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Case report

Villonodular synovitis of the shoulder joint. A case report

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SUMMARY. *Introduction.* Pigmented villonodular synovitis is a proliferative disorder affecting the joints, tendon sheaths and bursas. The most affected joints are the knees, hips and fingers. According to literature reviews, the shoulders are less affected. *Material and methods.* In this paper, we are reporting a case found on the glenohumeral joint, in a 77 year old female patient. She was also suffering from rheumatoid arthritis. Her clinical chart was characterized by profuse joint effusion. She underwent shoulder arthroscopy by biopsies and the histopathological diagnosis of diffuse pigmented villonodular synovitis was confirmed. *Discussion.* Given the rareness of occurrence in the shoulder, it is difficult to define the clinical findings and diagnostic support. However, our case, like nearly all those reported so far, shares a slow clinical course, general and non specific pain in the shoulder, increased volume of the joint, and region asymmetry. In all cases, anatomopathological findings are those showing long villi, extending to the peripheral osteochondral joint into the joint with a brownish color, sometimes bloody, which microscopically describes a proliferative synovia, fibroblasts or primitive mesenchymatous cells which, eventually provide a particular diagnosis.

Key words: synovitis, villonodular, pigmented, shoulder.

RESUMEN. *Introducción.* La sinovitis villonodular pigmentada es un desorden proliferativo que afecta articulaciones, vainas tendinosas y bursas. Las articulaciones más afectadas son: rodilla, cadera y dedos. De acuerdo a revisiones en la literatura, el hombro es el menos afectado. *Material y métodos.* En el presente artículo hacemos el reporte de un caso localizado en la articulación glenohumeral, en una paciente de 77 años de edad, portadora además de artritis reumatoide. Con un cuadro clínico caracterizado por derrame articular profuso se le sometió a artroscopía del hombro, mediante biopsias y se corroboró el diagnóstico histopatológico de sinovitis villonodular pigmentada difusa. *Discusión.* Dada la rareza de la presentación en el hombro resulta difícil definir los hallazgos clínicos y de apoyo diagnóstico, sin embargo, nuestro caso al igual que la generalidad de los hasta ahora reportados comparten: evolución clínica lenta, presencia de dolor generalizado e inespecífico en el hombro, aumento de volumen, asimetría de la región; en todos los casos, son entonces los hallazgos anatomopatológicos que muestran vellosidades largas, extendidas de la periférica unión osteocondral, hacia la articulación, con una coloración parda, a veces sanguinolenta y cuya descripción microscópica muestra una sinovial proliferativa, fibroblastos o células mesenquimatosas primitivas, la que finalmente da el diagnóstico específico.

Palabras clave: sinovitis, villonodular, pigmentada, hombro.

Introduction

Pigmented villonodular synovitis is a proliferative disorder of the synovia affecting the joints, bursas, and tendon sheaths. Chassaignac was the first to describe a nodular lesion of the membrana synovialis affecting the tendon sheaths of the fingers. Simon described for the first time the localized shape and Moser, was the first to describe the diffuse shape in the knee.^{3,4} Down was the first to suspect its malignant origin. The lesion has been described with

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Figure 1. AP X-ray of the shoulder.



Figure 3. Arthroscopic view. Synovial Villi.



Figure 2. Arthroscopic view. Synovial Villi.

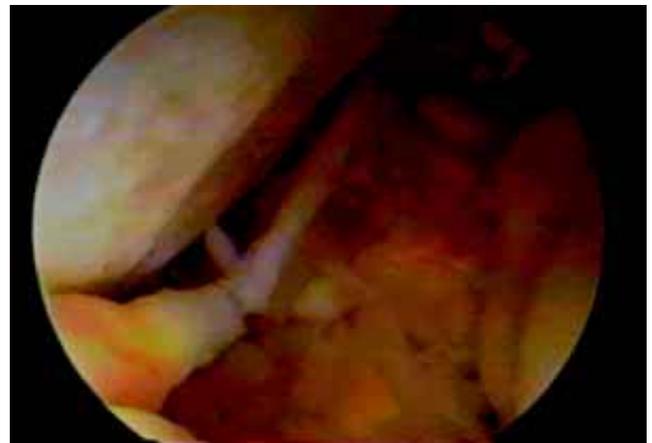


Figure 4. Arthroscopic view. Synovectomy in process.

