



Risk factors in Dupuytren's diathesis : is recurrence after surgery predictable ?

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In order to investigate the prognostic value of possible risk factors for Dupuytren's diathesis, clinical parameters on disease presentation in an operated group of patients were compared with self-reported recurrence after a minimum 2 years follow-up. In order of significance, the following factors were found to be significantly correlated with disease recurrence : age of onset under 50 years ($p = 0.01$), bilateral disease ($p = 0.01$), Ledderhose disease ($p = 0.01$), first ray involvement ($p = 0.02$), multiple ray involvement (more than 2 digits, $p = 0.02$), ectopic fibromatosis ($p = 0.02$), family occurrence ($p = 0.04$) and male gender ($p = 0.05$). No correlation of self-reported disease recurrence was seen with diabetes, frozen shoulder syndrome or epilepsy. An insight in the significance of the influence of specific risk factors on recurrence rates, helps in creating a clearer representation of Dupuytren's diathesis. This will help the surgeon to more accurately inform the patient and possibly to reconsider and adjust the choice in treatment options.

Keywords : Dupuytren's disease ; recurrence ; risk factors ; fibrosis diathesis ; self-reporting.

INTRODUCTION

Dupuytren's disease displays a high variability in clinical presentation and behaviour, with a spectrum ranging from benign and limited to widespread and progressing disease, recurring fast and severely after surgery. In benign forms, simple

resection of nodules can correct finger contractures without risk for recurrence. However, in the more aggressive forms, postoperative recurrence can be fast and extensile. In such cases, a simple resection might not be sufficient and more aggressive treatment methods may have to be considered. Predictability of risks for postoperative recurrence not only may be of interest to the surgeon to adjust treatment methods ; it may also help in informing the patient more appropriately before he consents to surgery.

Recurrence rates after surgery in Dupuytren's disease reportedly range from 0 to 71% (9). Patients with a severe diathesis have more aggressive forms of the disease with an elevated postoperative risk for recurrent contractures (6). A number of risk factors clinically associated with disease recurrence may indicate a Dupuytren's diathesis.

To investigate the prognostic value of possible risk factors for Dupuytren's diathesis, clinical parameters on disease presentation in a group of

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operated patients were compared with self-reported recurrence after a minimum 2 years follow-up.

MATERIAL AND METHODS

The files of 700 patients who had undergone simple fasciectomy for Dupuytren's disease in our department between 1983 and 2006 were studied. These patients were contacted by a questionnaire with specific questions on clinical presentation, risk factors and postoperative recurrence. A description of the disease presentation was done, taking note of the affected fingers and hands. The age of onset was noted, as was the family occurrence. Ectopic fibrotic lesions were noted: knuckle pads, frozen shoulder syndrome, Ledderhose and La Peyronie's disease. The score based on risk factors as presented by Abe *et al* was calculated (1). Associated conditions or factors such as diabetes, epilepsy and smoking were also asked for.

A correlation study was done to evaluate the influence of the different risk factors on recurrence rates with a regression analysis to include the impact of periods of follow-up and absence of recurrence.

Statistical methodology

No information about the moment of recurrence was included in the questionnaire. As a result, for patients with a reported recurrence it is only known that this occurred between the time of the operation and the

moment when the questionnaire was completed (so-called interval-censored data). Since there is a wide variability in the length of follow-up of the patients, comparing the observed proportions of recurrence is not appropriate. Instead, an accelerated failure time model allowing for interval-censored data is used to study the effect of various predictors on the time to recurrence. Spearman correlations are used to quantify the association between the Abe score and the total number of fingers.

RESULTS

We could include 342 files that were complete, related to patients who were still alive and completed the questionnaire. There were 279 male and 63 female patients. Recurrence was reported in 56% (table I): 58% in male patients versus 42% in female patients. Table I illustrates the most important risk factors and their overall presence. The respective occurrences of these individual factors in recurrent and non-recurrent disease are also shown. Results from univariable accelerated failure time models, on all subjects and on males only, show a time to recurrence approximately 5 times shorter (ratio = 0.19) for males than for females. The influence of the individual risk factors on recurrence is illustrated by showing the recurrence rates with or without the presence of these risk factors.

