risk).

It should also be noted that in the two observational studies,[78, 79] ECG and H&P assessment was done by more than one qualified health professional (including at least one cardiologist). In the study of Malhotra et al.,[78] the 2005 guidelines of the European Society of Cardiology on cardiovascular pre-participation screening in young athletes were followed while in the study of Menafoglio et al.,[79] the 2010 guidelines of the European Society of Cardiology were followed, with more strict criteria for ECG interpretation. Other studies were mostly based on the Veneto study, in which qualification required for screening assessment was high.

Finally, only the study of Halkin et al. assessed a yearly screening.[77]

Remarks

- **To be able to construct an efficiency frontier, all relevant strategies should be compared. Nevertheless, disagreements between studies were found concerning the identification of relevant strategies. Some studies considered that "no screening" should not be used as comparator because the current practice is H&P (also done for other reasons than cardiac screening). Some studies also considered a screening with ECG alone while some others considered that in practice, an ECG was always accompanied by an H&P.**
- **Only the study of Halkin et al. assessed the impact of a yearly screening[77] while the others focused on a one-time screening. Outcomes data are nevertheless usually based on the Veneto study[18] in which an annual screening was performed.**

[c] The abbreviation "ECG" in this chapter represents a rest-ECG

Table 7 – Description of interventions compared

Reference	No screening	H&P	H&P + ECG	ECG alone	Other	Yearly / one-time
Fuller 2000[76] (US)	-	x	-	x	Echo – general examination	One-time
Wheeler et al. 2010[81] (US)	x	x	x	x (in the supplement)	-	One-time
Malhotra et al. 2011[78] (US)	-	x	x	-	-	One-time
Leslie et al. 2012[3] (US)	x	-	x	-	-	One-time
Schoenbaum et al. 2012[80] (US)	-	x	x (ECG for negative H&P)	x (but included the cost of H&P)	-	One-time
Halkin et al. 2012[77] (US)	x	-	x	-	-	Yearly
Menafoglio et al. 2014[79] (CH)	-	x	x	-	-	One-time

(X): Investigated; (-): not investigated; CH: Switzerland; ECG: rest-electrocardiogram; Echo: echography; H&P: History and physical examination; US: United states.

4.2.4 Patient pathway and screening characteristics

Discrepancies were found among studies on patient pathways and on the performance of screening and diagnosis (second round) tests. Important variations in sensitivity and specificity values can be observed in Table 8. Moreover, compared to values reported in section 3, specificity values for H&P were quite high (i.e. a specificity between 69% and 85% was reported in section 3). For ECG, variations in specificity could partly be explained by an evolution of diagnostic criteria. According to the key points of section 3, imperfect data now suggest a specificity of 95% if the test is performed by expert sports electrocardiographists. Nevertheless, section 3 also highlighted that a combination of ECG and H&P might lead to a specificity of 70%, which is below values mentioned in Table 8. Concerning sensitivity values, there were rather in the lower range (i.e. in section 3, a sensitivity between 3% and 44% was reported for H&P and around 75% was reported for ECG).

The range of positive results in the selected studies after a first round screening varied as follows (see Table 8):

- With H&P: 2.2% to 5.9%
- With H&P + ECG: 5.8% to 30% (≈55% by including Leslie et al.[3])
- With ECG alone: 2.5% to 15.7%

Additionally to H&P and ECG, Fuller et al.[76] also analyzed the impact of using echography as first round screening and assumed a sensitivity of 80% and a specificity of 100% (positive results: 0.008%).

The study of Leslie et al.[3] assessed the impact of the screening per disease group (HCM, WPW, and LQTS) and used different specificity and sensitivity values for each disease. The cost of secondary testing and heart disease management was then assessed per group of diseases and summed to obtain a global cost. While an analysis per disease group is not a problem for true positives (the probability that patients suffer from two or more of these heart diseases is limited), this is not the case for false positives. Indeed, false positives for WPW could also be false positive for HCM and for LQTS, leading to an overestimation of positive results. By summing the positive rate for each disease group, 55% of patients seemed to be considered as positive.

Table 8 – First round screening: Performance of screening tests and percentage of positive results

	H&P			H&P + ECG			ECG alone			Source
	Sensitivity	Specificity	Positive results	Sensitivity	Specificity	Positive results	Sensitivity	Specificity	Positive results	
Fuller 2000[76] (US)	6%[1]	97.8%[2]	2.2%	-	-	-	70%[2]	84.3%[3]	15.7%	[1] Expert opinion ; [2] Fuller 1997 ; [3] Maron 1987
Wheeler et al. 2010[81] (US)	15%[1] (CI: 8-25%)	97%[2-3] (CI : 92-98%)	*3%**	68%[1-2] (CI : 50-73%)	95%[2;4] (CI : 93-97%)	*5.8%**	40%[5] (CI : 25-45%)	98%[5] (CI : 95.2-98.8%)	*2.5%**	[1]Fuller 1997 ; [2]Corrado 2006 ; [3]Nistri 2003 ; [4]Pelliccia 2007 [5]Not specified
Malhotra et al. 2011[78] (US)	-	-	5.9%	-	-	Around 30%	-	-	-	-
Leslie et al. 2012[3] (US)	-	-	-	HCM : 85%[1] WPW: 95%[2] LQTS: 80-85%[3]	HCM : 85%[1] WPW: 99.99%[2] LQTS: 60%[2]	*HCM : 15%** *WPW: 0.14%** *LQTS: 40%**	-	-	-	[1]Rodday 2012 (= ECG alone) ; [2]Expert opinion [3]Corrado 1998 ;
Schoenbaum et al. 2012[80] (US)	6%[1-2] (3%-40%)	95%[2] (85-100%)	5%	71%[3] (65%-77%)	91%[3] (72%-100%)	9%	70%[4] (65%-75%)	95%[2;4] (85%-100%)	5%	[1]Maron 1996 ; [2]Baggish 2010; [3]expert opinion ; [4]Fuller 1997 :
Halkin et al. 2012[77] (US)	-	-	-	-	-	9%[1] (20 years of annual screening)	-	-		[1]Corrado 2006
Menafoglio et al. 2014[79] (CH)	-	-	2.6%	-	-	6.3%	-	-	-	-

*(-) not investigated; *Own calculation; CH = Switzerland; H&P: History and physical examination; ECG: rest-electrocardiogram; HCM: Hypertrophic cardiomyopathy, LQTS: Long QT Syndrome; US: United States; WPW: Wolf-Parkinson-White.*

The patient pathway after a positive result during the first round screening was usually unclear. Globally, a selection of the following tests were identified:

- Cardiologist visit;
- ECG;
- Family testing with ECG;
- Echography;
- Exercise stress test;
- Holter;
- Blood pressure monitoring;
- Magnetic resonance imaging (MRI);
- Genetic testing;
- Cardiac electrophysiology study (EPS);
- Drug studies;
- Catheterisation;
- Coronary angiography.

Nevertheless, discrepancies between studies were found concerning the percentage of patients having these tests. Usually, every patients had at least a cardiology visit, an ECG and an Echo but the use of other tests varied greatly (see the Appendix 2.1.4 for more details).

At the end of the second round, the percentage of people with a diagnosed heart disease also varied (see Table 9). A global sensitivity and specificity of all the tests performed in the second round was only given in the study of Schoenbaum et al., i.e. a sensitivity of 0.9 (range: 0.8-1.0) and a specificity of 0.98 (range: 0.92 – 1.0) [d]. In the study of Fuller et al., the sensitivity and specificity of the second round was not specified but it seems that they assumed a sensitivity and a specificity of 100%.

[d] For a cardiologist visit (100% of patients), an ECG (100% of patients) and an echography if needed (90% of patients)

Table 9 – Second round: % of people diagnosed with a heart disease

	After H&P		After H&P + ECG		After ECG alone		Prevalence
	From the total of people screened	From people positive at screening	From the total of people screened	From people positive at screening	From the total of people screened	From people positive at screening	
Fuller 2000[76] (US)	Unclear	Unclear	-	-	Unclear	Unclear	0.01%
Wheeler et al. 2010[81] (US)	0.18%	5.7%	0.82%	14.2%	Not specified	Not specified	1.2%
Malhotra et al. 2011[78] (US)	0.34%	5.75%	0.88%	2.87%	-	-	-
Leslie et al. 2012[3] (US)	-	-	*Unclear* *HCM: ≈ 0.02%** *WPW: ≈0.14%** *LQTS: unclear*	Unclear	-	-	HCM : 0.05% WPW: 0.14% LQTS: 0.01%
Schoenbaum et al. 2012[80] (US)	0.11%	2.1%	0.24%	2.7%	0.16%	3.2%	0.1%
Halkin et al. 2012[77] (US)	-	-	2% (for 20 years)	22% (for 20 years)	-	-	-
Menafoglio et al. 2014[79] (CH)	0.65%	25%	1.03%	16%	-	-	-

(-): Not investigated. CH = Switzerland; H&P: History and physical examination; ECG: rest-electrocardiogram; HCM: Hypertrophic cardiomyopathy, LQTS: Long QT Syndrome; SCD: sudden cardiac death; US: United States; WPW: Wolf-Parkinson-White.

For people positive after the second round, the following interventions were identified (additionally to sport disqualification or restriction):

- Follow-up;
- Medication;
- Ablation;
- Cardiac surgery;
- Implantable cardioverter defibrillators (ICD);
- Pacemaker.

Discrepancies in treatment repartitions were found between studies (see in the appendix for this chapter for more details) and for an important part of patients (e.g. around 30% in the studies of Leslie et al.[3] and Schoenbaum et al.[80]), a simple follow-up seemed to be performed. This finding is in line with section 2.4, reporting that there was no agreement on the best management of asymptomatic individuals. It should also be noted that in the study of Schoenbaum et al.,[80] no ablation was performed, which could mean that they considered that Wolf-Parkinson-White (WPW)[e] was not a disease at risk of SCD or that these patients should not be treated. Conversely, in the study of Malhotra et al.,[78] an important part of patients positive after the second round had an ablation because the screening with ECG in this study mostly allowed to identify patients with WPW.

Remarks

- **Important discrepancies were found between studies concerning patient pathways as well as the performance of screening and diagnostic tests (both in terms of sensitivity and specificity). Concerning the number of positive results, no agreement was found between the studies (e.g. positive results after H&P + ECG varied between 5.8% and 30-55%).**

4.2.5 Quality of life data

Only two studies included the impact of screening on quality of life. Nevertheless, from these studies, it seems that no robust data could be identified. Wheeler et al.[81] used a study based on the HUI-III (Health Utility Index) to determine utility values of adolescents with normal health and adolescents with a heart disease but all other values were based on epxert opinions (consensus between the authors).
In the study of Schoenbaum et al.,[80] utility values were based on expert opinions, except the utility of patients with implantable cardioverter defibrillators (ICD) (see Table 10).
Moreover, Wheeler et al.[81] did not include the impact of sport disqualification on the quality of life while it is not clear if the study of Schoenbaum et al.[80] took this impact into account (the global decrement due to the diagnosis of a heart disease and sport restriction was based on a study analysing the quality of life of patients (average age 45 years old) with hypertrophic cardiomyopathy (HCM) in general and not of athletes with HCM).

Finally, Wheeler et al.[81] assumed that the quality of life of people with a heart disease returned to healthy levels within 4 years of diagnosis while Schoenbaum et al. assumed life-long decrements (with exponential trajectory to near normal levels over time).

[e] The treatment of WPW is usually either a follow-up, or an ablation.

Table 10 – Quality of life data

	Wheeler et al. (method)	Schoenbaum et al. (method)
Adolescents with normal health	• 0.94 - CI for simulation: 0.93-0.95 (HUI-III)	• 1 (expert opinion)
Adolescents with heart diseases	• 0.89 during four years - CI for simulation: 0.85-0.93 (HUI-III) • 0.94 - CI for simulation: 0.93-0.95 (expert opinion)	• Atheletes with heart disease and sport restriction: Decrement of 10% (range 5%-15%) during 0.5 year (range 0.1-1.0) (from another study also based on expert opinion) • Subsequent years (initial value): o If low risk of SCD (without treatment or on medication, also for false-positives): 0.98 (range: 0.97-0.99) (expert opinion) o If high risk of SCD and on medication: 0.97 (range: 0.96-0.98) (expert opinion) o If high risk of SCD and with ICD: 0.885 (range: 0.845-0.925), (disutility of battery replacement: 0.01 per event, every 7 years) (paediatric patients and parents self-reported preferences using the PedsQL, a generic instrument adapted to paediatric patients measuring health-related quality of life that can be transformed to a 0-100 scale, with 100 representing a perfect health) o These utilities recover on an exponential trajectory to near normal levels over time, except for patients who received an ICD implant.
Sport disqualification	• No impact, same utility values as people with heart disease.	• Stated above (athletes with heart disease and sport restriction). Nevertheless, the decrement is based on a study analysing the quality of life of patients (average age 45 years old) with hypertrophic cardiomyopathy (HCM) in general and not of athletes with HCM)
Athletes with a positive initial screening	• Decrement of 5% for 1 week compared to adolescent with normal heath, i.e. 0.939 for one year (expert opinion)	• Decrement of 10% (range 5%-15%) during 3.5 days (range 2.5-4.5), *i.e. 0.999 for one year** (expert opinion)
False negative	• No impact: 0.94 - CI for simulation: 0.93-0.95 (expert opinion)	• No impact: 1 (expert opinion)

**own calculation; ICD: Implantable cardioverter defibrillators; SCD: sudden cardiac death.*

Remarks

- **Only the studies of Wheeler et al.[81] and Schoenbaum et al.[80] expressed their results in terms of quality-adjusted life years (QALY) gained while the impact on the quality of life (QoL) is an important parameter to assess screening strategies. On one hand, only performing a cost-effectiveness analysis in this situation is therefore not appropriate but on the other hand, a cost-utility analysis cannot be done in an appropriate way because robust data are missing.**
- **The impact of sport disqualification or sport restriction is only partially taken into account in one study (Schoenbaum et al).[80]**

4.2.6 Other important input parameters for outcomes

The prevalence of the disease, the risk of SCD in athletes, and the effectiveness of patients' management had also an impact on the ICER and also for these input variables discrepancies were found between the studies (see Table 11). These important data were either based on expert opinion, or on the Veneto study (see section 3.6.1.1).

Table 11 – Risk of SCD per 100 000 athletes-years and effectiveness of patient management

Reference - Year (country)	SCD in athletes (total)	SCD in athletes with HD	Effectiveness of the management of patients with a heart disease	Sources
Fuller 2000[76] (US)	1	Not clear	It was assumed that 10% of those detected would live an additional 40 years and 90% would live an additional 20 years.	Expert (author) opinion.
Wheeler et al. 2010[81] (US)	*≈ 2.1**	174[$]	Impact of both disqualification and HD management: RR (SCD disqualified athletes with HD and under management / SCD in athletes with HD)= *0.16 (0.04-1)**	Corrado et al. 2006.[18]
Malhotra et al. 2011[78] (US)	-	-	-	-
Leslie et al. 2012[3] (US)	*1.5**	*824*[*$]*	Impact of disqualification: RR (SCD non-athletes with HD / SCD of athletes with HD) = *0.36*[*]* Impact of treatment: RR (SCD non-athletes with HD and treated / SCD non-athletes with HD and not treated)= 0 (except for WPW: RR *≈ 0.15** due to ablation failure)	Corrado et al. 2003.[52]Conservative assumption in favour of screening for the treatment impact
Schoenbaum et al. 2012[80] (US)	*2.25**	*2253*[*$]* (HD at risk)	Impact of disqualification: RR (SCD non-athletes with HD / SCD athletes with HD) = *0.41** Impact of treatment: RR (SCD non-athletes with HD under treatment / SCD non-athlete with HD and not treated) *≈0.85** for patients on medication (i.e. *72%**) and 0.05 for patients with an ICD (*1%** per year)	Corrado et al. 2003.[52] *Not clear for the treatment impact.*
Halkin et al. 2012[77] (US)	4	-	A linear reduction from 4 to 0.43 was applied on a 20 year period (*RR: 0.95 the first year; 0.108 after 20 years of annual screening**)	Corrado et al. 2006.[18]
Menafoglio et al. 2014[79] (CH)	-	-	-	-

*(-) Not investigated; *Own calculation; $Same sources but results differ because the definition of heart disease differed between studies (e.g. only heart disease at risk in the study of Schoenbaum et al. and every heart disease in the study of Wheeler et al.) CH: Switzerland; HD: heart disease; ICD: Implantable cardioverter defibrillators; RR: relative risk; SCD: Sudden cardiac death; US: United States.*

Remarks

- **In the study of Fuller[76], the effectiveness of the management of patients with a heart disease is only based on optimistic assumptions done by the author.**
- **In the studies of Schoenbaum et al.,[81] Leslie et al.[3] and Wheeler et al.,[81] important discrepancies were found concerning the risk of SCD for athletes with a heart disease and the effectiveness of patient management when they are detected. Discrepancies in SCD incidence rate for patients with a heart disease could in part be due to the fact that some studies only took into account heart disease at risk of SCD (Schoenbaum et al.)[80] while others took every heart diseases (Wheeler et al.)[81] or most important heart diseases (Leslie et al.)[3] into account.**
- **In these studies,[3, 80, 81] it was also assumed that athletes had a higher risk of SCD than non-athletes (at least 2.5) and that every detected patient had their risk of SCD reduced to the risk of SCD in non-athletes (also assuming that every detected patient complied with sport restriction/disqualification). Consequently, the higher the difference of SCD risk between athletes and non-athletes is, the higher the number of SCD cases is avoided by the screening (improving the ICER). Such an assumption is nevertheless not evidence based (see section 2.3 on sudden cardiac death in young sporty individuals).**
- **Most importantly, parameters used in these studies are based on the Veneto study. Additionally to the long list of limitations explain in the section 3.6.1 on the Italian experience, it should be noted that in the Veneto study, a yearly screening was done while in these studies, the screening was only performed one-time. Assuming that a one-time screening will have the same effect than a yearly screening is a very optimistic assumption.**
- **Finally, only the study of Halkin et al.[77] analyzed the impact of a yearly screening (also based on the Veneto study, with the limitation described in section section 3.6.1 on the Italian experience). Nevertheless, they assumed that the population in the non-screened group increased yearly by 2% and remained constant in the screened group, which inflated the number of people saved by the screening (more death without screening because more people practiced a sport and were at risk). They justified this assumption by the fact that in the Veneto study, 2% of athletes were disqualified each year, which was not the case. The disqualification rate of 2% in the Veneto study corresponds to the total number of people disqualified after 23 years of annual screening.**

4.2.7 Costs parameters

Four studies[76-79] only took into account the cost of screening tests and of the second round while others also included the management cost of patients with a heart disease.[76-79]

Important discrepancies was found in cost parameters between studies and in one study (Fuller),[76] the cost of screening tests were particularly low compared to other studies.

The cost of screening tests identified in the studies ranged as followed (see Table 12):

- H&P: $73-$130 (excluding the study of Fuller, i.e. $0)
- H&P + ECG: $107 - $360
- ECG alone: $34-$155 (excluding the study of Fuller, i.e. $10)

Discrepancies were also found concerning the average cost of the second round per person with a positive screening result at the first round. A minimum cost of $365 was identified (see Table 12).

It should also be noted that in one study (Leslie et al.),[3] second round and heart disease management costs were assessed separately per disease group and then summed. A second round cost was therefore taken into account for 55% of patients (sum of people positives for each disease), resulting in an overestimation of costs (at least for second round costs).

Remarks

- **Discrepancies were found between studies concerning cost parameters. Cost estimates used in the study of Fuller[76] were particularly low and second round costs in the study of Leslie et al. seemed overestimated.**

Table 12 – Cost parameters (in US $)

Reference (cost year)	H&P	H&P + ECG	ECG alone	Second round (average cost per people positive at screening)	HD management (average annual cost per patient with HD)
Fuller [76] (<1999)	$0	-	$10	After H&P: $500 After ECG: $365	Not included
Wheeler et al.[81] (2004)	$73 (including $12 of indirect costs) / CI: 55-84)	*$107**	$34 (including $7 of indirect costs) / (CI: 30-58)	$580 range: $330-$3000 (CI: $400-$780)	First year of diagnosis: $5000 (CI: $4000-$6000) Subsequent years: $350 (CI: $250-$450)
Malhotra et al.[78] (≈2010)	*≈$130** ($192000/1473)	Not clearly specified: *≈$170** (($192 000 + $54 095) /1473)	-	Average cost not clearly specified: *≈$1700* after H&P.* *≈$1000* after H&P + ECG.* (include ablation costs)	Not included
Leslie et al. 2012[3] (2010)	-	$195 (range: $107-$299)	-	Average cost not clearly specified: *At least $1000**	Average cost not clearly specified: *≈$11 500 the first year and ≈$600 the subsequent years**
Schoenbaum et al. [80] (≈2009)	*$112**	Not clearly specified: *≈$155**	Not clearly specified: *≈$155**	Average cost not clearly specified: *At least $543**	Average cost not clearly specified: *≈$1200* each year*
Halkin et al. 2012[77] (≈2012)	-	$263 ($360 including patient co-payment)	-	Average cost not clearly specified: *≈ $900** ($1170 with patients co-payment), mostly because the cost of an echo of $754 ($990).	Not included
Menafoglio et al.[79] (≈2012)	$83** (CHF 75.85)	$113** (CHF 102.8)	-	Average cost not clearly specified: ≈$780** (CHF 710)* after H&P + ECG (including ablation for 1.5%) and ≈$420** (CHF380)* after H&P (no ablation).	Not included

*≈approximation done according to the references; (-): Not investigated; *own calculation; ** converted using the exchange rate specified in the paper (CHF1 = $1.10). CH = Switzerland; CHF = Swiss Franc; HD: Heart disease; H&P: History and physical examination; ECG: rest-electrocardiogram; US: United States.*

4.2.8 Assessment of uncertainty

In four studies[76-79], uncertainty of results was not tackled. Fuller et al.[76] and Halkin et al.[77] did not perform a sensitivity analysis on uncertain parameters they used. Malhotra et al.[78] and Menafoglio et al.[79] were two observational studies and no relevant statistical parameters assessing the uncertainty of results (e.g. standard error) were reported.

Other studies performed either a one-way sensitivity analysis or both a one-way sensitivity analysis and a probabilistic sensitivity analysis (see Table 13).

Remarks

- **Uncertainty was not tackled in the studies of Fuller, Malhotra et al., Halkin et al., and Menafoglio et al.[76, 78, 79]**
- **Wheeler et al.[81] and Schoenbaum et al.[80] performed a probabilistic sensitivity analysis. Nevertheless, Wheeler et al.[81] did not follow standard recommendations for probabilistic analysis[82] because they reported they used normal distributions with the base case on the median and with asymmetric confidence intervals around the median.**
  Schoenbaum et al.[80] reported they used beta distributions for most parameters. Other distributions used were not specified. Moreover, they reported the probability of being cost-effective at threshold of $50 000/QALY and $100 000/QALY but did not report the credibility intervals of the ICERs.
- **Leslie et al.[3] decided to use a best case scenario as reference and limited the handling of uncertainty to a one-way sensitivity analysis (not on all uncertain parameters). Because their conclusion was not in favor of screening, using a best case scenario can be considered as an appropriate solution when the uncertainty is unknown and not evidence based (which is the case here).**

Table 13 – Assessment of uncertainty

Reference	Method
Fuller 2000[76] (US)	Not performed
Wheeler et al. 2010[81] (US)	One-way and probabilistic sensitivity analysis
Malhotra et al. 2011[78] (US)	Not performed
Leslie et al. 2012[3] (US)	One-way sensitivity analysis – presentation of a best case scenario as reference
Schoenbaum et al. 2012[80] (US)	One-way and probabilistic sensitivity analysis (CI not given)
Halkin et al. 2012[77] (US)	Not performed
Menafoglio et al. 2014[79] (CH)	Not performed

CI: credibility interval. CH: Switzerland; US: United States.

4.2.9 Results

A synthesis of the results of the economic evaluations is presented in Table 14 and Table 15.

4.2.9.1 Cost-consequences analyses

The studies of Malhotra et al. and Menafoglio et al.[78, 79] assessed a cost per detected heart disease abnormality and concluded that adding an ECG to H&P allowed to detect more cases at a reasonable cost (see Table 14).

Nevertheless, they only reported a cost per detected cardiac abnormality that was not especially at risk of SCD. By using a cost per detected heart disease at risk of SCD, the cost-outcome ratios would have been higher. Furthermore, in the study of Menafoglio et al.,[79] H&P alone would have been a dominated strategy compared to no screening (because no disease at risk of SCD were detected by H&P, see Table 14).

Moreover, authors specified that the sample size was limited, that physicians involved were highly experimented, and that ECG were interpreted by two experts (including at least one cardiologist). Therefore, they reported that their results may not be generalizable.

Halkin et al.[77] assessed a yearly screening with H&P and ECG and concluded that such a screening resulted in enormous cost per life saved of $10 600 000. They also added that the assumed SCD rate of 4 per 100 000 athletes-years in the non-screened population has been criticized as excessively high and that costs are probably underestimated. A lower SCD rate or higher costs will further increase the cost per life saved. Moreover, as stated in the section 1.1.1, the number of lives saved also seemed to be overestimated by the fact that the population in the non-screened group was assumed to increase each year by 2% while not in the screened group. A lower number of people saved would again have increased the cost per life saved. Conversely, they reported that results could be improved by using more restrictive definition of “abnormal” ECG, or in other words, by increasing the specificity of ECG. The uncertainty of results was nevertheless not handled. Finally, it would have been more appropriated to report a cost per life-year saved. If we assume that people could live an additional 100 years (overoptimistic), this would lead to an ICER of $106 000/LY (not discounted).

Table 14 – Results of the cost-consequence analyses

Reference - Year (country); CoI	Costs	Outcomes	Cost-outcome ratio
Malhotra et al. 2011[78] (US)	**Total cost:** • H&P: $343 725 • H&P + ECG: $894 870 ***Incremental cost*:*** • *H&P: -* • *H&P + ECG: +$551 145*	**Cardiac abnormalities detected:** • H&P: 5 of 1473 (0.34%) • H&P + ECG: 13 of 1473 (0.88%) ***Incremental outcome*:*** • *H&P: -* • *H&P + ECG: +8*	**Cost per cardiac abnormality detected:** • H&P: $68 745 per finding • H&P + ECG: $68 836 per finding ***Incremental cost-outcome ratio*:*** *H&P: -* *H&P + ECG: $68 893 per additional finding*
Halkin et al. 2012[77] (US)	**Incremental cost of screening (20 years):** • +$51 000 000 000 (+$69 000 000 000 including patient co-payment)	**Number of lives saved (20 years):** • +4813 lives	**Cost per life saved:** • $10 600 000 per life saved ($14 400 000 including patient co-payment)
Menafoglio et al. 2014[79] (CH)	**Total cost:** • H&P: $101 040.5** (CHF 91 855) • H&P + ECG: $173 210.4** (CHF 157 464) ***Incremental cost*:*** • *H&P: -* • H&P + ECG: +$72 170	**Cardiac abnormalities detected:** • H&P: *7 people detected, 6.5% of the total (own calculation)* • H&P + ECG: 11 people detected, 1% of the total **Cardiac abnormalities at risk of SCD:** 4 people detected (all by ECG, no by H&P), 0.4% of the total ***Incremental outcome*:*** • *H&P: -* • H&P + ECG: +4 (+4 at risk of SCD)	**Cost per cardiac abnormality detected:** • H&P: $14 434** per finding (CHF 13 122) • H&P + ECG: $15 747** per finding (CHF 14 315); *($43 303 per finding at risk of SCD)** ***Incremental cost-outcome ratio*:*** • *H&P: -* • *H&P + ECG: $18 042 per additional finding*

**Own calculation; ** converted using the exchange rate specified in the paper (CHF1 = $1.10). CH = Switzerland; H&P: History and physical examination; ECG: rest-electrocardiogram; SCD: Sudden cardiac death; US: United States.*

Summary of the cost-consequences analyses

- **The studies of Malhotra et al. and Menafoglio et al. concluded that adding an ECG to H&P allowed to detect more cases at a reasonable cost but that such results may not be generalizable. They nevertheless based this conclusion on a cost per cardiac abnormality detected not especially at risk of SCD. By assessing the cost per cardiac abnormalities at risk of SCD, results are less favorable. Moreover, it would be more appropriate to asses a cost per life-year saved. No clear conclusion can therefore be done from these two studies.**
- **The study of Halkin et al.[77] assessed a yearly screening with H&P and ECG and concluded, based on optimistic assumptions, that such a screening resulted in enormous costs per life saved. Based on own calculation, the ICER was superior to $100 000/LY.**

4.2.9.2 Cost-effectiveness analysis

H&P

Every study that analyzed H&P with other screening strategies and that have included "no screening" in the comparators agreed that such a strategy was either dominated or showed an extended dominance (see Table 15).[76, 81] Nevertheless, two studies considered that such a strategy should be done in practice for other reasons (not cited) (Wheeler et al.,[81] and Schoenbaum et al.[80]).

H&P + ECG, compared to no screening

Compared to no screening, a screening with H&P and ECG was usually not considered by the authors as a cost-effective strategy (or at the expensive end of the acceptable range assuming a willingness to pay threshold of $100 000 per life-year gained (LYG)). Leslie et al.[3] estimated an ICER of $90 828/LYG (QALYs not investigated) in the best case scenario and Wheeler et al.[81] found ICERs of $76 100/LYG (CI: $62 400/LYG – 130 000/LYG) and $111 000/QALY (CI: $58 800/QALY – dominated). The probabilistic analysis performed by Wheeler et al.[81] also showed the uncertainty in results. The probability of being cost-effective is 0% at a willingness to pay threshold of $50 000/LYG and 79.9% at a threshold of $100 000/LYG. Moreover, by taking into account the quality of life, the credibility interval showed that a screening with ECG and H&P could even be a dominated strategy. Their univariate sensitivity analysis also showed that results were mostly sensitive to:[81]

- The frequency of screening, i.e. an annual screening was unlikely to be cost-effective (one-time in the base case analysis), with an ICER around $500 000/LY;
- The prevalence of the disease. If the prevalence of heart disease was decreased to 0.3% instead of 1.2%, the ICER would become close to $200 000/LYG;
- The effectiveness of the patient management. If the risk of SCD in athletes with heart disase was 2.5 time higher than the risk for disqualified people (6 times in the base case analysis), the ICER of ECG+H&P would be equal to $93 000/LYG, and if the risk was only 1.6 times higher, the ICER of ECG+H&P would be close to $150 000/LYG;
- The risk of SCD in athletes. If the risk was dercreased to 38/100 000 person-years (instead of 174 in the base case), the ICER would become superior to $200 000/LYG;
- The cost of screening tests (impact not clearly reported);
- The specificity of screening tests (impact not clearly reported).

In the study of Leslie et al.,[3] results were most sensitive to the relative risk of SCD for athletes compared to non-athletes. If there is no difference, the ICER become superior to $200 000/LYG. The impact of less optimistic assumptions for other uncertain parameters was not reported. They did not perform a probabilistic sensitivity analysis but specified that their model represents a best case scenario unlikely to be achieved in real-world. Because they concluded that a screening with H&P and ECG was not cost-effective at a willingness to pay threshold of $50 000 ($90 828/LYG) using optimistic parameters, it can be expected that in a real world, such a conclusion will be maintained.

