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Original research

The NLstart2run study: Economic burden of running-related injuries in novice runners participating in a novice running program

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ABSTRACT

Objectives: To investigate the economic burden of running-related injuries (RRI) occurred during the 6-week 'Start-to-Run' program of the Dutch Athletics Federation in 2013. *Design*: Prospective cohort study.

Methods: This was a monetary cost analysis using the data prospectively gathered alongside the RRI registration in the NLstart2run study. RRI data were collected weekly. Cost diaries were applied two and six weeks after the RRI registration to collect data regarding healthcare utilisation (direct costs) and

absenteeism from paid and unpaid work (indirect costs). RRI was defined as running-related pain that hampered running ability for three consecutive training sessions.

Results: From the 1696 participants included in the analysis, 185 reported a total of 272 RRIs. A total of 26.1% of the cost data (71 RRIs reported by 50 participants) were missing. Therefore, a multiple imputation procedure was performed. The economic burden (direct plus indirect costs) of RRIs was estimated at €83.22 (95% CI €50.42−€116.02) per RRI, and €13.35 (95% CI €7.07−€19.63) per participant. The direct cost per RRI was €56.93 (95% CI €42.05−€71.81) and the indirect cost per RRI was €26.29 (95% CI €0.00−€54.79). The indirect cost was higher for sudden onset RRIs than for gradual onset RRIs, with a mean difference of €33.92 (95% CI €17.96−€49.87).

Conclusions: Direct costs of RRIs were 2-fold higher than the indirect costs, and sudden onset RRIs presented higher costs than gradual onset RRIs. The results of this study are important to provide information to public health agencies and policymakers about the economic burden of RRIs in novice runners.

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1. Introduction

Physical inactivity is a major risk factor for mortality and morbidity, resulting in high societal impact such as healthcare utilisation, reduced working participation and associated costs.^{2,3}

Many people seek running programs to start doing physical activity and to obtain the benefits of running practice. ^{4,5} Therefore, running programs for novice runners are of particular interest for public health, because they are a means to promote physical activity and thus to decrease the burden related to physical inactivity. However, running-related injuries (RRI) that occur during these programs may lead to running dropouts and/or failure to maintain an active lifestyle. ⁶ Early dropouts from running practice in novice runners could affect their motivation to continue running, or worse, this could lower their motivation to do any sort of exercise. In addition,

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RRIs may lead to health and economic burden for the novice runner and society.

Available data suggest that novice runners are at increased risk of RRIs when compared to more experienced runners.7 A metaanalysis has reported that the incidence of RRIs in novice runners was 17.8 RRIs per 1000 h of running (95% confidence interval [CI] 16.7–19.1). Determining the severity of sports injuries is an important step to injury prevention.⁸ A way to address the severity of sports injuries is investigating the burden of such injuries to the healthcare system in monetary costs (direct costs), but also the loss of productivity related to sports injuries (indirect costs).^{8,9} A previous study has reported the economic burden of RRI in runners participating in an organised training program preparing them for an event, with a total cost (direct plus indirect) estimated at €173.72 (95% confidence interval [CI] €57.17–€318.76) per RRI, and €104.89 (95% CI €30.51–€198.27) per runner. 10 This was a small study and, therefore, a larger and national-based study on the economic burden of RRIs in novice runners is justified.

Cost data of RRIs can be a useful measure to report the severity of RRIs,8 and it is also important to drive public health financial resources.⁹ The information provided by cost analyses can be used to set public health priorities and/or it could help in policy making towards injury prevention.¹¹ Therefore, the purpose of this study was to investigate the economic burden of RRIs occurred during a 6-week 'Start-to-Run' program for novice runners

2. Methods

This was a monetary cost analysis using the cost data that were prospectively gathered alongside the RRI registration in the NLstart2run study. The NLstart2run study is a multi-center prospective cohort study on the effects of a 6-week 'Start-to-Run' program in the Netherlands. The study was approved by the Medical Ethics Committee of the University Medical Center Groningen (2012/350) and it is registered in the Netherlands Trial Registry (NTR3676). For the purpose of this study, the participants were followed-up for the maximum period of 12 weeks (six weeks during the 'Start-to-Run' program and six weeks after sustaining an RRI). Detailed information of the design and methodology of the NLstart2run study has been published elsewhere.^{6,12}

