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## **The epidemiology of running for health: on health benefits, burden and costs of injuries, and injury prevention in running**

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# General Introduction

# Chapter 1



## Physical (in)activity and public health

The pandemic of physical inactivity is considered a global public health concern,<sup>1</sup> mostly because physical inactivity contributes to the worldwide epidemic of non-communicable diseases.<sup>2</sup> According to the 2009 report of the World Health Organization (WHO), physical inactivity is the fourth leading risk factor for mortality, being only behind of high blood pressure, tobacco use, and high blood glucose; and being ahead of risk factors like overweight and obesity, high cholesterol, unsafe sex, and alcohol use.<sup>3</sup> In 2010, insufficient physical activity (physical inactivity and low physical activity levels) accounted for 3.2 million deaths (95% uncertainty interval [UI] 2.7 to 3.7), and 69.3 million (95% UI 58.6 to 80.2) disability-adjusted life years (DALYs), representing 2.8% (95% UI 2.4 to 3.2) of the total DALYs.<sup>4</sup> This comprehensive study conducted with data of 187 countries has found that insufficient physical activity was ranked as the 10<sup>th</sup> leading independent risk factor for the global disease burden in 2010.<sup>4</sup>

On average, 31.1% (95% confidence interval [CI] 30.9 to 31.2) of the world's population presents insufficient physical activity levels.<sup>5</sup> A reduction of 10% in the prevalence of physical inactivity is estimated to lead to the prevention of approximately one million deaths per year.<sup>1</sup> Moreover, the health benefits of physical activity on mortality, morbidity, physiological, and cognitive outcomes are well documented,<sup>4,6-9</sup> suggesting that exercise may have effects comparable to medicines.<sup>9,10</sup> Therefore, the need for the development and implementation of physical activity programmes has been recognised and encouraged worldwide, as well as the inclusion of physical activity promotion in the non-communicable disease and obesity prevention agendas.<sup>1</sup>

### Running for physical activity promotion

Globally, 31.4% (95% CI 31.2 to 31.4) of the adult population reports practising vigorous-intensity physical activity at least three days per week.<sup>5</sup> The proportion of the worldwide physically active population who practises running is unknown. In the Netherlands, however, running was the fastest growing sport in 2010,<sup>11</sup> representing 12% of the sport population in 2012,<sup>12</sup> and was the third most practiced sport in 2013, with 2.1 million people reporting running at least once during 2013 (Table 1.1).<sup>13</sup> Running is very popular,

**Table 1.1** Sports participation and injury information in the Netherlands (2013)

Sport	Number of participants	Number of injuries	Number of injuries per 1,000 hours	Emergency department			Medical costs per injured participant
				Number of visits	Number of visits per 1,000 hours	Number of visits per 1,000 hours	
Total	–	4,500,000	2.0	130,000	0.054	€ 1,100	
Fitness	3,400,000	490,000	1.5	1,600	0.005	–	
Swimming	2,900,000	–	–	4,500	0.036	€ 1,400	
<b>Running</b>	<b>2,100,000</b>	<b>640,000</b>	<b>5.3</b>	<b>1,800</b>	<b>0.015</b>	<b>€ 1,100</b>	
Cycling	1,700,000	–	–	3,800	–	€ 2,600	
Outdoor soccer	1,400,000	850,000	4.3	37,000	0.190	€ 910	
Tennis	930,000	160,000	2.3	2,400	0.035	€ 1,500	
Skating	650,000	–	–	4,400	0.420	€ 1,400	
Skiing	580,000	–	–	1,200	–	€ 1,200	
Volleyball	470,000	210,000	5.1	3,100	0.076	€ 1,100	
Martial arts	470,000	180,000	4.3	4,400	0.100	€ 800	
Mountain biking	460,000	–	–	2,400	0.066	€ 2,000	
Equestrian	360,000	–	–	7,800	0.120	€ 1,600	
Indoor soccer	340,000	–	–	2,800	0.240	€ 980	
Hockey	280,000	–	–	6,900	0.230	€ 720	
Snowboarding	160,000	–	–	790	–	€ 920	
Motorsports	140,000	–	–	3,400	0.560	€ 1,800	

€-: Insufficient or no data available.

