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Frozen shoulder: unravelling the enigma

T D Bunker MCh(Orth) FRCS
Consultant Orthopaedic Surgeon
Princess Elizabeth Orthopaedic Hospital, Exeter

Frozen shoulder was one of the few conditions not studied by Hunter, for it was first described by Codman 141 years after Hunter's death in 1793. Hunter is justly remembered as the pioneer of scientific surgery. As a scientist he was puzzled by the conditions which he saw day by day and set out to unravel these enigmas by experiment and scientific enquiry. Now, 200 years later, there are very few diseases within the specialty of orthopaedic surgery which remain an enigma, but frozen shoulder is surely the most enigmatic of them all.

Codman (1934) coined the term frozen shoulder. He stated that patients with frozen shoulder shared certain common features. These features were: "a slow onset . . . of pain felt near the insertion of deltoid, inability to sleep on the affected side, painful and restricted elevation and external rotation, with a normal radiological appearance". The problem is that this definition is still too general for five of the six criteria are shared by impingement, the most common cause of shoulder pain. The difference lies in the loss of passive movement, and in particular external rotation.

Wiley (1991) arthroscoped 150 patients referred with a given diagnosis of frozen shoulder and 113 turned out to have some other pathology, which shows the scale of the problem! Lundberg (1969) suggested that frozen shoulder should be subdivided into two groups, both based on Codman's criteria. Primary frozen shoulders are those which, after an exhaustive search, have an unknown aetiology. Secondary frozen shoulders are those in which the condition is secondary to soft tissue injury, fracture, arthritis, hemiplegia or any other known cause. Our quest was to discover the elusive cause of primary frozen shoulder.

Frozen shoulder is also characterised by its course. Classically, it is said to have three phases, a painful phase, a stiffening phase and a phase of resolution. Codman stated that: "even the most protracted cases recover with or without treatment in about two years". Unfortunately, this has led to a somewhat nihilistic approach to the disease and this statement needs to be reviewed in the light of the following 60 years of investigation into frozen shoulder. Although Codman stated that the condition "clears up entirely", this is qualified in two ways; first, he states that "recovery is by degrees, it is pretty hard even for the patients to say when they are well"; second, what he meant by an entire recovery was that the joint was not left "deformed or otherwise permanently damaged". In other words, the condition does not lead on to arthritis. Contrast this with the statements of Simmonds: "complete recovery . . . is not my experience", and of DePalma: "It is erroneous to believe that in all instances restoration of function is attained".

The best paper on the natural history of frozen shoulder (Shaffer *et al.*, 1992) evaluated 62 patients both objectively and subjectively for an average follow-up of 7 years. They found that 50% still had either mild pain or stiffness of the shoulder, or both and that 60% still demonstrated some restriction of motion compared with study-generated control values. Marked restriction of movement, when present, was most common in external rotation. The authors concluded that complete resolution was not universal and led them to question whether this is a self-limiting condition, for half their patients remained symptomatic and more than half had a measurable loss of motion, although this caused little functional disability.

Over a 6-year period, more than 1600 patients were referred to my shoulder clinic in Exeter. Of the first 1324, 70 fitted the criteria of primary frozen shoulder, an incidence of 5%. Those patients with primary frozen shoulder who failed to improve with conservative treatment were advised to have manipulation. We elected to perform a shoulder arthroscopy on all patients undergoing manipulation under anaesthetic, unless there was a specific contraindication, or lack of theatre time, in order to exclude any other pathology, and to determine the arthroscopic appearance of frozen shoulder. In all, 35 patients underwent shoulder arthroscopy before manipulation and 13 were re-arthroscoped after manipulation. The major finding was a consistent abnormality arising out of the subscapularis bursa, at the base of the origin of the long head of the biceps. In all, 31 patients had an abnormal villous fronding of the synovium, which was clearly highly vascular, arising from the subscapularis bursa and spreading to a variable extent across the rotator interval area. The remaining four, whose disease was of longer duration, had dense scarring in this area. In some

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Correspondence to: Professor T D Bunker, Swallow Thatch, Kenn, Exeter EX6 7XJ

patients there was a nodular appearance of the synovium with finger-like projections, and in others a synovial fronding like seaweed. These changes are so consistent as to be pathognomonic for primary frozen shoulder. Thirteen patients were re-arthroscoped after manipulation. In 12 cases there was an avulsion of the capsule in the infraglenoid recess from the humeral neck. In one patient this tear extended into the inferior glenohumeral ligament.

