FROZEN SHOULDER — AN ALGONEURODYSTROPHIC PROCESS ?

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The frozen shoulder syndrome and the Sudeck syndrome are clinically in many aspects similar. Radioisotope bone scan shows an increased uptake in the affected areas in both diseases, while standard radiographs show a progressive demineralization. With measurement of bone-mineral density by quantitative digital radiography these local decalcification processes were diagnosed in an early stage of the frozen shoulder syndrome : of 12 patients with primary frozen shoulder 10 had a bone-mineral density decrease of more than 21% in the humeral head of the affected shoulder compared to the unaffected side. In the control groups (n = 32) the difference between affected and unaffected side (left and right humerus of the healthy probands) was in only one case each above 21%. There are several indications in the literature assuming the frozen shoulder to be an algoneurodystrophic process. Our observation supports this hypothesis, and may possibly lead to earlier diagnosis and improved therapeutic management.

Keywords : frozen shoulder ; algoneurodystrophy.
Mots-clés : épaule gelée ; algoneurodystrophie.

INTRODUCTION AND CLINICAL PICTURE

Historically the first term for a frozen shoulder-like condition was "périarthrite scapulo-humérale" described by Duplay in 1872 (4). More than 100 years later Nash (11) still called the frozen shoulder an "enigmatic rheumatological condition" which is difficult to define, difficult to treat and difficult to explain pathologically.

Primary (idiopathic) frozen shoulder is a condition of unknown etiology characterized by a spontaneous onset of shoulder pain accompanied by increasingly severe limitation of glenohumeral passive and active movements in all directions. Secondary frozen shoulder is an identical clinical syndrome, but an identifiable disorder such as a rheumatological or neurological disease is discernible.

There is a triphasic natural history of the frozen shoulder syndrome, the *painful phase* leading as the pain eases to the *adhesive/frozen phase*, when limitation of motion reaches its extreme. Then follows the *resolution phase* leaving significant numbers of patients with residual limitation of shoulder movement (12). A number of pathological mechanisms have been postulated for the frozen shoulder syndrome, including the hypothesis of an autoimmune mechanism as a response to degenerative changes in the rotator cuff tendons leading to a diffuse capsulitis (7), or irritation of the suprascapular nerve which contains a high proportion of sympathetic fibers supplying the shoulder joint (15) as basic pathological conditions. More than 20 theories have been put forward to explain the frozen shoulder etiology, but none have stood the test of time, leading Bunker (3) to propose the new term "HGAC" for the frozen shoulder syndrome ("Humeroglenoid Acromiocla-

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Table I. — Literature references assuming the frozen shoulder to be an algoneurodystrophic process

Diagnostic
Increased radioisotope uptake (14)
Osteopenia, erosions (9, 10)
Pathologic cutaneous microcirculation (Laser Doppler flowmeter (8))
Reduced skin temperature (thermography (13))
Abnormal temperature control after cold challenge (6)
Clinical
Dull night pain felt at rest (11)
Idiopathic condition, self-limiting disease (1)
Higher incidence in depressive (2) and neurotic (5) personalities
Bilateral frozen shoulder in hyperthyroidism (activation of sympathetic nervous system) (17)
Therapy
Stellate ganglion block (16)
Suprascapular nerve block (7)

vicular Syndrome" to the patient and "Haven't got a clue" to the doctor).

In the literature one can find quite a number of references assuming the frozen shoulder to be an algoneurodystrophic process (table I). Our study was undertaken to investigate the Bone Mineral Density (BMD) of the affected humerus in comparison with the unaffected side in an early stage of the disease in order to confirm the hypothesis that the frozen shoulder is an algoneurodystrophic process, where all tissues, including the bone, are involved and dystrophic.

