

Protecting the stars of tomorrow: Do international cardiovascular preparticipation screening policies account for the paediatric athlete? A systematic review and quality appraisal.

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## **ABSTRACT**

**Objective:** 1) Identify and review current policies for the cardiovascular screening of athletes to assess their applicability to the paediatric population; and, 2) evaluate the quality of these policy documents using the AGREEII tool.

**Design:** Systematic review and quality appraisal of policy documents.

**Data sources:** A systematic search of PubMed, MEDLINE, Scopus, Web of Science, SportDiscus, and CINAHL.

**Eligibility criteria for selecting studies:** An article was included if it was a policy/position statement/guideline/consensus or recommendation paper relating to athletes and cardiovascular pre-participation screening.

### **Results and Summary**

Of the 1,630 articles screened, 13 met the inclusion criteria. Relevance to paediatric athletes was found to be high in three (23%), moderate in 6 (46%) and low in 4 (31%), and only two provide tailored guidance for the athlete aged 12-18 years. A median 5 related citations per policy investigated solely paediatric athletes, with study designs most commonly being retrospective (72%). AGREEII overall quality scores ranged from 25%-92%, with a median of 75%. The lowest scoring domains were Rigour of development (median 32%) Stakeholder involvement (median 47%), and Applicability (median 52%).

### **Conclusion**

Cardiac screening policies for athletes predominantly focus on adults, with few providing specific recommendations for paediatric athletes. The overall quality of the policies was moderate, with more recent documents scoring higher. Future research is needed in paediatric athletes to inform and develop cardiac screening guidelines, to improve the cardiac care of youth athletes.

**Abstract word count:** 232

**Key words:** Preparticipation screening, Cardiovascular, Sudden cardiac death, Athletes, Paediatric, Guidelines.

**What is already known?**

- Cardiovascular screening of young athletes is supported by most leading sporting organisations and governing bodies around the world.
- Numerous policy documents exist yet guidance on how to perform cardiac screening has predominantly focused on adult athletes.
- Clear evidence-based guidance on how to perform cardiac screening in paediatric athletes is paramount to successful implementation and ensuring the health and well-being of this population, yet no methodological appraisal of current policies has been conducted.

### **What are the new findings?**

- Available policy documents for the cardiovascular screening of athletes typically has low to moderate relevance to the paediatric athlete, with only two policies providing specific guidance for the paediatric athlete: highlighting the need for new research and specific recommendations for this population.
- Data referenced within the policies were mostly derived from cohorts of adults only or combined cohorts of adult and paediatric athletes, suggesting caution is warranted in applying findings specifically to the paediatric athlete.
- The overall quality of the policies had a median score of 75%, with the lower scoring domains being rigour of development and stakeholder involvement. Methodology that follows the AGREE II instrument is needed in the planning and creation of athlete cardiac screening guidelines.

## INTRODUCTION

In the search to identify underlying cardiac pathology that may predispose an individual to sudden cardiac death (SCD), pre-participation cardiovascular screening of young athletes has long been advocated by the American Heart Association (AHA) and the European Society of Cardiology (ESC).<sup>1</sup>

<sup>2</sup> Among athletes with underlying pathology, exercise may trigger fatal arrhythmias, resulting in a 2.8 to 5.3-times greater incidence of SCD than their non-athletic peers.<sup>3 4</sup> SCD can affect young athletes of any age. While studies have typically reported an incidence of 1.25 to 2.5 per 100,000,<sup>5</sup> a recent study of UK adolescent footballers reported a sudden cardiac arrest (SCA) incidence of 6.8/100,000 person-years.<sup>6</sup>

Over the past decade there has been a notable increase in the awareness of SCD, the adoption of preparticipation cardiovascular screening, and the professionalisation of youth sport.<sup>7</sup> Today, most major sports organisations and governing bodies are advocating for or mandating cardiac screening. However, debate surrounds what defines an athlete, who should be screened, and from what age.