The 'Start-to-Run' program of the Dutch Athletics Federation is organised twice a year. The program aims to prepare participants without running experience for a continuous 20-min run over a 6-week period. All individuals registered to participate in this program in 2013 (March and September) were invited to partake in the NLstart2run study (n = 7660). Information about the study and procedures were sent to all 'Start-to-Run' participants. Individuals aged 18-65 years, who agreed with the digital informed consent and completed the baseline questionnaire were included in the study (n = 1772).

The training program consisted of two or three sessions per week (the third weekly session was optional). The first weekly session was a group training supervised by a licensed athletic trainer. The remaining weekly sessions were unsupervised individual training, and the participants were provided with a pre-defined schedule prepared by the Dutch Athletics Federation. Each session started approximately with a 15-min warm-up and finished with a 15-min cool-down consisting of walking, stretching and relaxing exercises. The supervised group sessions lasted approximately 90 min and consisted of a theoretical part (30 min) followed by a practical part (60 min). The unsupervised individual sessions lasted approximately 45 min and consisted only of running practice. The duration and intensity of the running training was gradually increased throughout the program.6

Cost categories and reference monetary costs used in the study.

Direct costs (healthcare utilisation)						
28.00						
72.00						
36.00						
9.27-36.41						
12.50						

Prices according to the Dutch Health Insurance Board (College voor Zorgverzekeringen).30

- ^a Costs related to absenteeism from paid work were estimated based on the mean income of the Dutch population according to age and gender.³⁰ The value for paid work represents the minimum and maximal values according to standardised prices by age and gender.
- b Costs related to absenteeism from unpaid work were estimated based on the mean working hours of the Dutch population according to age and gender.31

The baseline questionnaire was sent to the participants one week before the commencement of the 'Start-to-Run' program to collect personal data as age, gender, height, weight and educational level. Previous sports activities, previous injuries (last 12 months) and running experience were also registered.

A web-based training log was sent to the participants at the end of each week to collect data about running exposure and occurrence of running-related pain (self-reported pain attributed to running) experienced during the last week. Participants were asked not to report muscle soreness and blisters in the pain registration. 12 In the case of a reported running-related pain, the body region and hampering of running ability (i.e., a reduction in duration, distance, speed or inability to participate in running due to this runningrelated pain) were registered. An RRI was registered when running was hampered for three consecutive training sessions (i.e., at least a week) as a result of running-related pain at the same body part.^{6,12}

Two weeks after the initial registration of an RRI, a follow-up questionnaire was sent to the injured participants. This questionnaire asked about injury onset (sudden or gradual), injury body location (using a body chart), injury type (i.e., muscle-tendon unit, joint, ligament, or bone) and injury recovery. The follow-up questionnaire also included questions on healthcare utilisation (direct costs) and absenteeism from paid and unpaid work (indirect costs).

In the case of healthcare utilisation, questions were asked in order to specify which healthcare professionals were consulted (i.e., general practitioner, medical specialist or physiotherapist). The number of consultations for general practitioner and medical specialist were collected by answering the following alternatives: 1, 2, 3, 4, 5 or more consultations. For those participants who reported '5 or more', five consultations were considered in the analysis. The number of physiotherapy consultations was collected by answering the following alternatives: 1, 2, 3, 4, 5–9, 10–19 and 20 or more. For those participants who reported '5-9' or '10-19' the median was used (no participant reported '20 or more'). The number of consultations were categorized in order to facilitate answering the questionnaire (e.g., faster answer) and to avoid error and out-ofrange data entry. In the case of absenteeism, the participants were asked about the number of missing days and/or hours related to paid and unpaid work (e.g., household) since the RRI registration.

