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Epidemiological Evaluation Of Ledderhose Disease (Plantar Fibromatosis) In A Cohort Of Patients With Dupuytren's Disease: A Single Center Experience

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1. Abstract

There is limited and inconsistent description of the epidemiology of Ledderhose disease (LD). Our primary aim was to evaluate the epidemiology of LD in a cohort of patient's with Dupuytren's disease (DD), the secondary aim was to assess if any associations existed between LD and patients' risk factors for LD. We conducted a cross-sectional cohort study from May 2019 to May 2020 and data were collected from patients in a single DD clinic. There were 76 patients recruited. In all cases examined, the prevalence of LD was 42% (n=32) with a high proportion of males (78%) with LD. There were no statistically significant differences in LD risk factors between patients with DD with or without LD ($p>0.05$). There were no potential LD predictors determined. Knowledge of epidemiology of LD in DD patients can aid accurate counselling of patients with LD and subsequently provide a base for aetiological studies.

2. Keywords

Dupuytren's Disease, Epidemiology, Ledderhose Disease, Morbus Ledderhose, Palmar Fibromatosis, Plantar Fibromatosis, Foot, Hand.

3. Introduction

Plantar fibromatosis was first reported in 1875 by Madelung (Madelung, 1875)[1] and first diagnosed in 1897 by Dr. Georg Ledderhose (Ledderhose, 1897)[2]. Dr. Ledderhose recognized the nodules in the plantar aponeurosis as an equivalent of Dupuytren´s disease and characterized the disease as a proliferation of cells and blood vessels with a tendency to shrink. According to Leclerq, "plantar lesions occur as painless lumps in the non-weight-part of the sole, usually near the highest point of the arch. They usually produce no symptoms other than because of their size, and patients may be unaware of them" (Tubiana et al, 2000)[3]. The skin is generally mobile over the lump, which is fixed to the plantar fascia. Pain is infrequent, usually limited to mild discomfort after standing or walking for long periods (Allen, 1955)[4]. These may or may not be tender. In the literature, there are variable reports on the prevalence of Ledderhose disease (LD) in patients with Dupuytren's disease (DD) especially when dealing with different populations.

In an article by Mohede et al (2020)[5], for 730 men with Dupuytren disease, the surgeons' reported prevalence rate of Peyronie disease was 7.8 percent and of ledderhose disease was 16.1 percent. The participants self-reported prevalence rates of 8.8 percent for Pyronie's disease and of 22.0 percent for Ledderhose. Case studies of the disease have described occurrences between the ages of 2 and 83 years, but it has been most described in the 4th and 5th decades of life (Schmidt, 2019)[6]. There have been inconsistent reports of a sex bias in the prevalence of LD. Recent literature published in 2019 reported no significant differences between males and females (Fuiano et al., 2019; Schmidt, 2019) [6,7] while previous studies reported higher prevalence in males (Carroll et al., 2018; Gudmundsson et al., 2013; Neagu et al., 2018; Veith et al., 2013)[8-11]. Bilateral feet involvement ranges between 20-50% of cases (Fuiano et al., 2019)[7]. Reports suggest about 50% have coexisting palmar involvement (Pickren et al., 1951; Newman and McQuaid, 2019)[12,13]. Patients with DD are predisposed to developing severe contractures with rapid progression and higher risk of recurrence, estimated at 71% vs

23% (Gil et al, 2021)[14].

The aetiology of LD remains unknown. However, this condition is more frequently seen in patients with palmar fibromatosis (Dupuytren's disease, 5-21.3% association), penile fibromatosis (Peyronie's disease, 1-3% association), frozen shoulder and fibrous subcutaneous nodules (knuckle pads) on the dorsal aspect of proximal interphalangeal finger joints (Gudmundsson et al., 2013; Neagu et al., 2018; Schmidt, 2019 and Gil et al., 2021) [9,10,6,14]. There are also reports suggesting LD and frozen shoulder share a similar immunohistochemistry appearance with Dupuytren's disease (Bunker and Anthony, 1995; Carroll et al., 2018; de Palma et al., 1999)[15,8,16]. The risk of occurrence of LD is increased by alcoholic liver dysfunction, diabetes mellitus, epilepsy with long-term phenobarbital medication, nicotine abuse, repeated trauma, previous exposure to vibrations, vascular or autoimmune disorders, genetic inheritance and family history of LD (Neagu et al., 2018; Schmidt, 2019)[10,6].

In view of the variable reports on prevalence of LD, our primary aim was to evaluate the epidemiology of LD in an at-risk population of patients with Dupuytren's disease. In addition, our secondary aim was to assess any associations between LD and patients' risk factors for LD.