H&P + ECG, compared to H&P

It should be noted that some studies considered H&P as the current standard practice (also done for other reasons) and that a comparison with no screening was not considered appropriate. However, there is no evidence to support this (see chapter 3[f]).

Wheeler et al.[81] concluded that if H&P was the current standard practice, adding an ECG could be considered as cost-effective, with an ICER of $42 100/LYG (CI: $21 200 - $71 300) and $61 600/QALY gained (CI: $15 300-dominated). This is nevertheless above the usually accepted threshold of $50 000/QALY. Moreover, the probabilistic analysis showed the uncertainty of results. The probability of being cost-effective was 68% at a willingness to pay threshold of $50 000/LYG (but 99.9% at a threshold of $100 000/LYG). Probabilities for a threshold per QALY were not reported but the credibility interval show that by taking into account the quality of life, adding an ECG could even be a dominated strategy. Their univariate sensitivity analysis also showed that results were mostly sensitive to:[81]

- The frequency of screening. An annual screening was unlikely to be cost-effective (one-time in the base case analysis);
- The prevalence of the disease. If the prevalence of heart disease was decreased to 0.3% instead of 1.2%, the ICER would become superior to $100 000/LYG.
- The effectiveness of the patient management. If the risk of SCD in athletes with heart disase was only 1.6 time higher than the risk for disqualified people (6 times in the base case analysis), the ICER of ECG+H&P would be close to $200 000/LYG;
- The risk of SCD in athletes with heart disease. If the risk was dercreased to 38/100 000 person-years (instead of 174 in the base case), the ICER would become close to $200 000/LYG;
- The cost of screening tests (impact not clearly reported);
- The specificity of screening tests (impact not clearly reported).

Schoenbaum et al.[80] showed that compared to H&P alone, adding an ECG implied an ICER of $68 800/QALY gained and the probability to be cost-effective at a threshold of $50 000/QALY gained was only 30%. They concluded that a screening with H&P and ECG was not a cost-effective strategy compared to H&P alone. The univariate sensitivity analysis also showed that results were sensitive to the relative risk of SCD for athletes compared to non-athletes (2.5 time higher in the base case), the prevalence of heart disease in the population (0.1% in the base case), the specificity of ECG (0.95 in the base case) and H&P (0.95 in the base case) and the cost of ECG ($23 in the base case).

ECG alone

Fuller,[76] Wheeler et al.,[81] and Schoenbaum et al.[80] also analysed the impact of performing an ECG alone. Such an analysis has less sense because, as stated above, they firstly considered that H&P and ECG should be compared to H&P instead of to no screening because H&P should also be done for other reasons. If they consider that ECG alone could be performed, this means that they should also introduce no screening in the comparators.

Compared to no screening, Wheeler et al.[81] obtained an ICER of $62 100/LYG (CI: $43 900-$102 100) and of $94 600/QALY gained (CI: $24 400/QALY – dominated), which is above the usually accepted threshold of $50 000/QALY. No more details were given concerning the uncertainty of these results. The credibility intervals nevertheless showed that by taking into account the impact on the quality of life, ECG alone could even be a dominated strategy.

Fuller[76] obtained an ICER of $44 000/LYG but such ICER was only based on very optimistic assumption done by the author, such as a very low cost for the screening ($10) and high effectiveness of the screening (see section 4.2.6). No sensitivity analysis was performed in this study.

[f] Section 3.2 on the diagnostic performance of history-taking and physical examination and the discussion of the chapter 3 Clinical effectiveness of cardiac pre-participation screening

Because Schoenbaum et al.[80] considered that H&P should also be done for other reason, they compared a referral based on ECG alone to a referral based on H&P and included the cost of H&P in both strategies. They obtained an ICER of $37 700/QALY gained and a probability to be cost-effective at a threshold of $50 000/QALY of 66%. Nevertheless, their univariate sensitivity analysis showed that results were sensitive to:[81]

- The relative risk of SCD for athletes compared to non athletes. The ICER of ECG alone became superior to $50 000/QALY gained from a relative risk of 2.1 or less (base case: 2.5);
- The prevalence of heart disease in the population. The ICER of ECG alone became superior to $50 000/QALY gained from a prevalence of 0.075% or less (base case: 0.1%);
- ECG specificity. The ICER of ECG alone became superior to $50 000/QALY gained from an ECG specificity of 0.93 (base case: 0.95).
- H&P specificity. ECG alone dominates H&P when the H&P specificity falls below 0.87 (basecase: 0.95).

Finally, a question remains, i.e. can ECG alone be considered as good clinical practice (or, as assessed in the study of Schoenbaum et al., can we consider that base a decision on the ECG only, without taken into account the results of the H&P, is good clinical practice)?

Echo alone

Only one study (Fuller et al.)[76] assessed the impact of performing an echography alone during the first screening round and showed that such a strategy was not cost-effective (no sensitivity analysis was performed).

Table 15 – Results of the cost-effectiveness analyses

Reference - Year (country)	Costs	Outcomes	Cost-outcome ratio
Fuller 2000[76] (US)	**Incremental cost:** • No screening: - • H&P: + $11 000 per 1000 screened • ECG: + *$67 429 per 1000 screened** • Echo: + *$350 000 per 1000 screened**	**Incremental life-year saved:** • No screening: - • H&P: +0.13 per 1000 people screened • ECG: +*1.54 per 1000 screened** • Echo: +*1.76 per 1000 screened**	**Incremental cost-effectiveness ratio:** • *No screening: -* • H&P: $84 000/LYG *(extended dominance*)* • *ECG: $44 000/LYG** • *Echo: $1 285 000/LYG* (own interpretation, compared to ECG)* (reported: compared to no screening: $200 000/LYG)
Wheeler et al. 2010[81] (US)	**Incremental cost (compared to no screening):** • No screening: - • H&P: +$111 000 per 1000 athletes • H&P + ECG: +$199 000 per 1000 athletes From the supplement: • *ECG alone: around +$75 000*, based on figure 3*	**Incremental life-year saved (compared to no screening):** • No screening: - • H&P: +0.56 per 1000 athletes • H&P + ECG: +2.62 per 1000 athletes From the supplement: • *ECG alone: around +1.6*, based on figure 3*	**Incremental cost-effectiveness ratio (compared to no screening):** • No screening:- • H&P: Extended dominance • H&P + ECG: o $/LYG: $76 100/LYG (CI: $62 400/LYG – 130 000/LYG) o $/QALY: $111 000/QALY gained (CI: $58 800/QALY gained– dominated) *From the supplement (from probabilistic analyses, based on the median):* • No screening:- • ECG alone: o $/LYG: $62 100 (CI: $43 900-$102 100) o $/QALY gained: $94 600 CI: $24 400/QALY gained – dominated • H&P: Dominated • H&P + ECG (compared to ECG alone): o $/LYG:CI: *≈$117 000** (CI: $79 600-$233 200).

			o $/QALY gained: CI: *≈$190 000** (CI: $44 200 – dominated). **Incremental cost-effectiveness ratio (compared to H&P, i.e. considered as standard care):** • H&P: - • H&P + ECG: $42 100/LYG ($21 200 - $71 300) / $61 600/QALY gained ($15 300-dominated)
Leslie et al. 2012[3] (US)	**Incremental cost:** • No screening: - • H&P + ECG: +171 000 per 1000 individuals screened	**Incremental life-year saved:** • No screening: - • H&P + ECG: +1.9 per 1000 individuals screened	**Incremental cost-effectiveness ratio:** • No screening: - • H&P + ECG: $90 828/LYG
Schoenbaum et al. 2012[80] (US)	**Total cost:** • H&P: $169 270 per 1000 individuals screened. • ECG alone: $244 320 per 1000 individuals screened. • H&P + ECG: $286 170 per 1000 individuals screened. **Incremental costs:** • H&P: - • ECG alone: +$75 050 per 1000 individuals screened. • H&P + ECG: *+$41 850 per 1000 individuals screened, own calculation* (+$116 900 per 1000 individuals screened compared to H&P).	**Total QALYs:** • H&P: 28 475.237 per 1000 individuals screened. • ECG alone: 28 477.229 per 1000 individuals screened. • H&P + ECG: 28 476.938 per 1000 individuals screened. **Incremental QALYs:** • H&P: - • ECG alone: +1.992 per 1000 individuals screened. • H&P + ECG: *-0.291 per 1000 individuals screened, own calculation* (+1.701 per 1000 individuals screened compared to H&P).	**Incremental cost-effectiveness ratio:** • H&P: - • ECG alone: $37 700/QALY gained (probability to be cost-effective at a threshold of $50 000/QALY gained: 66%) • H&P + ECG: *Dominated (own interpretation)* (compared to H&P: $68 800/QALY gained, probability to be cost-effective at a threshold of $50 000/QALY gained: 30%)

4.3 Discussion

4.3.1 From the review of the literature

Even if the cost of an ECG seems a small price to save the life of a child, different arguments need to be analysed before concluding that adding an ECG would be a cost-effective strategy. Indeed, this depends a.o. on the performance of the ECG test to detect people at risk of SCD, the cost and risk associated with further evaluations (including false positive), and the impact of interventions being taken after a confirmed diagnosis. The low prevalence of heart disease at risk of SCD and the imperfect screening and diagnostic specificity result in a high number of healthy patients referred for additional tests.

According to results presented in the previous section, it seems that the cost-effectiveness of any screening strategy was quite doubtful. Among the three cost-effectiveness analyses that concluded in favour of a screening (i.e. Fuller and Schoenbaum et al. for ECG compared to H&P, and Wheeler et al. for ECG + H&P compared to H&P),[76, 80, 81] all of them falsely based their conclusion on the fact that H&P was the standard of care and analysed the impact of adding an ECG to H&P, or of basing the referral on ECG results alone. Nevertheless, according to chapter 3[g], it seems that there is no evidence showing that H&P should be the current standard practice. The study of Wheeler et al. showed that if no screening was considered in the comparators, H&P would result in an extended dominance, and adding an ECG to H&P would give an ICER superior to $100 000/QALY. All other cost-effectiveness analyses[h] concluded against screening.

Moreover, given the numerous concerns exposed in the methodological sections above, the validity of these results can be questioned and it seems that results would yet be worse in real practice. Indeed, every parameter for which results are sensitive (SCD risks, ECG specificity, and heart disease prevalence) have limited empirical basis and were quite optimistic in the models:

- The fact that SCD rate was assumed as higher in athletes than in non-athlete has an important impact on results. Nevertheless, the only comparative data supporting a higher risk of SCD in athletes comes from Padua (Italy). From other sources, it appears that the SCD risk is similar or may even be higher in non-athletes than in athletes (see section 2.3 on sudden cardiac death in young sporty individuals).[19, 20]
- Specificity of ECG assumed in the models was superior to 0.90 but it might be expected that a mass screening of athletes not always performed by a cardiologist would lead to a lower specificity (see section 3.3 on diagnostic performance of the ECG in pre-participation screening). Furthermore the overall specificity of screening that includes history-taking and physical examination in addition to an ECG inevitably decreases the overall specificity by adding additional false positives (see section 3.4 on diagnostic performance of history, physical examination and ECG combined). Most studies (Leslie et al., Schoenbaum et al., and Fuller)[3, 76, 80] also did not include all heart diseases that could cause SCD (e.g. Brugada syndrome, etc.). Seeking for more heart diseases could reduce the specificity of the screening and increase cost further (more false positive but also more treatment costs) making the ICER less favorable.
- Most importantly, effectiveness data are derived from the Veneto study, in which a yearly screening is performed. Assuming that a one-time screening would have the same effect as a yearly screening is very optimistic. Moreover, the findings in Italy have never been reproduced by other researchers (see section 3.6 on international experience with pre-participation screening). The study of Wheeler et al.[81] also showed that if a yearly screening was analysed instead of a one-time screening, all screening strategies were unlikely to be cost-effective.

[g] Section 3.2 on the diagnostic performance of history-taking and physical examination and the discussion of chapter 3 Clinical effectiveness of cardiac pre-participation screening.

[h] Or the same studies but with other screening strategies (e.g. H&P + ECG instead of ECG alone in the study of Schoenbaum et al.)[80]

It should also be noted that there is a lot of disagreement among experts about the treatment of those diseases in asymptomatic individuals (also reflected by the different pathways assumed in the models and in line with section 2.4). It is also not known whether avoiding vigorous exercise will prevent SCD to occur in other circumstances.

Moreover, negative implications of a (false) positive screening result was usually not taken into account (long term side effects of nontrivial therapies and lifetime medication or lifestyle restrictions and anxiety associated with living with a heart disease). A short term impact of false positive results and of heart disease diagnoses on the quality of life was only included in two studies; and the possible impact of sport disqualification was only partially taken into account in one study (Schoenbaum et al.).[81] Nevertheless, no robust data on quality of life seemed to be available, which makes it impossible to correctly assess such an impact. Beside negative impacts, benefits not related to cardiovascular health were also not included (e.g. identification and treatment of another disease).

Finally, it should be noted that cost estimates used in the studies are mainly based on US data and are not extrapollable to the Belgian setting. Nevertheless, when performed, sensitivity analyses showed that results were not sensitive to cost data, except concerning the cost of the first round. The impact of the cost of the first round was nevertheless not reported. A discussion on the impact of the screening with Belgian cost data can be found in the next section.

Summary of cost-effectiveness analyses

- **All three cost-effectiveness analyses that concluded in favour of a one-time screening (i.e. Fuller and Schoenbaum et al. for ECG compared to H&P, and Wheeler et al. for ECG + H&P compared to H&P)[76, 80, 81] falsely based their conclusion on the fact that H&P was the standard of care (not evidence based) and analysed the impact of adding an ECG or of basing the referral on ECG results alone. Compared to no screening, these strategies were not considered as cost-effective.**
- **In all other analyses,[i] a one-time screening was not considered as cost-effective.[3, 80, 81]**
- **All studies only analysed the impact of a one-time screening. The study of Wheeler et al. nevertheless showed in the sensitivity analysis that if a yearly screening was analysed, results were unlikely to be cost-effective.**
- **All sensitive parameters have limited empirical basis and all assumptions done for these parameters were very optimistic (in favor of a screening). More especially:**
  - **Effectiveness parameters were based on the Veneto study, in which the screening was annual (and not one-time). Moreover, the findings in this study have never been reproduced by other researchers (see chapter 3);**
  - **The risk of SCD in athletes was assumed higher than in non-athletes (assumption not supported by a number of studies, see section 2.3);**
  - **A high performance was assumed for screening tests;**
  - **The impact on the quality of life was not, or only in a limited way, taken into account in the analyses.**

4.3.2 *Cost considerations in a Belgian setting*

Because of the lack of robust data on every important parameter, we decided not to construct a model to estimate the cost-effectiveness of cardiovascular pre-participation screening strategies for athletes in a Belgian setting. This would not be efficient since it would be mainly hypothetical and have the same limitations as described above.

Nevertheless, the Superior Health Council (Hoge Gezondheidsraad / Conseil Supérieur de la Santé) concluded that even if there is currently no sufficient evidence in favor of screening, the screening of athletes between 14 and 34 years could be envisaged, with H&P every two years and arest-ECG every four years.[7] Therefore, we decided to describe here the general impact of such a screening approach using best case estimates and to

[i] or in the same studies but with other screening strategies (e.g. H&P + ECG instead of ECG alone in the study of Schoenbaum et al.)[80]

transparently discuss several important parameters in order to provide the different stakeholders insights in the possible costs and consequences of setting up a cardiovascular pre-participation screening program for athletes. It should be noted that we only discuss the impact of a first screening with ECG and H&P for athletes between 14 and 34 years. By repeating a screening program with H&P after two years and with an ECG after four years, the impact in terms of costs and harms would still be important while the impact on possible benefits would very probably be more limited. Results presented here would therefore be even worse by analysing the impact of repetitions. More details on calculations can be found in the appendix to this chapter.

4.3.2.1 Uncertain population

In terms of eligibility criteria, the Superior Health Council considers all young people who want to practice a sport for recreation or competition. These criteria are quite large and it is difficult to estimate the total number of people who would be eligible. If we assume that 1/3 of Belgians between 14 and 34 years could be eligible (i.e. 1/3[j] of 2 873 265 people aged 14-34 years in 2013), this gives a hypothetical cohort of around 1 million athletes to screen. We elaborate our example with such a hypothetical cohort since it is also very easy to transpose such a population size and all linked costs and consequences to another number.

4.3.2.2 Important costs

According to the Flemish Association of Sports Physicians (Vereniging van Sport- en KeuringsArtsen; SKA), on average, €60[k] per athlete will be asked for an organized cardiovascular pre participation screening in Belgium.[7] With a hypothetical cohort of around 1 million athletes to screen, this gives a cost of a first-round screening of around €60 000 000.[l]

The cost of the second round then depends on the percentage of positive results, which is determined by the prevalence of the heart disease in the Belgian population as well as the performance of the screening tests (sensitivity and specificity). As shown in the previous sections, all of these parameters are uncertain and an important variation between studies was found (e.g. between 5.8% and 30% positive results after H&P + ECG, see Table 8).

According to the Superior Health Council, in a very optimistic scenario, the sensitivity and the specificity of an ECG and H&P could be 0.75 and 0.95, respectively.[7] Based on the study of Magalski et al.,[54] they nevertheless recognized that for athletes and in real practice, these values could be lower and that up to 30% of athletes could have positive results. As reported in section 3.4 of this report, imperfect data suggest a sensitivity of pre-participation screening examination that includes history-taking, physical examination and ECG of 0.75 and a specificity of 0.70. For a prevalence of heart disease at risk of SCD of 0.3% in Belgium (as estimated by the Superior Health Council),[7] these two scenarios[m] give a percentage of positives results between 5.2% and 30.1%, which is similar to the range found in the literature review.

If we assume that these positive patients then, at the minimum, visit their cardiologist, have an additional ECG and have an echography to confirm the diagnosis, this gives a minimum second round cost of €117.72[n] per positive patient, i.e. a minimum second round cost that ranges between €6 133 212 and €35 474 922. This is a low cost compared to the minimum of $365 per positive patient identified in the literature (see 4.2.7). This cost also does not include other more expensive tests such as magnetic resonance imaging (MRI), genetic testing, or an electrophysiological examination, in some cases leading to catheter ablation and more rarely to prophylactic

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j No data found for Belgium but in France, 1/3 of young people have a licence for a sport.

k For H&P and ECG. This is low compared to the cost of screening (with H&P and ECG) identified in the models during the review of the literature on cost-effectiveness analyses (range $107 - $360).

l As recommended in the KCE guidelines,[83] we adopted the perspective of the health care payer. Because cost-sharing for such a screening between patients and governments (federal – federated entities) is not yet decided, costs are presented without distinction between payers.

m Scenario 1: Sensitivity: 0.75 – Specificity: 0.95; Scenario 2: Sensitivity: 0.75 – Specificity: 0.70;

n Official 2014 tariffs of the National Institute for Health and Disability Insurance that include official payments out of the government's health care budget as well as patients' co-payments.

implantation of a defibrillator. Other management costs such as medications were also not included.

Summary of potential costs

- **A first-round screening cost of around €60 000 000 for 1 million athletes screened**
- **A minimum second round cost[o] that ranges between €6 133 212 and €35 474 922 according to the performance of the screening tests.[p]**
- **Additional tests and the management of detected heart disease will further increase this minimal cost estimation.**
- **These costs will further be increased by taking into account repetition of screening.**

4.3.2.3 Potential harms

Because of the low prevalence of heart disease at risk of SCD in the population and the imperfect specificity of the tests, an important part of people will have a false positive test result after the first round. According to the performance of the tests (as assumed above), out of 1 million screened, up to almost 300 000 athletes may be false positives (i.e. between 49 850 and 299 100 according to the assumed performance of the tests), inducing unnecessary anxiety and costs.

By assuming a 100% sensitivity and specificity for the second round (i.e. again a very optimistic assumption), 2250 athletes on the 3000 with a heart disease at risk will be diagnosed and will have to live with such a diagnosis. The other 750 people at risk will be considered as "normal" and might therefore be less attentive to potential symptoms. Moreover, it should be noted that a specificity of 100% is unrealistic. With a specificity of 99% (still optimistic), between 2749 and 5241 will be diagnosed with a cardiac abnormality.

In addition, it is not clear what will happen after the second round and there is a lot of disagreement among experts about the treatment of those diseases in asymptomatic individuals. A number of screened will be disqualified for competitive sports participation or will be reoriented towards other kinds of sports. Still others will need life-long medical follow-up, repeat examinations, medical treatments, and so on.

It should also be noted that beside heart diseases at risk, an unknown number of cardiac abnormalities could be detected. Potential harms could therefore be even higher.

Summary of potential harms

- **According to the performance of the tests, between 49 850 and 299 100 athletes will be falsely positive at the first round, inducing unnecessary costs and short term anxiety.**
- **Around 2250 athletes will have to live with the knowledge of the disease, implying sport restriction and management of the disease (with a possible negative impact on the quality of life).**
- **This number (2250) can even be more important if we consider every cardiac abnormality that could be detected (and not especially heart disease at risk of SCD).**
- **Around 750 athletes with a heart disease at risk will not be detected and will be considered as "normal". They could therefore be less attentive to potential symptoms.**

4.3.2.4 Uncertain benefits

As discussed in the previous sections, no robust data exist concerning the incidence of sports related sudden cardiac death. Section 2.3 nevertheless showed that the risk can reasonably be estimated at around 1 per 100 000 athlete-years in European countries. According to these figure, each year, around 10 athletes on a million would therefore die suddenly for a cardiac cause. If we restricted the analysis to SCD during competition, the expected number of SCD would even be lower. Indeed, extrapolation of data from France, the Netherlands, Denmark, and US to the Belgian situation

[o] That only include a cardiologist visit, an ECG and an echography.

[p] Scenario 1: sensitivity of 0.75 and specificity of 0.95; scenario 2: sensitivity of 0.75 and specificity of 0.70.

suggested that maximum 2.5 athletes between 14 and 34 years would die during competition in Belgium (see section 2.3).

Moreover, among these people and according the assumed sensitivity of the tests, only 75% of them could be detected as being at risk. It is also not clear whether people detected will not anymore be at risk and whether avoiding vigorous exercise will prevent SCD to occur in other circumstances. Whereas the chance is low that pre-participation screening will save 1 of those individuals, it is also possible that 1 life will be lost because of overdiagnosis and overtreatment.

Summary of potential benefits

- **The incidence of SCD related to exercise is expected to be low, i.e. around 10 per 1 000 000 athlete-years. The number of SCD during competition would even be lower, i.e. maximum 2.5 per year for athletes between 14 and 34 years in Belgium.**
- **Not all of these athletes will be detected by the screening and be compliant with disease management. It is also not clear whether people detected will not anymore be at risk and whether avoiding vigorous exercise will prevent SCD to occur in other circumstances.**
- **There is no strong evidence that the risk of sudden cardiac death will be reduced by pre-participation screening of athletes.**

4.3.2.5 Uncertain ICER

According to an incidence of SCD related to exercise of 1 per 100 000 athlete-years, each year, around 10 athletes on a million would die suddenly for a cardiac cause without screening. In the most optimistic assumption, i.e. every detected patient is saved, and according to the sensitivity specified above (0.75), the screening could save maximum 7.5 patients each year. By assuming that until the next screening round with an ECG (i.e. four year), 30 patients could be saved (i.e. 7.5 x 4) and have the same life expectancy than people without heart disease (i.e. 56 additional years[q] according to the average life expectancy of people between 14 and 34 years old in Belgium)[84] and by discounting the life-years gained at 1.5%[r] as recommended in the KCE guidelines for economic evaluations,[83] this gives most optimistic ICERs that range at the minimum between €60 372/LYG and €87 157/LYG according to the performance of the screening tests (see the appendix to this chapter for more details).

It is important to note that figures presented here are not realistic and are minimum values not expected to be found with real data. Firstly, as specified above, cost estimations used represent a minimum that does not include all possible tests or patient management costs. Secondly, it is certainly too optimistic to consider that every detected patient will be saved and will have the same life expectancy than people without disease. Thirdly, the fact that some asymptomatic patients could not be compliant with sport restriction or disease management was not taken into account (best case assumption in favour of a screening). Fourthly, the ICERs would even be worsened by taking into account a repetition of screening tests (H&P every 2 years and ECG every four years) as the ratio of the costs versus potential benefits would increase. Fifthly, the ICERs would even be worsened by taking into account the impact on the quality of life.

In the worst case scenario, it can even be expected that a cardiavascular pre-participation screening would lead to important costs and harms for no benefit and would therefore be considered as a dominated strategy compared to no screening. Indeed, as specified in section 3.7, it is uncertain whether a cardiovascular pre-participation screening is beneficial in terms of SCD prevention and taking into consideration the very small risk of SCD in true-positives and the very small mortality risk associated with invasive testing and/or treatment of all positives, the harm induced by screening might - even in terms of mortality - be larger than the benefit.

Finally, it should still be noted that potential benefits not related to cardiovascular health were not taken into account in this analysis (e.g. identification and treatment of another disease).

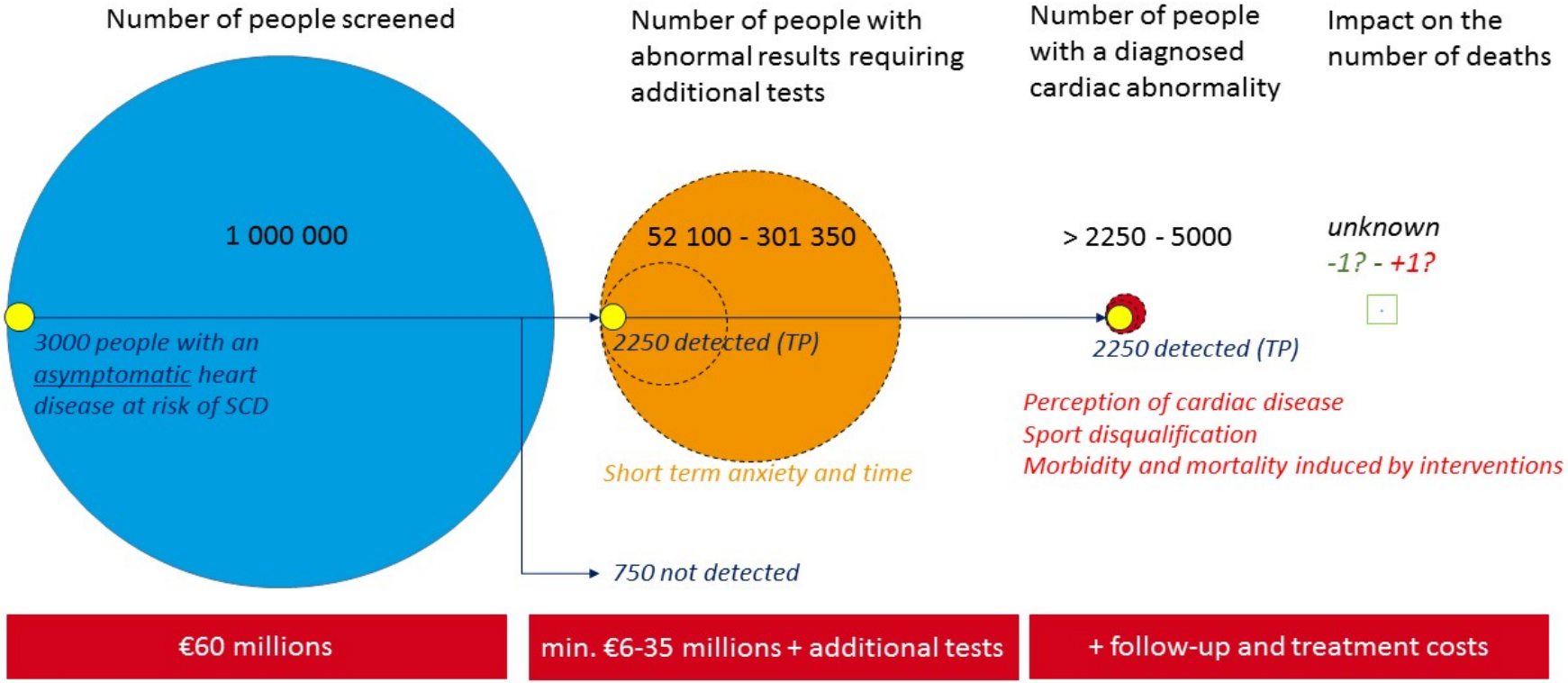
[q] The first year of the screening.

[r] And with half-cycle correction.

Summary of costs versus benefits

- **Even in the most optimistic scenario (best case deterministic analysis), the ICER would be relatively high (> €60 000/LYG). In the worst case scenario, a cardiovascular pre-participation screening would be a dominated strategy compared to no screening.**

Figure 7 – Impact of a cardiovascular pre-participation screening with H&P and ECG in Belgium (for a sensitivity of 0.75 and a specificity between 0.70 and 0.95)



TP: True positive; SCD: Sudden cardiac death.

4.4 Conclusion

According to results presented in the previous sections, it seems that the cost-effectiveness of any screening strategy was quite doubtful. All sensitive parameters (SCD risk, ECG specificity, and heart disease prevalence) have limited empirical basis and were quite optimistic in the models. In general, there are no reliable economic evaluations with convincing results showing that pre-participation screening in competitive athletes provides value for money. On the contrary, based on optimistic assumptions (or best case analysis in one study), economic evaluations usually showed that a one-time pre-participation screening was not cost-effective or might even be a dominated alternative if we take into account the impact on the quality of life. Two studies (Wheeler et al. and Halkin et al.) also showed that a yearly screening was unlikely to be cost-effective.

From the limited evidence available, it seems that performing a screening of athletes with ECG and H&P every four years (or more often) in Belgium would be expensive and potentially harmful compared to the uncertain benefits.

Key points

- **There are no reliable economic evaluations with convincing results showing that cardiovascular pre-participation screening in young athletes provides value for money. Based on optimistic assumptions, a one-time pre-participation screening seems not cost-effective or might even be a dominated alternative if we take into account the impact on the quality of life. Results are even worse for a yearly screening (unlikely to be cost-effective).**
- **In a Belgian setting, the screening of athletes with ECG and H&P every four years (or more often) would be expensive and potentially harmful compared to the uncertain benefits.**

5 LEGAL ASPECTS OF CARDIOVASCULAR PRE-PARTICIPATION SCREENING

5.1 Introduction

Sudden cardiac deaths in athletes are very visible, primarily due to media attention, and are compelling emotional reactions. Accordingly significant liability concerns exist for different parties involved in the decision making process of cardiovascular pre-participation screening. The first question relates to the government's duty to impose (or not) such tests and to define their content. Legislation can play a decisive role in the necessity and the modalities of pre-participation screening and in the responsibilities (and the related possible liability) of the parties involved. In Belgium, legislative initiatives have been taken by the respective competent Communities in Belgium. In a first part (5.3) the current legislation of the different Communities related to this issue will be discussed. These regulations describe the conditions for safe and healthy sports exercise in general and primarily aim at the prevention of sports-related injuries and the promotion of appropriate (medical) assistance and advice for athletes. For this report possible application of the relevant legislation to pre-participation screening for cardiac anomalies is focused on.

In the absence of binding legal requirements, sports clubs may rely on a physician or other medical personnel to determine appropriate medical screening procedures. Requests for medical examination also frequently stem from the athletes' initiative. In that scope, physicians may be faced to liability concerns. Who is liable if an athlete suddenly dies during sports participation while no ECG or any other pre-participation examination was carried out? Who is liable when an ECG was carried out but the result was false negative? Regardless of the question whether systematic cardiovascular screening is the best option to go for, physicians' (sports clubs' and other persons involved) fear for liability can lead to defensive medicine, involving the systematic screening and medicalization of sports participation. In a second part (5.4) the legal responsibilities and physicians' duties in the eligibility decision making process will be discussed.