METHODS

Measuring bone mineral content

During the past two decades, numerous procedures have been proposed for the measurement of bone mineral in different parts of the skeleton. We used the dual-energy x-ray absorptiometry (QDR-1000) / Hologic) which is more sensitive for osteoporotic processes, where standard radiographs show differences only if bone mass is decreased by more than 20 to 30%. The general components of this system are shown in fig. 1. To perform the bone-mineral density measurement, the operator has the patient lie comfortably on the table

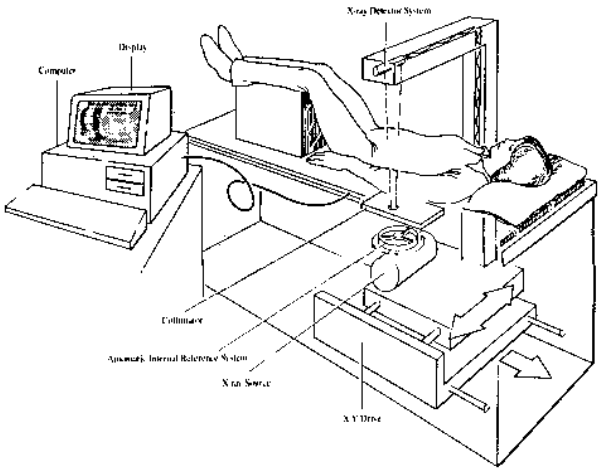

Fig. 1. — Components of the Hologic QDR-1000 x ray bone densitometer — measurement technique (see text).

and positions the scanning arm over the area of interest (fig. 2). The source and detector, which are controlled from the computer, scan across the area of interest in a serpentine fashion, and the information from the detector is stored and calculated by the computer unit. The detector is mechanically connected to the x-ray source so it always remains directly above the beam as the source scans. The results of the bone mineral content calculation from the displayed image are expressed as bone-mineral density (BMD) in grams/cm^2. The system uses two different energy levels to image and measure the bone-mineral content of the designated area of the body. The soft tissues that are contained within the area of interest are subtracted, and only the bones are imaged and measured.

Fig. 2. — Patient positioning for measurement of the BMD of the humerus on the densitometer table.

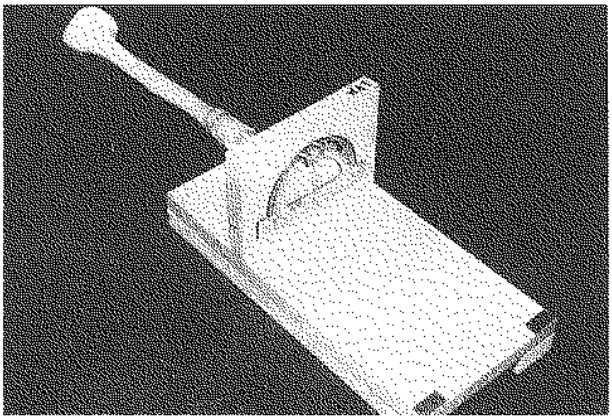

Fig. 3. — Excised human humeral bone fixed in the goniometer device for evaluation of the influence of rotation on the BMD result.

Standardization of the measurement method

Before using this technique on patients, we did *in vitro* measurements on excised human humeral bone (fig. 3) under various defined conditions to evaluate the influence of internal and external rotation of the humerus on the measured BMD. The influence of different surrounding media, namely air, water and oil bath, was tested, and the long-term precision of the method for determination of BMD was screened with the Hologic spine phantom. We performed measurements of BMD in 4 regions of interest (ROI) of the proximal humerus (ROI 1 : humeral head, ROI 2 : proximal metaphysis, ROI 3 + 4 : sections of humeral shaft — fig. 4). Absolute values for the BMD of the humerus are not available yet. Only comparison of the affected with the unaffected side (left and right sides of the healthy probands) concerning the same person was performed. These values or relative differences could then be used for interindividual comparisons.