If the injured participants indicated that the RRIs were not recovered at the time the follow-up questionnaire was sent after two weeks from the RRI registration, a second follow-up questionnaire with the same healthcare utilisation and absenteeism questions was sent four weeks later (i.e., six weeks after the RRI registration).

The main outcomes of this study were the monetary direct (healthcare utilisation) and indirect (absenteeism from paid and unpaid work) costs of RRIs. Cost categories that were registered

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during the study and their monetary costs used as reference in this analysis are presented in Table 1. Descriptive analysis was performed to summarise the demographic and cost data, and the results were expressed in mean and standard deviation (SD) for continuous variables and percentage for categorical variables.

A missing data evaluation was performed and direct and indirect cost values were imputed by multiple imputation using the multiple imputation by chained equations (MICE) procedure¹³, in order to avoid bias due to missingness.¹⁴ Also, it was necessary to impute data about the RRIs onset, which were missing in some cases. Twenty datasets were created with imputed data. The number of imputations followed previous recommendations.¹⁵

Mean direct, indirect and total costs related to RRIs were calculated per RRI and per participant. Differences in costs between RRIs with sudden and gradual onset, and between males and females were analysed based on mean differences and 95% CIs. ¹⁶ As cost data are nonparametric, bias-corrected and accelerated 95% CI was obtained by bootstrapping with 2000 replications. A complete-case analysis was first performed excluding those with missing data. In the imputed data analysis, the same procedure was applied in each imputed dataset and the results were further combined according to Rubin's rules. The cost analyses were conducted in R version 3.2.0, using the packages 'mice' and 'boot'. The bootstrapping results of the 20 imputed datasets were further combined according to Rubin's rules. In Microsoft® Excel® version 14.5.0.

3. Results

Of the 7660 individuals registered to participate in the 'Start-to-Run' program in 2013, 1772 participants were included in the NLstart2run study. Seventy-six participants were excluded from the analysis because no information was entered in the web-based training logs. Therefore, data of 1696 participants (22.1% of the source population) were analysed for the purpose of this study. The sample was composed of 364 (21.5%) males with a mean age of 48.3 (SD 8.9), and 1332 (78.5%) females with a mean age of 41.9 (SD 9.8). Additional characteristics of the participants can be found elsewhere. 12

A total of 185 participants (10.9%) reported 272 RRIs during the 'Start-to-Run' program. However, 71 RRIs (26.1%) presented

Table 2 Frequency and cost per category of direct and indirect costs for complete cases (n = 135).

Description	Frequency (%)	Cost (€)
Direct costs (healthcare utilisation), visits	305(100%)	11324.00
General practitioner, visits	20(6.6%)	560.00
Medical specialist, visits	14(4.6%)	1008.00
Physiotherapist, visits	271 (88.9%)	9756.00
Indirect costs (loss of productivity), h	185 (100%)	5820.43
Absenteeism from paid work, h	170 (91.9%)	5632.93
Absenteeism from unpaid work, h	15(8.1%)	187.50
Total costs	-	17144.43

missing data regarding RRI characteristics (including onset) and costs. Therefore, the descriptive and complete-case cost analyses were performed with 201 RRIs reported by 135 participants. Of the 135 injured participants, 66 (48.9%) consulted healthcare professionals (9.6%, n=13 by general practitioners; 6.7%, n=9 by medical specialists; and 45.2%, n=61 by physiotherapists), and 7 (5.2%) reported absenteeism from work due to RRIs (4.4%, n=6 from paid work; 0.7%, n=1 from unpaid work). Of the 201 RRIs with complete cost data, 52.7% (n=106) had a sudden onset and 47.3% (n=95) had a gradual onset.

In total, the direct cost related to RRIs was higher than the indirect costs (Table 2). Physiotherapy consultations were responsible for most of the healthcare utilisation, and absenteeism from paid work was responsible for most of the loss of productivity hours. Table 3 presents the total, direct and indirect cost results per RRI and per participant for the complete-case and imputed data analyses. The mean total cost per RRI with multiple imputed data (i.e., based on all 272 RRIs) was ≤ 83.22 (95% CI $\leq 50.42 - \leq 116.02$) and the mean total cost per participant (i.e., based on all 1696 participants) was ≤ 13.35 (95% CI $\leq 7.07 - \leq 19.63$).

