

# AN OBJECTIVE METHOD TO EVALUATE THE RISK OF RECURRENCE AND EXTENSION OF DUPUYTREN'S DISEASE

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**This study was undertaken to assess the influence of the factors related to Dupuytren's diathesis on the rates of recurrence and extension of Dupuytren's disease after surgery. The records of 65 patients who underwent surgery for Dupuytren's disease were retrospectively studied and the presence of factors related to diathesis were recorded. The sensitivity and specificity of each factor for predicting recurrence and extension were calculated. Odds ratios and 95% confidence intervals were also calculated and a discriminant analysis was performed to explore correlations between recurrence and extension and the significant variables.**

**Our results confirmed the prognostic value of diathesis. The results have been used to develop a new scoring system for evaluating the risk of recurrence and extension.**

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## INTRODUCTION

The term "Dupuytren's diathesis" was first described by Hueston (1963). Its features are an early onset of disease, bilateral hand involvement, a positive family history, and ectopic lesions. The concept of a diathesis has been confirmed in a large multicentre study (McFarlane, 1985) and many authors support the view that "the Dupuytren's diathesis" is a risk factor for the disease recurrence and extension. However, the influence of each factor of the diathesis on recurrence and extension has not been well documented.

The observation reported by Gordon (1957) and Hueston (1962) that recurrence did not occur beneath a skin graft popularized the use of dermofasciectomy in the treatment of Dupuytren's disease, especially for recurrent cases. However, Tubiana (2000) recommended that primary dermofasciectomy was reserved for patients with a high risk of recurrence, especially the young with rapid progressive disease. Unfortunately, there is no way of evaluating the risk of recurrence objectively before surgery.

The present study was undertaken to assess the influence of factors involved in, or associated with the Dupuytren's diathesis on recurrence and extension.

## PATIENTS AND METHODS

We retrospectively studied the records of patients with Dupuytren's disease who underwent surgery at one of four hand surgery units (Department of Orthopedic Surgery, Chiba University; Department of Orthopedic Surgery, Chiba Municipal Hospital;

Sakura Orthopedic Surgery; and Department of Orthopedic Surgery, Joto Social Hospital), and divided them into two groups. Group 1 consisted of 18 patients with recurrence and/or extension, all of whom had been treated initially with a subtotal fasciectomy and had later required revision surgery for recurrence and/or extension. Their mean length of follow-up was 5 (range, 3-12) years, and their average age at surgery was 59 (range, 38-73) years. Recurrence and extension of Dupuytren's disease were assessed according to the definition of Leclercq (2000). Group 2 consisted of 47 patients who had not had recurrence and extension after more than 5 years after surgery. All of these patients had also been treated with subtotal fasciectomy. Their mean follow-up was 6 (range, 5-14) years, and their average age at surgery was 62 (range, 42-76) years.

Dupuytren's diathesis was originally considered to consist of bilateral disease, early onset of disease, a positive family history, and frequent association with ectopic lesions (Hueston, 1963). We also assessed the influence of other factors on outcome. These included diabetes mellitus, alcoholism, epilepsy, little finger surgery, more than three digit involvement, and radial side involvement.

## Statistical analysis

Fischer's exact test or the chi-square test were used as initial screening tests to determine the risk factors for recurrence and extension. The sensitivity and specificity with regard to recurrence and extension were then calculated for each variable found to be significant in these initial analyses. Odds ratios and 95% confidence intervals were also calculated to assess the ability of

these factors to predict the risk of recurrence and extension. To compare our results with those of two other large series (Hueston, 1963; McFarlane and Botz, 1990), we calculated the sensitivity, specificity, odds ratio and P values for their original data. High sensitivity and high specificity were defined as values above 0.8 (McClure, 2001).

A discriminant analysis between group 1 and group 2 was performed to explore correlations between recurrence and extension and the variables that were found to be significant.

**RESULTS**

Bilateral hand involvement, knuckle pads, plantar fibrosis, early onset of disease, little finger surgery, and radial side involvement were all significantly associated with recurrence and extension. Among the factors included in Hueston's Dupuytren's diathesis, only a positive family history was not significantly associated with either recurrence or extension (Table 1).

Bilateral hand involvement and little finger surgery had high sensitivity and low specificity for predicting recurrence and extension. In contrast, knuckle pads, plantar fibrosis, radial side involvement and early onset of disease had low sensitivity and high specificity. Knuckle pads, plantar fibrosis and radial side involvement had odds ratio values which were well above 1.0 for the lower 95% confidence interval limit while early onset, little finger surgery, and bilateral hand involvement had an odds ratio value of close to 1.0 for the lower 95% confidence limit (Table 2).