Children are not “miniature adults”, and age specific normative values for cardiac testing are necessary to increase diagnostic accuracy as adult recommendations cannot be applied unequivocally.<sup>8</sup> Paediatric athletes do show physiological remodelling of the heart in response to exercise;<sup>9</sup> but owing to factors such as maturational development and variable disease penetrance and aetiology, the grey zone between physiological and pathological adaptation may be further blurred. In contrast to adults, children suffering a SCD event are more often found to have a structurally normal heart.<sup>10</sup> This may be a result of the growth and hormonal changes needed during adolescence to unmask genetic cardiac disease. In other words, prior to full biological maturation it could be more difficult to identify the early phenotypic manifestation of associated conditions.<sup>8</sup>

Concerns exist around the pre-participation evaluation of young athletes and whether current guidelines are suitable. To date the guidance on how to perform cardiac screening in athletes has predominantly used data from elite adult athletes.<sup>8</sup> Numerous international sports and medicine organisations have issued guidance on athlete screening, with variable degrees of overlap in the recommendations. Although age is now tentatively considered within athlete ECG<sup>11</sup> and echocardiogram guidelines,<sup>12 13</sup>

few policies consider paediatric athletes separately, and fewer target children specifically. The primary aim of this study was to review the current recommendations, guidelines, consensus, and position papers on athlete screening, with a focus on their relevance to the paediatric population. The secondary aim was to evaluate the quality of these policies using the AGREEII tool and identify potential areas of improvement.

## **METHODS**

### **Study design**

This systematic review was designed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.<sup>14</sup> University of York/PROSPERO was contacted prior to starting the review process, where they cited that registration was not required.

### **Study eligibility**

Inclusion criteria were: 1. Pertaining to athletes, defined as “One who participates in an organised team or individual sport that requires regular competition against others as a central component, places a high premium on excellence and achievement, and requires some form of systematic (and usually intense) training”;<sup>15</sup> 2. Policy, guidelines, scientific or consensus statement or recommendations paper by a medical organisation or sports governing body; 3. Relating to pre-participation screening; 4. English language.

Exclusion criteria were: 1. Studies including individuals that do not meet the criteria for athlete status; 2. Prior versions of policies by an organisation regarding the same population that have since been updated; 3. Guidelines for non-athlete screening (including individuals with cardiovascular disease or general population); 4. Non English language papers; 5. Editorials, letters to the editor, narrative reviews, theses, unpublished work, and conference abstracts; 6. Papers not clearly authored, endorsed or supervised by a medical organisation or governing body.

### **Database search**

PubMed, MEDLINE, Scopus, Web of Science, SportDiscus, and CINAHL databases were searched from inception to April 2020 and repeated in October 2021. Search terms were mapped to relevant MeSH terms or subject headings relating to the concepts of “guidelines”, “athletes”, “cardiology”, and

“screening”. Terms within each section were combined with the Boolean operator ‘OR’, and then concepts were combined with the ‘AND’ operator to produce the search strategy (The full search strategy (April 2020 output) is available in the supplementary material Table 1).

### **Study selection and data extraction**

Title, abstract and full text screening were performed using the Covidence® platform (Veritas Health Innovation Ltd, Melbourne, Australia), independently, by two reviewers (N.R.R and D.M.D.). Data extraction was conducted independently by N.R.R and D.M.D. and included paper and methodology description, data on the target population, the relevance to paediatric athletes, and level of evidence the policies are based on.

### **Relevance to paediatric athletes**

Relevance to paediatric athletes, defined as under 18 years old or mostly of high-school age, was evaluated by several methods, for each guideline, by two researchers (N.R.R and D.M.D.). An overall relevance grading was based on the rounded average of three items, scored from 1-3. An average of 0-1 was considered “Low relevance”, 1.1-2 was “Moderate relevance” and 2.1-3 was “High relevance” to paediatric athletes.

First, we determined whether the policies targeted the paediatric athlete – 1 (no stated mention of paediatric athletes in the policies remit), 2 (included in a broader age range) and 3 (paediatric athletes included in the policies remit). Second and third scores counted the number of paediatric references related to the topics of SCD and screening. Intervals were chosen arbitrarily, based on average number of references – 1 (1-4 references), 2 (5-9 references) and 3 ( $\geq 10$  references). The topic of “SCD” comprised all references evaluating the epidemiology, incidence and/or aetiology of SCA/SCD., “Screening” comprised all references evaluating the implementation, suitability and/or effectiveness of screening using any assessment modality. A reference could be assigned to both topics, if appropriate. The total number of original research papers referenced that included any paediatric athletes were counted, and a percentage from the total number of references per policy was calculated.

To evaluate the type of referenced studies, all original research papers were classified according to study design: case series (targeted population, no comparator), cross sectional, case-control, retrospective cohort, prospective observational, interventional (including randomised clinical trial),

systematic reviews/meta-analysis and economical simulation studies. Prospective observational studies were not further categorised, to avoid over-complicating the classification. The total mentions of paediatric related terms (“child\*”, “adolescen\*”, “p(a)ediatric”, “boy\*/girl\*”, “(high)school”) were counted.

