Origi<u>nal article</u>

Dupuytren disease is highly prevalent in male field hockey players aged over 60 years

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ABSTRACT

Background/aim Dupuytren disease is a fibroproliferative hand condition. The role of exposure to vibration as a risk factor has been studied with contradictory results. Since field hockey is expected to be a strong source of hand-arm vibration, we hypothesised that long-term exposure to field hockey is associated with Dupuytren disease.

Methods In this cross-sectional cohort study, the hands of 169 male field hockey players (IQR: 65–71 years) and 156 male controls (IQR: 59–71 years) were examined for signs of Dupuytren disease. Details about their age, lifestyle factors, medical history, employment history and leisure activities were gathered. Prior to the analyses, the groups were balanced in risk factors using propensity score matching. The association between field hockey and Dupuytren disease was determined using a subject-specific generalised linear mixed model with a binomial distribution and logit link function (matched pairs analysis).

Results Dupuytren disease was observed in 51.7% of the field hockey players, and in 13.8% of the controls. After propensity score matching, field hockey playing as dichotomous variable, was associated with Dupuytren disease (OR=9.42, 95% CI 3.01 to 29.53). A linear dose-response effect of field hockey (hours/week x years) within the field hockey players could not be demonstrated (OR=1.03, 95% CI 0.68 to 1.56). **Discussion** We found that field hockey playing has a strong association with the presence of Dupuytren disease. Clinicians in sports medicine should be alert to this less common diagnosis in this sport.

INTRODUCTION

Dupuytren disease (DD) is a chronic hand condition, characterised by fibroblasts of the palmar fascia that transform into myofibroblasts, proliferate, deposit matrix and form nodules. Later on, cords are formed that may contract, causing flexion contractures of the fingers.¹ In the general elderly population, the prevalence of this disease ranges from 0.6% to 31.6%.²

Several intrinsic risk factors have been associated with DD, such as genes,³ ⁴ age⁵ ⁶ and male gender.⁶ ⁷ Various extrinsic risk factors, such as alcohol consumption and vibration exposure, have been associated with DD as well.^{8–10} Vibration exposure to the hands due to the handling of vibration tools, for instance, can cause microtrauma and peripheral vascular changes.^{11–13} These two mechanisms are suggested to be involved in the pathogenesis of DD.

In the past decades, two literature reviews were conducted to elucidate the association between DD

and vibration.¹⁴ ¹⁵ Liss and Stock¹⁴ concluded that there is a strong indication for the presence of this association. Recently, a meta-analysis demonstrated that vibration was significantly associated with DD.¹⁵ Both reviews suggest that there is evidence for a dose–response relationship, but many included papers suffered from methodological flaws (eg, unjustified corrections for factors that are not confounders, no physical examination to diagnose DD, inconsistencies in cross tabs possibly leading to an overestimated effect size).¹⁰ ¹⁶ ¹⁷ So the reviews are inconclusive and more directed research towards vibration is needed.

Most studies investigating this association take only the vibration exposure during work-related activities into account.^{8–10} ¹⁸ ¹⁹ However, vibration exposure can also occur during leisure activities (eg, sculpturing, do-it-yourself activities)²⁰ and sports (eg, tennis, baseball, golf).^{21–24} If vibration exposure during these kinds of activities is associated with the presence of DD, this needs to be assessed when studying the influence of vibration on the occurrence of DD.

There is only one previous paper that reports a high prevalence of DD in sportsmen.²⁵ However, this study focuses on the repetitive strain to the palmar fascia, and not on the vibration exposure. Sports such as field hockey can result in vibrations with large amplitudes, especially when impacts are not located on the sweet spot of the stick.²³ Therefore, we expected that there might be an association between field hockey and DD. Furthermore, we assumed that the majority of the field hockey players are white collar workers in the Netherlands. Therefore, this population is suitable to investigate vibration exposure, since it is unlikely that the possible effect of vibration through hockey will be confounded by an effect of manual work. The aim of the current study was to determine whether field hockey is associated with the presence of DD in elderly male field hockey players. We hypothesised that this association is present, and that a dose-response relation exists.

METHODS

Design

The hypothesis was tested by a cross-sectional cohort study, including a group of field hockey players and a control group.

Participants

We performed a sample size calculation in G^*Power , V.3.1.2²⁶ using an OR of 2.36 for vibration and manual work in association with DD¹⁵ and a proportion of DD equal to 0.22.⁵ To obtain

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a power of 80% for a two-sided test with the significance level at 5%, 74 participants were needed in each group (hockey players vs controls) for a two-sample χ^2 test.