For athletes, different rights and duties arise once entered in a medical patient-physician relationship. Although pre-participation screening differs from the classical patient-physician relationship as it does not always take place on his/her demand and there are (mostly) no symptoms, patients' rights and privacy regulations play a significant role. These rights will be highlighted in a third part (5.5).

In a fourth part, the situation in a selection of other countries will be briefly summarized (5.6).

5.2 Methods

The study uses traditional legal research methods. Regarding the Belgian legal framework, the legal databases Juridat and Jura served as a data source. Additionally, websites of governmental bodies and relevant organisations were consulted. Oral information was obtained by several experts in the domain. A list of the contacted experts is available in colophon.

5.3 Sports Policy in Belgium: a competence of the Communities

In Belgium, the Communities are competent for person related matters such as preventive health care and for cultural matters such as sports. This implies that regulations regarding sports policy and the related health prevention related to sports participation differ in all Communities (Dutch, French and German speaking). The overlap between preventive health care and sports does not ease the situation, especially when different matters regarding sports participation (such as ethical sports participation and healthy sports participation) are incorporated in one Decree, such as for instance in the Flemish Decree regarding healthy and ethical sporting (cfr. Infra).[s] The Council of State stated in this matter that regulations regarding "medically responsible sporting" need to be considered as activities and services of preventive health care (person related matters).[t] The power to exercise the competences of the French Community regarding pre-participation screening have been transferred to the Walloon Region for the territory of the French Community.[u]

In Brussels-Capital decrees regarding person related matters apply to institutions that based on their organisation, need to be considered as belonging to one or the other Community (Flemish or French). The "based on their organisation" criterion refers to the language according to which the uni-community institution is organised. Yet, these uni-community organisations need to be accessible for anyone and are not allowed to limit their activities to one Community, even though the uni-community character will imply that particularly persons of the respective Community will turn to those organisations. The Flemish Community Commission and the French Community Commission exercise Community competences in Brussels. The Joint Community Commission has responsibility for matters considered as "bi-personal", the personal matters for which the Communities do not have competence on the bilingual territory of the Brussels-Capital Region. These matters concern, on the one hand, measures applying directly to individuals and, on the other hand, institutions which, because of their organisation, are not attached exclusively to one of the Communities. These bilingual institutions are either public institutions which are bilingual by definition, such as public hospitals, or private institutions which have not opted for either, such as bilingual sports clubs or sports events. This implies that the organs of the Flemish Community Commission, the French Community Commission, as well as the Joint Community Commission have competences in the domain of sports and preventive healthcare (pre-participation screening).

Although Communities have their proper competences, collaboration agreements enable collaboration on specific topics. In that scope, several collaboration agreements were contracted on the prevention and the doping

[s] In Brussels Capital decrees regarding cultural matters (such as sports) apply to institutions that based on their activities need to be considered as belonging to one or the other Community whereas decrees regarding person related matters apply to institutions that based on their organisation need to be considered to one or the other Community

[t] Advice 44/914/I/V of 26 August 2008 Council of State

[u] Art. 3 g Special Decree of 3 April 2014 regarding the transfer of the exercising of the competences of the French Community to the Walloon Region and the French Community Commission, *B.S./M.B.* 26 June 2014

control in sports.[v] For pre-participation screening, however, this option has not been used.

In the next chapter, the policy of the different Communities, focused on pre-participation screening is discussed.

5.3.1 Pre-participation screening in the Flemish Community

5.3.1.1 Scope of the Flemish Decree regarding medically and ethically responsible sporting

Sports pre-participation screening in Flanders is regulated by the Decree of 20 December 2013[w] (HES Decree) and its Executory Decree of 4 April 2014[x]. Originally, "medically responsible sporting" (medisch verantwoord sporten) and doping was regulated in a 1991 Decree.[y] Later on, topics on ethical sporting were added.[z] In 2012 the stipulations on doping were shifted to a separate Decree.[aa] The stipulations on medical and ethical sporting were updated and restricted, which resulted in the HES Decree. Whereas former regulations[bb] primarily focussed on the talented athletes, the underlying idea of the HES Decree is to provide a framework promoting and sustaining healthy and ethical sporting for all athletes (recreational, elite or top sport, specific sports), regardless whether they are participating in organised or structured sports or not. The key idea of the Decree is to respect and stimulate the autonomy of the sports community. The prevention of sports specific injuries and sports related health risks needs to be elaborated by sports organisations in collaboration with the government. Accordingly, the government opted for a supporting and facilitating policy with an increased responsabilisation of sports organisations (including sports clubs, sports federations, and organisations of sports manifestations) and athletes of all levels, exercising recreational, competitive or demonstration purposes.[cc] Sanctions or correcting measures are only imposed as a last option.

5.3.1.2 Pre-participation screening for cardiac anomalies = population screening for disease prevention?

During parliamentary works on the HES decree, the question was raised whether pre-participation screening falls in the field of application of the legislation regarding population screening. This legislation (Decree 12 December 2008 hereinafter called as Decree population screening[dd]) was implemented to promote the quality and the appropriateness of population screening. According to the Decree population screening, population screening on the initiative of a third party is submitted to the consent of the Flemish Minister of Public Health. The decision of the Minister needs to be based on the advice of the Flemish Working Group Population screening (Vlaamse Werkgroep Bevolkingsonderzoek). Population screening can solely be permitted if it is open for all persons of the respective target group, if there is a sound scientific basis for efficacy and efficiency and if it is very probable that the participants will have more benefits than disadvantages (art. 4 Decree population screening). In the explanatory memorandum of the HES Decree, it was stated that "aspects of pre-participation screening that

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v Collaboration agreement of 24 November 2011 between the Flemish Community, the French Community, The German speaking Community and the Common Community Commission regarding the prevention and control of doping in sports

w Decree of 20 December 2013 regarding healthy and ethical sporting, *B.S./M.B.* 31 January 2014

x Decree of 4 April 2014 of the Flemish government executing the decree of 20 December 2013 regarding healthy and ethical sporting, *B.S./M.B.* 1 July 2014

y Decree of 27 March 1991 regarding healthy, *B.S./M.B.* 18 June 1991

z Decree of 13 July 2007 regarding medically and ethically responsible sporting, *B.S./M.B.* 13 September 2007

aa Decree of 25 May 2012 regarding the prevention and control of doping in sports, *B.S./M.B.* 12 July 2012

bb S. Van Mulders, "De medisch verantwoorde sportbeoefening", in X. *Handboek voor sportrecht*, Brugge, Die Keure, 1995, 223 e.v.; R. Roelandt en R. Janvier, "Het Decreet inzake medisch verantwoorde sportbeoefening en de sportmedische keuring", *Medi-Ius* 1996, 20-25

cc Verslag bij het ontwerp van Decreet inzake gezond en ethisch sporten namens de Commissie voor Cultuur, Jeugd, Sport en Media, *Parl. St.* Vlaams Parlement 2013-2014, 2280/ 3, (https://docs.vlaamsparlement.be/docs/stukken/2013-2014/g2280-3.pdf)

dd Decree of 12 December 2008 of the Flemish government regarding population screening for prevention, *B.S./M.B.* 19 February 2009.

are not focussed on the detection of anomalies related to pathologies or diseases, are not considered as population screening in the sense of the Decree population screening, dealing with organised population or specific population groups screening actions for disease prevention". The Vlaamse Werkgroep Bevolkingsonderzoek also advised on this issue in 2012 and 2013. According to this working group, per analogiam with risk screening for labour participation, sports medical screening could be excluded from the scope of the legislation regarding population screening, if sound categories of specific risks per sports discipline can be defined and if the circumstances for which the exceptions apply are described (professional athletes, recreational athletes,…).[ee] In response to this, a Decree of the Flemish government of 16 May 2014 excluded particular types of sports screening from the field of application of the legislation regarding population screening. Pre-participation screening exclusively evaluating the aptitude of an athlete for sports and for which no medical intervention linked to the test result is expected is explicitly excluded from the scope of the legislation regarding population screening (art. 2, 7° Decree population screening[ff]). For example, a person with a very low weight beneath all boxing categories, with an increased risk for fractions, is physically not apt to go for boxing. A second exception is pre-participation screenings aiming to reduce health risks related to sports including a particular health risk, that is performed in line with the conditions and quality requirements set by the government (art. 2, 9° Decree population screening). It is questionable, however, whether screening for cardiovascular risks falls within these exceptions. For the first exception, the interpretation of the notion "exclusive" is problematic. Most screening will reveal results that can be used for the evaluation of health in daily life and not only for sports aptitude. Cardiovascular screening for instance, is not exclusively evaluating the aptitude for sports, it also applies to other activities in daily life. Moreover, if a positive test result is revealed, further medical examinations and possible medical interventions will be performed. The second exception relates to screening for health risks related to sports including a particular health risk. It is not clarified in the HES Decree, however, what should be understood by sports including a particular health risk. At least, it is not clear which sports categories imply such physical effort that there is a particular, increased risk for cardiac related accidents. Furthermore, it should also be mentioned that screening for cardiac anomalies is not a sports-specific risk screening at all. Cardiac anomalies can have the same (mortal) consequences during intensive daily activities (e.g. sprinting to catch a train) than during sports.[gg] As such, it could be argued that systematic pre-participation screening for cardiac anomalies does not fall into the field of application of the relevant exceptions. This implies that, as far as it is not defined for which sports categories there is a particular cardiac-related risk, pre-participation screening for cardiac anomalies could only be implemented with the consent of the Minister of Public Health.

5.3.1.3 Policy obligations of the government

The government has a general role in the definition of the preconditions for safe and healthy sports participation (art. 10 HES Decree). Governmental measures primarily intend to be focussed at support, facilitation, knowledge generation and dissemination in order to streamline quality and enhance uniformisation. Expert commissions can be created to provide advice to the government (art. 13 HES Decree and art. 7 and 8 of the Executory Decree). The expert commission for risk combat sports, for instance, is charged with the task to advice the Minister. Additionally, organisations for policy support and practice development can be set up (art. 14 HES Decree).

In relation to screening, the government is hold to protect citizens for needless or harmful screening. Accordingly, the implementation of population screening is submitted to several conditions (see 3.1.2).

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ee Advice nr. 12-01 of the Flemish working group population screeningregarding sportsmedical screening and the field of application of the Flemish legislation regarding population screening

ff Decree of 12 December 2008 of the Flemish government regarding population screening for prevention, *B.S./M.B.* 19 February 2009, modified by Decree of 16 May 2014 of the Flemish government regarding diverse stipulations for the execution of the Decree of 21 November 2003 regarding preventive health policy and modifying executory decrees of this Decree, *B.S./M.B.* 15 September 2014

gg Advice nr. 12-01 of the Flemish Working Group population screening regarding sports medical screening and the application of the Flemish legislation on population screening

5.3.1.4 Choice to impose or recommend pre-participation screening = responsibility of sports organisation

Sport organisations are held to stimulate healthy sporting by initiatives and measures enabling the prevention and management of sports related factors that could have a negative impact on the physical or psychological health of the athlete (art. 6 HES Decree). Athletes (or the parents or guardian of minor athletes – for the rights of minor patients) need to be informed on these initiatives and measures (art. 7 HES Decree). In that scope, sports organisations, regardless the competitive nature of the discipline are free to impose or recommend medical eligibility tests for athletes under their responsibility and to define the content of a possible test (art. 8 HES Decree). The legal relationship between the athlete and the sports organisation will most often be established by the enrolment as a member of the enrolment for an event. As such a test can be imposed or recommended by a sports club, a federation or the organizer of a sports manifestation. The test should be exclusively focussed on the evaluation of the physical aptitude of the athlete for the particular sports exercise (art. 9 HES Decree). Elements such as age, the nature of the sports, the intensity and the existing regulation on the international level need to be taken into account, when imposing such tests. Sports organisations have the duty to inform athletes (for minors also the parents or guardian) about the content and the effects of the test (art. 8, 3rd section HES Decree). Although in principle the sports organisations can freely determine the modalities of tests, quality standards regarding the content, the modus operandi or the required qualification to perform the test can be imposed or recognised by the government (art. 9, 3 HES Decree). In the preparatory works of the HES Decree it is stated that quality standards are norms, recommendations, procedures, operational techniques, etc.[hh] For sports involving a particular risk for the physical and psychological integrity of the athlete, the government can impose a particular medical eligibility examination (art. 10, 5° Decree HES Decree).

5.3.1.5 Any physician can perform a medical eligibility test

The Decree foresees that tests need to be performed by a physician. As sports medicine is not a legally recognised discipline, there is no specification on the qualification (GP, sports physician, etc) or the affiliation (part of the club, federation or not…) of the phsyican (art. 9, 2nd section HES Decree). In the parliamentary explanations related to the Decree, it was stated that GPs should be able to perform eligibility tests conform validated guidelines, standards and protocols if they have the necessary expertise. Training can be foreseen to ensure such expertise.[ii] In line with the explicit choice to focus on healthy sporting for all athletes and to appeal to standardised methods to ensure quality in prevention, the former system of medical examiners (keuringsartsen) and centers for medical sports screening of athletes has been reformed. Based on the idea to concentrate expertise, the Flemish government recognised (and subventioned) centers for sports screening and medical examiners having a specialisation in sports medicine. Top athletes could be medically screened and assisted for free by a recognised center for medical sports screening[jj]. The recognitions for these centers and the examiners stopped in 2014. From 2015 on, the elite athlete gets a budget for yearly testing by a physician fulfilling specific requirements related to amongst others diploma, experience and equipment.[kk] .

5.3.1.6 Guidelines and protocols as quality assurance

The overall idea to ensure quality by validated guidelines and protocols rather than reserving the practice of pre-participation screening to particular qualified physicians (e.g. medical examiners or sports physicians) also enables the uniformisation of the common practice. Whereas today, the content and the modalities of pre-participation screening vary, guidelines and protocols may help to harmonise practice (cfr.infra). According to the preparatory works, general standardised screening protocols and specific protocols adapted to the sports level (recreational, competitive or top sports)

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hh *Parl. St*. Vlaams Parlement 2013-2014, 2280/001, p.17

ii *Parl. St*. Vlaams Parlement 2013-2014, 2280/001, p. 8

jj https://www.bloso.be/TOPSPORT/ANDEREACTOREN/Pages/MedischVerantwoordSporten.aspx

kk Art. 18 § 3 HES Decree

or to a particular sports discipline could be used.[ll] Although the primary intention of the Decree is to give some guidance to sports organisations how to fill in their obligations related to the promotion of healthy sports participation[mm], quality standards can be recognised or imposed (art. 6, 4th section in general and art. 9, 3rd section for eligibility tests). In the preparatory works of the Decree, it is explicitly stated that the omission to apply quality standard for eligibility tests does not intend to create any legal consequences for physicians (see for the legal implications also 4. Liability).[nn] As such, quality standards need to be understood as guidelines, being a supporting tool the physician in decision making. However, legal implications can arise for sports organisations if they do not act according to an imposed quality standard. In that case, a sports organisation could be prohibited to organise the sports activities. Athletes failing to submit the requested medical certificates, drafted according to an imposed quality standard, could be excluded from participation to sports activities. It should also be noted that the initiator of practice guidelines or recommendations regarding pre-participation screening needs to obtain the consent of the Flemish Minister of public Health, unless the screening falls into the field of application of the exceptions as mentioned earlier (see 3.1.2). Even though guidelines on pre-participation screening will be followed (or not) for an individual athlete by an individual physician, they can be considered as population screening as the target population aimed at in a guideline is larger than the individual athlete (for the definition of population screening see art. 2 Decree population screening).

According to the preparatory works, quality standards should be elaborated in collaboration with the sports sector or with the organisations for policy support and practice development mentioned in the Decree (art. 14). These organisations can be recognised and subventioned by the Flemish Minister competent for sports (art. 14 §1). Since 1 January 2015, one organisation is recognised in the domain of healthy sporting: the Flemish Association for Sports Medicine (Vlaamse Vereniging voor Sportgeneeskunde (VVS)).[oo] The Minister will conclude an agreement with this organisation to specify its tasks.

Since 2013, the Flemish sports- and medical screening physicians (Vlaamse Sport- en Keuringsartsen SKA) promotes a Flemish protocol for sports medical eligibility testing (Vlaamse Aanbeveling sportmedisch onderzoek - VASO) that was subventioned and supported by the Flemish government.[pp] More details on this protocol can be found in section 6.4.1.

5.3.1.7 Policy obligations of sports clubs and sports federations

In general, sports organisations have the obligation to implement the policy of the Flemish government related to healthy and ethical sporting and to the initiatives and measures taken by the government. This implies amongst others that the recognised sports federations need to provide anonymised and statistic information related to the sports specific risks and physical injuries and the evolution in time (art. 2, 2nd section, 1° Decree 4 April 2014, hereinafter called as “Executory Decree”). At least one physician needs to be involved in a sustainable way in the process of policy setting and elaboration of a sports organisation (art. 2, 2nd section, 2° Executory Decree). Special attention is given to the position of minor athletes. The Decree specifies that sports organisations need to take age, needs and capacities of minors need to be taken into account in their policy setting (art. 5 HES Decree).

In their policy, sports organisations have to make an explicit choice whether or not to impose or recommend a pre-participation screening for their members. (art. 2, 1° Executory Decree). Sports organisations are also deemed to have a general policy including an analysis of the specific risks related to the participation to the sports discipline and the measures and initiatives taken to prevent and come forward to those risks (art. 2,

ll *Parl. St.* Vlaams Parlement 2013-2014, 2280/001, p. 21

mm Comment to art. 6, *Parl. St.* Vlaams Parlement 2013-2014, 2280/001, p.19

nn Comment to art. 6, *Parl. St.* Vlaams Parlement 2013-2014, 2280/001, p.19

oo Ministerial Decree of 28 November 2014 related to the recognition and the subvention of the “Vlaamse Vereniging voor Sportgeneeskunde” as organisation for policy support and practice development in the domain of health sporting, not published yet at the moment of writing the report; http://www.vvsport.be/index.php?menu=Home

pp https://cjsm.be/gezondsporten/themas/testen-op-geschiktheid/vlaamse-aanbeveling-sportmedisch-onderzoek-0; https://cjsm.be/gezondsporten/sites/gezondsporten/files/public/vaso.pdf

3°Executory Decree). As such, it can be presumed that the outcome of the analysis of the risks related to the participation to the particular sports discipline serves as an input for their healthy sporting policy and is in line with the decision to impose or recommend a test. On demand of the government, sports organisations need to report on the initiatives and measures enabling the prevention and management of sports related factors that could have a negative impact on the physical or psychological health of the athlete. At least the regulations on the policy on healthy sporting, the policy and the information transfer to the members of the organisation and the types of sports medical eligibility tests that are recommended or obliged, the categories of athletes that are subject of the possible eligibility tests and the context in which those test are applicable need to be reported on (art. 3, 3° Executory Decree). This justification is to be perceived rather as information than as a tool for control.[qq]

5.3.1.8 Sanctions

Several sanctions for the violation of the HES Decree and its executory decrees, ranging from a request to act conform the legislation, administrative fines up to 10.000 Euro to an interdiction for 2 weeks to maximum 12 months to organize sports, are foreseen (art. 17 HES Decree).

5.3.1.9 Other initiatives for the prevention or management of sports related sudden cardiac arrest

The action plan 'gezond sporten' of the Flemish government foresees in and subsidies training on reanimation and defibrillation for 225 sports clubs. The training sessions are provided by the Rode Kruis Vlaanderen and comprise a 3 hour course in resuscitation. Sessions are accessible for all persons linked to a club (athletes, parents, supporters, referees, etc.) and cost 75 euro (after subvention). In contrast with the French Community, the Flemish Community does not oblige sports infrastructures to be equipped with a subventioned automated external defibrillators or sport clubs to organize sports in infrastructures with defibrillators (cfr. infra).

Key points

- **According to the Flemish legislation, sports clubs, federations or organizers of a sports event are free to recommend or to impose pre participation screening to athletes.**
- **Yet, quality standards regarding the content, the modus operandi or the required qualification to perform the test can be imposed or recognised by the government.**
- **The Flemish government considers the Flemish protocol for sports medical eligibility testing (VASO), drafted by SKA, as a quality standard. Domus Medica, the Flemish association of general practitioners, is developping a guideline on "healthy exercising", based on the VASO. It is intended to provide an evidence-based answer for the physician which tests are indicated for pre-participation screening of healthy athletes.**

5.3.1.10 Discussion

The intention of the HES Decree is to allow sports organisations to self-regulate healthy sporting within a supporting and facilitating quality framework provided by the government. Quality standards play an important role in this process, for the sports organisations (and indirectly the athletes concerned) as well as for physician responsible to carry out the pre-participation screening. Hence, it is of an utmost importance that quality standards are elaborated via a transparent and scientifically valid process and by an independent organisation.[rr] However, no conditions related to scientific validity for the recognition of quality standards are defined in the HES Decree. Even though it was explicitly mentioned in the preparatory works that physicians are not bound by the quality standards in their decision making, guidelines are supposed to be scientifically valid and sound tools, especially if they are recognised or even imposed by the government. Furthermore, sports organisations will (probably) base their decision to impose (or not) pre-participation screening on guidelines. The HES Decree foresees in that scope that sports organisations need to be explicit on the

[qq] Personal Communication representative Flemish Community Department of sports

[rr] Information on guideline development and validation can be found on the website of http://www.cebam.be/nl/richtlijnen/Paginas/Hoe-een-richtlijn-maken.aspx

choice for a pre-participation screening. However, the HES Decree does not foresee any formal check on the scientific validity of the guidelines the sports organisation based its decision on. Yet, in practice, there are initiatives to support only scientifically valid guidelines. Domus Medica, the Flemish association of general practitioners, is currently developing a guideline on "healthy exercising", based on the Flemish protocol for sports medical eligibility testing (VASO). It is intended to provide an evidence-based answer for the physician which tests are indicated for pre-participation screening of healthy athletes.

It also has to be noted that the HES Decree is a generic framework for all sports participation modalities. Guidelines and quality standards, as referred to in the Decree, will often have a less invasive impact on the athletes' physical integrity than practice related to pre-participation screening for cardiac anomalies. In that scope, the focus on a sound process of quality control as for clinical practice guidelines may not be relevant for all types of guidelines and quality standards referred to in the Decree.

5.3.2 Sports eligibility screening in the French Community

5.3.2.1 Scope of the Decree regarding the prevention of health risk in sports of the French Community

Pre-participation screening in the French Community is regulated by the Decree of 3 April 2014 regarding health risk prevention in sports. In Brussels, the Decree is applicable to institutions that are exclusively linked to the French Community because of their activities and their organisation (cfr. Supra). If the sports discipline is exclusively practiced in French and the language of the organisation and functioning (e.g. in the statutes) is French, an institution can be considered to be exclusively linked to the French Community.[ss] Due to a transfer of the exercise of the competence for pre-participation screening to the Walloon Region for the territory of the French Community (and to the French Community Commission for Brussels), the Decree will need to be amended.[tt] As such the implementation of the Decree is unclear at the moment of writing the report (December 2014).

The Decree intends to respond to the (mortal) sports incidents with the obligation to provide medical eligibility certificates for sports participation. Sports includes organised as well as non-organised physical activities aiming at the expression or the improvement of the physical or psychological condition or to obtain results in competition of all levels, except in the private, family or school environment (art. 1, 4°).

The Decree applies to

- sports organisations, defined as sports federations, recreational federations and sports associations (associations coordinating and organising multidisciplinary sports activities for a group of persons[uu]),
- organizers of sports activities, included sports events, demonstrations or shows. As such sports clubs that are not affiliated to a sports federation can still fall within the scope of the Decree if they organise sports activities or organise events.
- sports circles (cercles sportifs), being an association of sports members affiliated to a sports organisation

The overall idea of the Decree is to make athletes and sports organisations, organizers and circles accountable for healthy sporting. At the same time, the government has a supporting role in providing a general framework to enable the actors involved to fulfil their tasks. A medical eligibility certificate, for which the model and the content is predefined is imposed for particular categories of athletes defined by the government (see 3.2.2.1). The intention is not to generally impose a medical examination of contra indication, but rather to limit it to the categories to athletes with an increased risk to injuries or medical accidents.[vv]

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ss Advice nr. 52.412 of the Council of State of 7 January 2013

tt Art. 3 g Special Decree of 3 April 2014 regarding the transfer of the exercising of the competences of the French Community to the Walloon Region and the French Community Commission, *B.S./M.B.* 26 June 2014

uu Definition see art.1, 10° Decree of 8 December 2006 related to the organisation and subventioning of sports in the French Community, *B.S./M.B.* 20 February 2007

vv *Parl St.* Parlement de la Communauté Française 2013-2014, 617/001, p. 9

5.3.2.2 *Medical eligibility certificate is obligatory for specific categories of athletes*

Categories with medical certificate

A medical certificate confirming the absence of any contra indication for the particular sports discipline is required for the participation to sports for a list of categories of athletes (art. 11). The primary intention is to impose pre-participation screening for athletes participating to competition sports. Other categories primarily relate to specific risk characteristics of the athlete's health condition.

The following categories need to provide a medical certificate:

- Sports with specific risks, extreme risks or some combat sports
- Intensive or competitive sporting with a frequency higher than the frequency set by the government, based on the advice of the Commission for the prevention of risks for health in sports (hereinafter called as 'the Commission')
- Athletes with personal or family history
- Athletes participating in competitive sports, including amateur sports with start in group and ranking. In case of mass events, such as for instance running events, the athlete needs to provide the medical certificate to the sports organizer at the enrolment to the event.[ww]
- Athletes with history of medical problems directly related to sporting
- Persons starting to sport after a long period of sedentary life. In the preparatory works it was mentioned that one year should be considered as a long period and that in case of doubt the athlete is responsible to get examined.[xx]
- Any person that never practised sports
- Athletes older than the age limit, set by the government
- Athletes having suffered from a disease, as defined by government
- Any top athlete, a promising athlete or a training partner
- Elite athletes

The Decree explicitly foresees that sports organisations, organisers and federations are not allowed to let athletes participate to a sports activity if they did not priorly obtain the medical certificate (art .10).

Content and model of medical certificate

The government defines, following the advice of the Commission, the content and the model of the medical certificate. The certificate should consist of a general part and specific parts that are applicable depending on the age of the athlete, the level, medical history or the risks inherent to the sports discipline. It is valid for 12 months (art. 11). Medical certificates are delivered by a physician who carried out the clinical examination. No specific qualification is required for physicians delivering medical certificates. If additional medical examinations are necessary, the medical certificate can only be delivered after successful completion of the examinations (art. 12). The government can organise information and awareness campaigns for physicians related to the content of the medical certificates. Moreover, the government will elaborate a guidance manual to inform physicians on the examinations that need to be performed to check the absence of a contra indication for a sports activity, taken into account the possible specific risks (art. 4). The Commission will elaborate the content and the model of the medical certificate as well as the guidance manual for physicians and advice the government regarding the content of the medical examination(s) (cfr. infra).

Declaration on honour

For participation to sports in situations other than the categories mentioned above (cfr. 3.2.2.1), the absence of a contra indication to participate to sports needs to be confirmed in a declaration on honour signed by the athlete or the parents or guardian for minors (art. 13). As such, a 40 years old athlete in good health condition, with no personal or family medical history who plans to participate to non-competitive sports activities, with a frequency of 3 times 1 hour a week will not be submitted to a medical examination of contra indication.[yy] The model of the declaration on honour will precise the

[ww] *Parl St.* Parlement de la Communauté Française 2013-2014, 617/001, p. 9

[xx] *Parl St.* Parlement de la Communauté Française 2013-2014, 617/001, p. 10

[yy] *Parl St.* Parlement de la Communauté Française 2013-2014, 617/001, p. 9

situations where a medical certificate confirming the absence of contra indications is necessary and that in case of doubt, a medical examination is recommended. Sports participation in the private, family or school environment can be practiced without a medical certificate or declaration on honour.

For some sports disciplines with lower risk and for some organizers -to be listed by the government-, the respective sports organisation or the organizer can ask a derogation of the obligation to provide a medical certificate. The government can then, based on the advice of the Commission, allow that the lack of contra indication is confirmed by a declaration on honour. The decision on this derogation is valid for four years and is renewable (art 13, 4th section).

5.3.2.3 Policy obligations of the government

The government is charged with supporting roles in the management of healthy sporting, such as information and awareness campaigns regarding risk prevention and the promoting of healthy sports participation targeted at athletes, personnel, sports organisations, circles and organizer of sports events (art. 3). Furthermore, the government can also organize information- and awareness campaigns for physicians regarding the content of the medical certificates. Additionally, the government drafts, based on a proposal of the Commission, a manual informing physicians on the examinations that need to be carried out to control the absence of a contra indication for a particular sports activity (art . 4). The government also defines the model and the content of the medical code of conduct (art. 7 and 8) and the medical certificates and drafts an overview of the general recommendations and medical contra indications for sports that will be included in the medical code of conduct (art. 6). The preparatory and advisory work for these tasks is done by the Commission.

5.3.2.4 Obligations of sports organisations and organizers

Sports organisations and organizers have a general obligation regarding the **prevention of risks** (art. 5). They are hold to protect and promote health during sports participation. Thereto the organisational conditions as well as the medical and sanitary support should be defined. An annual report on the measures for prevention and awareness for risks needs to be submitted to the government by the sports organisations (art. 22). Sports organisations also have an **information** duty towards the circles on their obligations included in the Decree and its executory Decrees to guarantee that these are complied with by the athletes and the personnel. Additionally, organizers and circles have a direct awareness and information duty to member athletes regarding the possible risks linked to the sports practice and the duties included in the Decree.

Sports organisations need to elaborate a **medical code of conduct**. To facilitate this task, a model is defined by the government (art. 7).[zz] A medical code of conduct contains at least an overview of the general recommendations defined by the government, the medical contra indications for the particular sport and rules for the organisation of medically justified sports exercise. The latter include:

- the age categories, the sex and the linked conditions for sports participation
- the minimum information to athletes regarding the healthy sporting, the athletes' duties and the duties imposed to the sports circles, in particular regarding the support of young athletes
- the health rules that need to be respected by the personnel of sports organisations and sports circles
- a procedure for the management of risks in case of an accident
- rules on the training of personnel for the management of the risks in case of an accident

For sports involving particular or extreme risks (art. 7 § 3) or combat sports (art .7 § 3 and 4) particular elements need to be specified in the code of conduct. A list of sports involving particular or extreme risks and combat sports is to be drafted by the government (art. 14). In the preparatory documents, ice hockey, American Football and delta planing and sea diving are mentioned as sports involving a particular risk.[aaa]

Organizers of sports events, that are not simultaneously sports federations, are not systematically held to draft a medical code of conduct. Organizers of sports events involving particular risks, extreme risks or combat sports,

[zz] *Parl St.* Parlement de la Communauté Française 2013-2014, 617/001, p. 8

[aaa] *Parl St.* Parlement de la Communauté Française 2013-2014, 617/003, p. 4

however, need to draft a medical code of conduct as well. Apart from the requirement to include specific measures for the prevention and the protection of minor athletes, the content of the medical code of conduct is similar to the code of the sports organisations. The code of conduct is approved by the government, based on the advice of the Commission.