All classifications were reviewed by N.R.R and D.M.D. In all steps above, a third reviewer (G.P.) arbitrated disagreements, and any discrepancies were resolved through team consensus.

### **Guideline quality assessment**

The quality and variability of policies were assessed independently by N.R.R. and D.M.D. using the Appraisal of Guidelines for Research & Evaluation II (AGREE II) tool.<sup>16</sup> This tool evaluates 23 items (Q1-Q23) grouped in 6 domains (Scope and Purpose, Stakeholder Involvement, Rigour of Development, Clarity of Presentation, Applicability, Editorial Independence), and an overall score, ranked on a 7-point scale. Domain scores from individual appraisers are summed and then scaled to a percentage of the maximum possible score for that domain using the formula (Reviewer 1 score + Reviewer 2 score - Minimum possible score) / (Maximum possible score–Minimum possible score)\*100.

### **Statistical analysis**

Categorical data are presented as frequency and proportion, while continuous values (including AGREE II domain scaled scores), are presented as median and range. Data were summarized according to the Synthesis without meta-analysis (SWiM) guidelines.<sup>17</sup> Descriptive statistical analyses were conducted using STATA/SE 12 (StataCorp LP, College Station, TX).

## **RESULTS**

After abstract and full text screening, 13 policies were included (Figure 1).<sup>2 18-29</sup> These were from a variety of organisations, ranging from general medicine, paediatrics, cardiology, sports medicine and athletic committees. Most were published after 2010 (n=10), two between 2000-2010 and one before 2000. Only one used systematic literature search methodologies, only two had paediatric specific recommendations, 8 offer recommendations on the frequency of preparticipation screening, with most discussing medical history, physical exam and/or ECG as screening modalities. Due to a lack of

interventional randomised control trials or meta-analyses of randomised control trials, none of the policies have a high grade level of evidence (see Table 1).



**Table 1.** Characteristics of included guidelines

<b>Document, year of publication</b>	<b>Organisation</b>	<b>Document type</b>	<b>Systematic search</b>	<b>Main target professionals</b>	<b>Athlete level</b>	<b>Age range</b>	<b>Age specific paediatric recommendations</b>	<b>Screening tools recommended</b>	<b>Frequency of screening</b>
Ghorayeb et al., 2019	Brazilian Society of Cardiology and Brazilian Society of Exercise and Sports Medicine	Guideline paper	No	Not stated	Amateur athletes and professional athletes	12-35 years or older	Yes	Medical history; physical exam; ECG	Before onset of training and then periodically
Preparticipation Physical Evaluation 5th edition, 2019	American Academy of Pediatrics (joint publication with multiple Organisations)	Monograph	No	Athlete's primary care provider	Athletes in organized sports or vigorous physical activities	Middle school, high school and college age	No	Medical history; physical exam	Comprehensive evaluation every 2 to 3 years; yearly update
Johri et al., 2019	Canadian Cardiovascular Society and Canadian Heart Rhythm Society	Position statement	No	Specialists and allied health professionals	Competitive or professional athletes	Not stated	No	Medical history; physical exam	Not stated
Drezner et al., 2017	American Medical Society for Sports Medicine	Position statement	No	Primary care sports medicine physician	Young competitive athletes	12-35 years	No	Physician autonomy	Every 1-3 years
Fritsch et al., 2017	Association of European	Recommendation paper	No	Not stated	Young competitive athletes	Not stated	No	Medical history; physical exam; ECG	Every 2 years

	Paediatric Cardiology								
Mont et al., 2017	European Heart Rhythm Association and European Association of Preventive Cardiology	Position statement	No	Not stated	All athletes, regardless of level	Any age	No	Medical history; physical exam; ECG	Not stated
Hainline et al., 2016	National Collegiate Athletic Association	Consensus statement	No	Sports institutions medical team	Collegiate student athletes	Collegiate student age	No	Medical history; physical exam	Yearly
Conley et al., 2014	National Athletic Trainers' Association	Position statement	No	Medical doctors, athletic trainers	All athletes, regardless of level	Not stated	No	Medical history; physical exam	At each new level of participation
Maron et al., 2014	American Heart Association and the American College of Cardiology	Scientific statement	Yes	Not stated	General population to competitive athletes	12-25 years	No	Medical history; physical exam	Not stated
Mahmood et al., 2013	American College of Preventive Medicine	Position statement	No	Not stated	High school or collegiate athletes	High school or collegiate age	No	Medical history; physical exam	Not stated
Ljungqvist et al., 2009	International Olympic Committee	Consensus statement	No	Sports team physicians	Elite athletes	Not stated	No	Medical history; physical exam; ECG	Not stated