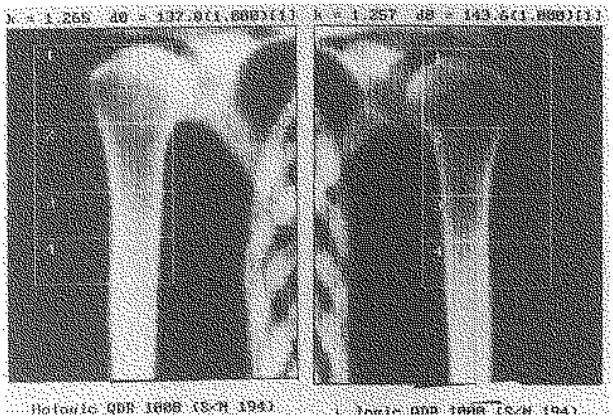

Fig. 4. — Regions of interest (ROI) of the proximal humerus for measurement of BMD (ROI 1 : humeral head, ROI 2 : proximal metaphysis, ROI 3 + 4 : sections of humeral shaft).

Clinical applications

Measurements were performed in three groups : 12 frozen shoulder patients, 12 patients with different shoulder disorders, and 20 healthy probands.

The 12 patients with idiopathic frozen shoulder (9 women, 3 men, average age 53 years) all showed an increased radioisotope uptake in the bone scan of the affected side (fig. 5) without involvement of the ipsilateral carpus. Pain in the affected shoulder lasted for an average of 9 weeks combined with a limitation of active and passive shoulder movements in all directions including at least a 50% reduction in external rotation. There were no identifiable internal disorders or precipitating events before the onset of complaints.

In the control group also immobilized of 12 patients with different shoulder disorders (degenerative changes of the rotator cuff, calcifying tendinitis and shoulder instability), the average age of 9 women and 3 men was 40 years, and symptoms lasted for an average of 8 weeks. In the group of 20 healthy probands there were 12 men and 8 women (average age 25 years).

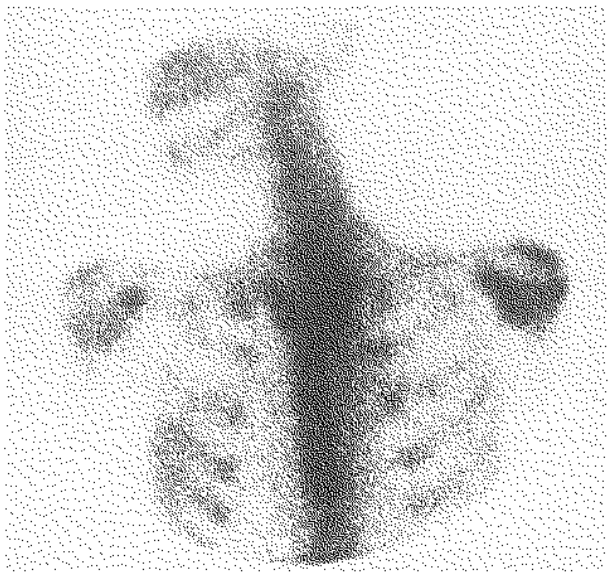

Fig. 5. — A 99-mTc diphosphonate bone scan of a frozen shoulder patient : increased radioisotope uptake on the affected side.

RESULTS

Standardization of the measurement method

Rotation of the humeral bone specimen in the goniometer device (fig. 3) resulted in a difference in the BMD value of 4.8% in 20° internal rotation and 2.8% in 20° external rotation compared to the neutral position. Measurements in an oil and water bath at 5, 10 and 15 cm depth showed a proportional decrease of the BMD value in correlation with the density and depth of the surrounding medium. The long-term reproducibility of the method for determination of BMD screened with the Hologic spine phantom showed a variation coefficient of 2.1% (standard deviation 2.1%).

Clinical applications

Of 12 frozen shoulder patients, 10 had a bone-mineral density decrease of more than 21% in the humeral head (ROI 1) of the affected shoulder compared to the unaffected side. In the control group also immobilized (n = 12) with degenerative changes of the rotator cuff, calcifying tendinitis and shoulder instability and in the group of healthy probands, the difference between the affected and unaffected sides (left and right humerus of the healthy probands) was above 21% in only one case each. Fig. 6 demonstrates the BMD quotients of the affected side to the unaffected side of the humeral head/ROI 1 (left/right side of the healthy probands); values below 1 indicate demineralisation of the affected side — the FS group having 10 out of 12 BMD values below 0.8. In the more distal regions of the humerus (ROI 2: proximal metaphysis, ROI 3 + 4: sections of humeral shaft), we found no significant differences of BMD values between left and right sides in all three groups.

