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Dupuytren's Disease Characteristics in Primorsko-Goranska County, Croatia

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A B S T R A C T

Dupuytren's disease predominantly occurs among Europeans, with higher reported prevalence in Northern than in Mediterranean European countries. We evaluated the recurrence and extension rates among Caucasian patients from Primorsko-goranska County in Croatia who underwent partial fasciectomy as treatment for Dupuytren's disease. Furthermore, we investigated the influence of diathesis factors on disease progression. Recurrent disease was observed in 68 out of 93 patients (73%). There where 48 (52%) patients with extension of the disease. Differences were found between patients with recurrent disease and those without recurrence regarding age at onset, age at operation and duration of disease. We compared patients younger and older than 50 years at disease onset, and found that older patients had a significantly higher recurrence rate. Characteristics of our patients fit into the picture of typical Dupuytren's disease except for the influence of early age at onset. Among our patients late age at onset proved to be a diathesis factor.

Key words: *Dupuytren's contracture, Dupuytren's diathesis, partial fasciectomy, follow-up study*

Introduction

Dupuytren's disease predominantly occurs among Europeans while it is rather sporadic among Africans or Asians[1,2]. It is commonly believed that the disease originated with and was disseminated by the Vikings, thus explaining the higher prevalence in Northern than in Mediterranean European countries[3]. Regardless of the familial component, suggesting an autosomal dominant pattern of inheritance with incomplete penetrance[4], the etiology of Dupuytren's disease is still far from being completely revealed. The Dupuytren's diathesis, a term first introduced by Hueston, describes certain characteristics of the disease with an aggressive course and higher level of recurrence after surgical treatment[5]. Its features are an early onset of disease, bilateral hand involvement, positive family history, and ectopic lesions. Numerous other factors like history of smoking, alcohol consumption, diabetes mellitus, epilepsy, manual labor, and hand trauma have been suggested with the cause of Dupuytren's disease, and are still a matter of discussion[6,7].

The goal of this study was to determine the Dupuytren's disease characteristics in Primorsko-goranska County in Mediterranean European country of Croatia, as it has been implied that Dupuytren's disease manifests itself differently in different countries[8]. Furthermore we aimed to assess the influence of diathesis factors on recurrence and extension of Dupuytren's disease among our patients.

Materials and Methods

Our research was conducted at the Department of Plastic and Reconstructive Surgery in University Hospital Center Rijeka in Croatia. University Hospital Center Rijeka is the largest hospital in Primorsko-goranska County and represents the central healthcare institution for over 300 000 inhabitants of the region.

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At our department 208 patients underwent partial fasciectomy as surgical treatment of Dupuytren's disease between January 1, 1996 and December 31, 2002. Of these, 203 were men (98%) and 5 women (2%), male:female ratio 41:1. Inclusion criteria were male patients who underwent only one operation during the mentioned period of 7 years. A total of 170 patients met the inclusion criteria and were invited by telephone to a follow-up examination. Among these, 77 (45%) patients could not be reached or refused to participate, while 93 (55%) patients agreed to enroll in the study. All of the enrolled patients were Caucasian of European ethnicity that underwent partial fasciectomy as treatment of Dupuytren's disease. Partial fasciectomy is the most common surgical procedure performed to treat Dupuytren's disease. During the procedure only diseased fascia is remove, while the one appearing healthy is left untouched. The age of the patients ranged from 43 to 80 years, with mean of 65 (SD =9) years. The median follow-up was 7 years (range, 3–12 years).

Patients' data including date of birth, age at onset of disease, family history of disease, presence of ectopic lesions, and bilateral hand involvement were extracted from our charts, as well as physical examination data gathered pre and postoperatively. Early onset of Dupuytren's disease was defined as initial manifestation of disease in patients younger than 50 years[9]. At follow-up evaluation each patient signed the consent form and was examined by the first author for signs of recurrence and extension of Dupuytren's disease. Additionally, during this visit the patient reviewed all the data extracted from our charts. Recurrence was defined as the development of new Dupuytren's disease lesions including the smallest palpable nodule irrespective of a presenting contracture in the same area where fasciectomy had been performed[5]. Hueston's table top test was used to identify reappearance of the contracture sufficient to require further surgery[10], combined with standard surgical criteria of metacarpophalangeal (MCP) joint contracture of greater than 30° or any proximal interphalangeal (PIP) joint contracture. Extension of disease was defined as the development of new Dupuytren's disease lesions irrespective of a presenting contracture outside the area where partial fasciectomy has taken place and in an area where no lesions were detected previously[5].