Corrado et al., 2005	Working Groups of the European Society of Cardiology	Consensus statement	No	Sports cardiology or sports physician	Competitive or professional athletes	12-35 years	No	Medical history; physical exam; ECG	Every 2 years
American Medical Association report, 2014	American Medical Association	Board of trustees report	No	Medical doctors	Adolescents participating in sports	Adolescents, not further defined	Yes	Medical history; physical exam	Yearly

### **Relevance to paediatric athletes**

Among all 13 included policies, there was considerable variance in the relevance to paediatric athletes. Ten (77%) had low-moderate relevance (Figure 2A). Four included the paediatric athlete within their remit, but just two provided specific direction for screening athletes between 12-18 years. Only one policy cited more than 10 articles based on paediatric only populations for both topics of athlete screening and SCD.

Citations of original related research including any paediatric athletes in their cohort ranged from 5-80 (median 27 per policy), a number representing between 10-53% (median 26%) of the total references in each policy (Figure 2B). Of these, the most commonly referenced studies were retrospective in design, (median 16 per policy; range 2-62), followed by prospective observational (median 7 per policy; range 1-13), (Table 2). The study design of this referenced paediatric original research differed considerably by topic and policy (Figure 3A).

A median of 5 citations per policy comprised only paediatric athletes (range 0-26). While comparable to that of adult only studies (median 4, range 0-24), it was lower than those using cohorts combining both paediatric and adult athletes (median 22 per policy, range 0-54), (Figure 3B). The median proportion of paediatric only references was higher in the screening topic (19%, range 0-100%) than in the SCD topic (5%, range 0-14%).

Mention of terms related to paediatrics ranged from 1-183 per policy (median 10 per policy), with the most common terms used being “adolescent” and “school/high school” (median 2 per policy; range 0-83). The terms “paediatric” and boy/girl were used rarely (median 0 per policy; range 0-11).

All data related to the relevance of paediatric athletes, type of research referenced, study designs and topics are summarized in Table 2.

<b>Table 2.</b> Relevance to paediatric athletes													
	Ghorayeb et al., 2019	Preparticipation Physical Evaluation 5th edition, 2019	Johri et al., 2019	Drezner et al, 2017	Fritsch et al, 2017	Mont et al, 2017	Hainline et al, 2016	Conley, et al 2014	Maron et al, 2014	Mahmood et al, 2013	Ljungqvist et al, 2009	Corrado et al, 2005	American Medical Association report, 1994
<b>Overall relevance to paediatric population</b>	Moderate	High	Low	High	Low	Moderate	Low	Moderate	High	Moderate	Low	Moderate	Moderate
<b>Targeted at paediatric athletes</b>	Included in remit	Included in remit	Not stated	Included in a broader age range	Included in a broader age range	Not stated	Not stated	Not stated	Included in a broader age range	Included in remit	Not stated	Included in a broader age range	Included in remit
<b>Total references</b>	324	223	68	118	26	180	76	138	275	23	122	36	37
<b>Citations of original research including only adult athletes (n, % from total)</b>	7 (2)	3 (1)	6 (9)	9 (8)	2 (8)	24 (13)	4 (5)	4 (3)	16 (6)	2 (9)	2 (2)	0 (0%)	1
<b>Citations of original research including any paediatric athlete (n, % from total) ‡</b>	37 (11)	51 (23)	27 (40)	62 (53)	5 (19)	47 (26)	22 (29)	33 (24)	80 (29)	11 (48)	12 (10)	16 (44)	7 (19)
<b>Number of citations evaluating SCD</b>	15	23	15	34	4	21	9	11	35	7	5	12	0
<b>Number of citations evaluating Screening</b>	23	30	14	31	2	28	13	24	52	8	9	5	7
<b>Reference study type</b>													
<b>Retrospective studies (n, % from ‡)</b>	28 (76)	39 (76)	16 (59)	45 (73)	2 (40)	32 (68)	14 (64)	24 (73)	62 (78)	8 (73)	6 (50)	14 (88)	5 (71)
SCD	8	16	9	25	2	13	5	8	24	5	2	10	0
<i>Case series</i>	5	5	6	10	1	6	2	3	13	1	1	6	0
<i>Cross sectional</i>	2	9	2	14	1	6	3	4	10	3	0	3	0