The indirect cost was higher for RRIs with sudden onset than for RRIs with gradual onset in the complete-case and imputed data analyses (Table 3). In the complete-case analysis, the total and direct costs were higher for males than for females, however these differences did not reach significance in the imputed data analyses. Nevertheless, the indirect cost was higher for males than for

 Table 3

 Monetary cost analysis of the running-related injuries (RRI) in novice runners.

Description	Total cost (€)		Direct cost (€)		Indirect cost (€)	
	Complete-case data	Imputed data	Complete-case data	Imputed data	Complete-case data	Imputed data
Costs per RRI	85.30 (64.46–138.60)	83.22 (50.42-116.02)	56.34 (45.44–71.47)	56.93 (42.05–71.81)	28.96 (10.87–73.71)	26.29 (0.00–54.79) ^a
	n = 201	n=272	n = 201	n=272	n = 201	n = 272
Sudden onset	104.33 (70.10–191.80)	100.25 (42.18–158.31)	55.32 (42.33–74.03)	57.59 (36.77–78.40)	49.01 (16.86–150.02)	$42.66 (0.00-95.25)^{a}$
	n = 106	n = 141	n=106	n=141	n=106	n = 141
Gradual onset	64.05 (43.54–89.67)	65.00 (35.29–94.71)	57.47 (39.38–83.41)	56.26 (34.64–77.87)	6.58 (0.00–19.74)	$8.74 (0.00-28.54)^{a}$
	n = 95	n = 131	n=95	n=131	n=95	n = 131
Difference	40.28 (-2.97 to 124.46)	35.25 (-26.03 to 96.52)	-2.15 (-29.48 to 20.37)	1.33 (-28.18 to 30.84)	42.44 (8.38 to 124.42) ^b	33.92 (17.96 to 49.87) ^b
Costs per participant	10.42 (7.01–17.71)	13.35 (7.07–19.63)	6.88 (5.02–9.29)	9.13 (6.08 to 12.18)	3.54 (1.12–9.77)	4.22 (0.00–9.17) ^a
	n = 1646	n=1696	n = 1646	n=1696	n=1646	n = 1696
Male	20.51 (9.19–57.60)	$24.09 (0.00-50.00)^a$	12.10 (6.02–23.22)	14.63 (4.81–24.46)	8.41 (0.00–33.20)	$9.46 (0.00-29.22)^a$
	n = 353	n=364	n=353	n=364	n=353	n = 364
Female	7.66 (5.40–13.27)	10.41 (5.27–15.55)	5.45 (4.03–7.36)	7.63 (4.79–10.47)	2.21 (0.59–8.97)	$2.78 (0.00-7.26)^a$
	n = 1293	n=1332	n = 1293	n=1332	n = 1293	n = 1332
Difference	12.85 (0.75 to 47.79) ^c	13.68 (-11.93 to 39.30)	6.65 (0.11 to 17.52) ^c	7.00 (-3.28 to 17.29)	6.20 (-1.80 to 40.41)	6.68 (0.28 to 13.08) ^c

All costs are presented in euros (€). Mean values are followed by the bias-corrected and accelerated (BCa) 95% confidence interval (CI) estimated by bootstrapping (2000 replications). Results in the imputed data analysis represent the pooled mean and BCa 95% CI estimates obtained by multiple imputations by chained equations (MICE) with 20 imputations combined according to Rubin's rules.

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^a Because of the large variability of the bootstrapping estimates among the 20 imputed datasets, the lower bound of the 95% CI calculated following Rubin's rules for these estimates was negative. As descriptive cost data cannot be negative, because there is no negative cost (except for differences), the lower bound of the 95% CI were replaced by zero when this occurred.

^b Statistically significant difference between sudden and gradual RRI onset.