In Hueston's (1963) and McFarlane and Botz's (1990) series, ectopic lesions, knuckle pads and plantar fibrosis were significant predictors of recurrence with low sensitivity and high or moderate specificity. In contrast, bilateral hand involvement had high or moderate sensitivity, low specificity, and an odds ratio value of

close to 1.0 for the lower 95% confidence limit. In the Hueston's and McFarlane's series, positive family history was not a significant factor for recurrence and extension and demonstrated high or moderate sensitivity (Table 3). Thus our results were similar to these reported in these two studies.

Based on the discriminant analysis between group 1 and group 2, the following function was developed:

$$Z = 1.46X_1 + 1.26X_2 + 1.44X_3 + 2.65X_4 + 3.36X_5 + 3.44X_6 - 5.75,$$

where  $X_1$  is bilateral hand involvement (with = 1, without = 0),  $X_2$  the little finger surgery (with = 1, without = 0),  $X_3$  the early onset of disease (with = 1, without = 0),  $X_4$  the plantar fibrosis (with = 1, without = 0),  $X_5$  the knuckle pads (with = 1, without = 0), and  $X_6$  the radial side involvement (with = 1, without = 0). If  $Z$  was greater than 0, the subject would be placed in group 1 and if  $Z$  was less than 0 the subject would be placed in group 2 ( $P < 0.001$ , discriminant error rate  $> 10\%$ , Mahalanobis distance = 5.59).

**Table 1—Demographic data of two patients groups**

	Group 1 (n=18)	Group 2 (n=47)	P-value
Diabetes mellitus	7	10	n.s.
Epilepsy	0	2	n.s.
Alcoholism	1	3	n.s.
Bilateral hand involvement	17	31	$P = 0.026$
Plantar fibrosis	12	7	$P < 0.001$
Knuckle pads	10	4	$P < 0.001$
Family history	2	0	n.s.
Early onset	5	3	$P = 0.032$
Radial side involvement	10	5	$P < 0.001$
Little finger surgery	17	31	$P = 0.026$
More than three digits involved	6	5	n.s.

Group 1 (recurrence and/or extension) and Group 2 (no recurrence and extension).  
n.s. = Not significant.

**Table 2—Sensitivity, specificity, and odds ratio for the six factors which were significantly associated with recurrence and/or extension in the present series**

	Se	Sp	OR (95% CI)
Bilateral hand involvement	0.94	0.34	8.8 (1.0–72)
Little finger surgery	0.94	0.34	8.8 (1.0–72)
Early onset	0.28	0.94	5.6 (1.1–27)
Plantar fibrosis	0.67	0.85	11 (3.2–41)
Knuckle pads	0.56	0.91	13 (3.3–54)
Radial side involvement	0.56	0.89	11 (2.8–40)

Se = sensitivity, Sp = specificity, OR = odds ratio, 95% CI = 95% confidence interval.

**Table 3—Sensitivity, specificity, P value, and odds ratio for recurrence and/or extension of the factors studied in Hueston's (1963) and McFarlane and Botz's (1990) series**

	Se	Sp	P-value	OR (95% CI)
<b>Hueston's (1963) series</b>				
Bilateral hand involvement	0.97	0.20	0.001	7.2 (1.9–26)
Knuckle pads	0.60	0.80	0.001	5.9 (2.8–12)
Plantar fibrosis	0.18	0.95	0.016	4.9 (1.3–18)
Positive family history	0.19	0.86	n.s.	1.4 (0.6–3.3)
<b>McFarlane and Botz's (1990) series</b>				
Bilateral hand involvement	0.74	0.35	n.s.	1.5 (0.9–2.5)
Ectopic lesions	0.36	0.79	0.034	2.1 (1.3–3.4)
Positive family history	0.32	0.73	n.s.	1.3 (0.8–2.1)
Early onset	0.40	0.71	0.039	1.6 (1.0–2.6)

Se = sensitivity, Sp = specificity, OR = odds ratio, 95% CI = 95% confidence interval,  
n.s. = not significant.

**DISCUSSION**

Our results confirm the prognostic value of the diathesis as bilateral hand involvement, the presence of ectopic lesions and an early age of onset all predict recurrence and extension.