5.3.2.5 Commission for prevention of risks for health in the domain of sports

A central advisory role and tasks regarding the elaboration of the model and the content of the medical certificates (art. 25 § 1, 7° and 9°) and the content of the medical examination of contra indication (art. 25 § 1, 8°) is given to the Commission for prevention of risks for health in the domain of sports (for the entire list of tasks see art. 25). The Commission will also draft a manual containing information for physicians on the examinations that need to be performed to exclude contra indication for sports participation (art. 25, § 1, 3°). Furthermore the Commission is also charged with the definition of a model for the medical code of conduct that sports organisations need to apply (art 25 § 1, 5°).

The Commission will be composed of maximum 20 members for a renewable period of 5 years (art. 26). The composition of the Commission will be defined by the government and will consist of representatives of the scientific, medical and sports sector, with at least one member representing the Belgian Olympic and interfederal Committee, the High Council for health promotion, the High Council for Sports and the association of sports federations, the recreational sports federations and the sports associations recognised in the French Community. Two members representing respectively the Minister of Health and the Minister of Sports and 2 members representing the General Direction of Health and the General Direction Sports of the Ministry of the French Community have an advising vote (art. 26). Decisions and deliberations are only valid if at least half of the members is present. An absolute majority of the present members is needed to decide in a valid way. If the necessary quorum is not attained a new meeting is organised within 14 days following the first meeting. Regardless the number of present members, a decision can be taken during that meeting (art. 29). At the moment of writing the report, the Commission is not composed yet. Due to a transfer of the exercise of the competence for pre-participation screening to the Walloon Region for the territory of the French Community (and to the French Community Commission for Brussels), the composition of the Commission is delayed. As long as the Commission was not set up, the French speaking Commission for health promotion in sports fulfils the tasks of the Commission (art. 34). Until today, however, no decisions have been taken regarding the elaboration of the Decree.

5.3.2.6 Sanctions

Governmental officials will control the respect of the decree and the executory decrees (art. 21). The acts performed by physicians will not be controlled, however. Besides civil and penal sanctions, administrative sanctions up to 10.000 Euro can be imposed to the sports organisations and organisers if they do not respect the Decree and its executory Decrees (art. 23). Specific sanctions are foreseen for violations related to sports activities with extreme risks or combat sports. If recognised federations violate the Decree or the executory decrees, recognition or subventions can be withdrawn (art. 24).

5.3.2.7 Other initiatives for the prevention or management of sports related sudden cardiac arrests

Since 31 December 2013, sports infrastructures falling within the field of competence of the French Community need to be equipped with an automated external defibrillator.[bbb] Subventioned and recognised sports infrastructures are also deemed to organise yearly information- and training meetings for the users of the sports infrastructure. Sports clubs that are not practicing sports in infrastructures complying with this obligation are no longer eligible for facultative subventions granted by the French Community. Furthermore sports clubs also need to provide training and information on the use of defibrillators and they need to make sure that the club members and/or the organisation participate to the training.

[bbb] Decree of 25 October 2012 on the availability of automated external defibrillators of category 1 in the sports infrastructures, *B.S./M.B.* 5 December 2012

Key points

- **According to the legislation of the French Community, a medical certificate confirming the absence of any contra indication for the particular sports discipline is required for the participation to sports for a for athletes participating to competition sports and other categories primarily relating to specific risk characteristics of the athlete's health condition.**
- **The content and the model of the medical certificate is defined by the government, based on an advice of the Commission for prevention of risks for health in the domain of sports. At the moment of drafting this report, the Commission is not operational yet.**
- **For participation to sports in situations other that the categories mentioned defined in the Decree, the absence of a contra indication to participate to sports needs to be confirmed in a declaration on honour signed by the athlete or the parents or guardian for minors.**
- **Sports participation in the private, family or school environment can be practiced without a medical certificate or declaration on honour.**

5.3.2.8 Discussion

The Decree sets a general framework for healthy sporting. It is clear that executory decrees will need to be issued to implement most of the dispositions. For instance, the definition of the categories for which a medical certificate is required (art. 11) is very imprecise. It is not clear, for instance from which sports frequency or intensity on a medical certificate will be required. Most of the clarifications to the categories are foreseen to be defined by the Commission. As long as the Commission has not been set up, however, there is legal uncertainty for athletes participating to sports and sports organisations or organiser offering sports activities without requesting a medical certificate. Furthermore it is doubtful if the listing of the risk categories is evidence based. Sports in a private or family environment or scolary activities are excluded from the obligation to have a medical certificate, whereas these persons can be expected to be exposed to the same risks as the categories defined in art. 11.

The Commission is apparently the central body responsible for the preparatory work for the government. It is not clear, however, how the Commission, once it will be operational, will proceed in the decision making. Will they appeal to external, independent bodies to study which should be the appropriate content for a medical certificate? Will they systematically base their decisions on scientifically valid recommendations or will they rather rely on the Commission representatives' expert opinion? According to oral information of the President of the current Commission for health promotion in sports, there is an intention to rely on scientifically valid recommendations of independent instances. Moreover, it was stressed that there should be a proper balance between the benefit of imposed examinations and the drop out of athletes or the administrative burden of sports organisations. Furthermore, the idea is to support the good initiatives that already exist in several federations, rather than starting and redefining everything from scratch. Sports federations today already have elaborated a policy regarding health prevention in their respective sports discipline.

According to the 2001 Decree regarding health promotion in sports[ccc], sports federations are hold to draft a medical code of conduct containing at least general recommendations and contra-indications for the respective sports discipline and the frequency of a medical examination. The absence of contra-indications needs to be checked by medical examination. Furthermore, the medical code of conduct contains rules regarding the organisation of sports practice, including amongst others the conditions of sports exercise linked to age categories, minimum information to athletes regarding health related rules, etc. The government approves the code of medical conduct. In addition, according to the recognition criteria, sports federations are hold to take all necessary measures to safeguard safety of

[ccc] Art. 5 Decree 8 March 2001 regarding health promotion in sports in the French Community, *M.B./B.S.* 27 March 2001

their members and other persons participating to the organisation or related to the sports activities.[ddd]

As such, the definition of the content and the model of a certificate, imposed by the 2014 Decree, should be the output of a collaboration of the actors in the sports domain. This idea is also translated in the representation of the respective actors in the composition of the Commission. Anyhow, a clarification of the decision making process, for instance in the code of conduct (art. 30) that the Commission will need to present to the government would enhance transparency.

5.3.3 Organisation of Sports pre-participation eligibility screening for bi-community organisations in Brussels Capital Region

5.3.3.1 No obligatory medical pre-participation certificate

For sport clubs, organizers of sports events, organising sports in Brussels and that have not opted for either Community, the regulations issued by the Joint Community Commission apply. Unlike the Decree of the French Community, the Ordonnance on health promotion in sports participation, the interdiction of doping and its prevention does not impose a medical eligibility certificate for sports participation.[eee] Most of the responsibilities with regard to prevention and promotion of healthy sporting remain with the sports club and the organisations and the government. As such the respective government (Verenigd College) is able to determine the modalities of health prevention and medical support of athletes, to define the promotion of adapted healthy sporting, by rendering athletes conscious of their responsibilities and by informing the physician (art. 5). Moreover, a medical code of conduct can be imposed to sport clubs of a specific sports discipline, organisations of certain sports manifestations and the operators of some sports infrastructures. In such case, the government can set the minimum conditions regarding health promotion and preventive healthcare included in those codes (art. 6). Finally, the government can draft a list with general guidelines and medical contra indications for the particular sports discipline. Sports clubs need to take all necessary measures for sports participation, including the physical and psychological wellbeing of athletes. Until today, however, little execution is given to the Decree.

5.3.3.2 Discussion

Similar to the legislation of the Flemish Community, the Ordonnance opted for a policy where responsibilisation of the athlete and the sports organisations in combination with a supporting and facilitating policy of the government gains over an obligatory certificate. The divergent legislation applicable in Brussels renders the uniformisation of pre-participation screening difficult (cfr. supra for the application of the Decrees of the respective governments in Brussels).

5.3.4 Sports eligibility screening in the German speaking Community

The Decree of the German speaking Community regarding the prevention of sports related health injuries applies to sports federations and sports clubs located in the German speaking Community and to athletes participating or training to sports in the German speaking Community.[fff] Sports clubs and sports federations have a general duty to transfer to their affiliated athletes, sports related information provided by the government. Particular conditions, such as amongst others obligatory medical certificates, are set for cycling competition and combat sports (chapter III and IV of the Decree). Athletes participating to cycling competition need to undergo a yearly medical examination, including amongst others an ECG in rest at the age of 12 to 18 and an ECG during exercise at the age of 19.[ggg] For all other sports disciplines, the government can impose a minimum age and other safety conditions according to the particularities of the sports discipline. If the government decides to impose a medical certificate for sports participation, the content and the frequency need to be set (art. 40). To our information,

ddd Art. 15 Decree of 8 December 2006 regarding the organisation and subventionning of sports in the French Community, *B.S./M.B.* 2 February 2007

eee Ordonnance of 21 June 2012 regarding health promotion in sports practice, prohibition and prevention of doping, *B.S./M.B.* 5 July 2012

fff Decree of 30 January 2006 regarding the prevention of sports related health injuries, *B.S./M.B.* 12 April 2006

ggg Decree of 7 July 2000 regarding cyclism, *B.S./M.B.* 11 October 2000

the government did not impose any pre-participation screening for cardiac anomalies for any other sports disciplines than those mentioned above.

5.3.5 *Intermediary discussion on the role of the Community governments and the policy related to the recommendation of pre-participation screening*

Although the legal translation is different, the underlying ideas of the Decrees of the French and Flemish Community and the Brussels' Ordonnance go into the same direction. The common denominator is the reference to guidelines regarding the best practices in pre-participation screening and healthy sporting in general. In principle, these guidelines do not (or should not) differ according to the sporting population of one or another Community, which pleads for a uniform strategy between all Community governments.

Today, there are no such sound guidelines, rendering a policy for the recommendation of pre-participation screening for cardiac anomalies difficult (cfr. Results section 3). Based on the currently available data it is impossible to establish evidence (and accordingly, proven measures) in reducing cardiac death in athletes.[hhh] Registries of sudden death during sports participation are lacking and the best available data indicate that the total number of athletic deaths in young individuals is extremely small. Moreover, there are no studies on the long term impact on health of restricting sports for athletes with a cardiac anomaly. Yet, it should be noted, that disqualification of athletes is probably not the standard option following a cardiac anomaly. Reorientation or an adapted level of sports participation may have a similar positive impact as participation to the original sports discipline, if the athletes decides to continue sporting.

Even if sound data on sports related cardiac deaths were available, the discussion on the sense of a policy recommending or imposing pre-participation screening seems to be a debate that exceeds the domain of sports participation. There is a considerable controversy in the screening of athletes participating to sports and the absence of screening in non-athletes. Non-athletes making intense physical strains in daily life (for instance a daily sprint to catch the train), as well as people with un unhealthy life style, making little effort have a risk for sudden cardiac arrest. Should we screen the healthiest population, i.e. those who are willing and can participate to sports activities, whereas others are not? Is it ethical to selectively screen athletes for cardiovascular conditions, given the impact on personal daily life (stress, impact on ability for insurance contract, labour contracts, etc.) and society (cost of (unnecessary) diagnostic tests etc.)? Based on these reflections, the most rational strategy for Community governments is to make aware and to inform the population on the clinical and ethical issues related to pre-participation screening. Rather than imposing unproven measures to calm emotional reactions, focussing on individual responsibility for (informed) decision making in screening for cardiac anomalies for the general population seems to be a good option. Similarly, in view of good clinical practice and increased legal certainty, physicians should be properly informed. Initiatives, supported by all Community governments could give more weight to the message and enable a uniform policy for the Belgian sports cardiac screening domain.

5.4 Liability

5.4.1 *Physicians' liability*

Physicians' liability concerns can rise when the physician attested that the athlete was apt to sport whereas the athlete happens to die during or immediately after sports or suffers from a sports related injury. In general, the law requires physicians to use skills and care consistent with good medical practice in evaluating an athlete's fitness to participate in sports.[iii] This implies on the one hand that physicians use reasonable care in detecting foreseeable medical abnormalities that may cause sudden cardiac death or serious injury to athletes participating in organized competitive sports. On the other hand physicians should also avoid unnecessary exclusion from sports participation. Physicians who allow participation in competitive sports or wrongly exclude athletes from sports are not legally liable per se for an injury or death caused by an undiscovered cardiovascular

[hhh] Mark S. Link et. Al, Sudden Cardiac Death in the Athlete: Bridging the Gaps Between Evidence, Policy, and Practice, Circulation. 2012; 125: 2511-2516 http://circ.ahajournals.org/content/125/20/2511

[iii] T. Vansweevelt, *De civielrechtelijke aansprakelijkheid van de geneesheer en het ziekenhuis,* Maklu, Antwerpen - Bruylant, Brussel, 1997, 960 p.

abnormality.[jjj] Malpractice liability for failure to discover latent, asymptomatic cardiovascular disease requires proof that the physician deviated from customary or accepted medical practice in his or her specialty (fault) and furthermore that proper utilization of appropriate methods would likely have disclosed the underlying medical condition before injury or death occurred (causality). In that scope scientifically valid and up to date guidelines can play an important role in the decision making and hence also in the assessment of possible liability of the physician. Guidelines may constitute some evidence of the medical standard of care for pre-participation screening of athletes. Compliance with qualitative, valid guidelines may establish a presumption, be it rebuttable, that a physician has met the appropriate legal standard of care. On the other hand, the unjustified deviation of a qualitative guideline can be a constituting element of the proof of fault.[kkk] Until today, however, the legal value of guidelines in Belgian jurisprudence remains unclear. To our knowledge, there are no malpractice cases where physicians were condemned for having omitted to perform an ECG test for sports participation.

If civil liability of physicians is established, penal liability will also play since physical injuries will be catalogued as (unintentional) assault and battery (art. 418 Penal Code). Moreover, physicians delivering medical certificates stating that no contra-indications were observed to participate to the respective sports discipline, without having examined the athlete can be sanctioned for forgery. Finally a disciplinary sanctions can also be imposed by the Council of Physicians.

5.4.2 *Athletes' duty of care*

In the assessment of liability for sports related medical accidents, athletes' behaviour also plays a role. Athletes (whether or not minor) have a general duty to use reasonable care to protect their health and safety as part of the pre-participation screening process. Hence, athletes should be aware of their capacities and their limits. Athletes' responsibility to assess their own physical condition and capacities was also stressed in the Decrees of the Flemish and French Community (cfr. supra). As such, athletes should refrain from exercises or competitions that are too demanding or for which they did not sufficiently train.[lll] Moreover, athletes are required to be truthful in providing their medical history, with accurate responses to the questions regarding personal and family history and any other material information that may be pertinent to their health. Omitting to collaborate or to behave as a normal careful athlete-patient, by withholding information or participation to intensive competition without any training for instance, can be qualified as a proper fault hampering qualitative performance of the physician. This can have an impact, if compensation for malpractice by a physician or by a sports club is claimed by the patient.

5.4.3 *Liability of sport clubs, sports organizations or their personnel*

Occasionally, sports injuries are the result of the sports organization's or persons' affiliated to the sports organisations (coaches, sports teachers, volunteers …) failure to take reasonable care for an athlete. Their duty of care can be described as the duty to take reasonable steps to avoid foreseeable harm for the athlete. This can consist of organizational measures guaranteeing safety of the athlete, specific measures for the prevention of harm such as a training scheme adapted to the athlete, advice on life style issues, diets etc. Several elements representing the general duty of care for sports organisations and s have already been integrated in the Decrees of the Communities (cfr. Supra). For instance, the Flemish Decree stipulates that sports organization need to take initiatives and measures related to the prevention and management of sports related factors that could have a negative impact on the physical or psychological health of the athlete.

[jjj] N.M. Panhuyzen-Goedkoop and J.L.R.M. Smeets, Legal responsibilities of physicians when making participation decisions in athletes with cardiac disorders: do guidelines provide a solid legal footing?, Br. J. Sports Med. 2014. 48:1193-1195

[kkk] Vinck I., Paulus D., Van Brabandt H., Ramaekers D. Medico—legale aspecten van klinische praktijkrichtlijnen. Brussel: Federaal Kenniscentrum voor de Gezondheidszorg (KCE) mei 2006. KCE Reports vol. 26A. Ref. D/2006/10.273/05.

[lll] L. Cornelis and F. Claeys, "Sport en aansprakelijkheid: een stand van zaken", *T.B.B.R.* 2003, p. 585

On top of the administrative sanctions that are already foreseen in the decrees, civil and penal liability could be established if the wrongful act of the respective club, organization or their personnel caused the injury or death of the athlete. It has to be noted that employees working for sports clubs or s of sports events can only be personally held civilly liable for intentional conduct, gross or customary negligence.[mmm] This limitation can be invoked against both the employer and the injured parties.

As mentioned earlier, athletes also have a duty of care, implying that they have to take all reasonable measures to prevent foreseeable injury. If it is established that the conduct of the participant was unreasonable or dangerous in the circumstances and contributed to the injuries, liability of organisations or their personnel can be alleged. As there is no sound evidence that cardiovascular pre-participation screening is beneficial, omitting to ask for a test or to refusing to participate in screening can probably not be seen as a failure of the athlete's duty of care.

Another defence that can be argued by sports organisations and their personnel is the voluntary assumption of risk implying that the participant voluntarily assumed the risk of injury.[nnn]

Key points

- **Physician's liability for failure to discover latent, asymptomatic cardiovascular disease by omitting pre participation screening is not probable since:**
  - **today there are no scientifically valid and sound guidelines proving that cardiovascular pre-participation screening is beneficial in terms of the prevention of SCD.**
  - **There are considerable potential harms induced by screening.**
- **Athletes also have a duty of care, implying that they have to take all reasonable measures to prevent foreseeable injury. As there is no sound evidence that cardiovascular pre- participation screening is benefical, ommitting to aks for a test can probably not be seen as failure of the athlete's the duty of care.**
- **Sports organisations and their affiliated personnel (coaches, sports teachers, …) need to take reasonable care for an athlete. Their duty of care can be described as the duty to take reasonable steps to avoid foreseeable harm for the athlete. Since there is a lack of evidence that cardiovascular pre-participation screening is beneficial, the ommission to impose screening to their affiliated athletes cannot be seen as a lack of duty of care.**

[mmm] Art. 18 Labour Contract Act of 3 July 1978*, B.S./M.B*. 22 August 2002

[nnn] H. Vandenberghe, H.," De aanvaarding van het risico", *TPR* 2010, afl. 4, 2099-2105

5.5 Patients' Rights and data protection

The position of athletes undergoing pre-participation screening for sports participation is different from the classical patient-physician relationship, since there are (mostly) no symptoms and health care is sometimes not primarily provided on the athlete's request but induced by the sports club. Athletes may also request advice or cardiac examination on their own initiative. The rights and duties of physicians and patients are regulated in Patients' Rights Act of 22 August 2002.[ooo] Patient is defined as 'the natural person to whom health care services are provided, whether at his request or not' (Art. 2, 1°). This implies that patients are not necessarily ill or that health care does not necessarily need to be provided on the patient's initiative. Health services also include health promotion (art. 2,2°). Accordingly, the explanatory memorandum of the Patients' Rights Act specifies that medical eligibility screening in a work situation or for insurance purposes is considered to fall in the scope of application of the Patients' Rights Act.[ppp] It can thus be argued that athletes undergoing medical examinations on the request of a sports club are also considered as patients as referred to in the patients' rights act. In the following section the patients' rights that are the most relevant for the topic will be discussed.

5.5.1 The right (not) to be informed

The patient has the right to receive from the health professional all relevant information necessary to assess his state of health and his prognosis (art. 7). It is question of all the relevant information that is necessary for gaining some idea of the patient's state of health and its likely progression. This may also concern information not yet available, which can be brought to light by the diagnostic tests following a positive result of screening. Communication with the patient must take place in clear language, which means that the method of providing information is adapted to each individual patient. The patient may request that the information be confirmed in writing. The ECG test and medical tests following a deviant ECG can reveal more medical data than strictly necessary for the sports participation assessment. There might for instance be a genetic deficiency, in which case the information is no longer solely related to the athlete him- or herself but also potentially to the relatives. Hence it is important that the patient knows which information will possibly come out of a test (cfr. also 6.2). Information is not provided to the patient if the latter explicitly requests not to know. If the patient exercises this right, the health care professional may not inform the patient: the duty to inform becomes a duty not to inform. The explicit request not to know can be given in writing, in which case it is annexed to the patient's medical record, or orally, in which case it is noted in the medical record.

5.5.2 The right to informed consent

Patients have the right to consent well informed, freely and in advance to any service provided by a health professional (art. 8). Consent must be given explicitly except when the health care professional, after having informed the patient adequately, can reasonably deduce consent from the patient's behaviour. Consent not given explicitly is also referred to as implicit, tacit or non-verbal consent. The consent shall be recorded and added to the patient's medical record at the patient's or health care professional's request and with the health professional's or patient's approval.

The information supplied to patients for the purpose of giving the consent relates to the objective and nature of the medical service, to the degree of urgency, the duration, the frequency, the patient specific contraindications, side-effects and risks involved in the service, and to the post-care, the possible alternatives and the financial consequences. In addition, this information relates to any other clarifications that the patient or health professional deems fit to make, including, if necessary, the legal provisions to be complied with in relation to a medical service. Additional Information should be considered for screening that is offered to a healthy person and not directly on his/her request. For ECG, It is of an utmost importance that the risk for false positive results and the related consequences are explained. Furthermore, athletes also need to be informed that a test can be false negative and is never a guarantee for a long term perfect health

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ooo Patients' Rights Act of 22 August 2002, *B.S./M.B.* 26 September 2002; For an extensive overview of patients' rights see T. Vansweevelt, "Definities en toepassingsgebied van de Wet Patiëntenrechten" *T.Gez*. 2003-04, afl. 2, 66-73; S. Brillon, S. Callens, V., Gauche, N. Noël, G. Schamps, M. Verhaegen, *Mémento des droits du patient et de la responsabilité médicale. La loi du 22 août 2002*, Brussel, Kluwer, 2003, 221 p.

ppp Explanatory memorandum Patients' Rights Act, *Parl. St*. Kamer 2002-2003, 1642/001, p. 16

condition.[qqq] Moreover, the physician should discuss with the patient the consequences of a positive test. These consequences may not only have an impact to sports participation but also to labour participation, the aptitude to contract insurance contracts, stress on daily life, etc. Patients always have the right to refuse consent or to withdraw from the right to be informed on the results (cfr. supra). In such case, the health care professional needs to inform the patient on the (medical) consequences of the refusal.

5.5.3 Right to free choice of the health care provider

Patients have the right to freely choose a health care professional and to change this choice, except for restrictions in cases determined by law (art. 6 Patients' Rights Act). Apart from the legal exceptions (e.g. Labour legislation), factual situations (e.g. patients in a coma) or organizational issues (e.g. there is only a limited number of surgeons in a hospital) can also restrict the right to choose freely.[rrr] Although several sports organizations have their own medical staff, athletes cannot be forced to be screened for cardiac anomalies or to undergo medical treatment or assistance by this staff.

5.5.4 Minors and incapacitated adults

The patients' rights act contains rules to protect the rights of patients who are legally or factually not capable of exercising their rights. For minor patients, patients' rights are exercised by the parents asserting authority over the minor or by the patient's guardians (art. 12 Patients' Rights Act). The minor patient will be involved in exercising his rights, bearing in mind his age and level of maturity. Minor patients who are deemed capable of reasonably grasping their situation may exercise their rights on their own behalf. As such information on the risks related to sports, content of pre-participation screening and the consequences of the results should be discussed with reasonable minors. Nowhere is it explicitly stated who is to judge whether the minor patient can be deemed capable of reasonably grasping the situation, but the most obvious course of action would be to leave it up to the health professional. This might be important in cases of minors acting under pressure of their parents or a trainer. As the physician may only act provided he has obtained valid consent, it is up to him to decide whether the conditions for a valid consent are present. The rights of adult patients who have the legal status of 'extended minority' or have been declared incompetent are exercised by their parents or guardians. The rights of other categories of adult patients who are not capable of exercising their rights (e.g. demented persons) are exercised by the person previously designated by the respective patient (art .14 Patients' Rights Act). This designated representative will act on the patient's behalf when and for as long as they are unable to exercise these rights themselves. If there is no patient-designated representative or if he fails to act, the rights of the incapable adult patient can be exercised by the cohabiting spouse, the legally cohabiting partner or the actual cohabiting partner. If this person refuses or if there is no such person, the rights can be asserted, in descending order, by an adult child, a parent or an adult brother or sister of the patient. If these persons refuse or if there are no such persons, the health professional concerned has to take care of the patient's interests, possibly after multidisciplinary consultation. This is also the case when there is a conflict between two or more representatives of equal rank, for instance a conflict between two children of the patient. An adult, incapacitated patient has to be involved as much as possible and depending on his comprehension, in the exercise of his rights.

5.5.5 Patient's duty of collaboration

According to the patients' rights act, patients have the duty to collaborate to allow physicians to their do their work appropriately (art. 4). This implies a.o. that patients should inform the physician as correct and as complete as possible on their medical history or medical family history. Omitting to collaborate as a normal careful patient, by withholding information for instance, can be qualified as a proper fault hampering qualitative performance of the physician. This can have an impact, if compensation for malpractice by a physician is claimed by the patient.

[qqq] Per analogiam for population screening see S.Callens and J. Ter Heerdt, Naar een normering voor het bevolkingsonderzoek, *T, Gez*. 2001-2002, 5-15

[rrr] P. Schoukens en F. Dewallens, "De vrije keuze van beoefenaar [van de geneeskunst", *T.Gez.* 2003-04, afl. 2, 151-159.

5.5.6 Health data protection and privacy

Several questions can rise when medical data of athletes are processed (e.g. storage, gathering, transfer to sports organisation, use for statistics etc.). Sports eligibility tests are comparable to medical eligibility examinations prior to insurance or labour contracts, where the same atypical triangular relation between patient, physician and third party (employer, insurer, sports club/federation) requires a specific approach to protect the patient's privacy. Labour physicians for instance are not allowed to transfer any medical data to the employer; solely the aptitude to the particular job is mentioned.[sss] As medical results remain with the physician, they need to be part of the patient file for which clear regulations (a.o. storage etc.) are available (cfr. infra). Furthermore predictive genetic testing is excluded in the scope of labour aptitude. Similarly, a description of a patient's current health condition can be transferred to the advising physician of an insurer. Predictive genetic data may not be transferred (art. 19 Patients' Rights Act modifying art. 95 of the Law on land insurance).

Unlike the medical eligibility examinations prior to insurance or labour contracts, several particular safeguards are lacking for pre-participation screening.[ttt] Today, there is great variety in the extent and the type of medical data that are asked for by sports clubs. Although most of the Community legislation (cfr. supra) mentions that the pre- participation screening needs to focus on the risks related to the sports participation, there is no specification of the type and the content of medical information (yet) and there is no explicit ban of genetic information. Furthermore, it is not defined which part of the medical information should be transferred to the club (all or just some results, the notion of aptitude or not), who should store the medical information and for how long. This can be problematic when medical data are transferred to sports secretariats where data are sometimes treated by non-medical personnel of volunteers. A definition and standardisation of the medical examinations that are necessary to assess the eligibility per specific sports discipline, as foreseen in the Decrees of the Flemish and the French Community could be a first step to come forward to this.

Yet, the absence of specific legislation does not imply that there is a legal vacuum. The basic data protection principles require that data are processed fairly and lawfully, legitimately, proportionately, accurately and up-to-date and for a limited duration (art. 4 Act on the on the protection of privacy in relation to the processing of personal data, hereinafter called as the Data Protection Act (DPA) and its executory Decree of 13 February 2001, hereinafter called as executory decree DPA).[uuu] Sensitive data such as health data have a special status and are subject to a more rigid regime. In principle, the processing of health-related personal data is prohibited (art. 7 DPA). Several exceptions to this principle are provided in law. If the data subject has given his written consent to the processing of those data, on the understanding that the consent may be withdrawn by the data subject at any time, medical data can be processed. Other legitimization bases for the processing of health data are summed up in the Data Protection Act. The processing of health data is allowed when the processing is necessary to protect the vital interest of the data subject or of another person if the data subject was not physically or legally capable of consenting (art. 7, § 2, f DPA) or for the management of health care services and if the data are processed under the supervision of a health care professional. Although one could argue that these options apply to eligibility sports screening there are

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sss art. 3 Law of 28 January 2003 related to medical examinations carried out in the scope of labour relations, *B.S./M.B.* 9 April 2003 F. Hendrickx, *Privacy en Arbeidsrecht*,Brugge, Die Keure, 1999, 358 p.

ttt H. Nys and U. PyPoPs, "Private verzekeringen en medische gegevens" in H. Nys et al., *Medische keuringen bij private verzekeringen juridisch "doorgelicht*", Brussel, Koninklijke Vlaamse Academie van België voor Wetenschappen en Kunsten, 2008, 19-40; H.Nys, "Overmaken van medische gegevens in verband met levensverzekeringen" in L. wostyn, K. Boucquey en F. Schockaert (eds.), *Overhandigen medische gegevens*, Gent, Academia Press, 2009, tweede geheel herziene druk, 171; L. wostyn, "Overmaken van medische gegevens aan hospitalisatieverzekeringen en reisannulatieverzekeringen" in L. wostyn, K. Boucquey en F. Schockaert (eds.), *Overhandigen medische gegevens*, Gent, Academia Press, 2009, tweede geheel herziene druk, 165; I.Plets, "Medische onderzoeken op het werk", *N.J.W.* 2003, 618-621

uuu Act of 8 December 1992 on the protection of privacy in relation to the processing of personal data, *B.S./M.B.* 18 March 1993 and Royal Decree of 13 February 2001 executing the Act of 8 December 1992 on the protection of privacy in relation to the processing of personal data, *B.S./M.B*. 13 March 2001

different caveats. Firstly, the legitimization bases are exceptions to the prohibition of processing of health data that have to be interpreted very narrowly. Moreover as patients will need to consent to the test as a medical intervention (cfr. infra), it seems obvious that they simultaneously consent to the data processing as an "all-in package".

According to the proportionality and legitimacy principles the kind and the amount of medical data that is processed needs to be specific for the particular well defined purpose, i.e. the aptitude of the athlete for participation to the particular sports and proportionate to evaluate the eligibility of the athlete. As only the aptitude of the athlete and not the detailed information is necessary information to decide on sports participation, one can argue that a certificate should only mention the positive or negative decision of the physician. Health-related personal data can only be processed under the responsibility of a health professional, except for the written consent of the data subject or if the processing is necessary for the prevention of a concrete danger or for the suppression of a specific criminal offence (art. 7 § 4 DPA). As storage of health data is also considered to be processing, medical certificates containing medical data transferred to the sport organisations or organizer should thus be stored under the responsibility of a health care professional, unless consent of the athlete. The fact that pre-participation screening needs to be performed by a physician and that health data can only be processed under the responsibility of a health care professional not only guarantees expertise but also creates an extra privacy protection of the athlete, as medical secrecy regulations apply (art. 458 Penal Code). The physicians' task is to decide whether the athlete is apt to participate to the particular sports. In that sense medical secrecy of the physician is linked to this specific task. One could thus argue that the particular task of the physician makes that he/she can notify to the sports organisation whether the athlete is eligible or not. All other medical findings fall within the scope of medical secrecy.

