

The association between frozen shoulder and Dupuytren's disease

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Fifty-eight patients with the diagnosis of primary frozen shoulder were independently examined by 3 surgeons for evidence of Dupuytren's disease. The disease was found in 52% (30/58) of the patients reviewed. These figures were compared with previously reported figures for a population of similar age. This showed that Dupuytren's disease is 8.27 (95% CI, 6.25-11.2) times more common in patients with frozen shoulder than in the general population; the difference between the two was highly statistically significant ($P < .001$, χ^2 test). We discuss the literature on the association between frozen shoulder and Dupuytren's disease and the implications of such a high proportion of patients sharing these two conditions. (J Shoulder Elbow Surg 2001;10:149-51.)

Frozen shoulder is a chronic fibrosing condition of the shoulder joint capsule.^{7,17,21} The predominant cells involved are fibroblasts and myofibroblasts.^{2,10} These cells lay down a dense type-III collagen matrix within the shoulder joint capsule, which subsequently contracts, leading to the typical clinical features of pain and stiffness. The histology is very similar to that of Dupuytren's disease of the palm (Figure), a condition with which it has many other features in common. The goals of this study were to quantify the incidence of Dupuytren's disease in a cohort of patients with a diagnosis of frozen shoulder and to discuss the implications of such an association.

METHODS

Of 2548 new patients with shoulder problems referred to the senior author, 163 had a diagnosis of primary frozen shoulder. Strict criteria were observed to come to such a diagnosis. Frozen shoulder was defined according to Codman's⁶ criteria (Table I); although, because we do not accept that patients with frozen shoulder show wasting of the spinati, we accepted a diagnosis based upon the remaining criteria. All our patients fulfilled these criteria and also had reduction of both active and passive movement.²² Limitation of passive external rotation was an important clin-

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1058-2746/2001/\$35.00 + 0 32/1/112883

doi:10.1067/mse.2001.112883

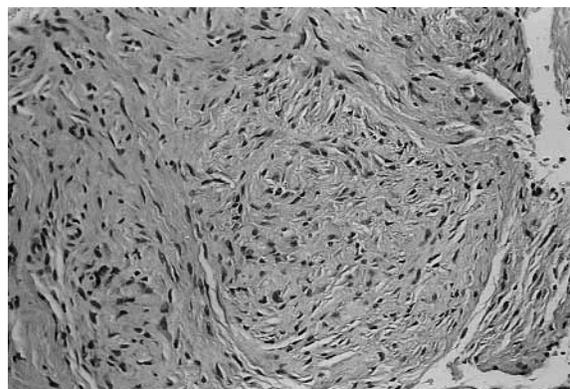


Figure Biopsy of coracohumeral ligament from a patient with frozen shoulder, showing features similar to those of Dupuytren's disease: highly cellular fibroblastic tissue arranged in lamellae and whorls (hematoxylin-eosin, original magnification $\times 300$).

ical feature present in all our patients. Great care was taken to exclude other diagnoses, including secondary frozen shoulder, on both clinical and radiologic grounds. The study was carried out in a district general hospital.

One hundred patients with primary frozen shoulder, identified from our shoulder database, were invited to a review clinic that was specially set up. Fifty-eight patients were reviewed (mean age 54.9 years; 28 men, 30 women).

Each patient was reviewed independently by 3 surgeons: 2 orthopaedic surgeons with a special interest in upper-limb surgery and 1 hand surgeon. Two of the surgeons had no previous contact with the patients. Each patient was examined for evidence of Dupuytren's disease of the hand, Garrod's knuckle pads, and Ledderhose's disease (plantar fibromatosis of the feet). In the patients with Dupuytren's disease, the type (flexion deformity, pits, nodules, or bands) and distribution were recorded on hand charts. The patients were also asked about any history of epilepsy, thyroid disease, and diabetes mellitus, and any family history of frozen shoulder or Dupuytren's disease.

RESULTS

Interobserver variation

The κ test was performed on the data to assess the interobserver variability to elicit the signs of Dupuytren's disease (Table II). The results were similar to previously published figures for the interobserver variation in the diagnosis of Dupuytren's disease¹³ and confirmed good interobserver correlation.

Table I Codman's⁶ criteria for frozen shoulder

Symptoms	Condition comes on slowly Pain is felt near the insertion of deltoid Inability to sleep on the affected side Able to continue daily habits and routines
Signs	Painful, restricted elevation Painful restricted external rotation Restriction of both the active and passive type Atrophy of the spinati Little local tenderness
Investigations	Normal results on radiography

Incidence of Dupuytren's disease

It was considered very likely that a patient had Dupuytren's disease when all 3 examiners found evidence of it. Similarly, when 2 examiners found Dupuytren's disease, with a close correlation in the distribution on the hand charts, it was concluded that the patient probably had Dupuytren's disease.