Table I. — Overview of the presence of most prominent risk factors in recurrent and non-recurrent disease. Percentages of recurrence with and without the presence of these individual risk factors are noted and the probability (p) of correlation with recurrent disease is calculated

Risk factor (RF)	Overall presence RF	In recurrent	In non-recurrent	Overall recurrence	Recurrence if factor absent	Recurrence if factor present	p
Under 50	48%	56%	37%	56%	47%	66%	0.01
Thumb	18%	23%	11%	56%	52%	73%	0.02
5 th finger	81%	84%	77%	56%	48%	58%	0.04
Over 2	18%	23%	13%	56%	53%	71%	0.02
Family	47%	52%	40%	56%	51%	62%	0.04
Fibromatosis	18%	24%	13%	56%	52%	71%	0.02
Ledderhose	14%	18%	9%	56%	53%	72%	0.01
Shoulder	36%	37%	36%	56%	56%	57%	0.9
Male	82%	86%	76%	56%	43%	59%	0.05
Bilateral	65%	79%	58%	56%	48%	61%	0.01

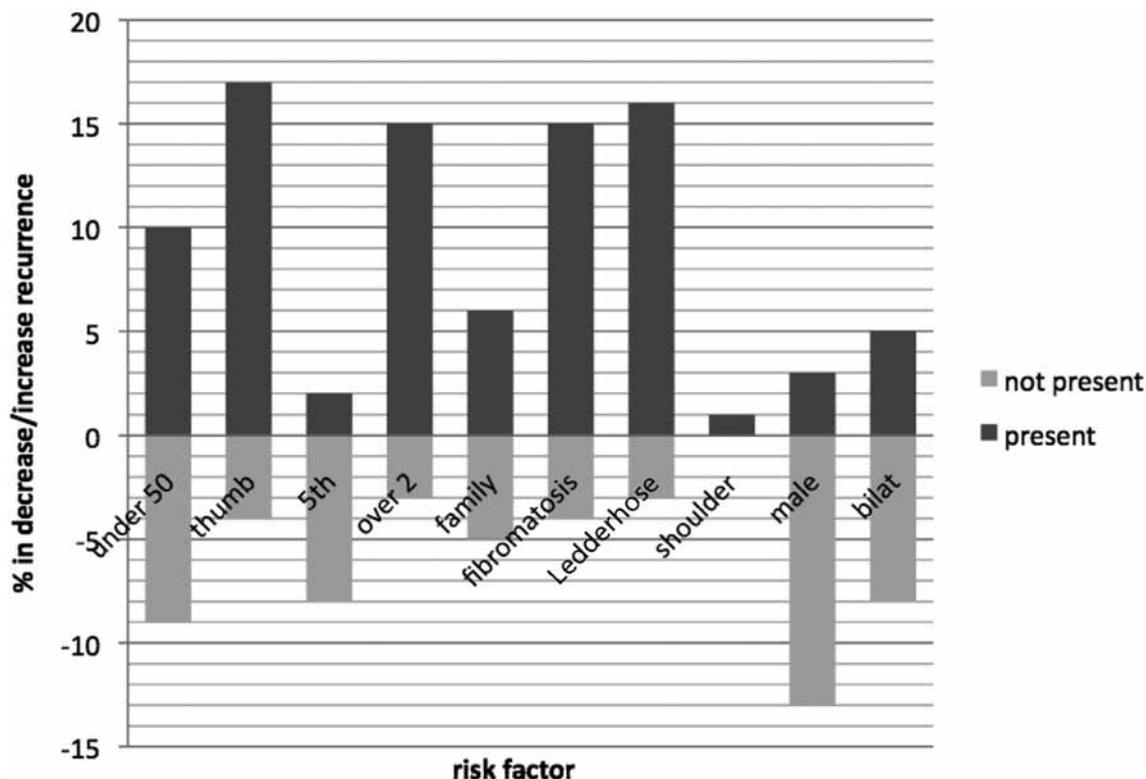


Fig. 1. — Illustration of the relative decrease/increase of recurrence if the individual risk factors for Dupuytren's diathesis were absent/present.