Wiley (1991) had 150 patients referred with frozen shoulder, 37 of whom qualified for the diagnosis of primary frozen shoulder and were arthroscoped. The appearance was uniform; there was a patchy, vascular, matted area of granulation tissue around the origin of the long head of the biceps tendon and the opening into the subscapularis recess. The scar tissue appeared as a small red nodular area of granulation tissue filling the subscapularis recess. Uitvligt *et al.* (1993) reported on 21 patients with primary frozen shoulder. Entry to the joint was difficult. There was a synovitis between biceps and subscapularis (the subscapular recess) in 19 of the 21 patients. Duralde *et al.* (1993) performed an arthroscopy on 11 patients with primary frozen shoulder, 14 with primary frozen shoulder who were diabetics, and 20 patients with secondary frozen shoulder. They found a synovitis in 42 shoulders. Hannafin *et al.* (1994) arthroscoped 15 patients with primary frozen shoulder. They found a diffuse vascular synovitis in the 13 patients. Esch (1994) arthroscoped 50 patients with primary frozen shoulder and found a thickened scarred rotator interval. Segmuller *et al.* (1995) assessed 24 patients with primary frozen shoulder and invariably found a proliferative synovitis just beneath the biceps insertion, that is in the subscapular recess.

These studies show a consistent pattern of arthroscopic findings in frozen shoulder. The major pathognomonic finding is an obliteration of the subscapularis recess with scar tissue, covered by a highly vascular papillary infolding of the synovium. The axillary recess is tightened and the joint is of a reduced volume. There are no adhesions. Whatever pathology there is appears to be extrasynovial or within the capsule and concentrated around the subscapularis bursa; this area of the capsule is a somewhat complicated area called the rotator interval.

Having seen arthroscopically that the pathology of frozen shoulder seemed to come from the rotator interval area of the capsule I elected, in those patients who failed to manipulate, to perform a surgical release of this area. In so doing I would be able to see whether there was a contracture of this area and, if so, study the pathology of this contracture. Twenty consecutive patients with primary frozen shoulder, who failed to manipulate, were explored surgically through a short deltoid splitting incision. In all of these patients the rotator interval area was found to be highly abnormal. Instead of the normal palpable sulcus there was a vascular area of nodular thickening and fibrous tissue. This fibrous tissue was noted to become tight if the arm was placed in external rotation, forming a checkrein to further movement.

There are five historical papers in the literature which describe the operative appearance of frozen shoulder. Neviaser (1945) found thickening and contracture of the capsule, and that the thickened capsule peeled from the humeral head like adhesive plaster from skin. Simmonds (1949) elegantly stated that the rotator cuff "looked like a vascular leathery hood with no obvious demarcation between the tendons". The usual demarcation between the tendons is what we now term the rotator interval, but which at that time was unnamed; this matches our findings. De Palma (1952) stated: "The coracohumeral ligament in these cases revealed significant alterations. It is converted into a tough inelastic band of fibrous tissue spanning the interval between the coracoid process and the tuberosities of the humerus. It acts as a powerful checkrein . . . division of the coracohumeral ligament allows early restoration of scapulohumeral motion". This is in perfect accord with the present study, and also with the findings of Neer *et al.* (1991) and Ozaki *et al.* (1989).