Source: VeiligheidNL, 2015<sup>13</sup>

probably because it is easily accessible, relatively inexpensive, time efficient and it has a social component.<sup>14-16</sup> There is evidence showing that running is effective on increasing levels of physical activity.<sup>15</sup> Ooms et al.<sup>15</sup> have shown that 4.5 months after the end of a 6-week start-to-run programme, 69% of the participants in the start-to-run group were still running, and they were spending 152 more min/week (95% CI 80 to 223) in vigorous-intensity physical activities, and 107 more min/week (95% CI 69 to 145) in sports activities, compared to the control group. Therefore, running can be a powerful ally in the promotion of physical activity.

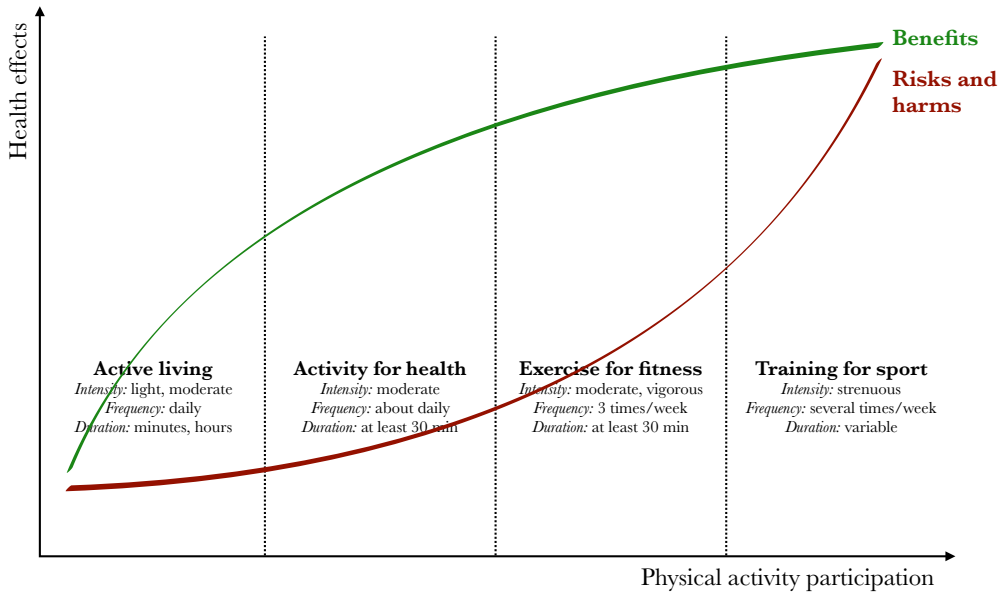
## **Health effects of running**

### ***Health benefits***

The effects of running on health have been studied mostly since the 1970s.<sup>17-24</sup> As physical activity in general can lead to health benefits, it is expected that running can also achieve such effects. However, this hypothesis should be investigated in order to describe in which outcome measures and to what magnitude these health effects may occur. This is relevant, because different types of physical activity or sports may result in specific health effects on physiological, psychological, and/or musculoskeletal outcome measures. Therefore, programmes on physical activity promotion may tailor the proposed activity or exercise to populations that would benefit most from these specific effects (e.g., sedentary, elderly, youth, and diseased). This was the main motivation for the study described in chapter 2 of this thesis. Chapter 2 describes a systematic review and meta-analyses on the health benefits of running. The purposes of the meta-analyses were to summarise the effects of habitual running on indices of health, and to describe the dose-response relationship between running exposure and selected health effects of running.