The hockey players were recruited from a field hockey club for elderly male field hockey players, most of whom have played at a high level for a long period of time. The measurements took place during a tournament, so all 204 elderly male field hockey players who were registered for the tournament were asked to participate in this study. The controls consisted of 250 males from an age-stratified random sample of the general elderly population in the city of Groningen, the Netherlands, that was drawn from the municipal administration. These participants were previously included in a prevalence study,⁵ and agreed to being approached again for further research. To control for the confounding effect of manual work, all manual workers were excluded, as well as all controls who were exposed to vibration during occupational, leisure or sports activities.

All participants gave written informed consent. Owing to the nature of this study, no approval of the medical ethics committee was needed.

Procedure

Prior to the start of the study, we asked the board of the field hockey club for permission to approach their members. After that, the field hockey players received an email with information about this study. On 16 May 2013, the measurements took place during a tournament. Before the start, the field hockey players were reminded of this study using a presentation.

All participating field hockey players were interviewed about their employment history, leisure activities, lifestyle factors, health and demographics. These interviews were conducted by employees of the department of Plastic Surgery, who received training beforehand on how to perform the interview, to ensure that all participants were interviewed the same way. After the interview, the hands of the participants were examined for signs of DD by medical doctors (RL and PMNW) with broad experience in diagnosing DD.

The same data were collected from the controls. Diagnosis of DD in the control group was already determined using physical examination in the study of Lanting *et al*⁵ using the same criteria, but controls were contacted again by telephone to collect detailed information about their employment history, leisure activities, lifestyle factors and other missing information.

Outcome measures

The primary outcome measure was the presence of DD in one or both hands, defined as nodules or cords with or without flexion contractures of the fingers. The severity of the disease was a secondary outcome measure and determined using an adapted version of the Iselin classification (figure 1).²⁷

We collected age, smoking habits and alcohol consumption as additional measures, including the amounts consumed represented by cigarettes, cigars or pipes per day or by glasses per week, respectively. All participants were also asked about their general health, including diabetes, epilepsy and whether they had sustained hand injuries in the past. The hockey players were asked about the intensity (hours/week) and duration (years) of field hockey during their life.

Statistical analyses

Descriptive statistics were presented by frequencies and percentages for nominal and ordinal data. For data at the interval or ratio level, descriptives were presented by medians and IQRs.



Figure 1 Iselin classification (adapted) indicating the different stages of Dupuytren disease.

Differences between the hockey players and controls were determined by Fisher's exact test for nominal and ordinal data, and by the Mann-Whitney U test for data at the interval or ratio level.

Since we expected the two groups to be significantly different on various characteristics, we used propensity score matching to balance the groups.²⁸ A propensity score can be considered as an a priori probability of a participant to be included in the experimental group, in this case the field hockey group, given a set of characteristics. By matching the participants based on the propensity score, we matched field hockey players with controls having the same likelihood to be part of the hockey group. The propensity score was calculated using a logistic regression model based on the variables age, diabetes, smoking, alcohol consumption and familial presence of DD.²⁹⁻³¹ Epilepsy was not included, since only one participant suffered from this disease. Then propensity score matching with replacement was carried out³² with exact match priority. We used a caliper of 0.2 SD of the logit score as the tolerance level for matching.³³ When multiple controls were equally eligible to be matched to a field hockey player, the participant to be matched was chosen randomly. To determine whether the matching procedure successfully balanced the two groups in descriptive characteristics, we did the same statistical comparisons as mentioned in the paragraph above.

A subject-specific generalised linear mixed model was fitted to determine the effect of field hockey as a dichotomous variable on the proportion of DD. A Bernoulli distribution with a logit link function was used. We entered hockey (yes/no) as a fixed effect, and a random effect for matched pairs to control for the correlated observations (resulting from the matching procedure).

Then, to examine the presence of a dose–response relation, we applied logistic regression within the field hockey group of the original (non-matched) database, with field hockey entered as a continuous variable. This variable was standardised to the absolute number of weeks that a participant had played ((hours/week×years)/168). Age was included in the model to account for its potential confounding effect. Additionally, we did an ordinal logistic regression analysis to determine the effect of field hockey on the severity of the disease, defined by the Iselin stage. A sensitivity analysis was performed to gain more insight into the effect of field hockey on the presence of DD.

The propensity matching procedure and analyses were performed in SPSS V.23. Missing values were excluded listwise from the analyses, and a significance level of 5% was used.