^c Statistically significant difference between males and females.

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females in the imputed data analysis, although the lower bound of the 95% CI revealed a small magnitude.

4. Discussion

The total cost of RRIs per novice runner participating in the 6week 'Start-to-Run' program of the Dutch Athletics Federation in 2013 was estimated at €13.35 (95% CI €7.07-€19.63). This indicates that, based on the total source population (n = 7660), one could expect to have an economic burden of about €102 thousand with the 6-week 'Start-to-Run' program. There are many more running programs for novice runners occurring annually in the Netherlands, therefore the costs related to RRIs in novice runners may be even higher. According to the 'Nederlands Olympisch Comité * Nederlandse Sport Federatie' (NOC*NSF), there were about 1 million people interested in starting to run in the Netherlands in 2013.¹⁸ If we consider that 10% of these people will participate in 'Start-to-Run' programs, one could expect to have a total cost related to RRIs in novice runners of about €1.33 million related to a 6-week program period, which represents about 0.2% of all annual costs related to sports injuries in the Netherlands. 19 However, running programs can have different purposes and training schedules, therefore this estimate is only for perspective purposes and it should be confirmed in future studies.

Direct costs related to RRIs were 2-fold higher than the indirect cost. This is in contrast with the available literature indicating the opposite in the case of sports injuries. 10,19-21 Hespanhol Junior et al.¹⁰ have reported that the indirect cost related to RRIs was 2fold higher than the direct cost. It might be that the period of six weeks after the RRI registration was a short period to account for productivity loss consequences, and this might have contributed to the discrepancy of the results found in the current study compared to the literature. Absenteeism from work may increase the costs on a larger scale than the healthcare utilisation, because a day of absenteeism from work $(\pm 6 \, h)$ contributes more to the economic burden than a consultation with a healthcare professional (Table 1). Therefore, a middle- or long-term follow-up might be necessary to adequately measure the economic consequences of productivity loss related to RRIs in prospective studies. In addition, the runners included in the study of Hespanhol Junior et al.¹⁰ were not novices, and maybe differences in the severity of RRIs between running populations may also explain the discrepancy in the results. With regard to direct cost, the results per RRI herein presented were similar to those reported by Hespanhol Junior et al. 10 Discrepancies were found for direct cost per participant (especially for males), but this difference may be due to few participants in the study of Hespanhol Junior et al.¹⁰ in comparison with the current study.

In general, the costs related to RRIs were higher for sudden than for gradual onset. However, the difference between sudden and gradual onset for indirect costs was the only statistically significant estimate in both complete-case and imputed data analyses. The data collected in this study were self-reported, therefore one could argue whether the novice runners were able to distinguish sudden from gradual onset. Most of RRIs reported in the literature are overuse injuries²², but in the current study 52.7% of the RRIs presented a sudden onset according to the self-reported data. Therefore, these results should be interpreted with caution. Also, the short period of follow-up could explain the cost differences between sudden and gradual onset. An acute injury (sudden onset) is supposed to be more severe in a short-term when compared with overuse injuries (gradual onset). On the other hand, overuse injuries may be more severe over time, and in fact studies have shown that cumulative severity (the sum of the severity measure over time) may be higher for overuse than for acute injuries.^{23,24} Therefore, it is possible that novice runners with acute injuries are more likely to seek health care (direct costs) and to have higher productivity loss (indirect costs) closer to the onset, whilst novice runners with overuse injuries are more likely to report these consequences spread over time. Future studies should investigate the costs of RRIs in novice runners in a long-term to explore this hypothesis.

Although the indirect cost was higher for males than for females in the imputed data analysis, the lower bound of the 95% CI was small, indicating that this difference may be considered trivial. A previous analysis of the NLstart2run study has shown no statistically significant association between gender and the incidence of RRIs. 12 These results indicate that the risk and costs of RRIs in novice runners may present no significant difference between males and females.