The distribution of age at onset and age at operation did not statistically differ from the normal distribution, so the results were presented by mean and standard deviation. To compare differences regarding age at onset and age at operation between recurrence and no recurrence group we used one-way ANOVA. The duration of disease was not normally distributed, so the results were shown by median and range (5^{th}–95^{th} percentiles). Man-Whitney test was used to determine the difference in disease duration between recurrence and no recurrence group.

The analysis of differences among our patients regarding age at onset was performed using Pearson, χ^2 test. All statistical values were considered significant at the P level of < 0.05. Statistical analysis of data was performed using Statistica for Windows, release 7.1 (Statsoft, Inc., Tulsa, OK, USA).

Results

The age at onset of Dupuytren's disease ranged from 28 to 69 years, with mean of 53 (SD=10) years. Initial onset of disease before the age of 50 years was experienced by 36 (39%) patients. At operation, the mean age was 58 (SD=9) years, and ranged from 37 to 72 years. The median duration of disease was 10 years (range, 5–27). The median time between onset and operation was 3 years, whereas for the highest number of patients (N=23, 25%) this period lasted for 2 years. Positive family history was stated by 30 (32%) patients, and bilateral hand involvement was present in 62 (67%) patients. Ectopic lesions were demonstrated in 13 (14%) patients. Out of these patients, 9 (10%) had Garrod's pads, 4 (4%) had Lederhose's disease, and none had Peyronie's disease (Figure 1).

Follow-up examination revealed that 68 out of 93 patients (73%) had recurrent Dupuytren's disease (Figure 2). Only 23 out of 68 patients (34%) with recurrence, or 25% of all patients, required further surgical treatment. There where 48 (52%) patients with extension of the disease to other fingers. Recurrence together with extension of the disease was observed in 39 (42%) patients.

We found several differences between patients with recurrent Dupuytren's disease and those without recurrence (Table 1). The mean age at onset in patients with recurrent disease was 55 years (SD=10), whereas in patients with no recurrence it was 48 years (SD=10). The difference in age at onset of Dupuytren's disease between these two groups was significant (P=0.003). Significant difference was also found regarding duration of disease between groups with and without recurrence (P=0.021). In patients with recurrence, the median duration of disease was 11 years (range, 6–27), and in patients without recurrence it was 14 years (range, 5–27). The mean age at operation in patients with recurrent Dupuytren's disease was 59 years (SD=10), which was significantly

TABLE 1
DIFFERENCES BETWEEN PATIENTS REGARDING RECURRENCE OF DUPUYTREN'S DISEASE

Variable	Recurrence (n=68)	No Recurrence (n=25)	P-value
Age at onset (years; mean, SD)	55 (10)	48 (10)	P=0.003*
Age at operation (years; mean, SD)	59 (10)	55 (8)	P=0.039*
Duration of disease (years; median, range)	11 (6–27)	14 (5–27)	P=0.021*

TABLE 2
Differences among Dupuytren's disease patients regarding age at onset

Variable	Patient Age < 50 Years (n=36)	Patient Age ≥ 50 Years (n=57)	P-value
Recurrence	21	47	P=0.011*
Extension	17	31	n.s.
Family history	11	19	n.s.
Bilateral hand involvement	24	38	n.s.
Garrod′s pads	2	7	n.s.
Lederhose′s disease	1	3	n.s.

n.s. = not significant

higher than in patients with no recurrence who on average had their first operation at 55 years (SD=8; P=0.039). No difference was found between these two groups regarding family history of Dupuytren's disease (P=0.974), bilateral hand involvement (P=0.185), presence of Garrod's pads (P=0.056), and Lederhose's disease (P=0.215).

Contrary to the findings concerning recurrence, there were no differences found between groups of patients with and without extension regarding age at onset of Dupuytren's disease (P=0.541), age at operation (P=0.777) and duration of disease (P=0.405). The same was true regarding family history of Dupuytren's disease (P=0.264), bilateral hand involvement (P=0.982), presence of Garrod's pads (P=0.803), and Lederhose's disease (P=0.048).