<i>Cohort</i>	0	2	1	1	0	0	0	1	1	1	1	0	0
<i>Case-control</i>	1	0	0	0	0	1	0	0	0	0	0	1	0
Screening	20	24	7	21	0	19	9	16	41	5	4	4	5
<i>Case series</i>	1	1	1	0	0	0	0	1	3	1	0	0	0
<i>Cross sectional</i>	5	19	5	12	0	9	4	14	29	3	4	4	5
<i>Cohort</i>	11	3	1	8	0	8	5	0	7	1	0	0	0
<i>Case-control</i>	3	1	0	1	0	2	0	1	2	0	0	0	0
<b>Prospective observational (n, % from ‡)</b>	7 (19)	10 (20)	7 (26)	13 (21)	2 (40)	8 (17)	7 (32)	5 (15)	13 (16)	2 (18)	4 (33)	2 (13)	1 (14)
SCD	5	6	4	8	2	6	4	2	9	1	1	2	0
Screening	2	5	4	6	1	3	3	4	5	2	3	1	1
<b>Systematic reviews/meta-analyses/interventional (n, % from ‡)</b>	2 (5)	1 (2)	4 (15)	4 (6)	1 (20)	3 (6)	1 (5)	2 (6)	1 (1)	1 (9)	2 (17)	0 (0)	0 (0)
SCD	2	1	2	1	0	1	0	1	1	1	1	0	0
Screening	1	0	3	4	1	3	1	2	1	1	2	0	0
<b>Economical simulation (n, % from ‡)</b>	0 (0)	1 (2)	0 (0)	0 (0)	0 (0)	4 (9)	0 (0)	2 (6)	4 (5)	0 (0)	0 (0)	0 (0)	1 (14)
<b>Population included by age*</b>													
<b>Paediatric only (n, % from ‡)</b>	2 (5)	17 (33)	3 (11)	19 (31)	0 (0)	11 (23)	5 (23)	11 (33)	26 (33)	3 (27)	2 (17)	3 (19)	7 (100)
SCD (n)	1	3	2	10	0	2	2	1	8	1	0	2	0
Screening (n; (Classification))	1	15	1	10	0	9	3	10	20	3	2	1	7
<b>Combined ages (n, % from ‡)</b>	35 (95)	34 (67)	24 (89)	43 (69)	5 (100)	36 (77)	17 (77)	22 (67)	54 (68)	8 (73)	10 (83)	13 (81)	0 (0)
SCD	14	20	13	24	4	19	7	10	27	6	5	10	0
Screening	22	15	13	21	2	19	10	14	31	5	7	4	0
Mentions of paediatric related terms	12	183	3	19	3	10	2	19	98	4	1	4	74
Child	4	30	0	3	2	2	0	3	36	0	0	0	0
Adolescent	4	66	0	5	1	2	0	4	15	0	0	1	51

Paediatric	0	1	0	2	0	2	0	0	6	0	0	0	1
Boy/girl	1	3	0	0	0	0	0	0	0	0	0	0	11
School/high school	3	83	3	9	0	4	2	12	41	4	1	3	11
*adult only studies shown in Figure 3B only, for comparison to paediatric and overlap paediatric-adult figures.													
‡ Citations of original related research including any paediatric athlete													

## **Quality assessment with the AGREE II tool**

The overall quality of the policies was between 25%-92%, with a median score of 75%. The higher scoring domains were Scope and Purpose (53-97%, median 83%) and Clarity (47-100%, median 81%), with most guidelines being well presented in a clear and concise manner. The lowest scoring domains were Rigour of Development (14%-78%, median 32%) and Stakeholder Involvement (22-97%, median 47%), owing to almost no use of systematic search methodologies and a lack of inclusion of non-medical stakeholders (All domain scores are shown in Figure 4 and Supplementary material Table 2).

### *Domain 1. Scope and purpose*

The median score in this domain was 83% (53-97%), with only one policy paper<sup>19</sup> being assessed with a low score of 53%. Lower scores were generally due to lack of clarity in defining the target population (Q3), especially in clearly defining the target age (with uses of un-descriptive terms such as “young” or “student”) or the type of athletic participation (amateur versus professional), but less so due to defining the clinical question or objectives discussed (Q1 and Q2).

### *Domain 2. Stakeholder Involvement*

The median score in this domain was 47% (22-97%), with two policies achieving high scores, of over 80%.<sup>21 29</sup> Various medical professions were well represented, still limited to sports medicine/connected professions in some cases (Q4). The input from public, patient or non-medical groups was sought in just 3/13 guidelines (Q5), while 6/13 policies had issues in clearly defining the professional target group (Q6).