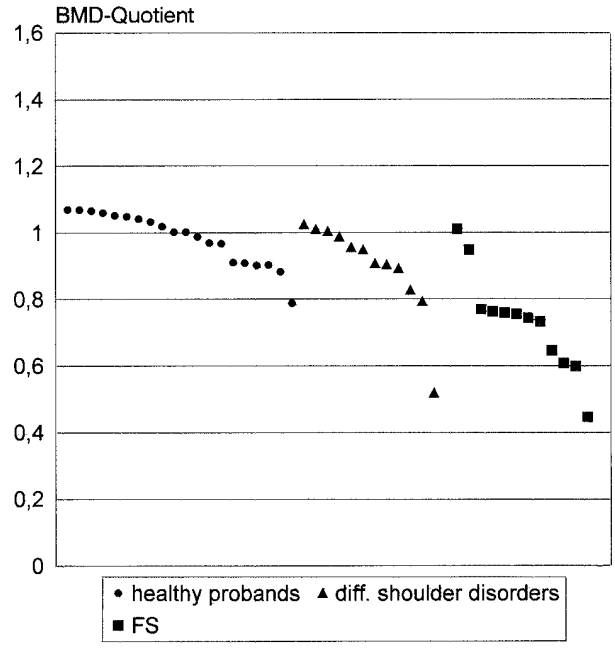

Fig. 6. — BMD quotients (left/right side of healthy probands, affected/unaffected side of patients) of the healthy probands (n = 20, round dots), patients with different shoulder disorders (n = 12, triangles) and frozen shoulder patients (n = 12, squares).

Case report

A 46-year-old secretary presented with painful active and passive limitation of shoulder movements in all directions on the nondominant side. The onset of symptoms 6 weeks before admission was spontaneous, the pain becoming worse at night and disturbing the sleep, the limitation of glenohumeral movement becoming gradually worse. There were no identifiable internal disorders or precipitating events before the onset of symptoms. A 99-mTc diphosphate bone scan showed an increased radioisotope uptake in the affected side without involvement of the ipsilateral carpus (fig. 5). We did 4 measurements of BMD in 1 year (fig. 7 and 8). On admission we found massive local osteoporosis of the affected humeral head (ROI 1), becoming even worse 3 months later, then gradually reaching the BMD of the unaffected side. Treatment included calcitonin (first 3 months), stellate ganglion blockade and physiotherapy.

The range of motion of the affected side on admission was 10-0-40° and 30-0-70° after one year of follow-up (external / internal rotations), and 70-0-30° on admission and 120-0-30° after one year of follow-up (abduction / adduction). Fig. 8 shows the BMD in the humeral shaft (ROI 4) of this patient, where no significant dif-

