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ORIGINAL ARTICLE

Prevalence of palmar fibromatosis with and without contracture in asymptomatic patients

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Abstract

Background: This retrospective study documents the proportion of hand clinic patients presenting with palmar fibromatosis with and without contracture. **Methods:** All “new” patients >18 years presenting to a single surgeon’s hand clinic over a 16-month period were included, and information was abstracted from chart review regarding patient demographics, reason for presentation, presence or absence of palmar fibromatosis, contracture, and prior known diagnosis of Dupuytren’s disease. The percentage of asymptomatic patients with palmar fibromatosis was calculated. **Results:** Of 827 patients, 306 had palmar fibromatosis. Among all patients, 33% of male and 40% of female patients had palmar fibromatosis. Only 8% had contractures, while 92% had palmar fibromatosis without contracture. Among those who had contractures, 81% presented with a primary complaint of Dupuytren’s disease (symptomatic contracture). Prevalence of palmar fibromatosis increased with increasing age. **Conclusion:** The findings demonstrate that Dupuytren’s palmar fibromatosis is common and often present without overt contractures.

Key Words: Flexion contracture, Dupuytren’s disease, Dupuytren’s contracture, morbus Dupuytren, palmar fibromatosis

Introduction

Dupuytren’s disease is a fibroproliferative disorder affecting the hand, which was first described by Felix Plater of Basel, Switzerland in 1614 and then by Henry Cline of London, England in 1777 [1,2]. It is characterised by a thickening of the palmar fascia, in which abnormal myofibroblasts lead to the formation of nodules in the proliferative stage of Dupuytren’s disease. The involutinal stage follows, whereby the myofibroblastic cells align along tension lines within the nodule and spread along the fascia into the fingers, laying down thick collagen and developing a cord. In the final, residual phase, the cord tightens and stiffens. The myofibroblasts and nodules diminish, leaving behind tissue with a sparse population of fibroblasts and thick bands of collagen. Palmar fascia is normally composed of Type I collagen and in Dupuytren’s this is slowly replaced with Type III [3], which is significantly thicker, tighter, and shorter. Ultimately, these Dupuytren’s cords may contract, leading to a permanent flexion contracture of the digits and deformity of the hand [4–7]. There are multiple factors involved in the development of Dupuytren’s disease, including genetic predisposition [8], environment, diabetes mellitus, alcoholism, trauma [3,9,10], and increased expression of growth factors, based on results that showed a higher percentage (vs control) of expression of growth factors in palmar fascia from patients with Dupuytren’s contracture using RT/PCR [3,11].

Existing literature regarding the epidemiology of Dupuytren’s disease suggests that the prevalence is highest in Caucasians of Northern European descent and that it is

more common in males [12–14]. Likewise, existing literature implies that patients with the nodular form of Dupuytren’s disease will commonly, but not inevitably, develop contracture of the hand [15,16]. The cases where palmar fibromatosis does not lead to contractures could be explained by a study conducted by Rayan and Moore [17], who describe what may be a variant of Dupuytren’s disease that is non-proliferative. This variant and the typical Dupuytren’s disease may represent two distinct clinical entities, with different courses and different prognoses. However, as DiBenedetti et al. [18] showed in a 2011 report, Dupuytren’s disease may be largely underdiagnosed when patients present with initial hand symptoms such as ropelike growths or hard bumps on the hand. This may be in part due to the usually painless and asymptomatic nature of palmar thickening, which patients may not notice (Figure 1).

Our current study was conducted to determine the prevalence of asymptomatic palmar fibromatosis without contracture in a group of patients presenting to a hand clinic without complaints of palmar fibromatosis. Our hypothesis is that asymptomatic palmar fibromatosis (without contracture) is common, increases with increasing age, and is much more common than palmar fibromatosis with contracture.

It is important to recognise palmar fibromatosis, since surgical procedures performed for other reasons or other trauma may worsen the palmar thickening or increase the risk of progression, and patients should be counselled appropriately [19–25]. Furthermore, environmental factors such as trauma and surgery have been shown to be important in the

pathogenesis of what is called Non-Dupuytren's palmar fascial disease [17].

Materials and methods

Following appropriate Institutional Review Board approval, a retrospective review was conducted of all consecutive patients who presented to the senior author's (JEA) hand clinic over a 16-month period (June 2012–September 2013). We excluded all patients <18 years of age and screened all of the patients to include only "new" patients in the study. The study was restricted to "new" rather than "return" patients in order to ensure that patients were counted once and only once in the study. This surgeon author practices in a metropolitan area at three sites that were included in the study, including a University tertiary referral centre, and two practices that include referral and community hand care; with an elective practice exclusively devoted to surgery of the hand, wrist, and elbow. These practices comprise patients from all economic strata. Data were abstracted from the medical record, including age at presentation, gender, presence or absence of palmar fibromatosis, presence or absence of digital contractures, if there was a prior known diagnosis of Dupuytren's, and reason for presentation to the hand surgeon's office.