We compared patients younger and older than 50 years at the time of initial disease onset, and found a difference between these two groups regarding recurrence (Table 2). Patients older than 50 years had a significantly higher recurrence rate than younger patients (P=0.011). There were no significant differences regarding extension of Dupuytren's disease (P=0.501), family history of Dupuytren's disease (P=0.780), bilateral hand involvement (P=0.998), presence of Garrod's pads (P=0.285), and Lederhose's disease (P=0.565) between these two groups.

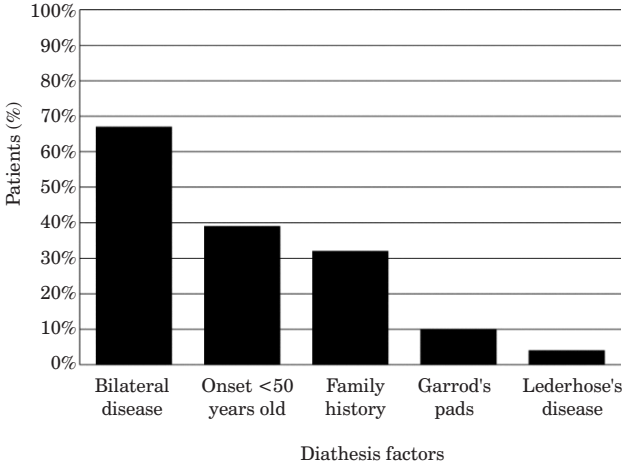

Fig. 1. Proportion of Dupuytren's disease diathesis factors among our patients.

Discussion

Our study, conducted among Dupuytren's disease patients in Mediterranean European country of Croatia, revealed until now unknown disease characteristics in our population. We report a high recurrence rate of 73% for all of our patients. The incidence of recurrence described in the literature varies widely, which is mainly influenced by the definition of recurrence and duration of postoperative follow-up. In our study we defined recurrence as the development of new Dupuytren's disease lesions including the smallest palpable nodule irrespective of a presenting contracture in the same area where fasciectomy had been performed[5]. Our median follow-up time was 7 years, while each patient underwent evaluation for recurrence after a minimum of 3 years following surgery. Originally it was believed that recurrence occurs shortly after surgery, which resulted in studies with short follow-up and low recurrence rates[5]. Later studies with long-term evaluation disproved this theory demonstrating higher recurrence rates and emphasizing duration of postoperative follow-up[11]. High incidence of recurrence among our patients is similar to those reported by other authors, all of which had a long-term follow-up. Tubiana and Leclercq reported a recurrence rate of 66% with a postoperative follow-up time of 10 years[12]. For the same follow-up time Norotte et al found the recurrence incidence to be 71%[13]. In the study conducted by Mantero et al recurrence rate was 77% after 30 years of follow-up[14].

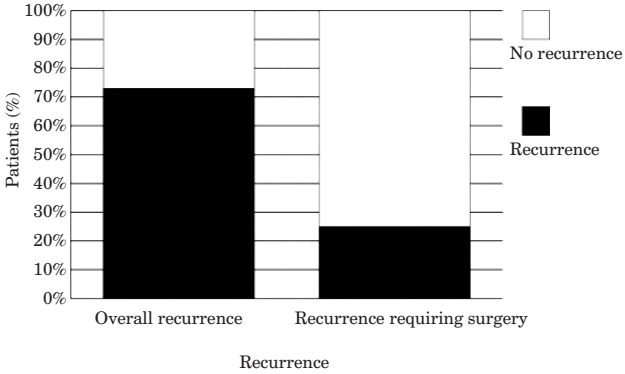

Fig. 2. Rate of overall Dupuytren's disease recurrence and recurrence requiring surgery among our patients.

High recurrence rate among our patients could be to some extent affected by the patient selection process. It could be possible that the incidence of Dupuytren's disease extension and recurrence among patients who refused to participate in the study was lower than among those who chose to enroll. Maybe they had no recurrence or extension making them less interested to undertake new physical examination. Despite of the high reported recurrence rate in our study, only 34% of patients with recurrent disease, or 25% of all patients, required further surgery. We could say that the disease among our patients is thus not predominantly aggressive, but rather that it is following a slow and steady progression course. Extension of disease was present in 48 out of 93 (52%) patients in our study, and this result is again similar to findings of Tubiana and Leclercq[12].