For pre-participation screening that can be considered as population screening, the respect of relevant dispositions of the privacy legislation related to the processing of medical data of athletes is included in the assessment prior to the consent of the Flemish Minister of Public Health.

In the governmental programme of the Flemish government, the elaboration of a database containing anonymised data on sports (medical) injuries of athletes, in collaboration with general practitioners and sports physicians is foreseen. This should serve as an input to define sports medical eligibility of athletes. In the long term this could be the basis for a personal sports passport. Such further processing of medical data is only possible if complied with several guarantees for privacy protection, as foreseen in the Data Protection Act and its 2001 Executory Decree (art. 13 and following).

Key points

The Patients' Rights Act applies to athletes involved in the pre-participation screening process.

- **Right to information and informed consent:**
  - **It is of an utmost importance that athletes are informed on the risk for false positive results and false negative results and the related consequences. Moreover, the physician should discuss with the patient the consequences of a positive test and other (genetic) information that may be revealed. These consequences may not only have an impact to sports participation but also to labour participation, the aptitude to contract insurance contracts, stress on daily life, etc.**
- **Freedom to choose the health care provider**

Athletes cannot be forced to be screened for cardiac anomalies or to undergo medical treatment or assistance by the (medical) staff of the sports organisation.

- **Minor's patients' rights are exercised by the parents asserting authority over the minor or by the patient's guardians. The minor patient will be involved in exercising his rights, bearing in mind his age and level of maturity. Minor patients who are deemed capable of reasonably grasping their situation may exercise their rights on their own behalf.**
- **Health data related to pre-participation screening fall within the scope of the Data Protection Act and need to be processed fairly and lawfully, legitimately, proportionately, accurately and updated and for a limited duration. Solely the aptitude of athlete for the particicular sports discipline can be legally communicated to the sports organisation.**

5.6 Pre-participation screening in a selection of European countries

5.6.1 France

In France, participation to competition sports or sports manifestations requires a medical certificate of absence of contra-indication.[vvv] Medical examination takes place at least annually. The content of the medical certificate is detailed for professional athletes and for athletes selected to be enrolled to the lists of high level lists (listes de haut niveau).[www] For professional athletes the content and the frequency are defined according to the sports discipline by the medical Commission of the Federation of the respective discipline. Athletes selected to be enrolled to the high level list need to undergo at least one medical examination each semester performed by a sports physician, an annual ECG in rest, a transthoracic ECG in rest, once in the sportive career (and repeated if the athlete is younger than 15 in the year of the first examination) and a maximal exercise test every 4 years during the sportive career.[xxx]

For all other athletes, the content of the medical certificate can be freely defined by the sports organization. It can be delivered by any physician. A standard questionnaire and the content of a physical examination is published by the Société Française de Médecine du Sport (SFMS).[yyy]

In 2014, the necessity of an annual pre-participation examination was subject of a discussion, initiated by the former Minister of Sports.[zzz] The idea was to increase personal responsibility of athletes and to simplify access to sports. Several alternatives for the annual medical examination were proposed:

- Medical examinations at predefined moments: first license for competition ou (re) starting to sport after the age of 35
- Making athletes fill out an annual auto-questionnaire; a positive response to one of the questions would imply medical examination.
- Taking benefit of the medical school examination to guarantee that children are supervised

Until today, however, the requirement for an annual medical examination for athletes in competition was maintained.

5.6.2 Italy

Italian law both mandates cardiovascular screening and holds physicians criminally negligent for improperly clearing an athlete with an undetected cardiovascular abnormality that ultimately leads to death during sports.[aaaa] Every citizen participating in official competitive sports activities must pass an annual screening protocol including at least a general physical examination, 12 lead scalar electrocardiogram and submaximal exercise test. If there is reasonable clinical suspicion of cardiovascular disease, additional testing may be requested. Although in principle any physician can perform the pre-participation examination, sports medicine in Italy is an accredited discipline. The training program includes postgraduate full time training for 4 years.

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vvv L. 231-2 à L. 231-2-2 Code du Sport, http://www.legifrance.gouv.fr/affichCode.do?idSectionTA=LEGISCTA000006167042&cidTexte=LEGITEXT000006071318&dateTexte=vig

www http://www.sports.gouv.fr/pratiques-sportives/sport-performance/Sport-de-haut-niveau/article/Le-sport-de-haut-niveau-c-est-quoi

xxx L'arrêté ministériel du 11 février 2004,J.O. n°41 du 18 février 2004 page 3275 texte n°46

yyy http://www.sfmes.org/sfmes/textes-utiles

zzz http://www.irbms.com/certificat-medical-obligatoire ; http://www.europe1.fr/france/sport-vers-la-fin-du-certificat-medical-annuel-1727213

aaaa Decree of 18 Februari 1982 of the Italian Ministry of Health regarding rules concerning the medical protection of athletic activities, Gazetta Ufficiale 5 March 1982. *Colucci M. Part I: organization of sport (Italy). §2, IV (Sports Doctors). In: Hendrickx F, ed. International Encyclopaedia of Laws: Sports Law. New York, NY: Aspen Publishers;* 2004*: 29–31*

5.6.3 The Netherlands

In the Netherlands, pre-participation screening in general (sportmedische keuring) was obligatory till 1983. Since 1984, risk assessment of sports participation is considered to be part of the athlete's individual responsibility. About 50 certified Sports medical advice centers perform about 30.000 pre-participation test per year at the athlete's request.[bbbb] In 2006 the working group Cardiovascular screening and sports advised to carry out cardiac screening in competitive (top) athletes aged between 12 and 35, based on the Lausanne Protocol. The Hoge Gezondheidsraad, however, gave a negative advice for a countrywide cardiac screening of competitive athletes.[cccc] Until today countrywide cardiac screening of competitive athletes is not implemented in the Netherlands.

A Dutch website SPORTCOR was created by the Association Sports Medicine and the Dutch Association for Cardiology to inform the population on sudden cardiac arrest in athletes.[dddd] The website also aims at registration of cardiac events in athletes. Everyone (parent, general practitioner, relative,…) having experienced death or successfully reanimated cardiac arrest of an athlete can notify this event via a secured line. The submitted data are controlled via medical files of autopsy reports.

bbbb http://www.henw.org/archief/volledig/id4351-de-sportkeuring-nuttig-of-noodzakelijk.html

cccc Jaarbericht bevolkingsonderzoek Gezondheidsraad Den Haag: Gezondheidsraad, 2006.

dddd https://www.juliuscenter.com/sportcor/nl-nl/informatie/inleiding/inleiding

6 CURRENT PRACTICES IN THE BELGIAN SPORTS WORLD

6.1 Introduction

Recently both the Flemish and the French-speaking Community set up new decrees and are elaborating these new regulations (see chapter 4). Meanwhile sports federations find themselves in an intermediate stage as the execution of the Decrees is not completed yet, in particular in the French Community. . This section aims to give an overview of the practices in the sports federations at the time of writing the report (December 2014), existing standards and guidelines, which health care providers are involved, if any reimbursement is available, role of the insurance companies and analysis of Belgian data.

The scope of this section is the pre-participation screening in general in young athletes (14-35y). This screening program can also include other tests, such as orthopaedic examinations.

6.2 Methods

For feasibility reasons, the section on current practice in the sports federations is limited to the most popular sports in Belgium. The selection of the sports is made based on a recent handbook on sports participation in Flanders[85] and contains following sports in random order: gymnastics, soccer, tennis, athleticism, volleyball, cycling, swimming, basketball, badminton, and dance. The data collection is restricted to the level of the licensed sports federations, no sport clubs were questioned.

In a first phase a standard email was sent to the 27 Flemish and French-speaking federations of the 10 most popular sports with a short presentation of the project and the following questions:

- Is a medical certificate compulsory? At which frequency does this pre-participation screening have to be performed?
- Are the medical tests specified?

- What are the consequences for the athlete if no medical certificate is not submitted?
- Is the current insurance extended with an additional insurance for sudden (cardiac) death?
- Are data on sudden cardiac deaths available?

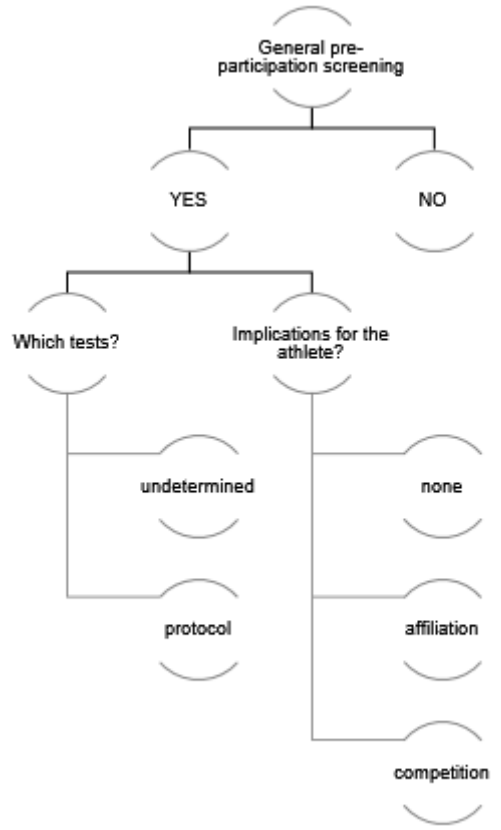
The majority of the sports federations gave already answers by e-mail to most questions. Nevertheless, they were all contacted by phone to validate the answers and to get more background information on the potential barriers and facilitators related to the implementation of a pre-participation screening program.

For the description of the other sections (current guidelines and standards, reimbursement, who is performing pre-participation screening, the role of the insurances and the analysis of Belgian data) a mixture of data sources was used: literature results on existing guidelines as described in chapter 3, detailed information on the VASO-protocol submitted by the representatives of SKA, information provided by the different stakeholders (representatives from sickness funds, insurance companies, sports federations etc) and grey literature (unpublished reports, websites etc).

6.3 Current practices in the Belgian sports federations

The results, collected by mail survey and phone interviews, are gathered in an extensive table. For clarity of the report, it was decided to move this table to the appendix. The main results are discussed in this section. Figure 8 gives an overview of the different scenarios of pre-participation screening mentioned by the sports federations. Two out of the 27 federations contacted did not answer to several mails and phone calls.

Figure 8 – Schematic overview of the current practices in the Belgian sports federations



6.3.1 Pre-participation screening program

Need for medical certificate: heterogeneous practice

The frequency of a medical certificate in the selection of Belgian sports federations who participated to the survey varied from no obligation (12 federations), only once at affiliation (5 federations) to once per year (5 federations) or more (3 federations).

Some sports federations differentiate between competitive and non-competitive athletes: the obligation for a medical certificate is sometimes linked to the competition license.

Only in one sports federation (Federatie Dans en Sport) it was mentioned that the sports clubs have the freedom to oblige the pre-participation screening to their members or not.

Uncertainty on the screening content

The majority of sports federations have not specified which tests should be performed in a pre-participation screening program. The physician has free choice of medical tests, which leads to a variety of approaches, and a lack of quality guarantee of these screening programs.

This can lead to a range of practices according to the physician, from only a short questioning of the family history to an extensive check-up by a specialist.

The requirement of the medical certificate which specifies the eligibility of the athlete for the specific sports activity, is merely seen as a (compulsory) administrative procedure. However, if the requirements are not fulfilled, this will have great implications for the athlete, notably no access to competition or even no affiliation to the sports club.

Some of the sports federations mention the online questionnaire of SKA (sportkeuring.be) on their website. One sport federation has chosen to follow the VASO-protocol (see paragraph 6.4.1) and encourages its members to submit their medical certificate via the electronic platform of SKA.

The Wielerbond Vlaanderen has already several years of experience in pre-participation screening and has implemented the most rigorous program: a yearly check-up with questionnaire, physical examination and rest ECG for competitive cyclists. At the age of 17-18y, an ECG during exercise is strongly recommended.

Within one sports domain, some differences can be noticed. Not only were there differences between the French and Dutch-speaking organisations, mainly due to the different regulations in each of the communities. Remarkable is also the difference between different federations within one sports domain in the same Community (e.g soccer, cycling).

Insurance

All sports federations, in both Belgian Communities, are obliged to have an insurance against sports injuries. The majority of sports federations (in 17 cases) have included in their insurance pack the additional insurance against sudden cardiac death (which in the contracts is erroneously labelled as “heart failure”.

More details on this additional insurance are explained in section 6.7.

Data on sudden cardiac deaths

The collection of data on sudden cardiac deaths was hampered by the lack of formal registration by the sports federations. In all sports federations no (or only a few) cases of sudden cardiac death were known. Often these persons were older than 35y. The data presented on the number of sudden cardiac deaths is not representative for the total number of sudden cardiac deaths during a sport activity, but aims to give a slight impression of the data collected by the sports federations.

6.3.2 Implementation of the new decrees

Current legal decrees stipulate that either the sports federations have to make a well-considered choice whether they will recommend or impose the pre-participation screening for their members or not (Flemish Community) or a medical certificate is required for competitive athletes and specific risk groups (French-speaking Community). The former legislation of the French Community already stipulated that sports federations were hold to draft a medical code of conduct with contra-indications for the respective sports discipline and the frequency of a medical examination checking the absence of these contra-indications. The 2014 Decree is more stringent and intends to define categories of athletes for which a medical certificate is obligatory and the content and the modalities of the certificates. However, the execution of the new decree has not yet taken place.

6.3.2.1 *Community initiatives*

In line with the newest decree of the Flemish Community, a decision tool for sports federations was developed by the Flemish Sports Federation (Vlaamse Sportfederatie) and the department of Culture, Youth, Sports and Media (Departement Cultuur, Jeugd, Sport en Media) (Checklist Gezond en Ethisch Sporten). This tool contains not only the legal requirements but also elements to facilitate the policy related to safe and ethical exercising. The checklist consists of following items:

- At least one physician should be involved in the elaboration of the sports federation-specific policy on safe exercising;
- Analysis (based on anonymous data registration of physicial injuries and/or literature) and prevention of sport specific risks with information towards the athltetes on the (prevention of) risks;
- pre-participation screening: choice between obligation and recommendation and for which specific groups of athletes. It is recommended to make this choice in collaboration with the involved physician or additional advice by SKA;
- Age limits: choice whether age limits are relevant for the specific sports federation;
- Information towards the members about the prevention of specific risks, the initiatives to facilitate safe exercising and the policy related to safe exercising;
- In contrast to previous decrees, the yearly reporting by the sports federations towards the Flemish community is replaced by a reporting on demand.

The implementation of the new decree of the French-speaking Community is less clear: each competitive athlete and a defined list of other athletes should yearly submit a medical certificate delivered by a physician, but the content and frequency of the pre-participation screening need to be determined by the Commission for prevention of risks for health in the domain of sports. As long as the members of the commission are not yet assigned, the implementation of the decree is on hold.

6.3.2.2 *Reported barriers for implementing a pre-participation screenings program*

The phone calls with the sports federations revealed that the major reason to have implemented a pre-participation screening program is due to the regulations as determined by decree rather than a medical need.

In some sports federations a pre-participation screening program was implemented but afterwards abandoned. Reasons mentioned for not (or not anymore) implementing a pre-participation screening program are:

- lack of quality control on the content of the screening program,
- the risk-analysis for sport injuries showed no added value of a pre-participation screening program,
- the related administrative burden and costs for implementation in sport federations with high number of members.

In line with these reasons, some sport federations were more convinced of the added value of resuscitation workshops and automatic external defibrillators (AED) rather than the implementation of a pre-participation screening program for all their members (competitive and non-competitive).

Main concerns related to a compulsory pre-participation screening program were:

- a potential barrier for the affiliation to the sports federation:
  - The additional costs of an annual pre-participation screening (which is not reimbursed by the NIHDI),
  - The additional efforts needed to perform these testswhich could demotivate potential members of a sports federation. Tthis concern was more pronounced in the sports federations in which the membership fee is already very low for the non-competitive athletes.
- the administrative burden related to the collection and follow-up of the medical certificates.

6.3.2.3 *Reported reasons for implementing a pre-participation screenings program*

On the contrary, some sports federations were in favour of a rigorous pre-participation screening of each of their members. However, they mentioned their lack of knowledge about the suitability of the medical tests and emphasized the need for scientific advice on this matter.

Each sports federation is obliged to set up a sport-specific analysis of the potential risks for injuries. However in practice this is rarely done yet. Some of the sports federations mentioned more sport injuries in the older population, not in the target population of this report. But this kind of citations are not based on data-analysis but rather on gut feeling.

The interviews with the Flemish sport federations showed that most of the respondents knew the SKA online screening tool. Some of them implemented or mentioned it on their website, whereas other sport federations mentioned the time-consuming aspect of performing the general protocol. Some federations also pointed out that any answer on the questionnaire ultimately led to an advice for a medical visit.

6.4 Guidelines and standards in Belgium

Currently there are no high-quality, validated guidelines on cardiovascular pre-participation screening in Belgium.

6.4.1 *VASO-protocol*

The Flemish sports and medical screening physicians (Vereniging voor Sport- en Keuringsartsen SKA) recommends a protocol for sports medical eligibility testing (Vlaamse Aanbeveling sportmedisch onderzoek - VASO) that was subventioned and supported by the Flemish government.

The VASO protocol prescribes that between the age of 14 and 34, athletes should perform a two yearly pre-participation screening to evaluate their ability to perform the sport they choose. This sports-medical examination is a global evaluation of the athlete: an internet-based questionnaire aiming at detection of multiple risk factors, including cardiac, pulmonary, neurological, orthopaedic, … is followed by a thorough physical examination (heart, lungs, blood pressure, orthopaedic examination, eyes, teeth,..) and a sports-specific functional orthopaedic testing battery (tests choice based on the most frequent injuries in that sport).

The VASO-protocol consists of the following components:

- History-taking (family and personal history)
- Clinical examination
- Rest ECG
- SCORE (Systematic Coronary Risk Evaluation in >35y): is out-of-scope of this report
- Functional orthopedic examination: is out-of-scope of this report
- Sport-specific tests: is out-of-scope of this report.

The specific cardiovascular recommendations are based on a non-systematic literature search completed with advice from a cardiologist panel (for those items where no definite guidelines were available). This panel consisted of adult and paediatric cardiologists, rhythmologists, interventional cardiologists and sports cardiologists.

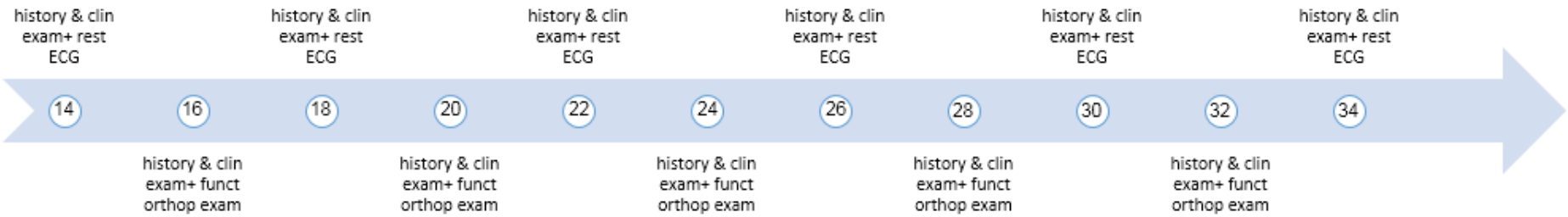
Following table gives an overview of the screenings algorithm (see Table 16) for all age groups. In this report we focus on the age group 14-34y.

Table 16 – screenings algorithm in VASO-protocol

Test	Age <14y	Age 14-34y	Age ≥35y
History and clinical examination	Every 2y	Every 2y	Every 2y
Rest ECG	/	At 14y + 18y Every 4y for very intensive sports	Once if not yet available from previous examinations
SCORE	/	/	Every 2y
ECG during exercise	/	/	After risk evaluation
Functional orthopedic examination	Every 2y	Every 4y	Every 4y

In practice this would mean that in the age group 14-35y, each athlete should have a medical check-up every 2years, resulting in a total of 11 medical visits containing 6 rest ECGs and 6 functional orthopedic examinations (see Figure 9) up to 35 years of age.

Figure 9 – Screenings algorithm in a timeline (14-34y) according to VASO



In order to avoid over-screening in a subpopulation of athletes who practice sports at very low intensity, the screenings algorithm takes also into account the intensity of the sports activities (based on MET-values). Three groups of intensity were defined: low intensity (<3 MET), moderate intensity (3-6 MET) and high intensity (>6 MET). Each group of intensity is further divided in subgroups based on the presence or absence of risk factors. Only in the low intensity group (<3 MET) with no risk factors, pre-participation screening is not recommended. In all other groups SKA recommends to perform a pre-participation screening with an alternating process of rest ECGs and functional orthopedic examinations.

The advantage of this VASO-protocol is the clear definition which tests at which frequency should be performed. However, so far this protocol has not been scientifically validated and cannot be regarded as a formal practice guideline.

The SKA was also involved in the report from the Belgian Superior Health Council,[7] on which the section on clinical effectiveness is mainly based.

6.4.2 *Development of new clinical guideline by Domus Medica*

Domus Medica, the Flemish association of general practitioners, is currently developing a guideline on “gezond sporten en bewegen bij mensen met een normal risico”. It is intended to provide an evidence-based answer for the physician which tests are indicated for pre-participation screening of healthy athletes. The final report of this guideline is foreseen for the end of 2016.

6.4.3 *Other initiatives*

We asked also for the point of view of the Belgian Olympic Interfederal Committee (BOIC) on the implementation of a pre-participation screening program. In line with the recommendations of the IOC (see section 3.5 on the Lausanne recommendations), they recommend a 2-yearly cardiovascular pre-participation screening program, consisting of history-taking, physical examination and rest ECG.

Key points

- **Currently no validated guideline on pre-participation screening is available.**
- **SKA developed, with financial support from the Flemish Community, a protocol for sports eligibility testing in which a rest ECG is recommend at the age of 14y and 18y.**
- **Domus Medica is developing a guideline on pre-participation screening.**

6.5 Reimbursement of pre-participation screening

Since a cardiovascular pre-participation screening program belongs to the domain of prevention, but may contain a diagnostic test (i.e. ECG), some misinterpretation can exist if reimbursement is foreseen and by whom. In this section we will discuss the stakeholders who might be responsible for any kind of reimbursement of cardiovascular pre-participation screening program.

6.5.1 Compulsory health insurance (NIHDI)

Currently, pre-participation screening is not reimbursed by the compulsory health insurance.[eeee] In practice, these tests are sometimes considered and reimbursed as a medical consultation.

Several test centres, physicians and sports federations provide information on the absence of reimbursement for the medical visit via their website or via a warning on the standard certificate (drawn up by some of the sports federations).

6.5.2 Community governments

The HES-decree from the Flemish Community states that support or financing for sports medical tests can be granted by the government for one or more sports categories according to the conditions foreseen by the government (art. 15, 2° Decree 20 December 2013). Currently, this article is the basis for the financing of the medical examination for talented athletes. Currently, there is no intention by the Flemish Community, however, to reimburse possible medical eligibility tests, such as cardiovascular pre-participation screening.[ffff] In the French-speaking Community there is no intention to reimburse cardiovascular pre-participation screening either.[gggg]

6.5.3 Sickness funds

One of the roles of the sickness funds is to provide information on a healthy lifestyle to their members. Each sickness funds has dedicated some space on their website on healthy moving and sport participation. Most sickness funds (CM, SM and OZ) refer to the website of SKA (sportkeuring.be).

Next to the provision of information, each sickness fund is, within the supplementary health insurance, free to offer a reimbursement for pre-participation screening.

Only two sickness funds offer a reimbursement to their members: the Flemish and Neutral Sickness Fund (Vlaams en Neutraal Ziekenfonds) (reimbursement of 30 euro every 2 years) and the Independent Sickness Fund (Onafhankelijk Ziekenfonds) (reimbursement of 30 euro every 4 years).

The independent sickness fund (Onafhankelijk Ziekenfonds) organises also free health checks (blood pressure, cholesterol, body composition) for their members, but no cardiovascular screening is included in the tests.

[eeee] L. Maroy, “Betaalt de ziekteverzekering een preventief sportgeneeskundig onderzoek terug?”, *Inf. RIZIV* 1994, 213-214

[ffff] Oral information representative Flemish Community Sports Departement www.cjsm.vlaanderen.be

[gggg] Oral information representative Flemish Community Sports Department

6.5.4 *Sports federations/clubs*

The sports federations and sports clubs are free to contribute to the costs related to pre-participation screening. In the interviews with the sports federations of the most popular sports in Belgium (see section 6.3), several options were mentioned, but the majority of the sports federations/clubs do not finance the pre-participation screening. Other (more rare) options which were mentioned, were: collaboration between sports club and a physician to perform the pre-participation screening at NIHDI-cost (without out-of-pocket payments); collaboration with the team physician; reimbursement of the medical visit; etc.

6.5.5 *Athlete*

If no reimbursement is foreseen by any of the above-mentioned stakeholders, the athlete will bear all costs.

Unfortunately the time resources of this project were too limited to set up a survey amongst the athletes to examine their perception and their experiences with pre-participation screening. This kind of research could be done by sports federation.

SKA advises a cost of €50 for a sports-medical examination comprising a medical questionnaire, clinical examination and sports-specific functional examination (taking about 40 minutes). The amount is €70 if a rest-ECG is added.

Key points

- **The Flemish Community nor the French Community governments have the intention to intervene in the costs of cardiac pre-participation screening.**
- **The athlete himself bears all costs related to pre-participation screening.**

6.6 Who is performing the pre-participation screening?

Healthcare providers

Both the Flemish and the French-speaking communities have regulated that only physicians have the competences to perform a pre-participation screening. In the newest decrees no specific competences are mentioned, therefore any physician is allowed to perform such kind of screening.

Due to the freedom of choice of the physician and the lack of quality control on the performance of the pre-participation screening, it could be estimated that this could lead to a variety in practices. However, the current lack of reliable data on who is performing the pre-participation screening, hampers to underpin this assumption.

In the phone interviews with the representatives of the sports federations a (non-exhaustive) variety of physician specialties were mentioned: GPs, team physicians, sports physicians, specialists (cardiologists, specialists in physical medicine and rehabilitation, etc), etc. The majority of the sports federations recommend their members to be tested by their general practitioner (GP), who is more familiar with the medical and family history of the athlete.

Currently SKA has set up a specific training for physicians (and in particular for GPs) in order to familiarize these physicians with their screening protocol (VASO-protocol) and with the use of its electronic platform. This kind of training facilitates the uniformity of practice, however the screening protocol is not yet scientifically validated (see section on current standards and guidelines). Also, after filling in the questionnaire on sportkeuring.be (which is financed by the Flemish community), the athlete is recommended to visit one of the listed physicians. However, until now this list is restricted to the list of licensed medical screenings physicians (keuringsartsen) and not open for every physician.

Training support centres

In contrast to the common equipment of a GP's office, athletes can also be tested in dedicated commercial laboratories. The medical examination centres (keuringscentra) (which statutes will be modified in 2015), are also accessible for non-professional athletes.

If an athlete would like to participate to a competitive or recreational sports event, a pre-participation screening could be required. Golazo, the main organizer of sports events in Belgium, demands for each of its sports events a proof of medical eligibility. Subsidiary company of Golazo is Energy Lab, a fully equipped training support centre for all kind of testing related to sports. Also a pre-participation screening is offered. This offer differ slightly from the VASO-protocol in frequency of testing and is dependent on the intensity of sports participation. In competitive athletes or in athletes who practice more than 6 hours per week, a 2-yearly pre-participation screening is recommended (in 14-35y athletes). This screening program consist of an anamnesis, clinical examination and a rest ECG. The functional orthopedic examination is not included. An ECG during exercise is only recommended in case of complaints or certain deviations on the rest ECG. In recreational athletes (14-35y) a similar protocol is recommended, but at a frequency of 4-yearly testing.

During the final expert meeting, representatives of SKA mentioned that in these commercial laboratories rarely a physician is present. Also they mentioned that the Belgian Cardiological League (Belgische Cardiologische Liga) has confirmed that by every performance test a physician should be present and these should be under monitoring of an EKG.

Key points

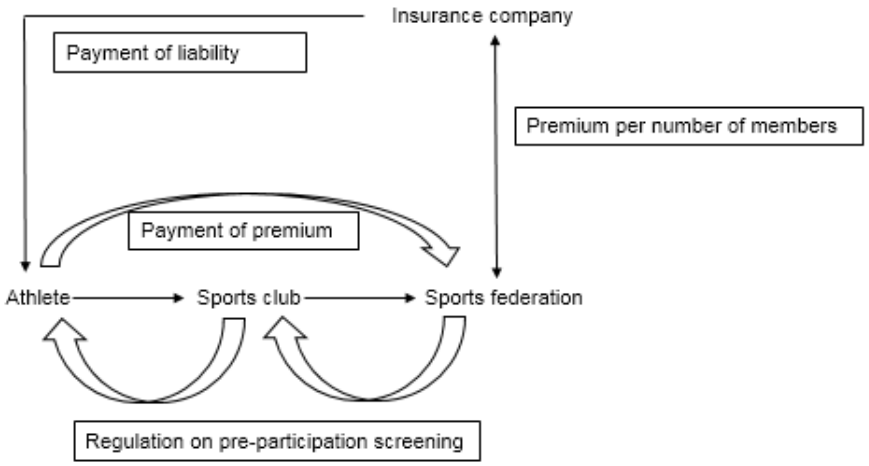
- **Only physicians are allowed to perform a pre-participation screening.**

6.7 Insurances

After consultation of Assuralia, three main insurance companies involved in the sport injuries were interviewed (Ethias, Arena, AG Insurance). However, due to the low number of insured sports federations, we decided to involve not further AG Insurance in this project.

In the following schematic Figure 10 the connections between the insurance company and the athlete are shown.

Figure 10 – Links between insurance company, the sports federation, the sports club and the athlete



Both Ethias and Arena offer an additional insurance against sudden death (designated as *heart failure* in the contracts) at a very low premium cost (≤1euro).

Since 2012, 8 insurance claims were submitted with 2 cases of SCD and 1 case of SCA with successful resuscitation. The other cases were mainly complaints about chest pain and cardiac arrhythmia. These data indicate the very low event rate of SCD in Belgium, but without solid numbers of the denominator (number of insured athletes), this assumption cannot be confirmed.

The insurance companies collect already some data on the number and kind of cardiac events, but a more systematic registration could facilitate data-analysis on the current Belgian situation.

Key points

- **A systematic registration of all sports injuries, including SCAs and SCDs, could facilitate the analysis of the current situation.**

6.8 Preliminary data in Belgium

The SKA provided a database with information on the medical examinations after filling in the questionnaire on the website sportkeuring.be. The current dataset contains only the number and kind of medical examinations, which were registered via the electronic platform. This is most probably an underestimation of the real number of medical examinations performed in 2013 and 2014.

In consensus with the representatives of SKA, it was decided not to present the preliminary data. In the current preliminary dataset, already some analyses could be performed on the number of persons who underwent a rest ECG (per age group) and which conclusions were linked to these rest ECGs. However, methodological aspects (e.g. the low number of participants per age group, bugs in the software etc) hampered a reliable interpretation. In the future more reliable and consistent data will become available.

6.9 Discussion

Within the framework of safe sports participation, policy makers stipulated several regulations with the intention to protect the athlete against potential harmful effects of exercising.

The clinical effectiveness of cardiovascular pre-participation (see chapter 3) will not be discussed in this section. This section describes the current situation on the field i.e. how the sports federations understand and implement the regulations and how these regulations are applicable (or not) to the athlete.