### ***Risks and harms***

All health promotion programmes or interventions should also consider the adverse effects that may be a result of their implementation (Figure 1.1).<sup>25</sup> Most of the risks and harms of running are associated with extreme situations, such as participating in ultra-marathon



**Figure 1.1** Hypothetical relationships between different strata of physical activity participation and health effects of physical activity. Used with permission of Ilkka Vuori.

running events.<sup>26-28</sup> Risks and harms of ultra-marathon running may include cardiovascular, musculoskeletal, dermatological, gastrointestinal, respiratory, and metabolic problems.<sup>26-28</sup> The risk of death during running is very low, with an estimated incidence rate of cardiac arrest of 0.39 per 100,000 runners.<sup>26</sup> As the running-related problems previously described present a low risk and occur in extreme situations like ultra-marathon running events, the main health problem related to running with the highest risk for the general running population is the musculoskeletal running-related injury (RRI).

## Running-related injury (RRI)

RRI can be defined as a disorder of the musculoskeletal system that has been sustained during participation in running.<sup>29,30</sup> A summary of RRI information in relation to other sports in the Netherlands can be found in Table 1.1. The risk of RRIs varies across different groups; for example, the incidence rate of RRIs is estimated at 17.8 (95% CI 16.7 to 19.1) and 7.7 (95% CI 6.9 to 8.7) RRIs per 1,000 hours of running for novice and recreational runners, respectively.<sup>23</sup> Prevalence and incidence measures for specific

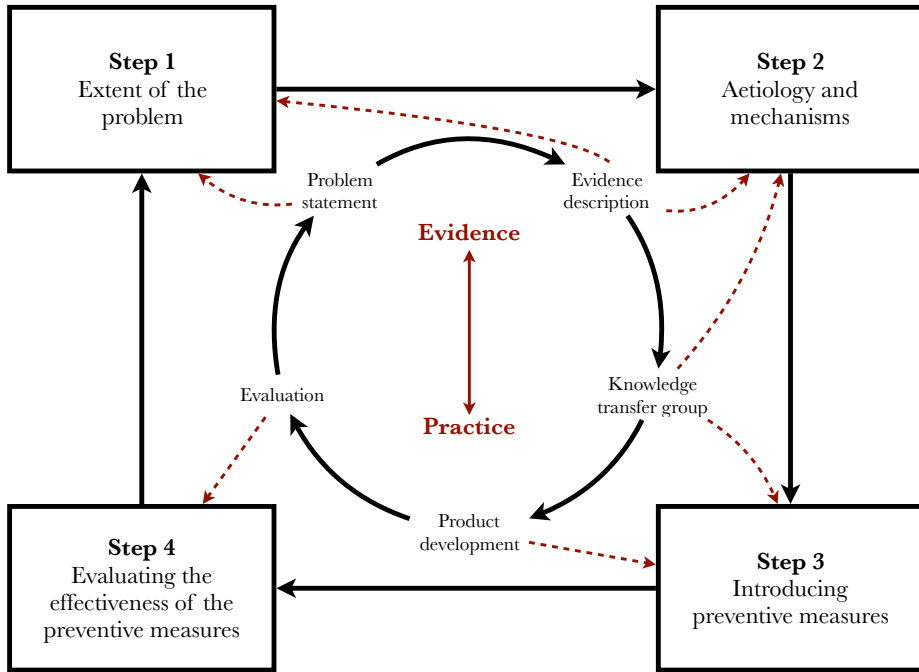
RRIs (e.g., diagnostic classifications of RRIs) are difficult to find and interpret because of differences in methods across studies,<sup>20</sup> but also because such studies are challenging to conduct due to methodological issues, such as sample size and prospective surveillance.<sup>31</sup> Summarising this evidence via a systematic review was, therefore, the main motivation for the study described in chapter 3 of this thesis. Chapter 3 presents a systematic review aimed at summarising the risk (prevalence and incidence) of the main RRIs reported in the literature.