Prevalence rates of diathesis factors among our patients were well within range of those reported in the literature. In the literature, just as in the case of recurrence, prevalence rates of diathesis factors are highly variable depending in the large part on the origin or ethnicity of the examined patients. Our rates were very similar, although somewhat lower, than those recently reported by Hindocha et al in a study conducted among patients in United Kingdom[9]. Issues arising in determining the true prevalence rates must also be taken into account. While an experienced physician usually gathers data about ectopic lesions and hand involvement during physical examination, the patient usually provides data about age at onset and family history. Sometimes patients cannot remember exactly when their disease started or they are not fully acquainted with their family history. Study conducted by Ling demonstrated that the rate of positive family history increased from 16% to 68% after examination of patients' relatives was conducted[4].

Furthermore we assessed the influence of diathesis factors on the incidence of recurrence and extension of Dupuytren's disease. None of the factors were connected to extension, and in the case of recurrence only age at onset demonstrated to be important. Numerous studies have shown that patients younger at the onset of disease are the ones with higher recurrence rates[6,7,15]. In our study the opposite was true, as higher recurrence rate was reported among patients older at the disease onset. Since the peak incidence of Dupuytren's disease is considered to be in the fifth decade of life, the same as in our study, we divided our patients regarding age at onset into two groups, younger and older than 50. Patients older than 50 years at disease onset demonstrated higher rates of recurrence. Early age at onset doesn't seem to count as a diathesis factor among our patients, however late age at onset does. Rayan suggested that there were two distinct clinical entities responsible for palmar fascial contracture, and he named them typical Dupuytren's disease and atypical Dupuytren's contracture[16]. Typical Dupuytren's disease is common among Europeans and its characteristics are peak incidence at fifth decade of life, positive family history, bilateral hand involvement, ectopic lesions and tendency for disease extension and recurrence. On the other hand in atypical Dupuytren's contracture age, family history, race, and ethnic background become irrelevant. Our patients' characteristics correspond to those with typical Dupuytren's disease, with exception of age at onset influence on disease recurrence. It could be said that Dupuytren's disease if behaving somewhat different in our population, which is consistent with McFarlane's suggestions that the disease could take different forms in different countries[8].

One of the limitations of our study is the fact that we only included patients who underwent one operation and excluded those who had two or more surgical treatments. Patients with more operations are the ones with a more aggressive course of the disease, and it would be interesting to see their impact on our findings. Preliminary study we conducted showed that, although major part of these patients did experience initial onset at an early age, their influence does not significantly change the characteristics of our population. Further investigation into this matter is required and we will address it in our future research.

Conclusion

Characteristics of our patients fit into the picture of typical Dupuytren's disease except for the role of early age at onset as a diathesis factor. Early age at onset did not demonstrate any influence on the disease progression, while on the contrary late age at onset was considered to be important.

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KARAKTERISTIKE DUPUYTRENOVE KONTRAKTURE U PRIMORSKO-GORANSKOJ ŽUPANIJI

S A Ž E T A K

Dupuytrenova kontraktura dominantno se javlja u Europljana, s višom prevalencijom u zemljama smještenim na sjeveru Europe. Cilj naše studije bio je procijeniti stupanj recidiva i ekstenzije Dupuytrenove kontrakture u pacijenata iz Primorsko-goranske županije liječenih u Kliničkom Bolničkom Centru Rijeka. Nadalje, željeli smo ispitati utjecaj čimbenika dijateze na progresiju bolesti. Recidiv je primjećen u 68 (73%), a ekstenzija bolesti u 48 (52%) od ukupno 93 pacijenta. Značajne razlike su opažene između pacijenata s recidiviom i onih bez recidiva s obzirom na dob pojave bolesti, dob pri operaciji i duljini trajanja bolesti. Usporedba pacijenata mlađih i starijih od 50 godina pokazala je značajno viši stupanj recidiva u starijih pacijenata. Karakteristike naših pacijenata uklapaju se u tipičnu sliku Dupuytrenove kontrakture, uz izostanak očekivanog utjecaja rane pojave bolesti na stupanj recidiva i ekstenzije. Upravo suprotno, pojava bolesti u kasnijoj životnoj dobi pokazala se kao čimbenik dijateze.