4. Methods

A cross-sectional study was nested in a large prospective cohort study (study protocol approved by the Human Research Ethics Committee. The study was conducted in an outpatient setting.
Participants aged 18 and above were recruited from the patients registered with the Dupuytren's clinic between May 2019 and May 2020. Written informed consent was obtained from all participants prior to commencement. Data collection included a questionnaire and clinical examination findings. The questionnaire was developed by the research team and consisted of questions about basic anthropometric data and relevant comorbidities (known personal history of LD, family history of DD and LD, alcohol consumption history, smoking status, Peyronie's disease, diabetes mellitus, frozen shoulder, epilepsy, alcoholic liver disease, repeated foot trauma, autoimmune disorder, vascular disorder) see Appendix 1.
Two plastic surgeons examined the participants' feet by palpating the plantar aspect of the feet. The surgeons were blinded to each other's findings. Diagnosis of LD was made when nodules were found in medial aspect of the foot, along the plantar fascia. Left or right foot, and the number of nodules were recorded.
Data were entered into an excel file and imported into STATA for statistical analysis (v16 StataCorp LLC, USA). Normally distributed continuous variables were expressed as mean and standard deviation (SD), otherwise as median (Interquartile range (IQR). Comparisons between data from Ledderhose and non-Ledderhose groups were performed using t-test or non-parametric equivalent Wilcoxon-Mann-Whitney test. Categorical variables were described as percentage frequencies (%) and analysed using Fisher's exact test. Odds ratios were calculated using linear or logistic regression analysis. P-values were considered significant if less than 0.05.

5. Results

Seventy-six participants with Dupuytren's disease consented to participate and fifty-seven (76%) of these were male. Thirty-two participants with Dupuytren's disease (42%) also had Ledderhose disease. There were no statistically significant differences in age, sex and body mass index (BMI) for the Dupuytren's disease patients with or without LD (Table 1). Ledderhose disease was observed to involve both feet in 53% of cases (n=17), 22% on the right foot (n=7) and 25% on the left foot (n=8). On average, participants with Ledderhose disease had two nodules on examination (Table 2). Only 18 of 32 (56.3%) of participants with LD were aware that of nodules were present on their feet (Table 1).

There were no statistically significant differences between participants with or without Ledderhose disease for the frequency of having a family history of Dupuytren's disease or Ledderhose disease. Nor was there a difference in their history of alcohol consumption or their smoking status, or the incidence of Peyronie's disease, diabetes mellitus, frozen shoulder, epilepsy, alcoholic liver disease, repeated foot trauma, autoimmune disorder or vascular disease (Table 2).

Increasing participants' age and elevated BMI were associated with lower odds of Ledderhose disease (OR 0.97, CI 0.83 - 1.1.3 and OR 0.76, CI 0.34 - 1.69 respectively) though not statistically significant (Table 3). All other variables (sex, family history of DD, family history of LD, current smoking status and current alcohol consumption) had no statistically significant association with Ledderhose disease development.

6. Discussion

Ledderhose disease epidemiology has not been precisely assessed. The present study aimed to evaluate the epidemiology of Ledderhose disease in a Dupuytren's disease cohort and to assess the associations between Ledderhose disease and risk factors for LD. The most interesting observation in our study is a higher prevalence of Ledderhose disease in patients with DD compared to the literature (42% vs 5-21.3%) (Schmidt, 2019; Neagu et al., 2018; Gudmundsson et al., 2013)[6,10,9], an important finding for future aetiology and interventional studies. The higher prevalence may well be because our cohort was selected from participants with known history of DD.

We have found that there is a sex difference in DD patients with a higher proportion of males in this group (76%), like other reports (Carroll et al., 2018; Gudmundsson et al., 2013; Neagu et al., 2018; Veith et al., 2013)[8,9,10,11]. However, a few recent articles reported no sex difference (51.7% males vs. 48.3% females) (Fuiano et al., 2019; Schmidt, 2019)[7,6]. The male distribution in those with or without LD was high as well (78% vs 74% respectively) although these percentages were not statistically significantly different. The mean age of patients affected by LD falls within the age range (2-83 years) described in the literature (Schmidt, 2019)[6]. Almost half of the participants with LD in our cohort were unaware of the

presence of their LD prior to examination. This highlights a very important point to also examine feet of patients presenting with DD since LD does coexist with DD in some cases. Bilateral foot involvement was observed in 46.9% (n=15) and this is consistent with what has already been reported (20-50%) (Fuiano et al., 2019)[7]. Overall, we found no statistical differences in LD and non-LD groups based on risk factors for LD (Table 1 and Table 2).

Previous studies have reported concomitance of LD with other conditions such as Peyronie's disease, frozen shoulder, diabetes, epilepsy, alcoholic liver disease, smoking and repeated foot trauma (Gudmundsson et al., 2013; Neagu et al., 2018; Schmidt, 2019)[9,10,6]. However, this was not observed in this cohort. We analysed age, sex, BMI, family history, smoking history and alcohol intake history as predictors of Ledderhose disease. However, none of the variables were found to be a statistically significant predictor (Table 3).

This study is limited by collection of data within single centre. This has the potential to affect the generalisability of results. A multi-centre study will be useful in the future to validate these findings. Researchers and clinicians should consider multisite data collection to increase data collection and improve the statistical power to better understand this condition.

7. Conclusions

The precise aetiology of LD is unknown. Our study found a higher prevalence of LD in patients with DD compared to what has been previously reported in the literature. There was a higher proportion of males with LD compared to females. LD was underreported by patients prior to examination. This calls for increased awareness and recognition of the condition. Studies with larger participant numbers are required to better understand the risk factors for developing LD.

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