Most of RRIs are overuse injuries.<sup>20</sup> More sensible methods for the registration and surveillance of overuse injuries have been suggested.<sup>32-34</sup> However, prospective and reliable data on overuse injuries are sparse in the literature. Regarding severity, RRIs can last months,<sup>35,36</sup> present moderate pain levels on average,<sup>36</sup> often lead to time loss (running sessions not fully accomplished, or completely missed due to RRIs),<sup>29,30,37</sup> medical attention,<sup>29,30,38</sup> productivity loss,<sup>29,30,38</sup> and costs for society.<sup>29,30,38</sup> However, prospective surveillance of RRIs using methods for registering and following-up overuse injuries, and also including measures of risk (prevalence and incidence), severity, and costs are sparse in the literature. Therefore, a review of concepts on sports injuries surveillance methods, including methodological issues on overuse injuries, was the main motivation for chapter 4 presented in this thesis. Chapter 4 describes a review of basic concepts on sports injuries surveillance, and discusses the implementation of such concepts in practice.

## **Prevention of RRIs**

Figure 1.2 presents the framework used in the studies on the prevention of RRIs included in this thesis. This framework is an interaction between the sequence of prevention on sports injuries suggested by van Mechelen et al.<sup>39</sup> (square form in Figure 1.2) and the Knowledge Transfer Scheme (KTS) framework suggested by Verhagen et al.<sup>40</sup> (round form in Figure 1.2). The sequence of prevention is a theoretical model aimed at guiding researchers on the steps necessary to effectively achieve sports injury prevention, whilst the KTS is a practical model aimed at collecting evidence and practice-based knowledge together, in order to translate such knowledge into practical actions for implementation in practice. The interaction of these two models can be a powerful tool for effective evidence-practice-based sports injury prevention actions.





**Figure 1.2** Framework used in the studies on the prevention of running-related injuries in this thesis. This framework is an interaction between the sequence of prevention on sports injuries suggested by van Mechelen et al.<sup>39</sup> (square form) and the Knowledge Transfer Scheme (KTS) framework suggested by Verhagen et al.<sup>40</sup> (round form).

### Step 1

The first step of the sequence of prevention of sports injuries (Figure 1.2) consists of identifying and describing the risk (prevalence and incidence) and severity of sports injuries in populations of interest (i.e., the target populations for RRI prevention). There is evidence suggesting that novice runners constitute one of the groups with the highest risk of RRIs.<sup>23,41</sup> The incidence rate of RRIs in novice runners is estimated at 17.8 RRIs per 1,000 hours of running (95% CI 16.7 to 19.1).<sup>23</sup> In 2014, 50 thousand people started to run in the Netherlands, and running was the second sport with the largest number of novice participants.<sup>42</sup> Therefore, novice runners should be one of the target groups for RRI prevention. However, data on the severity and consequences (especially costs) of RRIs in novice runners is sparse. This was the main motivation for the study described in chapter 5 of this thesis. Chapter 5 describes a nationwide and multicentre study on the economic burden of RRIs in novice runners.

The number of runners participating in running events has been growing in the Netherlands.<sup>43</sup> The participants of organised running programmes training for such events are usually inexperienced runners.<sup>29</sup> As discussed before, inexperienced runners (including novice runners) are at higher risk of RRIs compared to more experienced runners.<sup>23,41</sup> However, evidence is sparse on the risk and severity of RRIs in runners participating in organised running programmes preparing them to participate in events. This was the main motivation for the study described in chapter 6 of this thesis. Chapter 6 is a prospective cohort study aimed at investigating the health and economic burden of RRIs in runners training for an event in Tilburg, the Netherlands.

According to MudSweatTrails,<sup>44</sup> the trailrunning community is growing in the Netherlands. For example, there was no trailrunning event in the Netherlands and Belgium by 2010, and in 2015 there were over 150. Whereas inexperienced runners (including novice runners) present lower running exposure, trailrunners are usually more experienced and more exposed to running (i.e., higher running volume). Running exposure has been considered a necessary cause for RRIs, since one can only sustain an RRI when participating in running.<sup>45</sup> Therefore, inexperienced runners would be somewhere in the middle of the x-axis in Figure 1.1, whilst trailrunners would be more to the right. This hypothesis suggests that trailrunners could also have a high risk of RRIs, because they have a high running exposure. Therefore, trailrunners may also be one of the target groups on RRI prevention. Prospective data on the risk and severity (including costs) of RRIs in trailrunners are sparse. This was the main motivation for the study described in chapter 7 of this thesis. Chapter 7 is a prospective cohort study aimed at investigating the health and economic burden of RRIs in trailrunners.