Open surgical release gave us the opportunity to subject the excised tissue from 12 patients with primary frozen shoulder to histological and immunocytochemical examination. Specimens were stained with haematoxylin eosin and the empirical chemical stains van Gieson (for collagen III) and Martius-scarlet-blue. Sections were prepared for immunocytochemistry by the avidin biotin complex method using monoclonal antibodies against the following antigens: leukocyte common antigen (LCA, CD15) macrophage and synovial cell antigen (PGMI CD68), the common mesenchymal cell intermediate filament vimentin and α smooth muscle actin. For comparison, Dupuytren's tissue from the hand of six patients undergoing excision was similarly treated, sectioned and stained and the same immunocytochemical method and antibodies applied.

The tissue was made of nodules and laminae of dense collagen which was of the 'mature' type III. Nodules consisted of a collagen matrix among which there was a proliferation of fibroblasts arranged alongside layers or bundles of dense collagen. The cell population was moderate to high. A striking feature was increased vascularity (high or moderate) in seven cases. The histological appearance of tissues excised from Dupuytren's contractures of the hand were similar in all respects.

As regards the results of immunocytochemistry; vimentin was strongly expressed by the cells in the collagen matrix, which confirms that the cells are fibroblasts. Some of these fibroblasts also expressed α smooth muscle actin, thus displaying a differentiation or a change to a myofibroblastic phenotype. The myofibroblast, or contractile fibroblast, is the pathognomonic cell of contractile scar tissue such as is found in Dupuytren's and the other fibromatoses. Leukocytes and macrophages were scanty and were never seen in the nodules or laminae, only on the periphery and usually around small vessels. Synovium was present in seven cases and was either entirely normal or showed minimal papillary infoldings without hyperplasia. Tissues from Dupuytren's contractures produced the same range of results.

Neviaser's study of ten cases (1945) is still the most cited. He found a thickening and contracture of the

capsule with considerable or extensive fibrosis in six cases. Simmonds (1949) examined four cases which showed dense collagen fibres, an extraordinary degree of vascularity and the presence of fibroblasts. De Palma (1952) explored 32 cases surgically and studied some of these histologically. He noted degeneration of collagen fibres, pronounced round cell infiltration, increased vascularity, thickening of the synovial membrane and evidence of increased fibrosis. Lundberg (1969) studied the histology of the shoulder capsule in 14 patients with primary frozen shoulder. He noted that the capsule was compact or dense, with an increase in cells which were mainly fibroblasts. He stated that the findings were similar to that of Dupuytren's tissue, although he attributed this to a personal communication with Norden, his pathologist. Kay and Slater (1981), in a brief letter to the *Lancet*, stated that they had examined the shoulder capsule of one diabetic with frozen shoulder and that the histology was similar to Dupuytren's disease. They stated that the cells were purely fibroblasts and myofibroblasts. Ozaki *et al.* (1989) reported that the histological findings in some of his 17 cases showed fibrosis. Hannafin *et al.* (1994) showed a diffuse capsular fibroplasia, thickening and contracture.

So we have now come a long way in understanding the enigma of frozen shoulder. The pathology shows us that primary frozen shoulder is a fibrous contracture of the rotator interval area of the capsule. This consists of a dense collagen matrix. The tissue is highly cellular. The cells are fibroblasts and myofibroblasts. The pathology is similar to Dupuytren's. The contracture acts as a checkrein, causing a global restriction of movement.

The finding that frozen shoulder is a Dupuytren's-like disease does help in our understanding of the associations which occur with frozen shoulder. The relationship between diabetes and frozen shoulder is well documented. Diabetic patients have a 10% to 20% incidence of frozen shoulder, which rises to 36% in insulin-dependent diabetes. Of patients with bilateral frozen shoulder, 42% are diabetic.

Meulengracht and Schwarz (1952) found evidence of Dupuytren's contracture in 18% of their patients with frozen shoulder. Schaer (1936) found that 25% of his patients with frozen shoulder had Dupuytren's disease. The incidence was far higher in our series. Of 50 patients, 29 had a pit, nodule or band of Dupuytren's disease in their hands.