Diagnosis of palmar fibromatosis and presence or absence of digital contracture was based on physical examination of the hand by the senior surgeon.

Statistical analysis was performed with Chi-square analysis and Fisher's Exact Test. In addition, our analysis included a calculation of the prevalence of Dupuytren's disease as a function of gender and age. Statistical significance was defined as $p < 0.05$.

Results

Of the 827 "new" patients of age ≥ 18 years that were seen in the surgeon's hand clinic during the 16 month study period, 57% ($n = 474$) were women and 43% ($n = 353$) were men. A total (male + female) of 306 patients (37%) had palmar fibromatosis. In the female group, 40% ($n = 188$) had palmar fibromatosis compared to 33% ($n = 118$) of the male patients, resulting in a male-to-female ratio of 1:1.59 for patients with palmar fibromatosis. The average age of the female patients at presentation was 50.4 years (median = 51 years, range = 18–94 years), which is older than male patients (average = 46.0 years; median = 47 years; range = 18–98) ($p < 0.0001$). Table I shows the distribution of total female and male patients by age group, as well as the distribution of female and male patients who were diagnosed with palmar fibromatosis.

Among the 306 patients with palmar fibromatosis, 8% ($n = 26$) presented with contracture, while 92% ($n = 280$)



Figure 1. Asymptomatic palmar fibromatosis is noted in this female patient with subtle changes in the palm.

were asymptomatic (i.e. had palmar thickening but no contractures (Figure 1)). Among those who had contracture, nine were females and 17 were males. Furthermore, 21 patients presented to the hand clinic with a primary complaint of symptomatic contracture, while five presented for reasons other than Dupuytren's disease, but had Dupuytren's contractures. When age was considered, there was a statistically meaningful trend toward increasing prevalence with age by decade ($p = 0.0009$) (Figure 2), with the majority of the significance appearing to come from the males.

Discussion

These findings indicate that palmar fibromatosis is a common condition and that most patients (92%) with palmar fibromatosis do not have contracture and are asymptomatic ($p < 0.0001$). Based on this retrospective 16-month study in a single surgeon's practice, and considering that only 8% of palmar fibromatosis patients presented with contracture, this suggests the possibility that most patients may not go on to develop contractures, despite having palmar fibromatosis. Another possibility would be that the vast majority of these patients could have what has been

Table I. Distribution of Dupuytren's disease by age and gender.

	<30	30–39	40–49	50–59	60–69	70–79	>79
No. with Dups/total, males	6/76 (7.9%)	7/56 (12.5%)	19/59 (32.2%)	39/87 (44.8%)	29/50 (58.0%)	9/12 (75.0%)	9/13 (69.2%)
No. with Dups/total, females	11/55 (20.0%)	11/64 (17.2%)	36/91 (39.6%)	59/134 (44.0%)	46/79 (58.2%)	18/39 (46.2%)	7/12 (58.3%)
No. with Dups/total, males + females	17/131 (13.0%)	18/120 (15.0%)	55/150 (36.7%)	98/221 (44.3%)	75/129 (58.1%)	27/51 (52.9%)	16/25 (64.0%)