An athlete affiliated to a sports organization in Flanders or to a Flemish sports club in Brussels will, dependent on the choice that the sports federation made, be obliged or not to undergo a pre-participation screening program in order to become a member for the sports federation and/or to obtain his license for competition or sports event. In the French-speaking Community every athlete who participates in a competition either in a sports federation or during a sports event, will have to prove his eligibility to practice his sports activity. How, when and where he has to find this proof of eligibility is not yet decided. At that level the Flemish Community has already investigated in the quality assurance of this kind of screening by financing the sports physicians (SKA) to develop a protocol for sports eligibility testing. The validity of the VASO-protocol is questionable, but it defines clearly test modalities.

The scarcity of Belgian data on the incidence of SCDs during sports activities indicate that the policymakers took already some action to prevent sudden cardiac deaths during sports activities without any data on the current number of SCDs in Belgium (and abroad).

Currently, none of the respective competent governments of the French government have the intention to intervene in the cost of cardiovascular pre-participation screening. Yet, the non-reimbursement policy of an imposed (possibly repetitive) ECG test could therefore lead to a barrier for sports participation for specific populations. Furthermore this decision penalises a - in general - healthy population, who comply with recommended healthy lifestyle.

This section aimed to describe current situation. However, some methodological aspects hamper to draw solid conclusions. One of the main limitations is the lack of representative quantitative data on the current situation. Also due to time resources, the survey was not extended to a sample of athletes. The physicians involved in the expert meetings mentioned also the willingness to be tested by a certain amount of athletes. More in-depth research is needed to describe the perception of the athlete on pre-participation screening.

6.10 Conclusion

The current Belgian situation is characterized by a variety of practices. In the decrees from both Communities action has been taken to harmonize the different practices by either making the pre-participation screening compulsory for all competitive athletes and defined categories of athletes or by stimulating the sports federations to register all sport injuries and to analyse the sport-specific risks. However, both decrees are a framework and further elaboration is needed to implement these regulations.

Nowadays, the athlete can be obliged by the sports federation to undergo a pre-participation screening. No validated Belgian guidelines on cardiovascular pre-participation screening exist (yet) to inform the athlete and the physician for whom this test could be suitable, by whom it should be performed, which tests should be included and at which frequency the athlete should be tested.

7 ETHICAL CONSIDERATIONS - A TELEOLOGICAL APPROACH

Screening is intrinsically subject to ethical questions because it is a medical approach aimed at persons who, on the face of it, have no complaints concerning their health. And yet, the goal is to detect an element that increases the risk of disease or even death. Therefore, we are required to seriously consider whether the launch of such a programme is relevant since persons who consider themselves to be in good health will be faced with the fear of learning that their life may be changed by the discovery of a condition or a risk that they will have to take into account, whichever decision they take for the future. [86] If a person discovers that they have a disorder or that they run a particular risk of being affected by a certain condition, or even dying, this will inevitably modify their experience of health and their existence. Health in the neutral sense is a 'simple lack of somatic concern', where the 'body is completely forgotten'; in the positive sense, health is 'felt in the vigour of the body and in the happiness experienced in realising its strength'[hhhh]. This positive conception of health is particularly questioned when the screening concerns athletes 'in the prime of life'. Therefore, it comes as no surprise that in 1968, the World Health Organisation (WHO) already considered (1) that screening should relate to a major health problem[iiii]. Its document also specified (2) that the benefit of screening wasn't meant to be immediate, but that its purpose was to ensure a future health gain for those taking part in it. This international organisation also specified (3) that screening should be performed using reliable and valid methods. It also specified (4) that screening should respect the autonomy of individuals so that the programme would be accepted within the population. Finally, it insisted on (5) the 'cost-effective' nature of its results. This principle is well known. It forms the basis of the analyses that allow a recommendation to be given concerning the possible reimbursement of a drug, therapy, vaccination or screening. The fifth principle inevitably integrates the second one because the cost-utility analysis takes into account the known and quantifiable benefits of the care programme. It also integrates the third one because the evidence on which a cost-utility analysis is based depends on the quality assessment of the evidence used. The importance of the health problem leaves a certain amount of room for discussing, deliberating and taking into account both budgetary and ethical considerations. As regards autonomy, as we shall see, it should most certainly be taken into consideration in a context where the notion of empowerment is increasingly used to make the patient responsible for managing their health, illness, treatment and recovery.

7.1 Deontological or teleological ethics?

The cost-effectiveness analysis allows us to introduce a fundamental distinction between two types of ethical approach. One is qualified as 'deontological', and refers to duty and obligation. Assessing an attitude, a human action in the light of its compliance with a duty falls within the remit of deontological ethics. The most well-known representative of this way of conceiving ethics is probably Immanuel Kant, who left his mark on generations with his 'categorical imperative'. If, on the other hand, we only focus on the consequences of actions, we adopt an approach based on teleological ethics. The consequentialism that characterises these ethics can be found in the utilitarian doctrine. Utilitarianism can be defined as the approach comprised of the combination of the following three elementary principles[jjjj]: the theory or the economics of "well-being" according to which we assess a life situation in society in the light of the utilities the individuals composing this society derive from it; "consequentialism" according to which the judgement of acts and rules depends on the state of the things resulting from it, and the "sum-ranking" according to which the well-being of the members of a society is assessed according to the total sum of the utilities they derive from it. If we replace the concept of utility with that of QALY (quality-adjusted life-year), we arrive at the core of health technology assessments (HTA[kkkk]), the cost-utility analysis.

Even if deontological ethics may 'help us to live', discovering a path referred to as 'the good life', they can be difficult to integrate into the context of a world that is inevitably marked by material limits and the finiteness of

hhhh This is the double meaning of health given in the introduction to the recent book written by Jean-Claude Fondras [87], p.11.

iiii [2], quoted by [88] pp. 370-371.

jjjj See [89], pp.38 and 56 and [90], p.71.

kkkk HTA: Health Technology Assessment

existence. Some categorical imperatives can indeed enter into conflict with the clearly non-extensible nature of budgets. They can also make us forget that life is 'mortal', that death is inevitable despite all the means we may deploy to care for body and mind. Hence, the example of the imperative according to which '*we can't allow someone to die whose life we could save in a relative medium term*' could come into conflict with the concept of a limited budget. In concrete terms organising screening simply with the goal of satisfying the obligation to prevent a death, whatever the cost and whatever the possible harmful effects, rapidly contradicts the concept of the scarcity of available means. Besides the 'price', the sense of 'saving' a life inevitably takes on a different meaning depending on the person's state of health, their life expectancy and their quality of life. We could imagine another imperative that would consist of '*not informing a person that they run a greater risk of death than average if they don't have any symptoms*'. Contrary to the previous one, this 'obligation' would lie at the origin of a system that would exclude all forms of screening. A stance that would be difficult to accept in a world where medical technology allows us to anticipate more and more precisely the occurrence of certain health problems.

The difficulties illustrated by the two previous examples as well as the risk of imposing, on the whole of society, a particular concept of life and care through the choice of a particular imperative, leads us towards a teleological approach. But let us be clear, not adopting a deontological approach doesn't mean that we don't take into account humanistic values such as human dignity or respect for life. This means that we don't impose a duty or an obligation, however noble, on any system or society, simply in order to respect the different life options that everyone wishes to choose.

Even if a teleological approach in the field of healthcare suffers from certain aporias that characterise utilitarianism[llll], it would appear more fruitful to resort to consequentialist ethics in order to give a perspective on the effects of screening. It is precisely because of the consequentialism that characterises this approach that it is an ideal complement to the cost-utility analysis, which also reflects the 'consequences' in terms of the costs and benefits of a decision in the area of healthcare. The main interest of the teleological approach, unlike utilitarianism, is that it doesn't seek to maximise the sum of quantifiable values (utilities in the case of utilitarianism). In the following, it is therefore a question of presenting non-quantitative considerations which relativise (narrow down or reinforce) the results of the cost-utility analysis. To clarify, such an approach isn't definitive because it doesn't allow us to weight arguments and make the sum of these arguments that will remain qualitative. On the other hand, some could turn out to be determining factors when public decision-makers have to make choices. Therefore, the sole purpose of the following reflections is to contextualise, complete, relativise and enrich the conclusions that can be drawn when assessing the legitimacy of screening by considering the balance between its costs and its benefits expressed in QALYs. If the latter reveals a high cost per QALY, which is already a relative characteristic[mmmm], or if the evidence on which the analysis is based is rather weak, the ethical arguments that invite caution will reinforce the conclusions. Inversely, it will be necessary to imagine how we can balance quantitative arguments with essentially qualitative arguments. Recourse to the population's input could, in this case, turn out to be useful in order to discover trends, preferences and values across society.

Here, we propose distinguishing eight 'problems' within the framework of the teleological approach whose choice we have justified. We shall begin by considering the fundamental issue of freedom and the consubstantial issue of responsibility (point 2). This issue inevitably invites us to subsequently consider the importance of a socioeconomic and cultural gradient that characterises all areas of life (point 3). From a pragmatic point of view, we shall then deal with financial issues for people, in terms of accessibility and for the community, in terms of budgetary impact (point 4). We shall then take a step back to consider the meaning of screening for young athletes within the framework of an achievement society (point 5) before considering the dramatic consequences of the non-disqualification of a young person who

[llll] See [91], pp. 110-117 or [92], pp. 55-61

[mmmm] Belgium doesn't have a threshold beyond which this cost-utility ratio is considered as too high which could lead to the non-reimbursement of a health benefit (unlike Great Britain, which is one of the very rare examples in this domain). On the other hand, the calculation of a cost by QALY allows us to place this cost in a list that will grow as these kinds of analyses are made. It allows us to have an idea of the 'relative' cost of a service compared with others of a similar type (screening for another condition), or a completely different one (drugs).

dies during a physical activity (point 6). Within this context, we shouldn't forget the 'dangerousness' intrinsic to life (point 7). After considering the special case of minors (point 8), we shall end with a specific consideration for the important role of practitioners to inform correctly the athlete-patient (point 9).

7.2 Screening and freedom-responsibility

On the one hand, we shall consider the effect of screening on the 'direct' freedom of the individual to practice a sport, to do 'what they want' with their life and, on the other hand, its effect on 'indirect' freedom, i.e., the freedom we can deploy to practice a sport for the health benefits that characterise it, hence for the good state of health it allows us to maintain or achieve.

7.2.1 Limitation of 'direct' individual freedom

In the area in question, screening may not only be recommended but it may also become compulsory. Sports clubs or organisers of 'general public' events can choose to reduce the risk of accident to a minimum in order to maintain an image of 'the good management of their members' health. They may also be forced by law to oblige them to take an aptitude test. In this case, every person's freedom to practice a sport 'competitively' or 'recreationally' would of course be reduced. In order to justify such a stance, we could raise the argument according to which the measure would aim to protect individuals 'from themselves'. Screening could thus be presented as a way of preventing the athlete from practicing an activity that could 'kill' them, with a degree of certainty that obviously depends on the rate of false positives. At this stage, we may indeed question the relevance of imposing a test that wouldn't be very sensitive and/or specific. The argument of 'protecting the individual from themselves' is nonetheless open to attack because there are a considerable number of activities and ways of life that have been proven to be a health risk and can ultimately be fatal. For various reasons, the idea isn't to actually restrict the individual in their life choices. Here, we are talking about an unbalanced diet, smoking, excessive alcohol consumption, high-risk professions and sports and, also, a lack of physical activity. It is difficult to imagine how we could justify obligations/bans in all these areas simply with the intention of creating a healthy lifestyle. At the very most, we plan to use incentives whose results are – we might add – ambiguous[93-96]. Starting from an economic [97-100] or legal point of view [101-105], philosophers have examined this 'boundary line' between what falls under the scope of our freedom and thus our responsibility, and what falls under the scope of life circumstances for which we can't be blamed. Although interesting from an intellectual point of view, their solutions aren't very realistic from a technical standpoint, also giving rise to criticism from a moral point of view [106, 107]. Therefore, it isn't simply a question of knowing what is desirable or good for our health, we must also take into account what the actual possibilities are, everyone's actual freedom to be part of the proposed health process. We can therefore consider that screening of a restrictive and compulsory nature would be difficult to justify from a moral point of view owing to its negative effect on individual freedom. On the other hand, it could very easily be justified for reasons of civil and even moral responsibility on the part of sports clubs or organisers of sporting events.

Messages to promote health predominantly include an invitation to practice a physical leisure activity to make each of us 'responsible' for our overall health, which we are supposed to manage as a responsible person. These messages therefore seem to contradict a screening system that would ultimately exclude part of the population from this practice recognised as being beneficial to health. The extent of this contradiction should be measured in the light of false positives, i.e., by considering all those who will be excluded unnecessarily from physical sports activities.

7.2.2 Limitation of 'indirect' individual freedom

In this respect, the screening of young athletes affects individual freedom in another manner. Indeed, it isn't only likely to prevent people from doing what they want with 'their health' or their lives, it can also reduce the freedom of those who believe in the virtues of sport as a means to maintain good health. This limitation of freedom to practice sport becomes more important as the individual responsibility is nowadays questioned in order to differentiate the reimbursement of healthcare based on the individual's lifestyle[nnnn]. As underlined by the Nobel Prize winner in economic sciences, Amartya Sen, "freedom is the necessary and sufficient condition for responsibility"[oooo]; it is thus more difficult to attribute (at least in part) any responsibility to people

nnnn See, in particular, the recent Belgian survey by [108]

oooo [109], p.372.

who don't have the freedom to practice sport. Perhaps we could imagine a sort of immunisation of this responsibility for athletes who have been recommended to stop, or even banned from practicing a certain sport, or even any sport, on the basis of a test. However, there will still be all those who haven't dared to take this test or for whom the test results will be sufficiently ambiguous for them to be put off practicing their sport, even erroneously. The link between real freedom and responsibility, as well as the way of enabling everyone to achieve this real freedom and the ability to decide, is the subject of a body of literature that combines philosophy, anthropology, economics and epidemiology. The care given to patients to provide them with the information they need is likely to free them and allow them to take a decision autonomously[91, 110-114]. This 'informational care' requires time and we are quite right to wonder whether doctors would actually have enough time if they were to receive a sudden influx of athletes/patients. This clearly relates to the ethics of considering the consequences of the results of screening on the freedom to practice a sport and on responsibility, and whether or not to respect the recommendation, while also feeling 'responsible' for one's own health. It isn't sufficient to grant an athlete a dispensation from practicing sport for medical reasons for them to feel 'free of all responsibility'. The feeling or even the conviction that we are acting in a way that is beneficial to our health, especially by practicing sport, is reinforced by strong links, based on scientific evidence that endorses the positive aspects of physical activity. This means that being 'exempt' from sport isn't a 'doctor's note', like the one young pupils hope to obtain to avoid a swimming lesson in winter. This dispensation can be experienced as deep frustration at having to abandon a practice that is not only good for health, but also the source of personal satisfaction.

7.3 Screening and social, economic and cultural gradient

As we already underlined, this ethical problem, which relates to the freedom-responsibility duo, includes social, economic and cultural inequalities, which exacerbate the problem of losing freedom in a society that recognises individual responsibility. There is a large body of literature that demonstrates the presence of a socioeconomic gradient[pppp] as regards health, healthcare consumption, postponed healthcare, screening, and life expectancy[qqqq]. It was fairly recently completed by an epidemiological analysis that establishes a link between the degree of inequality in income distribution and a set of variables such as life expectancy, morbidity, criminality, teenage pregnancy, etc. [135]. We also know that "the knowledge, motivation and skills of individuals to access, understand, assess and use health information with a view to making judgements and taking decisions in everyday life as regards health, the prevention of illness and health promotion, in order to maintain or improve the quality of life", what we call health literacy, is also distributed according to a socioeconomic gradient [136, 137]. This means that not only do the less 'educated' or 'economically' disadvantaged adopt behaviours that are less propitious to maintaining health or recovery, they also don't have the same ability to understand and use the information they receive. It is therefore possible that these same people have difficulty understanding and interpreting the medical recommendations resulting from screening. This aspect of inequality is all the more important if the screening results are not categorical, or if they leave room for interpretation, if they require the person affected to make a decision. The educational gradient (according to the level of studies) that characterises physical activity in Belgium (see table below) very clearly quantifies the effects of the difficulty certain people have in 'integrating' the results of screening for young athletes. Indeed, their low level of literacy is compounded by insufficient practice to generate 'good health' or to avoid or at least reduce cardiovascular problems [138]. In other words, the most vulnerable people, whom we know are less likely to adopt a 'healthy' lifestyle, would also be among those to have the greatest difficulty in understanding the recommendations with regard to practicing a sport.

pppp Thanks to Pierre Bourdieu in particular, we know that cultural practices and preferences are closely linked to the level of education and social origin [115, 116], a reason for which it seems useful to add the cultural aspect to the economic and social aspects which are the subject of this gradient.

qqqq There are plenty of references concerning this subject. For instance, for Belgium: [117-134].

They could quite simply understand them as an irrevocable ban whereas a certain degree of assertiveness would perhaps allow them to envisage some physical activity under supervision.

Furthermore, it could be that the radicalism of these bans is dependent on the 'demanding' nature of the sport on a cardiovascular level. Therefore, it would be relevant, in terms of equity, to check the success of sports within the population according to its social, economic and cultural class because "a sport has all the more chance of being adopted by members of a social class if it doesn't contradict the relationship with the body at the deepest and most subconscious level, i.e., the body's map insofar as it is the depository of a complete vision of the social world, of the complete philosophy of a person and the body itself".[115] For instance, it is possible that the practice of a somewhat demanding sport like football is imbued with a social, economic and cultural gradient. The temptation of not taking any risks and 'rapidly disqualifying' young players to avoid possible deaths shouldn't be excluded.

However, we know that there are 2 000 football clubs in Belgium where 300 000 matches are organised every year for just over 415 000 members.[rrrr]

Hence, it is a very popular sport and, even if data on the subject is lacking, it may well be one of the sports that offers young people from less affluent social categories a way to channel their physical and mental energy, as well as social integration. The impact of screening in terms of depriving people of a sports activity must certainly be considered regarding the possible impact in terms of equity (equal probability for each athlete, whatever their social, economic and cultural class, of being 'disqualified' on equal objective medical grounds). As we have emphasised, even greater attention must be paid to this point if the screening isn't characterised by a high level of effectiveness.

If screening is set up, it is vital that those providing the service and sending out the results are fully aware that the 'terrain' receiving this information must be 'treated' in a very selective manner. This is a necessary condition to prevent screening from reinforcing the social inequalities already very present in the area of physical activity.[139]

Results of ISP 2013 survey relating to the practice of physical activities is presented according to the level of studies in Table 17.[138]

Table 17 – Percentage of the population of 15-year-olds and over

Educational categories	**Practicing** sufficient physical activity **to have a** positive impact on health	**Practicing** sufficient physical activity **to** avoid excessive weight **gain**	**Practicing** sufficient physical activity **to** reduce **the risk of developing a** cardiovascular disease	Risking **one's health owing to a** lack of physical leisure activities
No diploma or primary school diploma	15.6%	17.9%	23.8%	49.5%
Lower secondary education certificate	25.2%	26.4%	33.5%	39.1%
Higher secondary education certificate	31.0%	30.9%	44.3%	27.3%
Degree	25.9%	26.6%	42.0%	20.8%

[rrrr] http://www.belgianfootball.be/fr

7.4 Screening, accessibility of care and budgetary impact

The rate of cover for care and particularly ECG tests is such in our country that financial accessibility is not really a problem, contrary to countries where care is widely privatised.[56, 140] This financial accessibility could be fully guaranteed if the whole screening process (consultations and tests) was completely covered by medical insurance. However, we know that free care doesn't ensure equal access for everyone owing to barriers that can be described as 'cultural'. Moreover, we have seeing the development of medical centres that closely monitor every athlete whether they are high-level athletes or pure amateurs. The cost of this monitoring is of course dependent on the services provided, the time devoted and the complementary tests offered (for instance, determining the VO_2 max). If a 'simple' ECG can be envisaged as a means of determining who may or may not continue to practice their sport, those who have the (financial and informational) means could benefit from perhaps more specific, more adapted and more relative recommendations. In this case, fairness would be at stake. We also have to consider geographic accessibility, which we must ensure is sufficient given the high number of young athletes who will have to undergo tests every year and be seen to be given the results.

If it is indeed the decision-maker's wish to make everyone undergo screening, financial accessibility is clearly more or less a basic condition. And if screening is recognised as effective and its cost-efficiency ratio is considered socially acceptable, it would seem coherent to provide it for free. Here we can appreciate the importance of a medical, clinical analysis relating to efficiency and that of the cost analysis per QALY.

It is also part of ethical thinking to consider the opportunity cost of the entire approach. Indeed, it is always useful within the framework of an inevitably limited budget to wonder which other health interventions awaiting reimbursement won't be reimbursed owing to the budgetary impact of the screening of athletes. The cost-effectiveness ratio for which we have no threshold in Belgium offers an instant image of a societal cost that is rarely integrated into a comprehensive framework. Even if this ratio would be considered as acceptable at first sight, the extent of the budgetary impact may be such that the decision could be weighted against services with a similar impact but whose efficiency is based on stronger evidence or is simply higher (for instance, a lower rate of false positives).

7.5 An achievement-based societal context

At this stage, the ethical considerations proposed seem to plead in favour of caution before implementing screening for athletes aged 14 to 35 years. The following argument refers to the societal context within which the concept of achievement has established itself in nearly all areas of activity and at all ages. Escaping the pervasiveness of achievement in our lives is undoubtedly a matter of good sense and requires us to 'succeed' in our ability to resist 'social norms' [141]. The phenomenon probably isn't very new because in the 1970s already, Bourdieu wrote that "all groups run in the same direction, towards the same objectives, with those dictated by the group taking first place in the race" [115]. We could extend the plea for caution by arguing that banning the practice of a sport is equal, at least in part, to cutting an individual off from the society that requires them, through sport, to succeed. If the screening result turns out to be positive for someone attached to 'their achievements', it will be essential to redirect their quest for achievement towards other areas of action. The medical corps can help them in this matter but it can also help them to envisage a life without achievements. The individual 'saved' by screening can also understand and admit that they must benefit from this 'chance' to 'make the most' of life in another way. When a person who is not (yet) a patient is informed by a doctor that they risk becoming one, that they are risking their life by not respecting the recommendations given to them regarding caution, shouldn't the medical profession also provide informational care to help them cope with their new life? Informing someone passionate about sport or simply their physical condition that they must adapt, change pace and find derivatives, should not be taken lightly. It perhaps isn't incongruous to think that it is up to the medical profession, at least in part, to offer the necessary help so that this transition occurs in the most fruitful way possible.

In short, either we consider that the achievement-based society is a fact, even a constraint that we don't question and is even the source of integration and societal recognition. In this case, we must be able to manage the 'disqualification' of young athletes who will no longer be able to express themselves fully, in a 'recognised' way within society and think of options for these young people who will have to 'achieve' in other ways. Or, we express a fundamental criticism regarding the pervasiveness of achievement and we consider that 'disqualification' is a chance to escape it. In this case, we must adopt a view in accordance with this social criticism.

This attitude regarding achievement relates to anthropology; it is uncertain whether the neutrality decision-makers must show would allow this criticism and its consequences to be truly envisaged. Therefore, in general, we could say that disqualification will be experienced as the impossibility of pursing a 'quest for achievement'.

7.6 Exclusion from practicing sport versus the suffering of a grieving family

There is one last argument that, alone, is capable of rebalancing this analysis of teleological ethics, which, up until now, pleads at least in favour of caution and reflection before imposing screening on young athletes. It concerns the probability of the occurrence of sudden death which, even if it remains low, affects real victims and not only statistical victims. For the family of the victim in 'the prime of life' who dies suddenly as a result of a physical effort, the suffering cannot be alleviated by considerations of efficiency. The grieving family won't be sensitive to the argument of the freedom that we wanted to preserve for everyone, even at the risk of not informing a young person that their days were numbered if they continued to practice their sport. If we don't carry out screening for reasons of efficiency, those who have to live without their loved one may have the feeling that this person was a sacrificial victim on the altar of economic calculation. The injustice felt can be all the stronger when the family discover that the death was a result of an inherited malformation (which is generally the case in young athletes who suffer sudden death), that the victim wasn't at fault and that the parents, who are still alive, probably passed on a fatal genetic or congenital risk linked to a 'normal' life.

7.7 Living is dangerous

The sudden death of someone in the prime of life while practicing a sport is somewhat antinomical with physical activity, which is supposed to be good for our health. However, we may well ask ourselves whether the purpose of screening is to prevent deaths that cause a stir in the media or to warn all young people of the risk of dying suddenly during a physical effort of any kind. An example would be the sudden death of someone in their thirties running to catch their train. A life that ends too soon, a grieving family, but probably not even a paragraph in the press. As regards prevention, is it justifiable to make a distinction between this young man and the one who dies during the Brussels 20 km run? More fundamentally still, should we envisage a race for a zero-risk society that is probably already lost [142] or favour a certain lack of concern that also adds the 'pep' to life, which would lacking if we were to live in permanent fear of being the victim of an unforeseeable event. This issue obviously concerns the limits of prevention, the precaution society must apply to the lives of its members. An eminently societal issue that probably requires the opinion of those who are directly concerned.

7.8 The delicate issue of minors

The situation of minors with regard to the screening in question here is obviously subject to the same points of attention as those concerning the 'general population'. However, for adolescents between 14 and 18 years old, the "freedom", which we have stated as possibly being 'restrictive' or even lacking, is an even bigger stake. Indeed, the parents are not only legally responsible for their children, they also feel morally responsible for what happens to them. It wouldn't be surprising if their counterfactual attitude were tinged with greater caution than they would apply to themselves. The effects of exclusion from sports could therefore be greater in the case of children owing to a reinforced 'preventive' attitude among parents and the medical corps. Given that the problems we are looking for are malformations of genetic or congenital origin, the parents' feeling of responsibility may be exacerbated. The same is true of the impression of less freedom that the children might feel. Just like the recommendations for genetic tests [143], it would seem advisable not to consider the age of majority as a 'cut-off point', a threshold below which the young person has no say. It is indeed the true capacity of reflection, a certain form of maturity in young people that has to be taken into consideration to decide whether or not to proceed with the test. Indeed, we can't exclude the difficulty a young person may experience when they discover their inability to practice a sport, the effect on their self-esteem and the development of their personality. We can well imagine that they may prefer not to know (i.e. not be screened) and thus give up a sport.

7.9 The role of the practitioners stays very important

If a screening (per definition general and mandatory) is organized to receive the authorization to do sport, the practitioner is in charge of a very important mission. He/she has to practice an 'informational care' to inform the 'athlete' of all the possible consequences of the test. In a second time, he/she will have to inform the 'athlete' about the results of the ECG and try to help him/her first to understand and accept them. The practitioner will probably also be helpful to assist the athlete to find an alternative to the definitive cessation of the sport practice.

With or without the obligation to implement a mandatory screening, the practitioner will remain the most important interlocutor for a person who will engage in a sport activity and who feels a totally subjective anxiety or have concrete complaints when he/she makes an effort. It remains the responsibility of the practitioner to inform the 'athlete-patient' when there exist reasons to become cautious when he/she making an effort. It is also the responsibility of the practitioner to reassure the person about her possibilities to become an 'athlete'.

7.10 Conclusion

We know that the results of the cost-utility analysis do not seem very favourable for this type of screening. On the whole, the questions raised by a teleological approach accentuate the perplexity resulting from the economic and medical analysis. Nevertheless, it seems that the simple prospect of 'allowing' a young person 'to die', who could have been informed of the small risk they ran by practicing their sport, is the focal point, the argument which invites caution before deciding to abandon screening, even if its benefit is low. It would therefore appear that we are returning to a deontological approach, which proves that they are perhaps not as exclusive as they seem.

8 OVERALL CONCLUSION

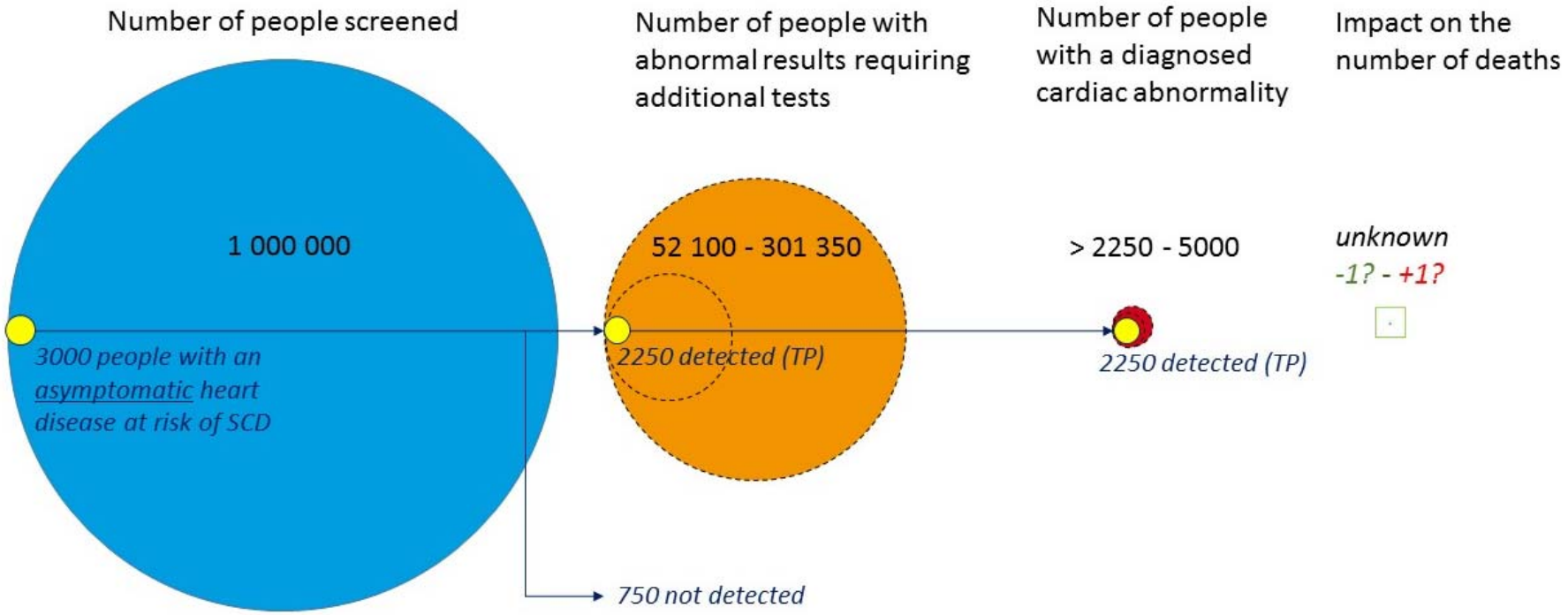
Sudden cardiac death in a young individual is a devastating event. Appropriate measures to prevent such tragedies would be more than welcome.

Within the framework of prevention and health promotion, the Flemish Community has stipulated in its legislation that sports federations should carefully consider whether or not to oblige or advise pre-participation screening for their members. In the French Community legislation, a medical certificate proving the absence of contra-indication for the particular sports discipline is compulsory for all competitive athletes or specific risk groups. The content of the certificate and the modalities of the medical examination are still to be defined by the government.

This report, which can be seen as an update of the report from the Belgian Superior Health Council (Hoge Gezondheidsraad - Conseil Superieur de la Santé),[7] has tried to give an answer on the question if cardiovascular pre-participation screening is useful.