RESULTS

A total of 325 individuals participated in this study. Among the field hockey players, 169 of the 204 hockey players who were asked to participate agreed to do so (83%). In the control group, this was 156 of the 247 (63%). Three of the 91 non-participating controls replied that they refused to participate, while the other 88 did not respond. In the full set of 325 participants, 24 of the 169 field hockey players (14%) were manual workers, compared to 67 of the 156 controls (43%). This difference was statistically significant (χ^2 =33.3, p<0.001). We excluded manual workers and controls exposed to vibration (figure 2). The propensity score matching procedure yielded 42 pairs, so the data of 84 participants in total were used in the analyses.

Table 1 shows the descriptive statistics of the field hockey players and the controls before and after propensity score matching. Before matching, there were significant differences between the two groups with respect to age and smoking habits. After matching, differences in characteristics reduced or vanished.

Table 2 shows the proportion of DD in the two groups, as well as the severity of the disease. DD was almost four times more prevalent in the field hockey group compared to the



Figure 2 Schematic representation of the inclusion and exclusion procedure.

controls. In both groups, mild disease (ie, nodules in the absence of contractures) was the most common disease presentation. Only a few participants in both groups showed DD with flexion contractures of the finger(s).

To visualise the proportion of DD with respect to the amount of field hockey exposure, we plotted the logit of the proportion of DD against different groups of field hockey exposure (figure 3). It can be seen that the logit of the proportion of DD was much lower in the control group compared to the field hockey players.

The results of the generalised linear mixed model after propensity score matching indeed show that field hockey playing as a dichotomous variable was significantly associated with the presence of DD (OR=9.42; 95% CI 3.01 to 29.53).

Within the field hockey group, figure 3 demonstrates that the highly exposed groups had a higher logit of the proportion of DD than the lower exposed groups. This indicates the possibility of a dose-response relation. However, applying logistic regression within the field hockey group showed that field hockey as a continuous variable (expressed as (hours/week×years)/168), corrected for age, did not demonstrate a significant dose-response relation with DD (OR=1.03; 95% CI 0.68 to 1.56). The same result was found when disease severity (Iselin stage) was used as an outcome variable (OR=1.00; 95% CI 0.67 to 1.49).

We did a sensitivity analysis to determine if the choice of the model (linear profile for field hockey and the log odds for DD) would affect the conclusion. Two additional analyses were provided. We tried different cut-off points for the amount of field hockey to see if field hockey beyond the cut-off point would entail a higher proportion of DD. The optimal cut-off point was determined at 217 hours/week×years, and provided a p value of 0.059. The second analysis tested a shift in distribution of field hockey between participants with and without DD using a Mann-Whitney U test (p=0.433). Both analyses did not demonstrate a dose–response relation either.

DISCUSSION

After propensity score matching, our main result shows that field hockey has a strong association with DD. This extends previous findings of vibration being associated with DD in the occupational setting.^{8–10} We did not find a continuous dose–response relation within the field hockey group. This seems to be in contrast to several other studies that report a linear dose–response relation between vibration and DD.^{8 16 17}

Why no dose-response relation?

There are several possible explanations that may account for this discrepancy. Previous reported studies may have overestimated the effect due to methodological issues, for example, no physical examination to diagnose DD, or inconsistencies in cross tabs possibly resulting in an incorrect OR calculation.⁸ ¹⁷

Further, we drew our sample among elderly field hockey players who were still playing. Therefore, severe cases of Dupuytren with large hockey exposure might not have entered our sample due to the sampling procedure, as patients with severe disease would not be able to play anymore. Another possibility is that hand injury, which might result from playing field hockey, contributes to the larger proportion of DD in the field hockey group, but not to a dose–response relation. Hand injury has previously been associated with DD,⁵ ^{34–36} and field hockey players might have a higher probability of injury as a consequence of the sport. We did inquire about hand injury to address this, but most participants had difficulties remembering

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Table 1 Characteristics of the field hockey players and the controls before and after the propensity score matching procedure