several terms. Among these, synovial xanthoma, synovial fibroendothelioma, benign fibrous histiocytoma, xanthomatous giant cell tumor, giant cell tumor of the tendon sheath, fibrohemorrhagic sarcoma and others were included. In 1941, Jaffe proposed the terms pigmented villonodular synovitis, pigmented villonodular bursitis, and pigmented villonodular tenosynovitis. With these terms clinically occurring variants were covered. In 1945, Jaffe formally published his studies.⁶ Granowitz subclassified two clinical forms by adding first the prefix (L) for pediculated or localized lesions and (D) for diffuse lesions.^{3,4,8}

Case report

This is a 77 year old female patient having a 6 year history of adult rheumatoid arthritis, medically controlled with azathioprine, 50 mg every 24 hours, prednisone 10 mg per day, and etodolac, 300 mg every 12 hours. In December 1977, the patient was first seen at the orthopedics service when she noticed an increased volume of her left shoulder, secondary to joint effusion

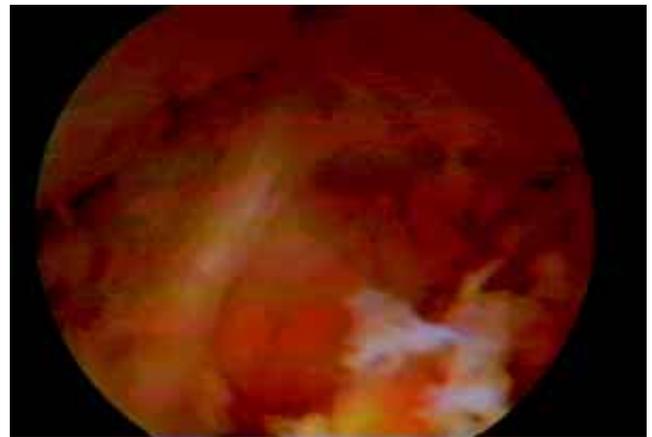


Figure 5. Arthroscopic view. Synovectomy in process.

(++++), and associated to pain on the anterior face of the shoulder, a feeling of distension and limitation of the arches of motion. Previously, the rheumatology service performed two arthrocenteses drawing 75 and 150 cc of

fluid respectively. The arthrocentesis cytochemical report showed dark yellow, lustrous and turbid fluid, total proteins 4.88 g/dl, 9,700 cells/LU leukocytes, 6,810 LU, erythrocytes, 2,150 LU, PMN 80%, MN 20%. The monthly report in September, 6 months later, showed turbid yellow fluid, total proteins 2.50 g/dl, 7,910 cells/LU, 2,940 leukocytes LU, 3,970 erythrocytes LU; PMN 90% and MN 10%. Clinically the patient did insidiously with functional limitation of her shoulder: abduction 80°, adduction 10°, flexion 70° and pain was exacerbated when trying to go beyond these ranges. In addition, a major increased volume with anatomical deformity accompanied these symptoms. The X-ray film showed an AP projection with osteopenia of the humeral head, thinned corticals, and the presence of subchondral bone cysts (*Figure 1*).

An ultrasound of the joint was taken to confirm the fluid collection. The patient underwent shoulder arthroscopy through a posterior portal from which a total 350 cc of yellow brownish fluid with lumpy detritus was removed. Arthroscopy also revealed a hypertrophic, abundant and diffuse synovia, involving the entire glenohumeral joint showing gross digitations or elongated nodes with a pink base and brown streaks, and distension of the anterior joint capsule (*Figures 2 and 3*).

Later, with the arthroscope the author conducted a broad synovectomy and cleansing of the joint (*Figure 4 and 5*). The material taken was sent to the pathologist who made the following report: hyperplastic synovial cells, subsynovial nodular proliferation with elongated architectures, isolated findings of hemosiderin. The final diagnosis was: pigmented villonodular synovitis.

After the arthroscopic surgery, the patient had pain remission with gradual recovery of her arches of motion of the joint and, of course, anatomy restoration.

Discussion

Pigmented villonodular synovitis is a proliferative disorder of the synovia. As we said before, it affects the joints, bursas and tendon sheaths. Since 1941, with papers by Jaffe,^{4,6} a variety of names for this disease were devised. With more recent papers, it was possible to identify a localized and a diffuse variant of the disease. The localized nodular form is an isolated or circumscribed lesion that may be pediculated and commonly affects the tendon sheaths of the hand. The diffuse form is characterized by a general involvement of the synovia and is often found in large joints.^{2,8,9} According to Myers,¹² the annual incidence of pigmented villonodular synovitis is 1.8 patients per one million people, affecting men and women equally. It is more common during the 3rd and 4th decades of life.^{4,15} Typically, the lesion involves only one joint with the knee being the most affected and accounting for 80% of cases. Other joints affected are the hips, ankles, large toes, and rarely the shoulder and temporomandibular joints.^{2,4,12}

