

DIFFUSE PIGMENTED VILLONODULAR SYNOVITIS OF THE SHOULDER A CASE REPORT & REVIEW OF LITERATURE

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The combination of a diffuse pigmented villonodular synovitis and a rotator cuff tear of the shoulder in a 64-year-old man is described. The patient was treated by complete synovectomy, open repair of the rotator cuff tear and a Neer acromioplasty. Six months after this surgical treatment the patient was free of pain, and clinical examination revealed an almost normal range of motion.

Keywords : diffuse pigmented villonodular synovitis ; shoulder.

Mots-clés : synovite villonodulaire pigmentée ; épaule.

INTRODUCTION

Pigmented villonodular synovitis (PVS) is a relatively rare condition first described in 1941 by Jaffe (2).

Histologic lesions consist of a fibrous stroma, pigment deposition, histiocytic infiltrate and giant cells, with lesions occurring in a diffuse or circumscribed form.

The condition affects mainly young adults with a peak incidence in the third and fourth decade. In order of decreasing frequency the disease is encountered in the knee, hip, ankle and shoulder joint. The symptoms are mild pain, swelling and limitation of motion. Radiographic examination is normal in cases of circumscribed villonodular synovitis. In diffuse pigmented villonodular synovitis, however, radiographic evidence of osteoarthritis is almost always present.

Typical characteristics of an involved shoulder joint are risk of avascular necrosis, and evidence of osteoarthritis with lytic erosions in the humeral head and glenoid.

A definite diagnosis is made at surgery and confirmed histologically. Most authors advocate synovectomy in moderately affected shoulders. In severe destruction treatment consists of synovectomy and a Neer total joint replacement.

CASE REPORT

A 64-year-old man presented with a painful left shoulder, present for more than 1 year.

There was a spontaneous onset without history of acute trauma. During the preceding month the clinical picture was complicated by acute painful swelling of the joint. Symptoms improved after aspiration, which revealed a brown-stained fluid. On the third flare-up, the patient was referred to the orthopedic department.

Clinical examination showed active abduction of 110° (passive 160°) and a weakened, passive abduction score between 70 and 100°, external rotation of 40° (55°), and internal rotation of 40° (50°), retropulsion was 50° (75) and propulsion

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Fig. 1a

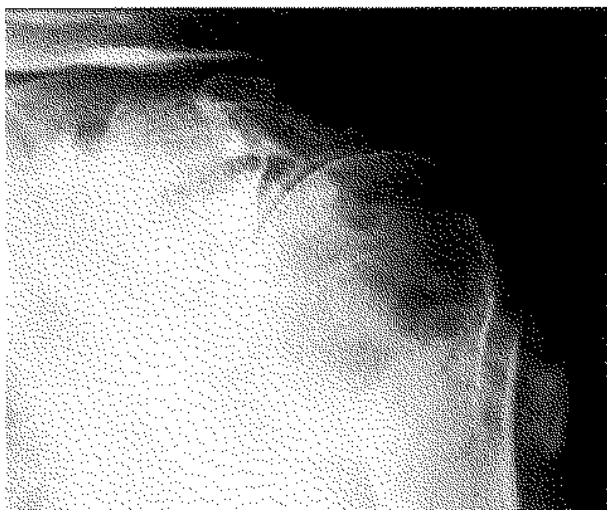


Fig. 1b

Fig. 1a and b. — Standard roentgenogram with no signs of bony involvement, subacromial and coracoclavicular calcifications, and acromioclavicular osteoarthritis.

115° (160°). Mild cervical osteoarthritis with diminished lateral flexion of the neck was noted. Blood tests, including a sedimentation rate and coagulation panel, were normal. Standard roentgenography and CT-arthrography showed a large rotator cuff tear, subacromial calcifications and acromioclavicular osteoarthritis (figs. 1 and 2).

As this active man had already suffered for a long period of time, it was decided to do an open Neer acromioplasty with complete synovectomy.

Histopathological examination confirmed the pigmented villonodular synovitis (figs. 3 and 4).

The patient had recovered well 4 months after the operation, with a good range of motion of the shoulder showing 130° of abduction, 60° of external rotation and 50° of internal rotation. Previous complaints of pain and swelling had resolved.

DISCUSSION

Diffuse PVS of the shoulder is a very rare condition. Ten cases were described in detail in the literature (1, 3, 4, 5, 6). The simultaneous occurrence of PVS with a rotator cuff tear has, to our knowledge, not yet been described. In this case we believe that the rotator cuff tear was caused by a classic impingement syndrome and not by invasion of this locally aggressive neoplasm. Nonetheless one might suspect such an etiology,

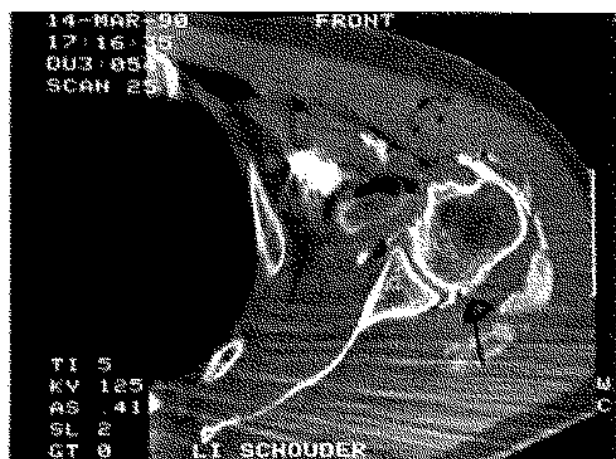


Fig. 2. — CT-scan with some possible bony involvement of the posterior side of the humeral head (arrow).