A test with high sensitivity and low specificity can be utilized for screening but other tests are needed to exclude normal cases. In contrast a test with low sensitivity and high specificity is important for deciding on the treatment of an individual (McClure, 2001). A test with high sensitivity and high specificity is ideal, but no factor assessed in the current study had scores of more than 0.80 for both sensitivity and specificity. Bilateral hand involvement and little finger surgery have high sensitivity and low specificity while plantar fibrosis, knuckle pads, radial side involvement, and early onset have low sensitivity and high specificity for predicting recurrence or extension.

With regard to odds ratios, plantar fibrosis, knuckle pads, and radial side involvement had a stronger influence on recurrence and extension than bilateral hand involvement, little finger surgery, or early onset. Although the risk of recurrence and extension cannot be represented by a single parameter, it may be represented by a combination of these factors.

In light of our results, we suggest a scoring system to evaluate the risk of recurrence and extension. The discriminant function shown in the Results section can be transformed to the following formula:

$$2/3Z + 3.83 = 0.97X_1 + 0.84X_2 + 0.96X_3 + 1.77X_4 + 2.24X_5 + 2.29X_6,$$

which approximates to  $D = a + b + c + d + e + f$ , in which  $D$  is the diathesis score and  $a$  = bilateral hand involvement (with = 1, without = 0),  $b$  the little finger surgery (with = 1, without = 0),  $c$  the early onset of disease (with = 1, without = 0),  $d$  the plantar fibrosis (with = 2, without = 0),  $e$  the knuckle pads (with = 2, without = 0), and  $f$  the radial side involvement (with = 2, without = 0) (Table 4). The clinical distribution of the diathesis scores is shown in Fig 1. When  $D$  was

**Table 4**—Allotted points in the scoring system for the evaluation of the risk of recurrence and extension

Features of variables	Variables	Point
High sensitivity and low specificity	(a) Bilateral hand involvement	1
	(b) Little finger surgery	1
Low sensitivity and high specificity	(c) Early onset of disease	1
	(d) Planter fibrosis*	2
	(e) Knuckle pads*	2
	(f) Radial side involvement*	2

\* = demonstrated odds ratio values more than 2.0 for at the low 95% confidence limit.

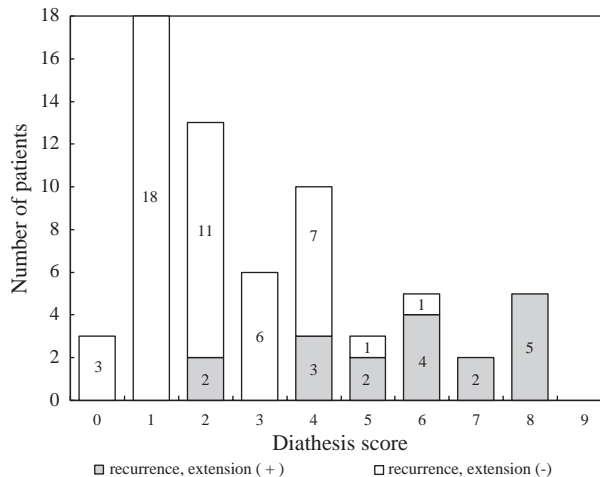


Fig 1 Clinical distribution of Dupuytren's diathesis score in 65 patients.

greater than 4, there was a high risk of recurrence and extension in our series, whereas there was little risk of recurrence and extension when  $D$  was less than 4.

In our series only two patients had a positive family history and this was not significantly related to recurrence and extension. A positive family history was also not a significant factor in the Hueston's, McFarlane and Botz's series, and these results are in line with those of Foucher et al. (1992), Moermans (1991), and Vigroux and Valentin (1992). Tubiana (1982) reported that patients with severe radial involvement which had occurred early in the disease process have most of the criteria of Hueston's "Dupuytren's diathesis". Our previous report (Abe et al., 2004a, 2004b) suggested that radial side disease, whether severe or not, is associated with the Dupuytren's diathesis and is a risk factor of recurrence. It is therefore reasonable to include radial side disease as an element of the diathesis score and exclude positive family history.

Rombouts (1989) reported a histologic classification for the prediction of recurrence. However, it is impossible to predict recurrence prior to surgery with this method.

A prospective cohort study is necessary to verify our diathesis score's ability to predict recurrence and extension, but it appears to allow the surgeon to evaluate the risk of recurrence and extension objectively before surgery.

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