## *Step 2*

The second step of the sequence of prevention on sports injuries (Figure 1.2) consists of establishing the aetiology and mechanisms of sports injuries. Investigating the aetiology and mechanisms of RRIs is challenging, especially because of the overuse nature of most RRIs. Overuse injuries are characterised by a non-identifiable and gradual onset, with fluctuation of symptoms over time.<sup>32</sup> Malisoux et al.<sup>45</sup> suggested that running exposure

is the only necessary cause for RRIs. In fact, one is only at risk of sustaining an RRI when exposed to running. Also, the main risk factor for RRIs described in the literature is the history of RRIs.<sup>21</sup> The second step of the sequence of prevention was achieved by summarising the available scientific evidence on RRIs, mainly with systematic reviews,<sup>20-22,46</sup> and then contrasting the evidence with the results of the cohort study presented in chapter 7<sup>30</sup> in order to develop an intervention on RRI prevention for trailrunners (chapter 8).

### *Step 3*

The third step of the sequence of prevention on sports injuries (Figure 1.2) consists of developing or introducing preventive measures. Therefore, the KTS framework<sup>40</sup> was used to combine the evidence and practice-based knowledge in order to develop an intervention on RRI prevention for trailrunners. A group composed of trailrunners, trainers, a stakeholder of the MudSweatTrails, and RRI researchers was established with the purpose of discussing the aetiology and mechanisms of RRIs in trailrunners (second step of the sequence of prevention), and strategies to implement such knowledge in practice (i.e., participatory research<sup>47</sup>). The outcomes of a brainstorm meeting with this group resulted in the development of an evidence-practice-based intervention on the prevention of RRIs for trailrunners (chapter 8 describes the development of the intervention).

### *Step 4*

The fourth step of the sequence of prevention on sports injuries (Figure 1.2) consists of evaluating the effectiveness of the preventive measures. Effective interventions or programmes on RRI prevention are scarce.<sup>46,48-50</sup> Some of the challenges on the development of effective prevention programmes for RRIs are: (1) missing information on RRIs for target groups; (2) runners usually do not have a coach, club, or medical staff to guide them in their training on a regular basis in contrast with other sports, making it difficult to implement and maintain adherence to prevention programmes, and also to receive feedback about their health (injury) status; and (3) the interventions or programmes should be developed following a behaviour change approach, which has not been done in most

of the interventions on RRI prevention described in the literature. These challenges were the main motivation for the development and evaluation of the evidence-practice-based intervention on RRIs in trailrunning described in chapter 8. Chapter 8 is a randomised controlled trial aimed at evaluating the effectiveness of an online tailored intervention on the determinants of preventive behaviour, on promoting preventive behaviour towards RRI prevention, and on the prevention of RRIs in trailrunners.

## **General objectives of the thesis**

The general objectives of this thesis were: (1) to investigate the health benefits of running; (2) to investigate the health and economic burden (i.e., direct and indirect costs) of RRIs in different running population; and (3) to evaluate the effectiveness of an intervention developed by following the KTS on the prevention of RRIs in trailrunners.

## **Specific objectives of the thesis and related chapters**

- To summarise the evidence on the effects of endurance running on biomedical indices of health, including the dose-response relationship (chapter 2).
- To summarise the evidence on the incidence and prevalence of diagnostic classifications of RRIs (chapter 3).
- To describe concepts of sports injury surveillance and discuss their implementation in practice (chapter 4).
- To investigate the health and economic burden (i.e., direct and indirect costs) of RRIs in different running populations (chapters 5, 6, and 7).
- To develop an evidence-practice-based intervention aimed at promoting preventive behaviour towards RRI and, consequently, to prevent RRIs in trailrunners (chapter 8).
- To evaluate the effectiveness of the developed intervention on the determinants and actual preventive behaviour, and on the prevention of RRIs in trailrunners (chapter 8).

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