The probability of correlation of the risk factors with disease recurrence is calculated. In order of significance, the following factors are significantly correlated with disease recurrence : age of onset under 50 years ($p = 0.01$), bilateral disease ($p = 0.01$), Ledderhose disease ($p = 0.01$), first ray involvement ($p = 0.02$), multiple ray involvement (more than 2 fingers, $p = 0.02$), ectopic fibromatosis ($p = 0.02$), family occurrence ($p = 0.04$) and male gender ($p = 0.05$). No correlation of self-reported disease recurrence was seen with diabetes, frozen shoulder syndrome or epilepsy.

The influence of these individual risk factors on the rate of disease recurrence is illustrated in figure 1, illustrating the specificity of these risk factors. The sensitivity is illustrated in figure 2, where the overall presence of these risk factors in all patients and the recurrent and non-recurrent group is shown. The Abe score, which combines

these risk factors, also correlates well with disease recurrence ($p = 0.004$).

In the bivariable accelerated failure time models, each predictor is combined with the total number of fingers and next with the score of Abe, both showing an increasing significant correlation. The order of the significance of the most important individual risk factors is illustrated in figure 3.

In multivariable accelerated failure time models, all predictors with a $p < 0.1$ from the bivariable models are combined with total number of fingers, with and without including the Abe score.

Accumulating the risk factors progressively, starting with the most significantly correlated one, as shown in figure 4, reveals a low self-reported recurrence of 34% if no risk factors are present rising in a linear way to recurrences as high as 94% if all factors are present.