	Before propensity score matching			After propensity score matching		
	Hockey players (n=145)	Controls (n=58)	p Value	Hockey players (n=42)	Controls (n=42)	p Value
Age in years (median)	67.0	61.5	0.001*	66.0	62.5	0.122*
IQR	65.0–71.0	58.3-68.3		(62.0–69.3)	(59.0–71.0)	
Missing (n)	3	0		0	0	
Alcohol consumption in units/week (median)	10.0	5.0	0.166*	8.5	6.50	0.603*
IQR	5.0-20.0	0.2-11.3		(3.8–14.0)	(2.0–14.0)	
Missing (n)	3	0		0	0	
Smoking in pack years (median)	2.5	6.4	0.001*	3.0	0.0	0.334*
IQR	0.0–13.0	0.0-23.0		(0.0–15.0)	(0.0–11.2)	
Missing (n)	3	0		0	0	
Diabetes (n)	11 (7.6%)	5 (8.6%)	0.779†	4 (9.5%)	3 (7.1%)	1.000†
Missing (n)	0	0		0	0	
Epilepsy (n)	0 (0.0%)	1 (1.7%)	0.286†	0 (0.0%)	0 (0.0%)	-
Missing (n)	0	0		0	0	
*Mann-Whitney II test						

†Fisher's exact test.

 Table 2
 Proportion of Dupuytren cases and disease severity in the field hockey group compared to the control group

	Hockey players (n=145)	Controls (n=58)
Dupuytren (n)	75 (51.7%)	8 (13.8%)
Missing (n)	0	0
Iselin stage (n)		
1	63 (43.4%)	5 (8.6%)
2	5 (3.4%)	1 (1.7%)
3	6 (4.1%)	2 (3.4%)
4	1 (0.7%)	0 (0.0%)
Missing (n)	0	0

the type of injury, and which hand or finger was injured. Thus, we could not evaluate this hypothesis any further.

The results of this study suggest that the onset of DD may be triggered by playing field hockey, and that it does not influence the disease course. One theory is that mechanical stress (of field hockey) produces microtrauma that may trigger the Wnt-signaling pathway, which is involved in cell proliferation and differentiation.³⁷ By activating this pathway through mechanotransduction,³⁸ proliferation of fibroblasts occurs. This theory is supported by the findings that the anomalies in DD tissue resemble the early stages of wound repair.³⁶ ³⁹ This would support our findings of an effect of field hockey with the absence of a dose–response relation.

Strengths and limitations

The strength of this study is that all the participants were physically examined by medical doctors experienced in diagnosing DD. An inter-observer agreement study indicated that experienced observers reach an agreement of 95–100% for diagnosing the disease.⁴⁰ This guarantees the reliability of our observations.

Another strength is the use of propensity score matching. In observational studies like this, randomisation is not possible. Therefore, several sources of bias are introduced that can result in incorrect conclusions.²⁸ After matching, the differences between the two groups were reduced or vanished, indicating

that propensity score matching provided a more balanced view on the characteristics between the field hockey players and controls. Additionally, by excluding all manual workers, we had a relatively homogeneous group of participants who were all exposed to vibration in the same way. This further extends the reliability of our results. Finally, using a sensitivity analysis, we tested whether we chose the correct model to evaluate the association. This was not significant, so we can assure that there is no dose–response relation between field hockey and the presence of DD in our sample.

There are several limitations in our study. First, recall bias was probably present in our study, since many variables were gathered using an interview. Unfortunately, there are no alternatives to determine, for instance, the lifetime exposure to vibration as in field hockey.

Second, the sample size calculation showed that we needed 74 participants in each group, while we had 42 participants in each group after matching. However, this sample size calculation was performed for a two-sample χ^2 -test and not for matched samples. Propensity score matching increased our effect size compared to an analysis without correction.

Third, it is likely that the surface on which the field hockey players have played, and the material of which the sticks have been manufactured, can have large influences on the biomechanics of a field hockey shot. We did not gather this kind of information, so it might be that the lack of a dose–response relation can be explained by this.

Finally, we probably missed severe cases due to the sample selection, since hockey players with severe DD may not be able to play anymore. Missing severe cases has probably resulted in an underestimation of the effect size in our sample. Although we examined 83% of the hockey players, it might be that those who did not participate were not affected and therefore did not feel the need to participate. However, by emphasising the importance of participation before the start of the tournament, especially if the players were thought to be unaffected, we tried to limit the selection bias. Unfortunately, selection bias can never be ruled out in observational studies like this.

In conclusion, we found that field hockey playing has a strong association with the presence of DD. Clinicians in sports medicine should be alert to this less common diagnosis in this sport.

Figure 3 The effect of field hockey on the logit of the proportion of Dupuytren disease.



What are the findings?

- ► Field hockey is strongly associated with Dupuytren disease.
- ▶ No dose-response relation was found.
- Field hockey seems to trigger the onset of Dupuytren disease, but appears to have no influence on the course.

How might it impact on clinical practice in the future?

Given the high prevalence of Dupuytren disease in male field hockey players, clinicians should be alert to this less common diagnosis in sports medicine.

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