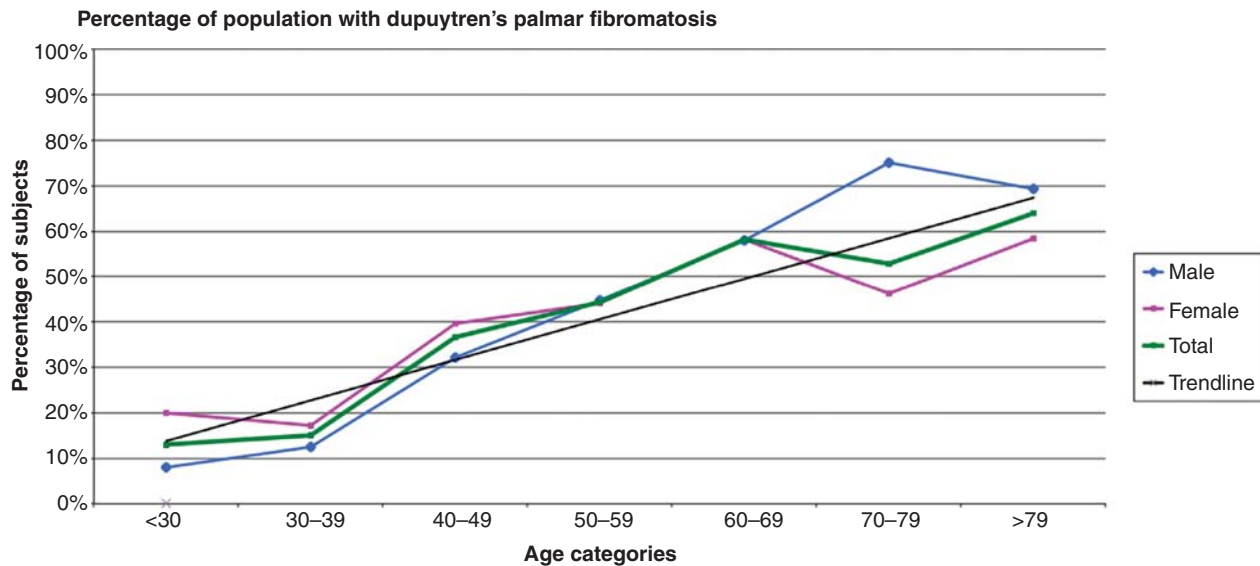


Figure 2. Prevalence of Dupuytren's disease as a function of age and gender. There is a statistically significant increase in palmar fibromatosis with increasing age by decade. Total population (green)*: $R^2 = 0.91$, $p = 0.0009$; Male population (blue)*: $R^2 = 0.95$, $p = 0.0002$; Female population (pink)*: $R^2 = 0.79$, $p = 0.007$; Statistically significant trend line²⁵ (black) for total population. *Statistically meaningful trend.

called non-Dupuytren's fascial palmar disease, which would explain the absence of contracture, although only a prospective study would allow one to make this conclusion and make the determination if patients have Dupuytren's or non-Dupuytren's fibromatosis. Interestingly, although the great majority of patients with palmar fibromatosis and contractures (21 out of 26) presented to the hand clinic with symptomatic Dupuytren's contracture as their primary complaint, a curiously high remainder (five out of 26) presented for reasons other than the contractures. This may suggest that many patients, even those with contractures, adapt well to their condition and may go undiagnosed despite contracture as they or their referring providers may not notice it. In addition, the prevalence of palmar fibromatosis significantly increases with increased age, in both females and males (Figure 2).

However, in contrast to prior reports in the literature [26-28], the prevalence of palmar fibromatosis in our study was similar between the genders. In three different studies, Zemel [28], Zemel et al. [29], and McFarlane [27] reported a male-to-female ratio ranging between 6:1-15:1, while the ratio in our study was 1:1.59. Our divergent results might be explained by differing criteria for defining "Dupuytren's disease". It is possible that prior studies documented only Dupuytren's palmar fibromatosis with contracture (failing to document cases of palmar fibromatosis without contracture), while in the current study we document both palmar fibromatosis with and without contractures and document the same. Some prior studies fail to specify if palmar fibromatosis without contracture was included in documentation of prevalence of Dupuytren's disease. In the present study, however, when Dupuytren's fibromatosis with contracture (rather than fibromatosis without contracture) was considered, more males than females were affected, as is seen in other studies.

One reason Dupuytren's contracture is believed to affect more males than females was provided by Pagnotta et al. [29,30], who showed a correlation between an abnormally

high number of androgen receptors and palmar fibromatosis. Because men have higher androgen levels, which target the receptors associated with palmar fibromatosis, it was proposed that males, thus, have greater likelihood of contractures. The high number of female patients with palmar fibromatosis without contractures in our study could be due to the higher average age of female patients (50.4 years) vs male patients (46.0 years) as a confounding factor; alternatively, the prevalence of fibromatosis without contracture in both genders may be related to baseline genetic or environmental factors, with development of contractures being more likely in males in part related to hormonal influences. In fact, in our series, fibromatosis with contracture was more common in males (65%) over females (35%), suggesting that the prevalence of Dupuytren's fibromatosis is similar across genders, but women are less likely to develop contractures (i.e. be symptomatic).

One limitation of this study is that it represents a subset of patients in a single metropolitan geographic area in the upper Midwest who presented to a single surgeon's practice; the age and gender distribution in the study are not representative of the US population in general. In particular, Minnesota, where the study was carried out, has a greater proportion of individuals of Northern European ancestry than the US population in general, and Dupuytren's disease is more common in that demographic group (US Census [32]). Further investigations conducted on a larger geographical scale will likely provide further information about the prevalence and natural history of Dupuytren's, including asymptomatic palmar fibromatosis and palmar fibromatosis with contractures.

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