The total cost (direct plus indirect) of an RRI in novice runners found in the current study (€83.22) was lower than the total cost of an ankle sprains injury in Dutch volleyball athletes during one season $(\le 360.60)^{20}$, and it was lower than the total cost of a physical activity-related injury in Dutch children aged 10-12 years during one school year (€188).¹¹ The Dutch Athletics Federation seems to have a 'Start-to-Run' program that leads to RRIs with low economic burden when compared to injuries in other populations. However, this result should be interpreted with caution since the current study followed-up the costs of RRIs over a period of six weeks after the RRI registration, whilst the other studies were conducted during an entire season²⁰ or a year.¹¹ More importantly, the definition of injury in the current study (running-related pain that hampered running ability for three consecutive training sessions) may be considered more inclusive when compared with the studies of Collard et al.¹¹ and Verhagen et al.²⁰ Both studies included mainly acute injuries, whereas 47.3% of the RRIs registered in the present study were overuse injuries, which may be considered less limiting and severe from a healthcare perspective. This might have influenced the lower costs found in the current study relative to the other published literature.

Although the health benefits^{4,5} and economic savings of running related to reducing the risk of diseases^{2,5} outweigh the health and economic burden of RRIs, the consequences of RRIs in a population that is starting to exercise can be worrisome, since dropouts from physical activity could be a result of such injuries.^{2,6} This is relevant for public health since the promotion of physical activity is a matter of concern worldwide^{2,7}, and preventing RRIs could avoid dropouts from running and/or physical activity practice.

This is the first study to report the economic burden of RRIs in novice runners participating in an organised and national-based 'Start-to-Run' program. The NLstart2run study is a multi-center prospective study that has been conducting in the Netherlands. Therefore, the results of this study are generalizable for the Dutch population who decide to partake in the 'Start-to-Run' program of the Dutch Athletics Federation, which includes thousands of participants each year. Another strength of this study is the prospective nature of the cost data collected using cost diaries, which decreases the risk of recall bias. ^{9,28} Cost data collected from medical records could underestimate the costs of RRIs not severe enough to result in medical attention ¹¹, and the validity and reliability of such databases may not be very high. ²⁹

This study had also limitations. A total of 26.1% of the cost and RRI onset data was missing. Cost values were imputed by MICE in order to avoid bias due to missingness¹⁴, however the amount of missing data might have still introduced bias in the results. Data about medicines taken and diagnostic tests due to RRIs were not collected, thus these costs were not included in the direct cost analysis. This could have underestimated the direct costs of RRIs. The number of general practitioner, medical specialist and physiotherapy consultations were categorised when more than four consultations were reported. This could have introduced bias in

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the exact number of healthcare consultations. The diagnoses of RRIs were not possible to confirm due to logistic reasons. This excluded a stratified cost analysis by RRI diagnosis. Also, the data collected in this study were self-reported, and the ability of novice runners in distinguishing sudden and gradual onset of RRIs may be a matter of concern.

5. Conclusions

This study showed that the economic burden (direct plus indirect costs) of RRIs in novice runners participating in a 6-week 'Start-to-Run' program was estimated at \in 83.22 (95% CI \in 50.42– \in 116.02) per RRI, and \in 13.35 (95% CI \in 7.07– \in 19.63) per novice runner. Direct costs were 2-fold higher than the indirect costs. Indirect costs were higher for RRIs with sudden onset than for RRIs with gradual onset. The results of this study are important to provide information to public health agencies and policymakers about the economic burden of RRIs in novice runners.

6. Practical implications

- A running injury was estimated to have an economic burden of €83.22 (95% CI €50.42-€116.02) in novice runners participating in a 6-week 'Start-to-Run' program.
- The estimated cost related to running injuries per novice runner of a 6-week 'Start-to-Run' program was €13.35 (95% CI €7.07-€19.63).
- Direct costs of running injuries (related to healthcare utilisation) were 2-fold higher than the indirect costs (related to absenteeism from paid and unpaid work).
- On average, the indirect cost per running injury was €33.92 (95% CI €17.96-€49.87) higher for injuries with sudden onset than for injuries with gradual onset.

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