### *Domain 3. Rigour of development*

The median score in this domain was 32% (14-78%), with one policy scoring a satisfactory 78%.<sup>24</sup> Only one policy described a systematic approach to the literature search (Q7), presented clear criteria for selecting included studies (Q8) or the methodology for selecting the recommendations (Q10). The strengths and limitations of the discussed literature was generally mentioned in all policies, but only in 3/13 was this written in a clear, separate subsection (Q9). Benefits, risks and side effects were discussed in most policies, and in 5/13 this was conducted in clearly identifiable sections or paragraphs (Q11). Just two policies linked key references or offered level of evidence data for each individual



recommendation. In all other cases it was either generally discussed in the text or otherwise difficult to assess (Q2). Although most of the policies were published in peer review journals, in just two instances was external peer review before publication explicitly mentioned in the manuscript. (Q13). None of the included policies offered any clear framework for updates, at most suggesting discussions for future directions (Q14).

#### *Domain 4. Clarity of presentation*

The median score in this domain was 81% (47-100%). Two policies scored low, under 70%,<sup>27 28</sup> attributed to ambiguously formulated recommendations (Q15), which were not clearly separated from the majority of the discussions (Q17). An issue present in most policies was the lack of an exhaustive discussion regarding all screening modalities and relevant issues, but rather limiting this to physical examination and ECG (Q16).

#### *Domain 5. Applicability*

The median score in this domain was 52% (27-77%), with two policies scoring satisfactory.<sup>21 26</sup> In only 3/13 policies were facilitators and barriers to implementing the recommendations clearly discussed in a separate section, while in most other statements this featured throughout the text (Q18). Tools for aiding recommendations were either provided or referenced in 7/13 policies (Q19). There was a separate discussion of the resource utilization in 6/13 of the policies, with most other still tangentially discussing the topic (Q20). In only two guidelines was there any mention of mechanisms for overseeing the implementation of the recommendations (Q21).

#### *Domain 6. Editorial independence*

The median score in this domain was 67% (17-100%), with three policies scoring very low.<sup>2 25 28</sup> The scores varied significantly due to missing editorial independence statements from the supporting organisation in 8/13 cases (Q22) and incomplete documentation of the conflict of interest of authors in 4/13 cases (Q23). As such, these scores did not reflect a lack of editorial independence but rather a lack of uniform documentation in the published manuscript.

## **DISCUSSION**

In this systematic review of available policies published in the peer reviewed literature for the cardiovascular screening of athletes, we demonstrate that out of the 13 included, just 3 were found to be highly relevant to paediatric athletes. The quality judgement of the policy, as evaluated using the AGREE II tool, varied greatly, with particular improvement needed in the Rigour of Development, Applicability, and Stakeholder involvement domains, which concurs with the limitations found regarding paediatric relevance. Newer documents did score better overall, but as these findings show, more work is required both in conducting original research and the methodology to develop consensus to improve athlete screening recommendations for paediatrics (Central figure).

### **Relevance of current policies to paediatric athletes**

*“We recognise that young competitive athletes (<18 years) require specific expertise in the evaluation, interpretation of findings and management”.*<sup>26</sup>

This statement from the International Olympic Committee in 2009 shows that for over a decade the need for paediatric specific guidance has been apparent. Yet within our review in 2022, there is still ambiguity with the applicability of screening recommendations for this age group, as only two provide specific guidance for athletes aged 12-18 years. The remaining documents either failed to state the age range or included one that incorporated both children and adults. One reason for the limited tailoring of the policies to the paediatric athlete is the sparse availability of research data to provide evidence-based guidance. Concurrently, there is also a lack of a systematic approach in the literature search, with seven policies citing less than 4 original articles investigating cardiac screening in this age group.

### **Quality appraisal of current screening policies**

The 13 policies we identified in the present review performed generally well in each domain, yet the limited search strategies used within the policies was highlighted in finding that “rigour of development” was the poorest performing domain within the AGREE II analysis. This domain assesses the processes used to gather and synthesize the evidence, and the methods used to formulate the recommendations. When formulating recommendations, the Institute of Medicine describes literature searching as the key step in developing valid guidelines.<sup>30</sup> Yet the search strategy was reported in only

one policy within our review. Not performing a systematic review to guide policy creation may ultimately introduce certain biases, such as selective citation, producing less valid, reliable, and accurate recommendations.<sup>31</sup> The 2014 AHA policy,<sup>24</sup> which according to the organisations methodology manual, did include a systematic search, cites 20 articles directly relating to the screening of paediatric athletes, emphasising that more recent policies have failed to account for sections of the available literature.