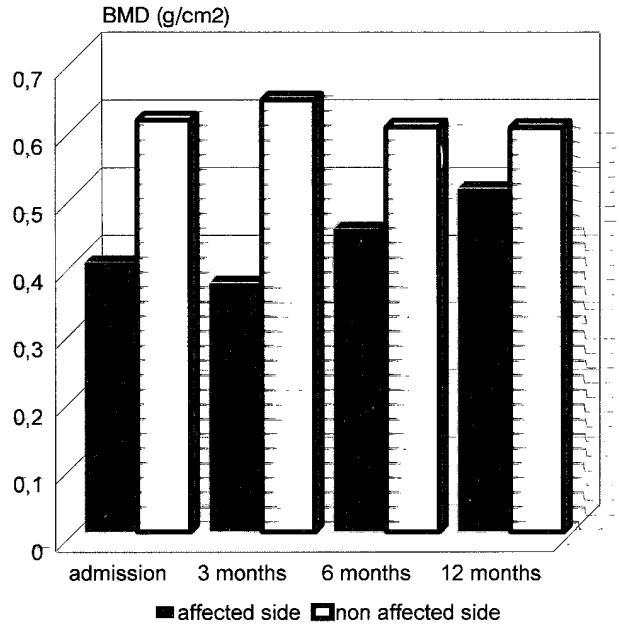

Fig. 7. — One-year follow-up of a frozen shoulder patient of the BMD (g/cm²) in the humeral head (region of interest 1). Black bars denote the affected side, open bars the unaffected side.

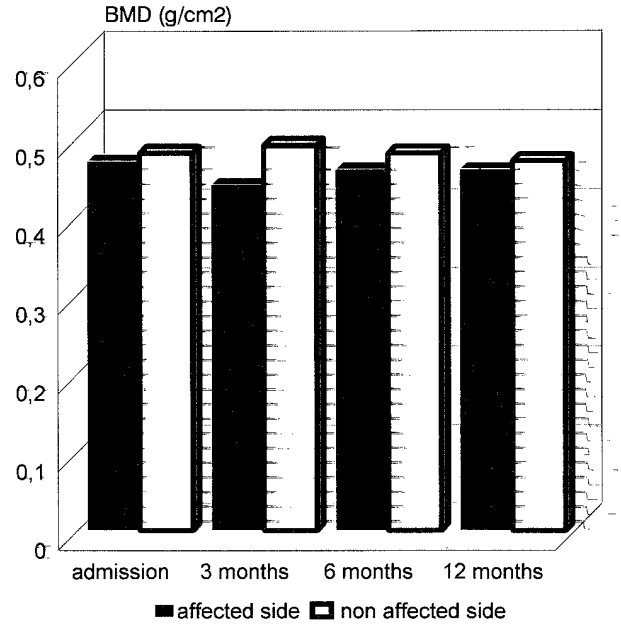

Fig. 8. — One-year follow-up of a frozen shoulder patient (see fig. 7) of the BMD (g/cm²) in the humeral shaft (region of interest 4). Black bars denote the affected side, open bars the unaffected side.

ferences between affected and unaffected sides were found.

DISCUSSION

The frozen shoulder syndrome is a condition of unknown cause characterized by severe pain and progressive limitation of movement of the shoulder. Many theories have been put forward to explain its etiology, but none have stood the test of time. Pathologic material is scanty, and there is no uniformity of opinion as to the exact nature of the lesion. The role of autonomic dysfunction in the etiopathology of frozen shoulder has been hypothesized by many authors, and there are several indications in the literature assuming the frozen shoulder to be an algoneurodystrophic process (table I).

Waldenburger *et al.* (14) found in 82% of idiopathic frozen shoulder patients an increased radioisotope uptake in the affected side without involvement of the ipsilateral carpus. Physical therapy associated with administration of subcutaneous salmon calcitonin had a statistically significant increased effect on pain compared with physiotherapy alone. The author concludes that the adhesive phase of the frozen shoulder syndrome corresponds to the retractile phase of algodystrophy, Steinbrocker stage I-II. Mani *et al.* (8) measured the reflex changes in the cutaneous microcirculation in the C-5 dermatome on the lateral aspect of the shoulder overlying the deltoid muscle by laser Doppler flowmeter. Sixteen out of 22 frozen shoulder patients had absent laser Doppler flowmeter responses to inspiration / expiration (clinical test of autonomic dysfunction including the Valsalva response), leading the author to the conclusion that sympathetic fibers in the ventral cervical nerve root might alter the vascular supply to the shoulder and result in atrophic changes in the perivascular tissues. By thermography, which is an adjunct to clinical diagnosis in algodystrophy, Vecchio *et al.* (13) screened 28 frozen shoulder patients, finding differences in skin contribution in 82% of the subjects, nearly three-quarters of whom had reduced skin temperature.