Of the patients reviewed, 47% (27/58) were found to have evidence of Dupuytren's disease by all 3 examiners, and in 5% (3/58), 2 examiners found evidence of Dupuytren's disease, with a close correlation in the distribution on the hand charts. Therefore, the overall incidence of Dupuytren's disease in our frozen shoulder patient group was 52% (30/58). This information is represented in Table III.

We compared our results with those of Early,⁸ who examined 2524 persons between the ages of 45 and 64 years and found an incidence of Dupuytren's disease of 6.2% (156/2524). The difference between the two groups was highly statistically significant ($P < .001$, χ^2 test), with a relative risk ratio of 8.27 (95% CI, 6.25-11.2). Even when only the patients in whom all 3 examiners found Dupuytren's disease were included (27/58), the relative risk ratio was 7.53 (95% CI, 5.50-10.32).

It should be noted that, of the 30 patients found to have evidence of Dupuytren's disease, only 3 had moderate or severe disease causing fixed joint contractures of the metacarpophalangeal or interphalangeal joints. The remaining patients had mild disease that affected the palmar fascia but did not cause joint contracture.

Other associations

Garrod's pads were found in 16 patients (28%); whereas evidence of Ledderhose's disease was only found in 1 patient (2%). Of the patients reviewed, there was no association with epilepsy or epilepsy treatment (0/58). Twelve percent (7/58) of patients gave a history of thyroid problems (5 hypothyroid, 1 hyperthyroid, 1 thyroid cyst). Thirty-one percent (18/58) had diabetes mellitus, of whom 15 patients were insulin dependent; the relative incidence of Dupuytren's disease in those patients with and without diabetes mellitus is represented in Table IV. As would be expected, the incidence of

Table II Kappa values for interobserver error

	Kappa value
Examiners 1 and 2	0.724
Examiners 1 and 3	0.896
Examiners 2 and 3	0.828

Dupuytren's disease in those with diabetes mellitus is higher than in those without. Twelve percent (7/58) of patients stated that they had a first-degree relative with a diagnosis of frozen shoulder, and 7% (4/58) had a first-degree relative with Dupuytren's disease.

DISCUSSION

Schaer²⁰ first noted the association between frozen shoulder and Dupuytren's disease in 1936, finding Dupuytren's disease in 25% of his patients with frozen shoulder (although he did not comment on patient numbers). In 1941, Lund¹⁴ found 14 patients with frozen shoulder in a review of 360 epileptic inpatients, and 12 of whom had Dupuytren's disease. Meulengracht¹⁶ reviewed 78 patients with "frozen shoulder," finding Dupuytren's disease in 18% (14/78). It should be noted, however, that his patient mix was somewhat heterogeneous and included a group of patients with pyrexia, weight loss, and a raised erythrocyte sedimentation rate.

We found evidence of Dupuytren's disease in 52% (30/58) of the patients with frozen shoulder that we reviewed. This is a considerably higher incidence than has been previously reported and, in comparison with an age-matched sample from the general population,⁸ is highly statistically significant ($P < .001$, χ^2 test).

Although the cause of frozen shoulder and Dupuytren's disease remains an enigma, the two conditions do share a number of striking similarities. Histologically, both show deposition of type-III collagen in nodules and bands, with a similar distribution of fibroblasts and myofibroblasts.^{2,12,15} Clinically, both may lead to joint contracture, and both may follow trauma. There are also metabolic associations, with a significantly increased incidence of diabetes mellitus and hyperlipidemia in both conditions³ as well as cytokine abnormalities, which have been shown to have a similar pattern in both diseases.^{1,4,19} Frozen shoulder is known to have abnormalities of matrix metalloproteinase and metalloproteinase inhibitors,⁴ and, interestingly, a recent trial that examined the use of a matrix metalloproteinase inhibitor as a treatment for inoperable stomach carcinoma noted the development of bilateral frozen shoulder in 6 of the 12 treated patients and of Dupuytren's disease in 3.¹¹ Finally, chromosomal studies have shown evidence of trisomy 7 and 8 syndrome in cell cultures of tissue taken from both conditions.⁹

The two conditions are generally understood to have very different natural histories—Dupuytren's disease being a progressive condition, and frozen shoulder