Frozen shoulder has been associated with cardiac disease. Many patients with cardiac disease have raised serum lipids. We tested 43 of our patients' fasting serum lipid levels and found a significant elevation of cholesterol and triglyceride in frozen shoulder. The mean serum cholesterol concentration in patients with frozen shoulder was 5.92 mol/l compared with 5.14 mmol/l in the control patients. The mean serum triglyceride concentration in the frozen shoulder patients was 2.24 mmol/l and in the controls 1.62 mol/l. Raised serum lipids are also found in patients with Dupuytren's contracture (Sanderson *et al.*, 1992) and in patients with diabetes (Havel, 1979).

If frozen shoulder is a similar disease to Dupuytren's contracture, then it would fall into the group of diseases which are termed the fibromatoses. The superficial fibromatoses are Dupuytren's contracture of the palm, Garrod's knuckle pads, plantar fibromatosis of Ledderhose, and Peyronie's penile fibromatosis. Clonal chromosomal abnormalities have been found in many of these conditions. In Dupuytren's contracture, clones of cells trisomic for chromosome 8 have been noted. Trisomy 8 has also been found in Peyronie's disease.

Ten consecutive patients with primary frozen shoulder underwent surgical release and cell cultures were derived from this tissue. Chromosomal abnormalities were found in six of the ten patients. Clonal abnormalities were detected in four of these patients. Two patients had trisomy 7 one patient had trisomy 8 and the final patient with clonal abnormalities had both trisomy 7 and trisomy 8. Non-clonal abnormalities were found in a further two patients, one of whom was 47,XY,+8 and the other had three different chromosomal abnormalities, 47,XY+7, 47,XY+8 and 47,XY+21. Clearly this is a small series and we are still collecting the controls. However, the fact that the same clonal chromosomal abnormality has been noted, in the same proportion of cases, to the studies on Dupuytren's disease, would seem to add weight to the hypothesis that frozen shoulder is caused by a pathological fibrosis and should be classified within the group of diseases termed the fibromatoses.

For the first time we are beginning to understand the cause of frozen shoulder and this gives our generation an immense advantage over our forebears. Treatment must be aimed at releasing this contracture by manipulation or surgical release. Manipulation has attained the status of the most predictable form of treatment. But what of the resistant 20% who are not benefited by manipulation? We elected to perform a surgical release in 20 consecutive patients with resistant frozen shoulder. Pain scores measured on a visual analogue score improved from 8.0 to 2.4. Function improved from a score of 6.7 to 18.9 out of 30. Flexion improved from a preoperative 89.2° to 125.5° and external rotation from 7.5° to 36.7°. We would concur with Ozaki *et al.* (1989) that surgical release is logical and simple and gives good results in patients who fail to improve with conservative treatment or manipulation, but urge caution in insulin-dependent diabetic patients.

Thus the enigma of frozen shoulder has been unravelled. We have shown that the cause of frozen shoulder appears to be a fibrous contracture of the rotator interval and coracohumeral ligament of the shoulder joint. Histologically, this tissue is composed of a dense collagen matrix, consisting mainly of mature type III collagen. The tissue is highly cellular, and the cells are fibroblasts and contractile myofibroblasts. These findings are similar to Dupuytren's contracture.

The two conditions share similar abnormalities of growth factor and cytokine expression, clonal chromosomal abnormalities and serum lipid elevation. Both conditions are common in diabetes mellitus and can occur after minor trauma.

Elucidating the pathology of frozen shoulder allows us

to comprehend the clinical features and natural history of the condition and to formulate a logical plan of treatment. Clinically, the contracted tissue acts as a checkrein to external rotation and causes a global loss of active and passive movement.

In the more contracted cases, if treatment is to be effective it must be aimed at stretching, breaking or surgically dividing the abnormal tissue and preventing it from recurring. Surgical release, and in particular arthroscopic surgical release, show promise in the management of this common, disabling, protracted and painful condition.

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