We also identified several aspects for improvement, particularly in relation to document development and rigour, applicability, and stakeholder involvement. Prior to formulating any document, a key question here is who should participate.<sup>32</sup> Despite dissent and discussion being stated as the foundations on how we improve science,<sup>33</sup> just one policy<sup>22</sup> reported areas of disagreement among the panel. There have also been calls for panel members to disclose their personal leaning on contentious issues prior to consensus meetings.<sup>34</sup> One policy that did this was that of Drezner *et al*,<sup>27</sup> by purposefully selecting the panel to provide a balanced view on ECG utilisation. Beyond panellists with different views, a multi-disciplinary panel is also necessary to limit any issues of equity, diversity and inclusivity.<sup>32</sup> This is particularly important within athlete screening policies where there are numerous stakeholders involved. Yet only the AHA policy addressed the important role of both patient and legal representation.<sup>24</sup>

### **Do we need separate paediatric guidelines?**

Up to 40% of all children are registered in official teams,<sup>35</sup> with the paediatric age group representing the largest population of all athletes. Despite this representation, of the research cited in the policies that included paediatric athletes, 77% used cohorts that included a combined sample of paediatric and adults. This action of grouping young athletes into the broad category of 12-35 years is common not only in the available literature but the policies themselves. Thus, care must be taken in applying these findings to the specific paediatric cohort, as there is no indication that they are transferable, and may even mask potentially important nuances that arise in the maturing athlete.

Child athletes can develop profound physiological cardiac remodelling;<sup>9</sup> and like the adult athlete, create the diagnostic dilemma of differentiating between physiological and pathological adaptation. Age is now considered within the athlete ECG<sup>11</sup> and echocardiogram guidelines,<sup>12 13</sup> and although we know that the paediatric athlete differs in several aspects to the mature adult athlete, no policy discussed in any detail the nuances of screening this population; with mention of paediatric related terms being as infrequent as one per policy.

Adolescence is a period when genetic cardiac disease may present for the first time, however, the beginning of the adolescent growth spurt can range from 9.5-16 years,<sup>36 37</sup> whilst skeletal maturation may not be reached until the age of 19 years or beyond.<sup>38</sup> Together with the unpredictability of disease penetrance during adolescence,<sup>39</sup> it means that a single screening before this time may allow a cardiac condition to go undetected.<sup>6</sup>

Additionally, to ensure diagnostic accuracy for this age group, age specific normative values and tailored approaches to disease management are also needed.<sup>8 40</sup> One such consideration is that of biological maturation. While not acknowledged within any screening policy, using biological instead of chronological age may improve diagnostic accuracy substantially.<sup>41</sup> While caution is warranted in applying ECG and echocardiographic criteria without due attention to maturational status, its assessment is not without challenges. The Tanner assessment is easily conducted but has ethical and child-protection considerations, while the wrist X ray of biological assessment elicits exposure to radiation<sup>42</sup> and would require increased resource utilisation, introducing potential barriers to access.

Within this systematic review, just the policy of Ghorayeb et al,<sup>20</sup> provides tailored guidance specifically for the paediatric athlete with the recommended use of ECG. These recommendations from Brazil however do not discuss their tailored approach to age in detail, and cite just one article related specifically to the screening of paediatric athletes.<sup>43</sup> It is outside the scope of this review to assess the quality of the research cited, however we found the majority of cited literature to be of a cross sectional design. With robust data lacking that asserts cardiac screening to be effective in reducing the incidence of SCD, in not only paediatric but adult athletes, the debate between screening approaches continues. This is exemplified by a clear continental split in policies recommending (5) versus not recommending

(7) the use of ECG with the screening remit. In such situations where there is conflicting evidence, it has led to current policies generally coming from consensus or position statements, as opposed to clinical practice guidelines.<sup>44 45</sup>

### **Moving forward**

As found in our review, the timing of initial cardiac screening varies significantly depending on the policy, with a potential 9-year discrepancy. Several policies recommend cardiac screening to begin at the onset of competitive activity, meaning athletes as young as 5-8 years old would be screened.<sup>46-48</sup> Overall, the lack of consensus is evident in our systematic review, with little guidance on the topic. In addition, some recommendations altered their own lower age range from 14 to 12 years old, despite no change in the research base.<sup>11 49</sup>

With policies recommending screening to be undertaken in the preadolescent years, a thorough understanding of the issues relevant to paediatric cardiac screening is required, including the rapid yet highly variable pubertal changes, legal responsibilities of sports governing bodies, and managing the complex relationship between physician, parent, and patient. Both the ESC<sup>50</sup> and AHA<sup>51</sup> have outlined core curricula required for effective sports cardiology practice, yet these do not involve documenting competency in the evaluation of paediatric athletes. While American College of Cardiology (ACC) curriculum<sup>51</sup> does state that collaboration with paediatric cardiologists is needed, there is a distinct lack of such specialists available to interpret all paediatric screenings.<sup>24 52</sup> Consequently, forming valid and reliable guidelines together with certified educational pathways is imperative (Table 3).