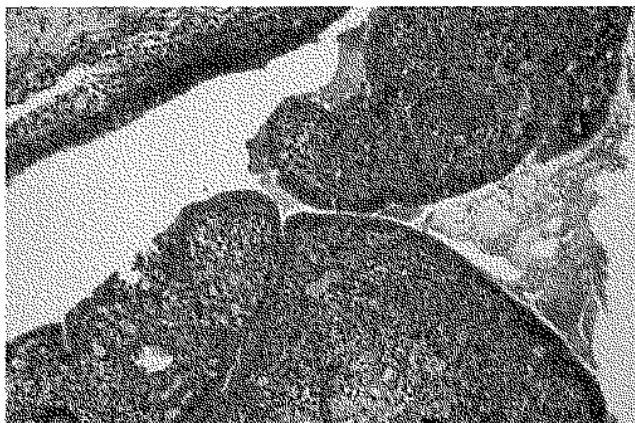


Fig. 3

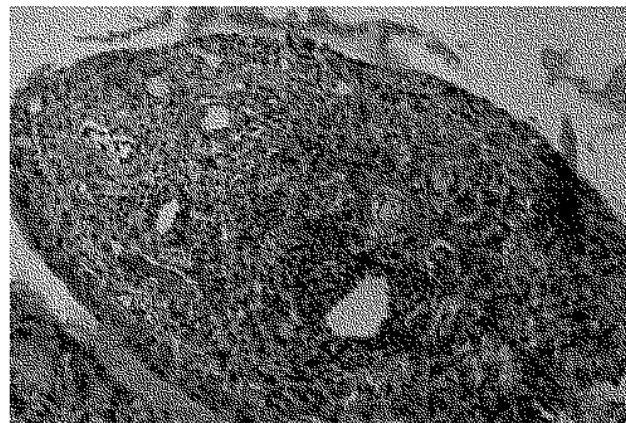


Fig. 4

Fig. 3 and 4. — Histologic lesions in pigmented villonodular synovitis.

f = fibrous stroma (nodule) ; p = pigment deposition (hemosiderine) ; g = giant cells and histiocytic infiltrate.

as local destruction of bone by villonodular synovitis has often been described (4, 5).

An important consideration in the treatment of PVS is that the disease is not malignant (2), and should therefore not be treated as such. Awareness of the condition is an important factor in early diagnosis, when there is almost no destruction, and treatment is still easy.

In most cases the correct diagnosis is only made after three or more years of symptoms, when there is significant osteoarthritis and cyst formation (1, 9). The majority of cases described in the literature have been treated with a Neer total joint replacement (1, 3, 6). Treatment of diffuse PVS of the hip and shoulder by synovectomy alone is generally not successful (3, 6, 7). In severe joint destruction, procedures should include extensive synovectomy and total joint replacement.

Recurrence after operation in PVS of the shoulder is very rare, in contrast with the knee (1, 3, 5, 7). Reasons for this fact are not clear, although it may result from the easier surgical procedure in the shoulder (1, 3).

CONCLUSIONS

1. PVS of the shoulder is a locally invasive disease. Most commonly of the diffuse type, it is

a synovial proliferative disorder. If untreated, it destroys the joint surface. Early awareness is important.

2. Surgical procedures that do not remove all the diseased synovium are likely to be followed by recurrence. Although no clear analysis can be made, it appears from the literature that recurrence of PVS in the shoulder is less frequent than in the knee, probably because surgical synovectomy is easier in the shoulder.

3. On the contrary, local and bony aggression seems to be more important in shoulder involvement with humeral and glenoidal destruction.

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SAMENVATTING

T. MULIER, J. VICTOR, J. VAN DEN BERGH en G. FABRY. Diffuus gepigmenteerde villonodulaire synovitis van de schouder.

De combinatie van een diffuus gepigmenteerde villonodulaire synovitis en een rotatorcuff-scheur van de schouder bij een 64-jarige man wordt beschreven. De patiënt werd behandeld door middel van een volledige synovectomie, open hechting van de rotatoren cuff scheur en een acromioplastie volgens Neer. Zes maand

na de chirurgische behandeling is de patiënt pijnvrij en het klinisch onderzoek toont een nagenoeg volledige mobiliteit.

RÉSUMÉ

T. MULIER, J. VICTOR, J. VAN DEN BERGH et G. FABRY. Synovite villonodulaire pigmentée de l'épaule.

Les auteurs rapportent l'association d'une synovite villonodulaire pigmentée de l'épaule et d'une rupture de la coiffe des rotateurs chez un homme de 64 ans. Le patient fut traité par synovectomie totale, et réparation à ciel ouvert de la coiffe des rotateurs associée à une acromioplastie selon Neer. Six mois après le traitement chirurgical l'épaule est indolore avec récupération d'une mobilité normale.