Table III Number of patients with Dupuytren's disease

	Evidence of Dupuytren's disease found by:				Total
	All 3 examiners	Only 2 examiners	Only 1 examiner	No examiners	
Number of patients	27	3	5	23	58
Percentage of total patients	47	5	9	40	100

being a self-limiting disorder that fully resolves with time. However, the available literature shows that although most frozen shoulder patients have little long-term functional disability, most do have ongoing discomfort and a measurable limitation of movement (particularly external rotation).^{5,7,15,16,18,21}

This study confirms that Dupuytren's disease is commonly seen in patients with frozen shoulder and suggests that the two conditions may share a common biochemical pathway that leads to contracture.

REFERENCES

- Baird KS, Crossan JF, Ralston SH. Abnormal growth factor and cytokine expression in Dupuytren's contracture. *J Clin Pathol* 1993;46:425-8.
- Bunker TD, Anthony PP. The pathology of frozen shoulder. *J Bone Joint Surg Br* 1995;77:677-83.
- Bunker TD, Esler CNA. Frozen shoulder and lipids. *J Bone Joint Surg Br* 1995;77:677-83.
- Bunker TD, Reilly J, Baird K, Hamblen D. Cytokines and Growth factors in frozen shoulder. Presented at the Combined Meeting of the Orthopaedic Associations of the English Speaking World, Auckland, New Zealand; Feb 1-6, 1998.
- Charnley JC. Periarthritis of the shoulder. *Postgrad Med J* 1959;35:384-8.
- Codman EA. Tendinitis of the Short Rotators. In: Ruptures of the supraspinatus tendon and other lesions in or about the subacromial bursa. Boston: Thomas Todd and Co; 1934. p. 1-35.
- DePalma AF. Loss of scapulohumeral motion (frozen shoulder). *Annals of Surg* 1952;135:194-204.
- Early PF. Population studies in Dupuytren's contracture. *J Bone Joint Surg Br* 1962;44:602-13.
- Feldman A, Bunker TD, Delmege D. Clonal chromosomal abnormalities in frozen shoulder [personal communication].
- Hannafin JA, DiCarlo EF, Wickiewicz TL. Adhesive capsulitis: capsular fibroplasia of the shoulder joint. *J Shoulder Elbow Surg* 1993;2:36-8.
- Hutchinson JW, Tierny GM, Parsons SL, Davies TR. Dupuytren's

Table IV Incidence of Dupuytren's disease in patients with frozen shoulder with and without diabetes mellitus

	Incidence of Dupuytren's disease
Frozen shoulder with diabetes mellitus	72% (13/18)
Frozen shoulder without diabetes mellitus	43% (17/40)

- disease and frozen shoulder induced by treatment with a matrix metalloproteinase inhibitor. *J Bone Joint Surg Br* 1998;80:907-8.
- Kay NRM, Slater DN. Fibromatosis and Dupuytren's disease. *Lancet* 1981;2{8241}:303.
- Lennox IAC, Murali SR, Porter R. A study of the repeatability of the diagnosis of Dupuytren's contracture and its prevalence in the Grampian region. *J Hand Surg [Br]* 1993;18:258-61.
- Lund M. Dupuytren's contracture and epilepsy. *Acta Psychiatr Neurol* 1941;16:465-92.
- Lundberg BJ. The frozen shoulder. *Acta Orthop Scand* 1969;119:1-59.
- Meulengracht E, Schwartz M. The course and prognosis of periarthritis humeroscapularis with special regard to cases with general symptoms. *Acta Med Scand* 1952;143:350-60.
- Neviaser JS. Adhesive capsulitis of the shoulder. *J Bone Joint Surg Br* 1945;27:211-21.
- Reeves B. The natural history of frozen shoulder. *Scand J Rheumatol* 1975;4:193-6.
- Rodeo SA, Hannafin JA, Tom J, Warren RF, Wickiewicz TL. Immunolocalization of cytokines and their receptors in adhesive capsulitis of the shoulder. *J Shoulder Elbow Surg* 1997;15:427-36.
- Schaer H. Die atologie der periarthritis humeroscapularis. *Ergebn Chir Orthop* 1936;29:11.
- Simonds FA. Shoulder pain: with particular reference to the frozen shoulder. *J Bone Joint Surg Br* 1949;31:426-32.
- Zuckerman JD, Cuomo F, Rokito S. Definition and classification of frozen shoulder: a consensus approach. *J Shoulder Elbow Surg* 1994;3(1):S5.