The **clinical benefit** of pre-participation screening in reducing SCD of young athletes is presently not fully clear. There is however no doubt that SCD in this population is a very rare event. Therefore, in the absence of a perfect diagnostic test, pre-participation screening inevitably induces a huge number of false-positives, leading to **overdiagnosis and overtreatment** of the healthiest segment of the population.

Figure 11 – Impact of a cardiovascular pre-participation screening with H&P and ECG in Belgium (for a sensitivity of 0.75 and a specificity between 0.70 and 0.95)



TP: True Positive; SCD: Sudden Cardiac Death

Based on the scientific data provided in the present report (summarised in Appendix 4 and in Figure 11) we can assume that out of 1 million screened young people, a cohort of 50 000 to 300 000 individuals will be identified that will be suspected of having a disease that may lead to SCD. Further investigations will identify 2250 among them that are affected by a disease. The large majority of those will never die suddenly or will even never have any symptom of the disease.

The most prevalent diseases are WPW (1360 per million) and HCM (450 per million). Some experts argue that asymptomatic people with one of those conditions do not need to be treated. Other experts will proceed to catheter ablation or ICD implantation in selected individuals. They argue that, although it has not been shown that these interventions reduce the risk of SCD, there is a pathophysiological rationale for it. However, those treatment modalities have their proper mortality risk which appears to be of a similar magnitude of the SCD risk of asymptomatic affected individuals. This means that it cannot be taken for granted that lives will be saved because of the detection of those diseases at screening. This statement is confirmed by the fact that the incidence of SCD is not lower in Italy, where screening is mandatory, as compared to other countries (US, France) where there is no systematic screening.

In Belgium it can be expected that yearly up to 10 young people may suddenly die during sports. Whereas the chance is low that pre-participation screening will save 1 of those individuals, it is possible that 1 life will be lost because of overdiagnosis and overtreatment.

Among the initially suspected 50 000 to 300 000 individuals, a final diagnosis will remain unclear in an estimated 2000 to 3000 of them because downstream diagnostic techniques are not 100% performant.

Eventually, around 5000 of the original 1 million screenees (i.e. 0.5%) will be labelled as suffering from a cardiac disease. This may lead to temporary or lifelong disqualification from competitive sports, psychological harm, and (lifelong) medical follow-up and treatment with unknown benefit.

David Sacket, the father of Evidence-Based Medicine, argues that curative and preventive medicine are absolutely and fundamentally different in their obligations and implied promises to the individuals whose lives they modify.[144] He further explains that, when patients are looking for help, the doctor promises to do his best without guaranteeing that his interventions will succeed. In contrast, in preventive medicine, the fundamental promise a doctor makes must be that, on average, the involved symptomless individual will be the better for it. Accordingly, the presumption that justifies the medical intervention must be based on the highest level of randomised evidence that the preventive manoeuvre will do more good than harm. Without evidence from positive randomised trials it cannot be justified soliciting the well to accept any personal health intervention.[144]

The lack of solid evidence of the benefit of cardiovascular pre-participation screening, and the certainty of the harms it induces, makes that such screening cannot be defended.

The Belgian Superior Health Council acknowledged the absence of hard scientific evidence favouring cardiovascular screening and recommended against mandatory screening.[7] However, based on the conviction of professionals involved in the report, and the presumed societal support for screening, it positively recommended a strictly supervised cardiovascular pre-participation screening in young people who want to participate in competitive sports, with the inclusion of an ECG. In a recently published assessment of the ECG as a screening test in young individuals, the American Heart Association concludes that there is insufficient information available to support universal ECG screening for cardiovascular disease in asymptomatic young people, both in competitive athletes and in the general youthful population.[9]

Concerning the **cost-effectiveness**, there are no reliable economic evaluations with convincing results showing that pre-participation screening in young athletes provides value for money. Based on optimistic assumptions, a one-time cardiovascular pre-participation screening seems not cost-effective or might even be a dominated alternative if we take into account the impact on the quality of life. Moreover, results are even worse for a yearly screening (unlikely to be cost-effective).

Our calculations for the Belgian setting showed that a cardiovascular screening consisting of the combination of history, physical examination and a rest-ECG (every four years or more often) [ssss], would be expensive and potentially harmful, compared to the uncertain benefits.

The implementation of a screening program can also have consequences on the extent of **liability** of the physician. Malpractice liability for failure to discover latent, asymptomatic cardiovascular diseases requires proof that the physician did not act in line with the accepted medical practice in his/her specialty (fault) and that the proper utilisation of the appropriate methods would likely have discovered the underlying medical condition (causality). Up to now the inconsistency between existing guidelines, are likely to exonerate the physician from any proof of fault.

Athletes involved in a pre-participation screenings program are considered as patients, as defined in the Patients' Rights Act. This implies amongst others that an athlete should correctly be informed about the potential benefits and harms of cardiovascular pre-participation screening.

Practices among sports federations vary largely. Currently the athlete can be obliged to undergo a pre-participation screening, but rarely a clear testing protocol is recommended by the sports federations. Also financial support for the athlete is lacking.

In accordance to a **teleological approach**, the following points of attention should be considered if a general screenings program would be implemented:

- Risk of limitation of 'direct' individual freedom i.e. to be free to practice sport
- Risk of limitation of 'indirect' individual freedom i.e. to be free to do an activity which is good for the health
- Risk of reinforcement of social, economic and cultural inequalities
- Financial consequences, i.e. financial accesibility for the popluation and budgetary impact

Furthermore is screening in minors is a delicate issue, with a specific limitation of their freedom by the parents, who are legally and morally responsible for their children.

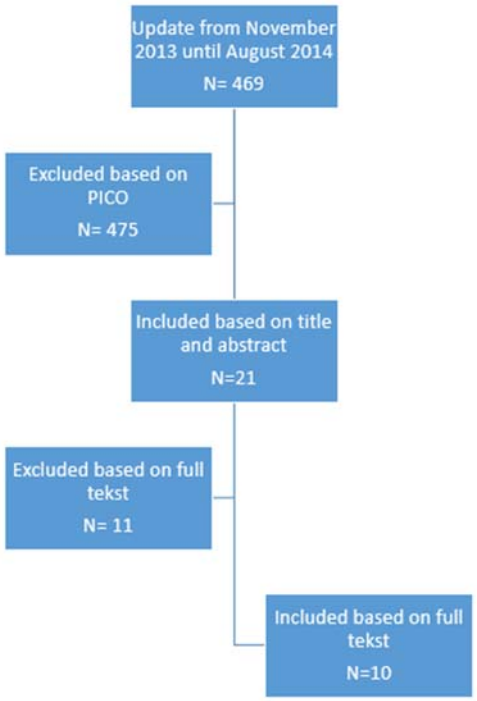
With or without the obligation to be screened, it remains the responsibility of the physician to inform the athlete on the potential benefits and harms linked to a cardiovascular pre-participation screening program.

[ssss] As specified in the report of the Belgian Superior Health Council

APPENDIX

APPENDIX 1. CLINICAL EFFECTIVENESS – LITERATURE SEARCH RESULTS

Appendix 1.1. Selection sequence of relevant primary studies



Appendix 1.2. Result of search for guidelines and systematic reviews

Source	Search terms	Number of hits	Relevant titles
National Guideline Clearinghouse	Sudden death Screening and cardiac and sport	112 23	Casa DJ, Guskiewicz KM, Anderson SA, Courson RW, Heck JF, Jimenez CC, McDermott BP, Miller MG, Stearns RL, Swartz EE, Walsh KM. National Athletic Trainers' Association position statement: preventing sudden death in sports. J Athl Train. 2012 Jan-Feb;47(1):96-118. [207 references] U.S. Preventive Services Task Force. Screening for coronary heart disease with electrocardiography: U.S. Preventive Services Task Force recommendation statement. Ann Intern Med. 2012 Oct 2;157(7):512-18 . [13 references] Singapore Ministry of Health. Screening for cardiovascular disease and risk factors. Singapore: Singapore Ministry of Health; 2011 Mar. 101 p. [189 references] Mahmood S, Lim L, Akram Y, Alford-Morales S, Sherin K, ACPM Prevention Practice Committee. Screening for sudden cardiac death before participation in high school and collegiate sports: American College of Preventive Medicine position statement on preventive practice. Am J Prev Med. 2013 Jul;45(1):130-3. [23 references]
GIN	Sudden death Cardiac screening Sport Screening and sport	15 2 1 0	Mahmood S, Lim L, Akram Y, Alford-Morales S, Sherin K, ACPM Prevention Practice Committee. Screening for sudden cardiac death before participation in high school and collegiate sports: American College of Preventive Medicine position statement on preventive practice. Am J Prev Med. 2013 Jul;45(1):130-3. [23 references] Casa DJ, Guskiewicz KM, Anderson SA, Courson RW, Heck JF, Jimenez CC, McDermott BP, Miller MG, Stearns RL, Swartz EE, Walsh KM. National Athletic Trainers' Association position statement: preventing sudden death in sports. J Athl Train. 2012 Jan-Feb;47(1):96-118. [207 references]
EBMPracticeNet	Cardiac screening Sports	0 0	/
SIGN	Full list of guidelines	139	/
ESC	Full list of guidelines	35	/
Medical Journal of Australia	Full list of guidelines	94	/

National Health and Medical Research Council (NHMRC)	Sudden death Cardiac Screening Sports	2 0 2 0	/
The New Zealand Guidelines Group (NZGG)	Full list of guidelines	55	/
NICE	Sudden death Cardiac screening Sports	63 63 60	/
HAS	Dépistage	171	/
Google custom search	Sudden death Sudden death and sports	668 34	Casa DJ, Guskiewicz KM, Anderson SA, Courson RW, Heck JF, Jimenez CC, McDermott BP, Miller MG, Stearns RL, Swartz EE, Walsh KM. National Athletic Trainers' Association position statement: preventing sudden death in sports. J Athl Train. 2012 Jan-Feb;47(1):96-118. [207 references] Mahmood S, Lim L, Akram Y, Alford-Morales S, Sherin K, ACPM Prevention Practice Committee. Screening for sudden cardiac death before participation in high school and collegiate sports: American College of Preventive Medicine position statement on preventive practice. Am J Prev Med. 2013 Jul;45(1):130-3. [23 references] Singapore Ministry of Health. Screening for cardiovascular disease and risk factors. Singapore: Singapore Ministry of Health; 2011 Mar. 101 p. [189 references]

Appendix 1.3. Overview of selected 20 guidelines and systematic reviews

#	year	First author	Quality appraisal	Recommendations relevant for the present report
1	1994	Strong et al. (Group on Science and Technology, American medical Association)	Consensus statement	The usefulness for identifying adolescents at risk for sudden cardiac death or who have previously undiagnosed chronic medical disorders that could affect athletic participation, however, is not substantiated. No specific recommendation on the usefulness of ECG in the pre-participation screening.
2	1996	Maron (AHA)	No methods section	Non-invasive testing can enhance the diagnostic power of the standard history and physical examination; however, it is not prudent to recommend routine use of such tests as 12-lead electrocardiography, echocardiography, or graded exercise testing for detection of cardiovascular disease in large populations of young or older athletes.
3	2004	Wingfield 2004	Systematic review, AMSTAR score Y, can't answer, Y, Y, N, Y, N, Y, Y, N, N	ECG in particular is favored by some authors but not others, though poor evidence actually exists for its efficacy.
4	2005	Corrado	Consensus statement	The consensus document recommends the implementation of a common European screening protocol essentially based on 12-lead ECG, in addition to history and physical examination.
5	2006	Zipes (ACC/AHA/ESC)	Consensus statement	Twelve-lead ECG and possibly echocardiography may be considered as pre-participation screening for heart disorders in athletes (level of evidence: B, class IIb)
6	2006	Bille (IOC)	Systematic review, AMSTAR score Y, Y, Y, Y, N, Y, N, can't answer, Y, N, N	Within the Lausanne Recommendations, a 12-lead rest-ECG is recommended for all participants (after the onset of puberty).

7	2007	Maron (AHA)	Consensus statement	A large population pre-participation screening initiative for US athletes that mandate a 12-lead ECG, such as that proposed by the ESC and IOC, is probably impractical and would require considerable resources that do not currently exist, as well as substantial long-term federal government subsidization. Such screening could also be potentially deleterious to many athletes by virtue of false-positive test results that would lead to unnecessary further evaluations and testing, anxiety, and possibly disqualification without merit. On the other hand, the panel does not arbitrarily oppose volunteer-based athletic screening programs with non-invasive testing performed selectively on a smaller scale in local communities if well designed and prudently implemented.
8	2009	Perez	Systematic review, AMSTAR score Y, N, N, N, N, Y, N, N, NA, NA, N	Whether or not an ECG should be routinely added to the athletic PPE remains highly controversial. Therefore, a large-scale trials in the USA is necessary.
9	2011	Borjesson	No methods mentioned	The sensitivity of screening with ECG is vastly superior to, and the cost-effectiveness significantly better than, screening without ECG. Cardiac screening without ECG is not cost-effective and may be only marginally better than no screening at all and at a considerable higher cost. The current evidence suggests that the ECG should be mandatory in pre-participation screening of athletes.
10	2011	Drezner	No methods mentioned	A comprehensive personal and family history and physical examination are recommended components of cardiovascular screening in athletes but offer little sensitivity in identifying athletes at risk for SCD and the value of these measures alone is questionable. Electrocardiogram should be recommended and offered to athletes as part of a pre-participation cardiovascular screen.
11	2011	La Gerche	No methods mentioned	A systematic collection of Australian data is required before routine pre-participation screening can be introduced in Australia.
12	2011	Steinvil	Limited search in newspapers	Mandatory ECG screening of athletes had no apparent effect on their risk for cardiac arrest.

13	2011	Singapore Ministry of Health	No methods mentioned	For pre-participation screening, a two- or more stage screening process is encouraged, where the first stage consists of personal and family history taking and physical examination. Based on the findings of the first stage, further tests such as a rest-ECG (if not already done), chest X-ray, exercise stress test, echocardiogram, blood investigations, urine tests, etc. may be ordered if indicated.
14	2012	Estes	No methods mentioned	The optimal strategy for advancing toward the widely accepted goal of prevention of sudden death in the athlete is not yet accompanied by sufficient evidence regarding outcomes, risks, benefits, and cost to resolve ongoing debates related to screening and intervention strategies. Currently, including the ECG in screening is potentially harmful to many athletes because of false-positive test results that would lead to unnecessary further evaluations and testing, anxiety, and, possibly, disqualification without merit.
15	2012	Asif	No methods mentioned	The addition of ECG to cardiovascular screening greatly increases the ability to detect athletes at risk and to meet the primary objective of the pre-participation evaluation (detection of those at risk). Electrocardiogram screening in athletes can be performed accurately and with a low false-positive rate through application of modern standards for ECG interpretation.
16	2012	Casa	Consensus statement	The pre-participation physical examination should include the completion of a standardized history form and attention to episodes of exertional syncope or pre-syncope, chest pain, a personal or family history of sudden cardiac arrest or a family history of sudden death, and exercise intolerance. Further research is needed to understand whether additional tests, such as ACG, improve sensitivity and can be performed with acceptable cost-effectiveness and an acceptable false-positive rate.
17	2012	Moyer	Consensus statement	The USPSTF recommends against screening with rest- or exercise ECG for the prediction of coronary heart disease (CHD) events in asymptomatic adults at low risk for CHD events.
18	2013	Casa	Consensus statement	A resting 12-lead electrocardiogram may be used in many pre-participation screening programs. A rest-ECG may increase identification of athletes with cardiac conditions associated with sudden death. Questions and limitations regarding sensitivity and specificity, preclude universal ECG screening for all athletes at this time. Proper

				physician education in ECG interpretation in athletes and appropriate cardiology resources for secondary evaluations when indicated are important.
19	2013	Mahmood (ACPM)	Consensus statement	The American College of Preventive Medicine recommends against routine screening for potential sudden cardiac death with ECG, echocardiography, and genetic testing in individuals without personal risk factors.
20	2013	Dvorak	Consensus statement	The Precompetition Medical Assessment (PCMA) as recommended by FIFA involves at least a focused player medical history (PMH), family medical history (FMH) and cardiac specific physical medical examination. A rest-12-lead ECG should be undertaken as part of the PCMA on all players at the beginning of their playing career and then once every year.

APPENDIX 2. ECONOMIC EVALUATION

Appendix 2.1. Literature search cost-effectiveness

Appendix 2.1.1. First stage

In September 2014, the websites of HTA institutes (Table 18) and the Health Technology Assessments (HTA) database of the Cochrane Library were investigated to identify HTA reports. The research was updated in December 2014. Finally, one potential HTA was identified. Table 18 up to Figure 12 provide an overview of the applied search strategies.

Table 18 – List of INAHTA member (and ex or non-members) websites searched for HTA reports

Abbreviation	Institute	Country
AETS	Agencia de Evaluación de Tecnologias Sanitarias	Spain
AETSA	Andalusian Agency for Health Technology Assessment	Spain
AGENAS	The Agency for Regional Healthcare	Italy
AHRQ	Agency for Healthcare Research and Quality	USA
AHTA	Adelaide Health Technology Assessment	Australia
AHTAPol	Agency for Health Technology Assessment in Poland	Poland
AQuAS (CAHIAQ)	Agència de Qualitat i Avaluació Sanitàries de Catalunya - Catalan Agency for Health Information, Assessment and Quality (formerly CAHTA)	Spain
ASERNIP-S	Australian Safety and Efficacy Register of New Interventional Procedures -Surgical	Australia
AVALIA-T	Galician Agency for Health Technology Assessment	Spain
CADTH	Canadian Agency for Drugs and Technologies in Health	Canada
CDE	Center for Drug Evaluation	Taiwan
CEDIT	Comité d'Évaluation et de Diffusion des Innovations Technologiques	France
CEM	Inspection générale de la sécurité sociale (IGSS), Cellule d'expertise médicale	Luxembourg
CENETEC	Centro Nacional de Excelencia Tecnológica en Salud Reforma	Mexico
CMERC	Department of Internal Medicine	South Africa
CNHTA	Committee for New Health Technology Assessment	Korea
CONITEC	National Committee for Technology Incorporation	Brazil
CRD	Centre for Reviews and Dissemination	United Kingdom
DAHTA @DIMDI	German Agency for HTA at the German Institute for Medical Documentation and Information	Germany
DECIT-CGATS	Secretaria de Ciëncia, Tecnologia e Insumos Estratégicos, Departamento de Ciência e Tecnologia	Brazil

ETESA	Department of Quality and Patient Safety of the Ministry Health of Chile	Chile
FinOHTA	Finnish Office for Health Care Technology Assessment	Finland
G-ba	The German Health Care System and the Federal Joint Committee	Germany
GÖG	Gesundheit Österreich	Austria
GR	Gezondheidsraad	The Netherlands
HAS	Haute Autorité de Santé	France
HCT-NHSRC	Division of Healthcare Technology, National Health Systems Resource Center, New Delhi	India
HealthPACT	Health Policy Advisory Committee on Technology	Australia
HIQA	Health Information and Quality Authority	Ireland
HIS	Healthcare Improvement Scotland	United Kingdom
HQO	Health Quality Ontario - Evidence Development and Standards Branch (formerly MAS)	Canada
HSAC	Health Services Assessment Collaboration	New Zealand
HTA-HSR/DHTA	HTA & Health Services Research	Denmark
IECS	Institute for Clinical Effectiveness and Health Policy	Argentina
IHE	Institute of Health Economics	Canada
IQWiG	Institut für Qualität und Wirtschaftlichkeit im Gesundheitswesen	Germany
INESSS	Institut national d'excellence en santé et services sociaux (INESSS)	Canada
KCE	Belgian Federal Health Care Knowledge Centre	Belgium
LBI of HTA	Ludwig Boltzmann Institut für Health Technology Assessment	Austria
MaHTAS	Health Technology Assessment Section at Ministry of Health of Malaysia	Malaysia
MSP-Uruguay	Ministry of Public Health	Uruguay
MTU-SFOPH	Medical Technology Unit - Swiss Federal Office of Public Health	Switzerland
NCCHTA (NETSCC, HTA-NIHR)	National Coordinating Centre for Health Technology Assessment	United Kingdom
NECA	National Evidence-based healthcare Collaboration Agency	Korea
NHC	New Zealand National Health Committee	New Zealand
NHMRC CTC – NHMRC	Clinical Trials Centre	Australia
NIHR-HSC	National Institute for Health Research – Horizon Scanning Centre	United Kingdom
NOKC	Norwegian Knowledge Centre for Health Services	Norway
OSTEBA	Basque Office for Health Technology Assessment	Spain

RCHD-CS	Ministry of Public Health of the Republic of Kazakhstan, Republican Centre for Health Development, Centre of Standardization, HTA department	Kazakhstan
SBU	Swedish Council on Technology Assessment in Health Care	Sweden
UCEETS	The National Coordination Unit of Health Technology Assessment and Implementation	Argentina
UVT-HTA	HTA Unit in A. Gemelli University Hospital	Italy
VASPVT	State Health Care Accreditation Agency under the Ministry of Health of the Republic of Lithuania	Lithuania
ZINL	Zorginstituut Nederland (Formerly CVZ – College voor Zorgverzekeringen)	The Netherlands
ZonMw	The Medical and Health Research Council of The Netherlands	The Netherlands
Ex or non-member websites		
BCBS TEC	Blue Cross and Blue Shield Association's Technology Evaluation Center (TEC)	USA
CHE	Centre for Health Economics	United Kingdom
ICTAHC	Israel Center for Technology Assessment in Health Care	Israel
HITAP	Health Intervention and Technology Assessment Program	Thailand
MSAC	Medicare Services Advisory Committee	Australia
NHS HIS	Healthcare Improvement Scotland	United Kingdom
NHS	National Health Service	United Kingdom
NICE	National Institute for Clinical Excellence	United Kingdom

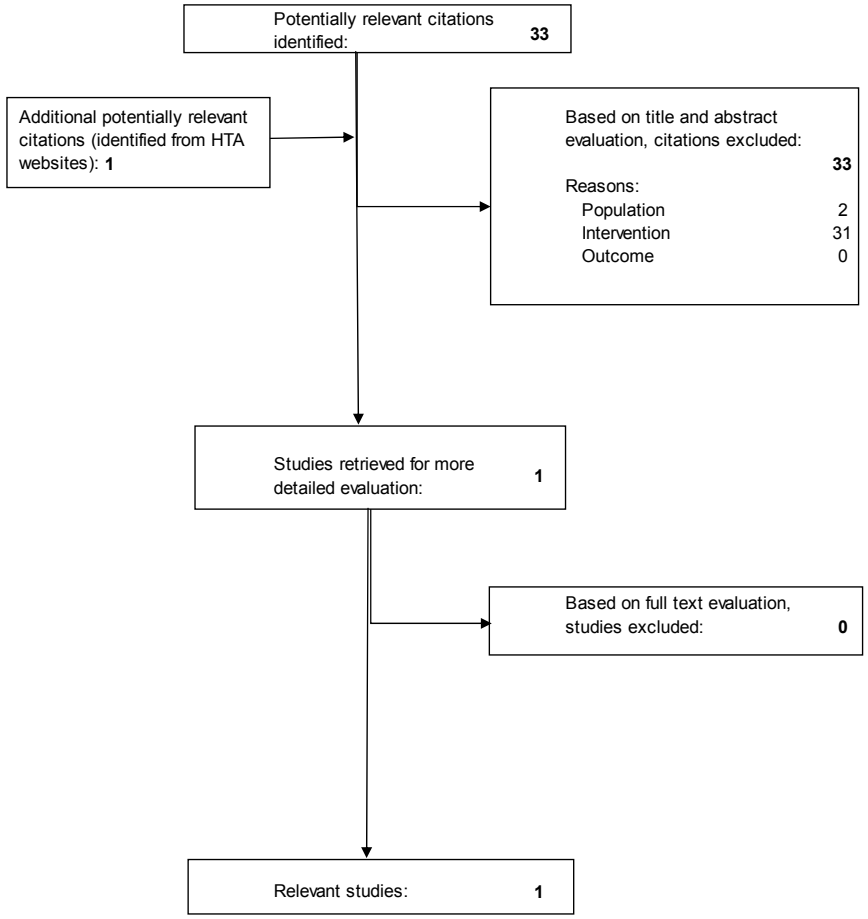
Table 19 – Search strategy and results for HTA database (Cochrane Library - results of 24 December 2014)

Date	17 September 2014, updated on 24 December 2014
Date covered	All
Search Strategy	#1 MeSH descriptor: [Death, Sudden, Cardiac] explode all trees 19 #2 sudden near/3 death:ab,ti 19 #3 MeSH descriptor: [Heart Arrest] explode all trees 37 #4 SCD:ab,ti 2 #5 MeSH descriptor: [Cardiomyopathy, Hypertrophic] explode all trees 7 #6 MeSH descriptor: [Arrhythmias, Cardiac] explode all trees 124 #7 (hypertrophic cardiomyopathy or HCM or Long QT Syndrome or LQTS or Wolff Parkinson White or WPW):ab,ti 11 #8 #1 or #2 or #3 or #4 or #5 or #6 or #7 164 #9 MeSH descriptor: [Mass Screening] explode all trees 629 #10 MeSH descriptor: [Cardiovascular Diseases] explode all trees and with qualifier(s): [Diagnosis - DI]128 #11 MeSH descriptor: [Medical History Taking] explode all trees 4 #12 MeSH descriptor: [Electrocardiography] explode all trees 46 #13 MeSH descriptor: [Physical Examination] explode all trees 270 #14 MeSH descriptor: [Physical Exertion] explode all trees 1 #15 MeSH descriptor: [Diagnostic Techniques, Cardiovascular] explode all trees 230 #16 screen*:ab,ti 676 #17 electrocardiogra*:ab,ti 10 #18 examination:ab,ti 22 #19 history:ab,ti 15 #20 #9 or #10 or #11 or #12 or #13 or #14 or #15 or #16 or #17 or #18 or #19 1346 #21 #8 and #20 27 #22 MeSH descriptor: [Sports] explode all trees 24 #23 MeSH descriptor: [Sports Medicine] explode all trees 1 #24 MeSH descriptor: [Athletes] explode all trees 0 #25 sport*:ab,ti 3 #26 athlet*:ab,ti 2 #27 football:ab,ti 0 #28 soccer:ab,ti 1 #29 basketball:ab,ti 0 #30 volleyball:ab,ti 0 #31 tennis:ab,ti 2 #32 squash:ab,ti 0 #33 badminton:ab,ti 0 #34 swimming:ab,ti 0 #35 swimmer*:ab,ti 0 #36 running:ab,ti 0 #37 runner*:ab,ti 0 #38 gymnastic*:ab,ti 0 #39 bicycling:ab,ti 0 #40 #22 or #23 or #24 or #25 or #26 or #27 or #28 or #29 or #30 or #31 or #32 or #33 or #34 or #35 or #36 or #37 or #38 or #39 30 #41 #20 and #40 6 #42 #8 and #40 0 #43 #21 or #41 or #42 33

Table 20 – Results of search strategy (first stage, i.e. HTA reports)

Database	References identified
	24 December 2014
HTA (via the Cochrane Library)	33
Websites of HTA institutes	1
Total (excl. duplicates)	34

Figure 12 – Flow Chart of the first stage



Appendix 2.1.2. Second stage

In September 2014, the following databases were searched: NHS Economic Evaluation Database (NHS EED - via the Cochrane Library), Embase, Medline (via Ovid), Psychinfo (via Ovid), and EconLit (via Ovid). Table 21 up to Figure 13 provide an overview of the applied search strategies. The research was updated in December 2014.