Occurrence of risk factors

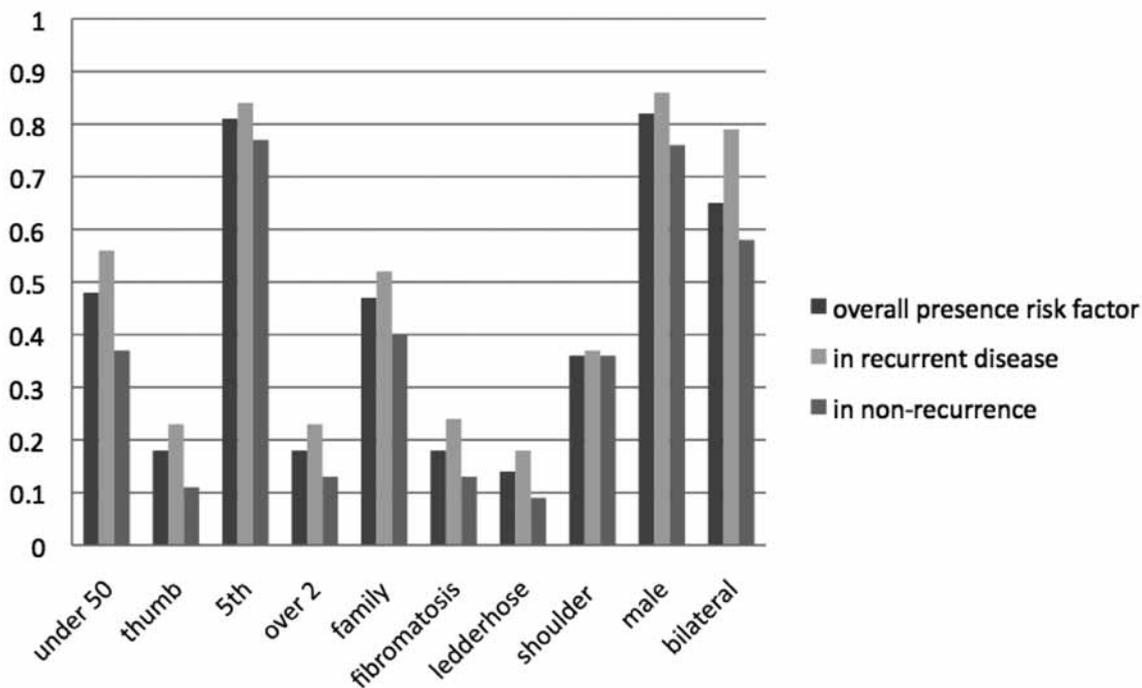


Fig. 2. — Illustrative table showing the overall presence of the specific risk factors in all patients and specifically in the group with self-reported recurrence or persisting absence of symptoms.

correlation recurrence - risk factor

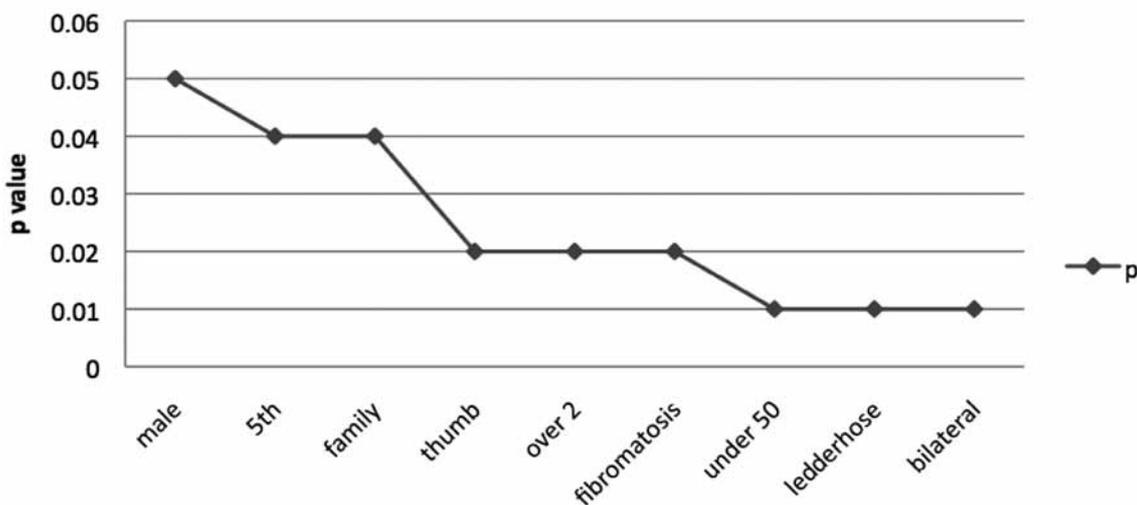


Fig. 3. — The probability of the correlation of specific risk factors with an increasing significant correlation with disease recurrence is illustrated in this graph.