### **Limitations**

By including only policies written in the English language, some national policies may have been omitted. There is currently no validated methodology of performing systematic searches of non-indexed sources, and therefore policies that were not published, peer reviewed, or indexed would not have been included. This may have been overcome by implementing an online search of known sporting organisations, yet this would be logistically difficult to avoid selection bias. To account for this, it may

be prudent for sporting organisations to hereafter publish their screening policies; to not only allow for inclusion in future systematic reviews but to publicise their policy more broadly. The method of evaluating relevance was not based on previous work, nevertheless, we sought to adopt a more objective way of quantifying how the general body of literature was used in each policy paper, and how the paediatric sub-set of work was used. Finally, the AGREE II tool was designed to evaluate the quality of guideline papers, with a goal to ensure uniformity in approach and reporting. Having found areas scoring poorly among the papers reviewed in this study, it is less a reflection of the authors and more a reflection of the limitations of the field, which extends to the referencing of the original research. Identifying these limitations, and showcasing how more recent documents have improved, may encourage future work is of even higher quality.

## **Conclusion**

Cardiac screening policies predominantly focus on adult athletes, with the data guiding these recommendations chiefly coming from studies that involve cohorts of athletes that combine both children and adults. There is an immediate need for paediatric specific research that accounts for age, sex, training status and maturation, to develop a database of normal and abnormal findings that inform screening guidelines. Expanding paediatric cardiology expertise in the care of young athletes and ensuring the development of certified educational pathways in cardiac screening for primary care and sports medicine physicians should be considered. For future guidelines, more robust development methods are needed in their planning and creation, using systematic and reproducible methodologies, with involvement of all key stakeholders, to improve cardiac care for paediatric athletes.

Table 3. Future policy recommendations for the screening of paediatric athletes.

<b>Policy Recommendations</b>	
<p><b>Paediatric specific research:</b></p> <ul style="list-style-type: none"> <li>• More research focusing on the paediatric athlete is needed.</li> <li>• Specific attention should be paid to age, sex, training status and maturation.</li> <li>• The development of normative values for cardiac testing which appropriately accounts for body size is fundamental.</li> <li>• The creation of an international paediatric athlete registry should be sought.</li> </ul>	<p><b>Education:</b></p> <ul style="list-style-type: none"> <li>• Too few paediatricians and paediatric cardiologists are sufficiently trained to provide expert opinion on the paediatric athlete.</li> <li>• Certified training pathways and a stronger engagement of paediatric and sport’s governing bodies should be developed.</li> <li>• Work towards a child athlete-centred paediatric sports cardiology specialty is merited.</li> </ul>
<p><b>Clinical practice</b></p> <ul style="list-style-type: none"> <li>• There is a need to ultimately develop and use paediatric athlete specific screening recommendations.</li> <li>• Maturation and puberty should be considered when assessing the paediatric athlete.</li> </ul>	<p><b>Stakeholder involvement:</b></p> <ul style="list-style-type: none"> <li>• A synergistic approach for guideline development is needed between paediatric and sports cardiologists, sports medicine physicians, exercise physiologists, policymakers, sporting organizations, coaches, and parents.</li> <li>• Consideration for the paediatric athlete should be at the centre of guideline development.</li> </ul>

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## FIGURE LEGENDS

**Central Figure.** Summary of review process, key findings, and future considerations.

**Figure 1.** PRISMA flowchart of screened, included and excluded studies.

**Figure 2.** Proportion of policies by relevance to paediatric athlete's score (Panel A). Proportion of original research citations including paediatric athletes from total, sorted from highest to lowest (Panel B).

**Figure 3.** Proportion and number of original related research citations by study design (Panel A), and by age group (panel B), on the topic of screening (left side) and sudden cardiac death (right side). Overlaid values are the number of citations, with the horizontal scale representing percentage from topic relevant citations. PPE - Preparticipation Physical Evaluation; AMA - American Medical Association.

**Figure 4.** Quality appraisal scores calculated using the AGREE II instrument.<sup>16</sup> Overall scores (left panel), each of the 6 domain sub-scores (right panel). Sorted from highest to lowest, with median value in red.