In 1994 Tong reported a case of a shoulder joint. In reviewing the literature he found 14 cases reported so far.¹⁸ In 1999 Müller presented a case of a young patient with a shoulder lesion studied by MRI. The lesion appeared to simulate a malignant tumor. Upon review of the literature, he found 25 cases of shoulder villonodular synovitis.¹¹ In our review, in addition to the above papers, we have found one more case reported by Joseph Cheng in 1997, referring to a patient subject to two prior procedures for glenohumeral instability. The first procedure involved arthroscopy and the second, was open surgery using the anterior capsulolabral reconstruction technique. After these two approaches and three asymptomatic years, revision arthroscopy was performed and two fibronodular, vascularized lesions of the anterior glenoid were found. The histopathological analysis revealed localized villonodular synovitis.² Konrath reported yet another case, also in 1997, although this is an extra-articular lesion found in the subacromial bursa. Coincidentally, Saw Miller reported another localized case in the subacromial and deltoid bursa, that very same year.

With regards to the etiopathology of this disorder, there are major ongoing controversies at this time. Some authors propose synovial hyperplasia related to idiopathic inflammatory components as the cause for etiology. Other authors relate the lesion to benign neoplastic processes and a reaction to repetitive trauma. More recent histopathological analyses and DNA densitometry lead towards a neoplastic cause supported also by abnormal findings of aneuploidism and cytogenic findings. There are reports such as those by Bertoni with three documented cases of malignant transformation.^{1,2,4,5,7,13}

Because this disorder very rarely occurs in the shoulder, it is difficult to define the clinical findings and supporting diagnosis. However, our case, as most of those reported so far, shares the following information: slow clinical course, general and non specific shoulder pain, increased joint volume, and region asymmetry. In this case, given the profuse effusion there was also a venous mesh, shiny skin and, of course, a history of previous arthrocenteses. Impairment of the joint function will depend, at any rate, on the extension of the effusion and, naturally, the tolerance of the patient to pain. In cases where the subacromial bursa was swollen, motion was limited thus preventing sliding during abduction and elevation of the shoulder.^{4,5,12,15}

With regards to diagnostic support, imaging, even if sensitive, is very non specific especially when related to the shoulders. In a review of 11 villonodular cases of the knee joint reference is made, from the X-ray point of view, to increased density and irregularities of the cortical. Also, extra-articular imaging mentions calcium depositions pointing at a malignancy. Breimerg and Freibergger added erosion to Lewis' criteria, bone cysts, and sclerous bone lesions attributing them to changes in increased intra-articular pressure due to entrapment of soft tissues. These chang-

es are currently much more related to degenerative processes and even disuse of the joint. In a review paper of the Mayo Clinic, the presence of cysts and bone erosion are correlated in 33% of cases of diffuse synovitis and 25% in the nodular form.^{2,4,9,16,17} In our case reported, X-ray imaging was basically demineralization and thinned corticals from the humeral head to the glenoid in addition to suchondral bone cysts.

Even if cytology and cytochemical analysis of the synovial fluid, usually altered, may lead to a diagnosis, they have not been very specific in cases reported and responded more to an inflammatory chronic process, a pathognomonic correlation. Some authors mention the increased intra-articular fluid usually bloody or of xanthochromic aspect, mustard yellow colored, low glucose, high proteins, and a frequently low white cell count. Some of these data are also shared by our case.^{4,8,16}

Finally, the anatomopathological description of lesions show long villi extending into the osteochondral union towards the joint with a brownish coloring sometimes bloody showing under the microscope a proliferative synovia, fibroblasts or primitive mesenchymatous cells prone to collagen production and histiocytic type cells with a phagocytic function. Other studies discuss multinucleated giant cells corresponding to hemosiderin depositions. These findings have been proposed to support the notion that this is a proliferative neoplastic process of synovial fibroblasts and histiocytes. Flandry, however, in his series contradicts these findings.^{1,2,15} In our case the report was the presence of hyperplastic synovial cells, subsynovial nodular proliferation with elongated architecture, isolated hemosiderin findings which are shared with published reports.

Managing villonodular synovitis has also been the cause of controversy. Marginal resection in localized forms and broad synovectomy in diffuse presentation have been suggested. Most authors agree with open surgical resection of the nodular type representing a better option. Other authors have proposed to replace the joint, primary with radiotherapy and in cases of postoperative relapse. More recently, arthroscopic approach has been considered as the method of choice for the resection of the lesions. The latter is the choice carried out in our patient, with a broad resection of the hypertrophic synovial tissue.^{2,10,14}

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