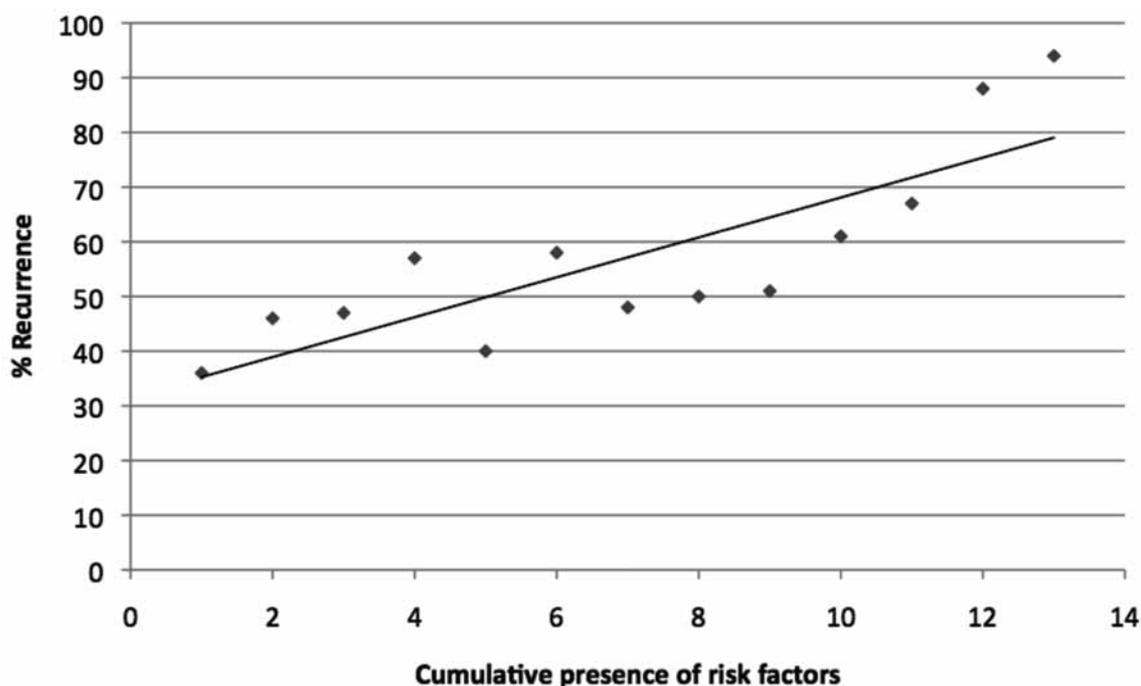


Fig. 4. — Accumulating the risk factors, initiating this multivariate analysis with the most significantly correlated risk factor shows an increasingly high recurrence risk ranging from 36% if no factors are present towards 94% in case of all risk factors present.

DISCUSSION

In this study, risk factors appear to give a significant indication for Dupuytren's diathesis. The relative significance of all individual factors is illustrated in order of significance: most importantly these are the young age of onset under 50 years, bilateral disease and association with Ledderhose disease. Then, first ray and multiple ray involvement (over 2 fingers) indicate a higher recurrence risk. Finally, the presence of ectopic fibromatosis and the family occurrence of Dupuytren's disease are associated with a higher recurrence, as appears to be the male gender. The family aggregation may point towards a genetic background and it has been suggested that Dupuytren's disease has an autosomal inheritance pattern with a variable expression of the disease (5).

Although some factors appear to be related with the occurrence of Dupuytren's disease, like smoking, epilepsy, frozen shoulder and diabetes, no relation was found with the aggressiveness or the post-

operative recurrence of the disease (4,7,8,10). This indicates that these disorders may very well induce a Dupuytren-like disorder, but are not necessarily related to Dupuytren's diathesis with an aggressive form and high recurrence rates.

Although finger contractures can be corrected by surgery, Dupuytren's disease itself is incurable. Bulstrode *et al* even suggested that, if the patient lives long enough, recurrence of the contractures is inevitable (2).

A weakness of this study is the method of self-reporting. Although postal questionnaires are somewhat less accurate than outcome scores given by a clinical observer, they are used more and more, since they allow an evaluation of outcome without a visit and restrict observer bias. The patient's satisfaction is verified by his personal perception, which is the motivation for operating the contractures in the first place (3). Self-reporting of recurrence implies that the patient observes a loss of the surgical gain. Whether this actually means extension, a

true recurrence or even scar tissue formation, this does not make a difference to the patient and has no effect on how we should judge the outcome (3).

Diverging figures have been reported for recurrence rates in Dupuytren's disease (9). An insight in the significance of the influence of specific risk factors on recurrence rates, may help creating a clearer representation of Dupuytren's diathesis. This will help the surgeon to more accurately inform the patient and possibly, to reconsider and adjust the choice in treatment options.

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