Table 21 – Search strategy and results for NHS EED (Cochrane Library- results of 24 December 2014)

Date	17 September 2014 updated on 24 December 2014
Date covered	All
Search strategy	#1 MeSH descriptor: [Death, Sudden, Cardiac] explode all trees 44 #2 sudden near/3 death:ab,ti 16 #3 MeSH descriptor: [Heart Arrest] explode all trees 70 #4 SCD:ab,ti 1 #5 MeSH descriptor: [Cardiomyopathy, Hypertrophic] explode all trees 5 #6 MeSH descriptor: [Arrhythmias, Cardiac] explode all trees 223 #7 (hypertrophic cardiomyopathy or HCM or Long QT Syndrome or LQTS or Wolff Parkinson White or WPW):ab,ti 8 #8 #1 or #2 or #3 or #4 or #5 or #6 or #7 282 #9 MeSH descriptor: [Mass Screening] explode all trees 1173 #10 MeSH descriptor: [Cardiovascular Diseases] explode all trees and with qualifier(s): [Diagnosis - DI] 300 #11 MeSH descriptor: [Medical History Taking] explode all trees 6 #12 MeSH descriptor: [Electrocardiography] explode all trees 113 #13 MeSH descriptor: [Physical Examination] explode all trees 659 #14 MeSH descriptor: [Physical Exertion] explode all trees 3 #15 MeSH descriptor: [Diagnostic Techniques, Cardiovascular] explode all trees 483 #16 screen*:ab,ti 1243 #17 electrocardiogra*:ab,ti 17 #18 examination:ab,ti 40 #19 history:ab,ti 26 #20 #9 or #10 or #11 or #12 or #13 or #14 or #15 or #16 or #17 or #18 or #19 2674 #21 #8 and #20 71 #22 MeSH descriptor: [Sports] explode all trees 60 #23 MeSH descriptor: [Sports Medicine] explode all trees 2 #24 MeSH descriptor: [Athletes] explode all trees 2 #25 sport*:ab,ti 1 #26 athlet*:ab,ti 5 #27 football:ab,ti 1 #28 soccer:ab,ti 1 #29 basketball:ab,ti 0 #30 volleyball:ab,ti 0 #31 tennis:ab,ti 1 #32 squash:ab,ti 0 #33 badminton:ab,ti 0 #34 swimming:ab,ti 0 #35 swimmer*:ab,ti 0 #36 running:ab,ti 1 #37 runner*:ab,ti 0 #38 gymnastic*:ab,ti 0 #39 bicycling:ab,ti 0 #40 #22 or #23 or #24 or #25 or #26 or #27 or #28 or #29 or #30 or #31 or #32 or #33 or #34or #35 or #36 or #37 or #38 or #39 65 #41 #20 and #40 18 #42 #8 and #40 5 #43 #21 or #41 or #42 86

Table 22 – Search strategy and results for EMBASE (results of 24 December 2014)

Date	17 September 2014 updated on 24 December 2014
Date covered	<1946 to Present>
Search Strategy	#68. #64 AND #65 AND #66 AND #67 557 #67. #52 OR #55 OR #56 OR #57 OR #58 OR #59 OR #60 OR #61 OR #62 OR #63 3,711,509 #66. #40 OR #41 OR #42 OR #43 OR #44 OR #45 OR #46 OR #47 OR #48 OR #49 OR #50 OR #51 459,142 #65. #21 OR #22 OR #23 OR #24 OR #25 OR #26 OR #27 OR #28 OR #29 OR #30 OR #31 OR #32 OR #33 OR #34 OR #35 OR #36 OR #37 OR #38 OR #39 401,476 #64. #1 OR #2 OR #3 OR #4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10 OR #11 OR #12 OR #13 OR #14 OR #15 OR #16 OR #17 OR #18 OR #19 OR #20 1,541,938 #63. history:de,cl,ab,ti 937,984 #62. examination:de,cl,ab,ti 967,406 #61. electrocardiogra*:de,cl,ab,ti 239,094 #60. screen*:de,cl,ab,ti 834,512 #59. 'cardiovascular system examination'/exp 777,436 #58. 'physical examination'/exp 157,741 #57. 'electrocardiography'/exp 155,212 #56. 'anamnesis'/exp 174,229 #55. 'cardiovascular disease'/exp AND 'diagnosis'/exp 1,072,494 #54. 'diagnosis'/exp 4,909,947 #53. 'cardiovascular disease'/exp 3,122,101 #52. 'mass screening'/exp 161,799 #47. 'wolff parkinson white':de,cl,ab,ti 7,196 #46. 'lqts':de,cl,ab,ti 1,765 #45. 'long qt syndrome':de,cl,ab,ti 8,936 #44. 'hcm':de,cl,ab,ti 4,994 #43.'hypertrophic cardiomyopathy':de,cl,ab,ti 18,297 #42. scd:de,cl,ab,ti 10,446 #41. (sudden NEAR/3 death):de,cl,ab,ti 62,097 #40. 'sudden cardiac death'/exp 3,721 #39. swimmer:de,cl,ab,ti 925 #38. runner:de,cl,ab,ti 3,334 #37. running:de,cl,ab,ti 56,701 #36. bicycling:de,cl,ab,ti 975 #35. gymnastic*:de,cl,ab,ti 2,846 #34. soccer:de,cl,ab,ti 5,238 #33. badminton:de,cl,ab,ti 295 #32. squash:de,cl,ab,ti 2,388 #31. basketball:de,cl,ab,ti 3,384 #30. volleyball:de,cl,ab,ti 1,510 #29. football:de,cl,ab,ti 8,116 #28. tennis:de,cl,ab,ti 4,778 #27. swimming:de,cl,ab,ti 28,480 #26. sports:de,cl,ab,ti 52,185 #25. sport:de,cl,ab,ti 70,745 #24. 'exercise'/exp 221,073 #22. 'sports medicine'/exp 15,604 #21. 'sport'/exp 105,553 #20. pricing:de,cl,ab,ti 5,503 #19. price*:de,cl,ab,ti 32,691 #18. financ*:de,cl,ab,ti 189,028 #17. budget*:de,cl,ab,ti 36,234 #16. econom*:de,cl,ab,ti 508,416 #15. cost*:de,cl,ab,ti 687,058 #14. 'cost minimization analysis'/exp 2,581 #13. 'funding'/exp 21,083 #12. 'finance'/exp 9,708 #11. 'hospital cost'/exp 26,703 #10. 'health economics'/exp 631,409 #9. 'health care financing'/exp 11,546 #8. 'health care cost'/exp 210,174 #7. 'financial management'/exp 308,868 #6. 'economic aspect'/exp 1,158,632 #5. 'cost control'/exp 50,194 #4. 'cost of illness'/exp 14,316 #3. 'cost effectiveness analysis'/exp 102,403 #2. 'cost benefit analysis'/exp 65,378 #1. 'socioeconomics'/exp 180,888

#51. 'heart arrest'/exp	56,280	#23. 'athlete'/exp	31,700
#50. 'hypertrophic cardiomyopathy'/exp	18,519		
#49. 'heart arrhythmia'/exp	365,432		
#48. 'wpw':de,cl,ab,ti	1,856		

Table 23 – Search strategy and results for Medline (OVID) and MEDLINE In-Process & Other Non-Indexed Citations (OVID) (results of 24 Dec 2014)

Date	17 September 2014 updated on 24 December 2014		
Date covered	<1946 to Present>		
Search Strategy	1 economics/ (27435) 2 exp "Costs and Cost Analysis"/ (190940) 3 economics, dental/ (1868) 4 exp "economics, hospital"/ (20295) 5 economics, medical/ (8895) 6 economics, nursing/ (4026) 7 economics, pharmaceutical/ (2645) 8 (economic* or cost or costs or costly or costing or price or prices or pricing or pharmacoeconomic*).ti,ab. (447777) 9 (expenditure* not energy).ti,ab. (18551) 10 value for money.ti,ab. (939) 11 budget*.ti,ab. (17832) 12 or/1-11 (577303) 13 ((energy or oxygen) adj cost).ti,ab. (2765) 14 (metabolic adj cost).ti,ab. (822) 15 ((energy or oxygen) adj expenditure).ti,ab. (17450) 16 or/13-15 (20295)	23 swimming.tw. (17662) 24 tennis.tw. (2369) 25 squash.tw. (1176) 26 badminton.tw. (200) 27 soccer.tw. (3901) 28 football.tw. (4479) 29 volleyball.tw. (975) 30 gymnastic?.tw. (1728) 31 basketball.tw. (2075) 32 bicycling.tw. (847) 33 running.tw. (37308) 34 runner?.tw. (6252) 35 swimmer?.tw. (2738) 36 or/18-35 (208163) 37 Death, Sudden, Cardiac/ (11704) 38 exp "Heart Arrest"/ (35755) 39 SCD.tw. (5822) 40 (sudden adj3 death).tw. (32910) 41 Cardiomyopathy, Hypertrophic/ (11850) 42 exp "Arrhythmias, Cardiac"/ (172274)	45 Mass screening/ (86072) 46 exp cardiovascular diseases/di (336291) 47 medical history taking/ (17473) 48 electrocardiography/ (170058) 49 physical examination/ (30498) 50 physical exertion/ (53975) 51 exp diagnostic techniques, cardiovascular/ or exp heart function tests/ (678625) 52 screen*.tw. (459851) 53 electrocardiogra*.tw. (66908) 54 examination.tw. (487740) 55 history.tw. (428378) 56 or/45-55 (2163984) 57 36 and 44 (3436) 58 36 and 56 (45978) 59 44 and 56 (115333) 60 57 or 58 or 59 (160169) 61 17 and 60 (2447) 62 letter.pt. (857710) 63 editorial.pt. (358555)

	17 12 not 16 (572914) 18 exp Sports/ (135430) 19 sports medicine/ (9718) 20 Athletes/ (4110) 21 sport?.tw. (37133) 22 athlet*.tw. (33744)	43 (hypertrophic cardiomyopathy or HCM or Long QT Syndrome or LQTS or Wolff Parkinson White or WPW).ti,ab. (17228) 44 or/37-43 (234130)	64 historical article.pt. (313233) 65 62 or 63 or 64 (1513925) 66 61 not 65 (2383)

Table 24 – Search strategy and results for EconLit (OVID) (results of 24 December 2014)

Date	17 September 2014 updated on 24 December		
Date covered	1886 to November 2014		
Search Strategy	1 (economic* or cost or costs or costly or costing or price or prices or pricing or pharmacoeconomic*).ti,ab. (417845) 2 (expenditure* not energy).ti,ab. (22568) 3 value for money.ti,ab. (510) 4 budget$.ti,ab. (18199) 5 sport?.tw. (1955) 6 athlet*.tw. (427) 7 swimming.tw. (71) 8 tennis.tw. (100) 9 squash.tw. (20) 10 badminton.tw. (0) 11 soccer.tw. (369) 12 football.tw. (874)	13 volleyball.tw. (5) 14 gymnastic?.tw. (6) 15 basketball.tw. (310) 16 bicycling.tw. (38) 17 running.tw. (2537) 18 runner?.tw. (99) 19 swimmer?.tw. (3) 20 (sudden adj3 death).tw. (44) 21 SCD.tw. (20) 22 (hypertrophic cardiomyopathy or HCM or Long QT Syndrome or LQTS or Wolff Parkinson White or WPW).ti,ab. (10) 23 screen*.tw. (2688) 24 electrocardiogra*.tw. (5)	25 examination.tw. (9293) 26 history.tw. (22940) 27 1 or 2 or 3 or 4 (439803) 28 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 (5902) 29 20 or 21 or 22 (72) 30 23 or 24 or 25 or 26 (34446) 31 28 and 29 (6) 32 28 and 30 (305) 33 29 and 30 (1) 34 31 or 32 or 33 (312) 35 27 and 34 (131)

Table 25 – Search strategy and results for Psychinfo (OVID) (results of 24 December 2014)

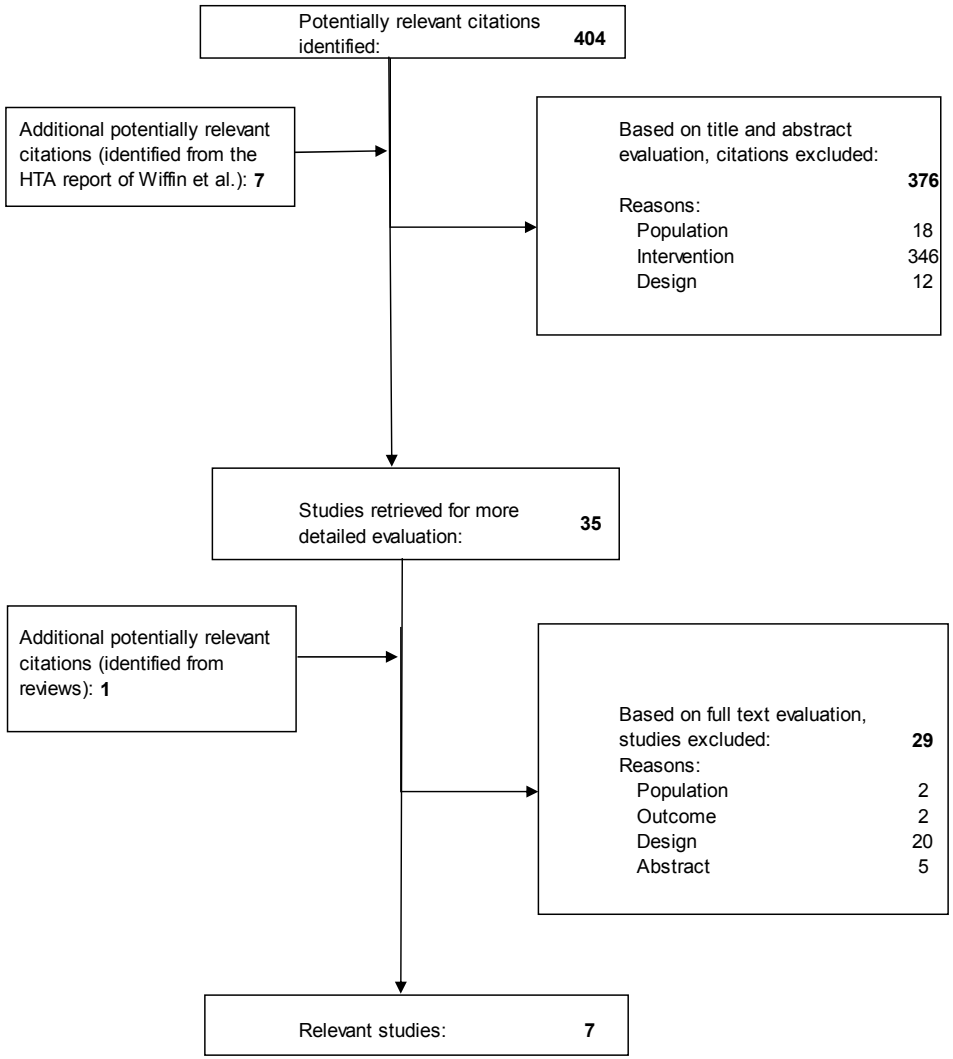
Date	17 September 2014 updated on 24 December 2014
Date covered	1806 to December Week 4 2014
Search Strategy	1 (economic* or cost or costs or costly or costing or price or prices or pricing or pharmacoeconomic*).ti,ab. (149051) 2 (expenditure* not energy).ti,ab. (5547) 3 value for money.ti,ab. (318) 4 budget$.ti,ab. (6080) 5 sport?.tw. (22280) 6 athlet*.tw. (14601) 7 swimming.tw. (4317) 8 tennis.tw. (1192) 9 squash.tw. (71) 10 badminton.tw. (128) 11 soccer.tw. (2063) 12 football.tw. (2606) 13 volleyball.tw. (565) 14 gymnastic?.tw. (665) 15 basketball.tw. (2167) 16 bicycling.tw. (232) 17 running.tw. (12340) 18 runner?.tw. (984) 19 swimmer?.tw. (579) 20 (sudden adj3 death).tw. (2164) 21 SCD.tw. (713) 22 (hypertrophic cardiomyopathy or HCM or Long QT Syndrome or LQTS or Wolff Parkinson White or WPW).ti,ab. (163) 23 screen*.tw. (66314) 24 electrocardiogra*.tw. (1871) 25 examination.tw. (93899) 26 history.tw. (163429) 27 1 or 2 or 3 or 4 (156408) 28 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 (48795) 29 20 or 21 or 22 (2969) 30 23 or 24 or 25 or 26 (308916) 31 28 and 29 (51) 32 28 and 30 (3648) 33 29 and 30 (448) 34 31 or 32 or 33 (4117) 35 27 and 34 (217)

After removal of all duplicates, a total of 404 papers were identified for the years 2012-2014 (Table 20). Finally, based on our inclusion/exclusion criteria, 7 economic evaluations were selected (see Figure 13).

Table 26 – Results of search strategy (second stage)

Database	References identified
	24 December 2014
NHS EED (via the Cochrane Library)	86
Embase	557
MEDLINE(R) and MEDLINE(R) In-Process & Other Non-Indexed Citations (via Ovid)	2383
Econlit (via Ovide)	131
Psychinfo (via Ovide)	217
Total (incl. duplicates)	3374
Duplicates	230
Total (excl. duplicates)	3144
Time limit 2013 - 2014	404

Figure 13 – Flow Chart of the second stage (2013-2014)



Appendix 2.1.3. Data extraction sheet

Template

Table 27 – Data extraction sheet

1	Reference (including all authors)
2	Conflict of interest and/or study funding
3	Country
4	Study question
5	Type of analysis (analytic technique) • e.g. cost-effectiveness analysis, cost-utility analysis, …
6	Design • e.g. Markov model, decision tree, …
7	Population
8	Intervention
9	Comparator
10	Time horizon
11	Discount rate • For costs and/or effects
12	Perspective
13	Costs • Cost items included • Measurement of resource use • Valuation of resource use • Data sources • Currency and cost year • Other aspects…
14	Outcomes • Endpoints taken into account and/or health states • Valuation of health states

	• Treatment effect and Extrapolation • Utility assessment (Quality of Life) • Data sources for outcomes • Other aspects…
15	Uncertainty • Scenario analysis • Sensitivity analysis
16	Assumptions
17	Results • Cost-effectiveness and/or cost-utility (base case) • Scenario analysis • Sensitivity analysis • Other aspects…
18	Conclusion of the authors
19	Reported limitations
20	Own remarks

Appendix 2.1.4. Tests performed in the second round

Table 28 – Details on the second round

Reference	Second round
Fuller 2000[76] (US)	**After H&P:** • Cardiology consultation, treadmill testing, echo, ECG, thallium stress testing, EPS, left heart catheterization, coronary angiography and/or event monitor recording (*percentages not clearly reported*). • % and sensibility/specificity not given. **After ECG:** • Echo for 98%, not clear for the remaining 2%. • Echo: Sensitivity: 80% - Specificity: 100%. **After Echo:**

	• No other test.
Wheeler et al. 2010[81] (US)	• 1 clinic visit and Echo (100%) + exercice stress test or Holter monitor (percentage not reported) + in few cases MRI or commercially available genetic testing (percentage not reported) • Sensitivity and specificity not given
Malhotra et al. 2011[78] (US)	NB: tests not performed in the centre (e.g. some echo) or already performed previously are not taken into account. **After H&P:** • ECG (30%), Echo (30%), EPS (6%) • Sensitivity and specificity not given • Positive results: 6% (5/87) – 0.34% (5/1473) **After ECG:** • *(own calculation) Echo (51%, 227/443), MRI (10%, 44/443), Exercise Stress test (2%, 10/443), Drug studies (1.6%, 7/443), Holter (1.4%, 6/443)** • Sensitivity and specificity not given • Positive results: 1.8% (8/443) – 0.55% (8/1463)
Leslie et al. 2012[3] (US)	<u>For HCM:</u> • Echo for 100% + genetic test for both ECG and H&P positives or for Echo positive, i.e. ≈ *4% based on own calculation*: • Echo (Rodday et al.): o Sensitivity: 60% o Specificity: *around 99.9% (own calculation)** • Genotic test: (Expert opinion) o Sensitivity: 54% o Specificity: not clear, seems *100% (own calculation)** • Combination of tests: *Positive results: 0.13% of people positive at the screening – only 42% of people with a HCM seem detected (19/45), own calculation.** <u>For WPW:</u> • Echo - 100%, followed by ETT or EPS or both in function of results • *People screened positive at the screening seems to be considered as having the disease and the following tests determine the intervention.** <u>For LQTS:</u>

	• ECG on parents + genetic test for 100% + Echo ≈ 46% or exercise stress test ≈ 54%* • Sensibility ans specificity: not given • After 6 months, 5% are confirmed positives, 5% are confirmed negative and 90% remain unconfirmed.
Schoenbaum et al. 2012[80] (US)	• Cardiologist visit (100%) + ECG (100%) + Echo if needed (90% - range 85-95%) • Sensitivity: 90% / range : 80-100% • Specificity: 98% / range : 92-100% • Positive results (own calculation): o *H&P: 2.1 % (from sreening +) / 0.11% (from general population) / FDR = 95%** o *H&P + ECG: 2.7 % (from sreening +) / 0.24% (from general population) / FDR = 74%** o *ECG alone: 3.2 % (from sreening +) / 0.16% (from general population) / FDR = 61.3%** • Annual reclassification of FP to TN: 20% (for the first 3 years).
Halkin et al. 2012[77] (US)	• Echo for 100%, exercise stress test for 82%, Holter for 41%, and MRI, catheterization and/or EPS in 5% (*Corado 2006 and Pellicia 2008*). • Positive results: 22% (resulting in a disqualification rate of 2% per year (9%*22%)
Menafoglio et al. 2014[79] (CH)	**After H&P:** • *Echo for 84%, exercise stress test for 52%, Holter for 20%, Blood pressure monitoring for 24%, MRI for 0%, ECG (with exposure to adenosine, signal averaged or other) for 64%, Genetic testing for 0%, EPS with ablation for 0% (own calculation, on 25 patients positives at screening)** • Sensitivity-sepcificity not given • *Positive results: 28% (7/25, own calculation)** **After H&P + ECG:** • *Echo for 89.6%, exercise stress test for 64.2%, Holter for 29.9%, Blood pressure monitoring for 10.4%, MRI for 7.5%, ECG (with exposure to adenosine, signal averaged or other) for 13.4%, Genetic testing for 1.5%, EPS with ablation for 1.5% (own calculation, on 67 patients positives at screening)** • Sensitivity-sepcificity not given • *Positive results: 16% (11/67, own calculation)**

** Own calculation*

Appendix 2.1.5. Interventions performed

Table 29 – Details on interventions

Reference	Interventions
Fuller 2000[76] (US)	*Not taken into account*
Wheeler et al. 2010[81] (US)	• Cardiac surgey for 1% of positive results • EPS and ablation for 2% of positive results • ICD for 2% of positive results • Medication: % not given
Malhotra et al. 2011[78] (US)	**After H&P:** • *EPS and ablation for 100% of positive results (5/5).** **After ECG:** • *EPS and ablation for 50% of positive results (4/8)** • *Follow-up for 50% of positive results (4/8)**
Leslie et al. 2012[3] (US)	For HCM: • Follow-up: 48% of positive results • Medication: 40% of positive results • ICD: 10% of positive results • Surgery / cathetorization: 2% of positive results For WPW: • EPS and Follow-up: 4% of positive results • EPS and Ablation: 96% of positive results For LQTS: If confirmed positive: • Follow-up and Beta blokers: 100% • ICD: 10% If remain unconfirmed: • Follow-up: 100% • Beta blokers: 50% • ICD: 0%

	Total (own calculation): • *Follow-up only: not clear (around 35%?)** • *Medication: Not clear (around 11.9%?)** • *Ablation: Not clear (around 52%?)** • *ICD: Not clear (around 2.4%?)** • *Surgery / cathetorization: Not clear (around 0.4%?)**
Schoenbaum et al. 2012[80] (US)	If HCM (50%) • Follow-up: 44% of positive results • Medication: 56% of positive results • ICD: 12.5% per year if high risk (around 6%) If arrhythmias (25%) • Follow-up: 5% of positive results • Medication: 95% of positive results • ICD: 12.5% per year if high risk (around 15%) If other heart disease (25%) • Follow-up: 18% of positive results • Medication: 92% of positive results • ICD: 12.5% per year if high risk (around 12%) Overall, 70% of high risk patients get an ICD within 10 years. Total: • *Follow-up: 28% of positive results** • *Medication: 72% of positive results** • *ICD: 1% per year**
Halkin et al. 2012[77] (US)	• Not taken into account in the calculations.
Menafoglio et al. 2014[79] (CH)	• Not taken into account in the calculations. • Only mentioned: EPS with ablation in 9% of patients with cardiac abnormalities (1/11 patients)

** Own calculation*

Appendix 2.2. Cost considerations in a Belgian setting

Appendix 2.2.1. Parameters

Table 30 – Average cost of the second round

Test	%	Unit Cost
Cardiologist visit (102594)	100%	€ 36.74
ECG (475075)	100%	€ 17.77
ECHO (469814)	100%	€ 63.21
Total		€ 117.72

Table 31 – Other parameters

Parameters	Scenario 1	Scenario 2
Number of people screened	1 000 000	1 000 000
Unit cost of screening (ECG + H&P)	€ 60.00	€ 60.00
Average cost of the second round	€ 117.72	€ 117.72
Prevalence of heart disease	0.3%	0.3%
Sensitivity (ECG+H&P)	0.75	0.75
Specificity (ECG+H&P)	0.95	0.70
SCD rate per 1000 000 athlete-year	10	10
Effectiveness of patient management	100% of people detected are saved	100% of people detected are saved
Average life expectancy for people aged 14-34 years old	56 years	56 years

Appendix 2.2.2. Results

Table 32 – Costs

	H&P+ECG (scenario 1)	H&P+ECG (scenario 2)
Screening cost	€ 60 000 000.00	€ 60 000 000.00
Second round cost	€ 6 133 212.00	€ 35 474 922.00
Total cost	€ 66 133 212.00	€ 95 474 922.00

Table 33 – Life-year saved (discount rate of 1.5 and half-cycle correction)

1	7.389163	7.5	7.444581
2	14.55993	14.77833	14.66913
3	21.51713	21.83989	21.67851
4	28.26553	28.68951	28.47752
5	27.84781	28.26553	28.05667
6	27.43627	27.84781	27.64204
7	27.0308	27.43627	27.23353
8	26.63133	27.0308	26.83107
9	26.23777	26.63133	26.43455
10	25.85002	26.23777	26.04389
11	25.468	25.85002	25.65901
12	25.09162	25.468	25.27981
13	24.72081	25.09162	24.90622
14	24.35548	24.72081	24.53814
15	23.99555	24.35548	24.17551
16	23.64093	23.99555	23.81824

17	23.29156	23.64093	23.46624
18	22.94735	23.29156	23.11945
19	22.60822	22.94735	22.77779
20	22.27411	22.60822	22.44117
21	21.94494	22.27411	22.10953
22	21.62063	21.94494	21.78278
23	21.30111	21.62063	21.46087
24	20.98632	21.30111	21.14371
25	20.67617	20.98632	20.83125
26	20.37062	20.67617	20.5234
27	20.06957	20.37062	20.22009
28	19.77298	20.06957	19.92127
29	19.48077	19.77298	19.62687
30	19.19287	19.48077	19.33682
31	18.90923	19.19287	19.05105
32	18.62979	18.90923	18.76951
33	18.35447	18.62979	18.49213
34	18.08322	18.35447	18.21885
35	17.81598	18.08322	17.9496
36	17.55269	17.81598	17.68434
37	17.29329	17.55269	17.42299
38	17.03773	17.29329	17.16551
39	16.78594	17.03773	16.91183

40	16.53787	16.78594	16.6619
41	16.29347	16.53787	16.41567
42	16.05268	16.29347	16.17307
43	15.81545	16.05268	15.93406
44	15.58172	15.81545	15.69858
45	15.35145	15.58172	15.46658
46	15.12458	15.35145	15.23801
47	14.90106	15.12458	15.01282
48	14.68085	14.90106	14.79096
49	14.46389	14.68085	14.57237
50	14.25014	14.46389	14.35702
51	14.03955	14.25014	14.14484
52	13.83207	14.03955	13.93581
53	13.62765	13.83207	13.72986
54	13.42626	13.62765	13.52695
55	13.22784	13.42626	13.32705
56	13.03235	13.22784	13.1301
Total	1087.277	1103.586	1095.431

Table 34 – ICER

H&P+ECG (scenario 1)	H&P+ECG (scenario 2)
€ 66 133 212.00 / 1095.43 = €60 372/LY	€ 95 474 922.00 / 1095.43 = €87 157/LY

APPENDIX 3. CURRENT PRACTICES IN THE BELGIAN SPORTS WORLD

Appendix 3.1. Overview of the pre-participation screening programs in the selection of sports federations

Table 35 – Characteristics of pre-participation screening programs in the 10 most popular sports in Belgium

Sports federation	Pre-participation screening program (+frequency)	Tests	Consequences in case of absence	Specific insurance against SCD	Numbers of SCD*
Gymnastics					
Gymnastiek Federatie Vlaanderen	Compulsory VASO-protocol for A-gymnasts, recommendation to sports clubs to apply the SKA-protocol for B-gymnasts, all other levels no compulsory medical certificate	Currently ECG at 16y for A-gymnasts, from next season on the tests cited in the VASO-protocol, but mainly emphasis on the musculoskeletal examination, ECG less important	No access to competition	no	no SCDs known
Fédération francophone de Gymnastique et de fitness	Only yearly medical certificate for gymnasts in division 1 and 2, other levels no obligation	/	/	no	Since 2008 (year of fusion): no SCDs known
Soccer					
Koninklijke Vlaamse Voetbalbond	Medical certificate, only at time of 1st affiliation	No specific tests	No affiliation	Yes	no SCDs known
Koninklijke Belgische Voetbalbond	No medical certificate needed	/	/	Yes	Dataset from >10y but not yet analysed
Association Liégeoise de Football Amateur	No medical certificate needed	/	/	No	no SCDs known
Vlaamse Zaalvoetbalbond	No medical certificate needed	/	/	Yes	1 SCD known (<35y)

Ligue Francophone de football en salle	No medical certificate needed, starting from 2015-2016 medical certificate needed	No specific tests	No access to competition	No	No SCDs known
Tennis					
Tennis Vlaanderen	No medical certificate, only noncommittal advice on website in case of health problems of doubts	/	/	Probably yes (still in negiotation)	No SCDs known
Association Francophone de Tennis	No medical certificate needed	/	/	Yes	No SCDs known
Athleticism					
Vlaamse Atletiek Liga	No obligation only advice on the documents for affiliation	No specific tests	No consequences	Yes	2 SCDs (known heart patients >35j)
Ligue Belge Francophone d'Athlétisme	Medical certificate, 1x/year	No specific tests	No access to competition	no	No SCDs known
Volleyball					
Vlaamse Volleybalbond	Medical certificate, only by affiliation or in case of absence during 2 seasons	No specific tests	No affiliation	Yes	No SCDs known
Association Interprovinciale Francophone de Volleyball	No info	No info	No info	No info	No info
Cycling					
Wielerbond Vlaanderen	Medical certificate needed for competitive athletes, 1x/year	Modified SKA questionnaire (with UCI questions in), physical examination and ECG yearly,	No access to competition	Yes	No SCDs known

		Advice for exercise test at 17-18y			
Vlaamse Wielrijdersbond	No medical certificate	/	/	Yes	No SCDs known
Vlaamse Bond voor Rijwieltoerisme	No medical certificate	/	/	Yes	No SCDs known
Fédération Cycliste Wallonie-Bruxelles	Only medical certificate for competitive athletes, 1x/year	No specific tests, questionnaire from UCI	No access to competition	Yes	No SCDs known
Swimming					
Vlaamse Zwemfederatie	Medical certificate for competitive athletes (only by affiliation), no obligation for non-competitive athletes, only by affiliation	No specific tests	No access to competition	Yes	No SCDs known
Fédération Francophone Belge de Natation	No info	No info	No info	No info	No info
Basketball					
Vlaamse Basketballiga	Medical certificate needed for all members, 1x/year	No specific tests	No access to competition	Yes	No data gathered
Association Wallonie-Bruxelles de Basketball	Only medical certificate for competitive athletes, 1x/year	No specific tests	No access to competition	Yes	No SCDs known
Badminton					
Badminton Vlaanderen	No medical certificate	/	/	Yes	3 SCDs >35j
Ligue Francophone Belge de Badminton	No medical certificate	/	/	No	1 SCA<35y
Dance					
Dansliga Sportfederatie	No medical certificate, but referral to VASO-protocol	/	/	Yes	No SCDs known

Belgische DansSportFederatie	Medical certificate by affiliation and every 2y	No specific tests	No access to competition	No	No SCDs known
Federatie dans en sport	No obligation but determined by sports clubs, frequency between 2 and 5 years	No specific tests	None	Yes	No SCDs known
Association Francophone des Clubs de Danse	Medical certificate, 1x/year	No specific tests	No affiliation	No	No SCDs known

**The mentioned data on number of SCD are not representative for the total number of SCD in Belgium. These data should be interpreted with great caution.*

APPENDIX 4. DATA SUPPORTING CLINICAL EFFECTIVENESS SUMMARY

In the final conclusion of this report, a summary of the clinical effectiveness of cardiovascular pre-participation screening is presented in a format that is accessible for a non-scientific audience. For the sake of clarity, rounded numbers are provided. Below, justification for those numbers are depicted.

- 1 million = 1/3 of 2.8 million 14-34 years-old Belgians, i.e. those practicing organised sports or participating in mass sports events
- Up to 10 SCDs per year: cf. Table 1
- WPW prevalence: Rodday data (cf. Table 3): 1360/million. ECG sensitivity: >0.90.
- HCM prevalence: Rodday: 450/million. ECG sensitivity 0.85.
- 50.000 à 300.000 (in fact 52.100 to 301.350): specificity of combined Hx/P/ECG = 0.70 or 0.95 (2 scenarios).
- “above 2000” = 2250 true positives (sensitivity of combined Hx/P/ECG = 0.75); the number 0.75 is a mean of 1.00 and 0.50. These numbers may somewhat underestimate the true sensitivity, given the almost perfect performance of the ECG in diagnosing WPW. Following this reasoning, the number 2250 below might in fact be 2500. Given the wide uncertainty surrounding all the numbers provided, this will not affect the final estimations. Furthermore, diagnosing WPW has no clear impact on overall mortality in the population considered here.
- Performance of downstream investigations hyper-optimistically put at sensitivity of 1.0 and specificity of 0.99. Hyper-optimistic number of false positives: 498 out of 52,100 or 2250 out of 301,350. Hence “a few thousands”.
- “above 2000” + “a few thousands” = 5000.

	First round specificity 0,70	First round specificity 0,95
	1000000	1000000
Prevalence	0,30%	0,30%
Affected	3000	3000
Not affected	997000	997000
sens. 1st round	0,75	0,75
spec. 1st round	0,7	0,95
TP	2250	2250
FN	750	750
TN	697900	947150
FP	299100	49850
sens. 2nd round	1	1
spec. 2nd round	0,99	0,99
TP	2250	2250
FN	0	0
TN	296109	49352
FP	2991	498,5

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