In view of the possibility that sympathetically mediated pain could be responsible for frozen shoulder symptoms, Jeracitano *et al.* (6) compared vasomotor control of frozen shoulder patients with normal subjects using computer-assisted thermography before and after a cold challenge. During the rewarming phase the average skin temperature in the frozen shoulder patients was 1.3°C lower than in normal controls. The author interprets this result as significant abnormalities in the mechanisms controlling shoulder-skin temperature originating from a sympathetic dysfunction in the dermatome serving the pain-affected area.

As in Sudeck disease, frozen shoulder patients often suffer from dull night time pain (11), and there are idiopathic and posttraumatic causes with the disease being self-limiting (1). Fleming *et al.* (5) found increased neuroses with a higher incidence of free-floating and somatic anxiety and depressive personalities in frozen shoulder patients compared to patients in general practice. Wohlgetan (17) postulated that the close resemblance of hyperthyroidism to activation of the sympathetic nervous system may underlie its association with frozen shoulder ; he presented a case of bilateral frozen shoulder in a patient with unrecognized primary hyperthyroidism. Wassef (15) obtained highly significant improvements by treating frozen shoulder patients with a suprascapular nerve block. The choice of blockade of the suprascapular nerve as a new concept for the management of frozen shoulder being interpreted as reflex sympathetic dystrophy was based on the fact that the nerve contains a high proportion of sympathetic fibers supplying the shoulder joint.

Our study was undertaken to investigate the bone-mineral density of the affected humerus in comparison with the unaffected side in an early stage of the disease in order to confirm the hypothesis that the frozen shoulder is an algoneurodystrophic process where all tissues, including the bone, are involved and dystrophic. Of 12 patients with primary frozen shoulder, 10 had a bone-mineral density decrease greater than 21% in the humeral head of the affected shoulder compared to the unaffected side. In the control group also immobilized with degenerative changes of the rotator cuff, calcifying tendinitis and shoulder instability, and in the group of healthy probands, the difference between affected and unaffected sides (left and right humerus of the healthy probands) was above 21% in only one case each. This was a static study, and no measurements were made to assess the effects of therapy, though the results of this limited study may lead to a potentially worthwhile therapeutic approach for the frozen shoulder syndrome by interpretating the disease as an algodystrophic process. Therapeutic management by mobilization under anesthesia and intensive physiotherapy was improved by calcitonin medication and stellate ganglion blockade. This procedure still has to be evaluated by a prospective randomized study. The development of effective therapy will require studies of well-selected patient groups at similar stages of the frozen shoulder syndrome.

Given that our understanding has advanced little since the first description of the self-limiting disease of frozen shoulder (4, 11), at present we do little more than practice the art of medicine which, as Voltaire would suggest, "is to keep the patient occupied while the disease runs its inevitable course" (11).

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RÉSUMÉ

L. P. MÜLLER, M. RITTMEISTER, J. JOHN, J. HAPP, F. KERSCH BAUMER. L'épaule gelée : un processus algoneurodystrophique ?

Le syndrome de l'épaule gelée et le syndrome de Sudeck présentent de nombreuses similitudes cliniques. La scintigraphie osseuse démontre une augmentation de la captation au niveau des régions atteintes dans les deux affections, et la radiographie conventionnelle est caractérisée par une déminéralisation progressive. Les auteurs ont mesuré la densité minérale osseuse par radiographie digitalisée quantitative lors de la phase précoce du syndrome d'épaule gelée : parmi les 12 patients atteints du syndrome, 10 présentaient une diminution de la densité minérale de plus de 21% au niveau de la tête humérale de l'épaule atteinte, par comparaison au côté sain controlatéral. Dans les groupes témoins comprenant 32 patients, la différence entre côté pathologique et côté sain ne dépassait 21% que chez un patient dans chaque groupe. La littérature suggère que le syndrome de l'épaule gelée correspond à un processus algoneurodystrophique. Notre étude va dans ce sens, et pourrait éventuellement permettre un diagnostic plus précoce et un traitement amélioré de l'affection.