Um caso de sinovite vilonodular do ombro em adolescente: diagnóstico por imagem e anatomopatológico

A case of villonodular synovitis of the shoulder in an adolescent: imaging and pathologic diagnosis

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RESUMO

Em quadros de monoartrite crônica devem ser investigadas doenças inflamatórias como a artrite reumatoide (AR), doenças infecciosas como a tuberculose e outras doenças que causem espessamento sinovial e derrame articular como sinovite vilonodular pigmentada (SVNP), hemangioma sinovial, osteocondromatose sinovial e lipoma arborescente sinovial. Relatamos o caso de uma jovem paciente com quadro de monoartrite em ombro, cujo exame por imagem mostrou sinovite e cujo exame histopatológico obtido através de artroscopia com biópsia revelou tratar-se de SVNP. **Relato do caso:** J C M, 15 anos, sexo feminino, branca, estudante, foi encaminhada ao reumatologista com hipótese diagnóstica de artrite idiopática juvenil (AIJ) pauciarticular. Apresentava, há um ano, dor no ombro D, que melhorava com o uso de anti-inflamatório não-esteroidal (AINE) em dois dias. Teve nesse período de cinco a seis destes episódios que duravam poucos dias. Negava outras queixas articulares ou sistêmicas. Trazia exames normais ou negativos: hemograma, VHS, proteína C reativa, fator reumatoide, sedimento de urina. O FAN era positivo 1/80, pontilhado fino. Trazia Ressonância Magnética do ombro indicativa de sinovite glenoumeral com derrame articular com conteúdo expansivo de tecidos moles na bursa subescapular, segundo o laudo, podendo corresponder a *pannus*. Como história e exame físico não eram compatíveis com AIJ, foi realizada outra RM que mostrou aumento da lesão já descrita. Foi então encaminhada à

ABSTRACT

Chronic monoarthritis demand an investigation of inflammatory diseases, such as rheumatoid arthritis (RA), infectious diseases like tuberculosis; and other diseases that cause synovitis and joint effusion, such as pigmented villonodular synovitis (PVNS), synovial hemangioma, synovial osteochondromatosis and arborescence lipoma. We report the case of a young patient with chronic right shoulder monoarthritis, who's magnetic resonance imaging (MRI) showed synovitis. Arthroscopy was performed and the biopsy revealed PVNS. ***Case report:*** *J C M, 15 years-old, female, Caucasian, student. She was sent to a Rheumatologist along with a diagnosis of juvenile idiopathic arthritis (JIA). The patient presented, for one year, a mild pain of insidious onset in her right shoulder, with relief of the symptoms in two days under nonsteroidal anti-inflammatory therapy (NSAIDs). During this year, the patient presented five or six episodes of pain with the same characteristics. No other signs and symptoms were related. The following tests showed normal or negative results:complete blood count, ESR, C-reactive protein, rheumatoid factor and urinalysis. The antinuclear antibody (ANA) was 1/80 speckled pattern. The MRI of the shoulder showed glenohumeral synovitis with joint effusion and soft tissue swelling in the subscapular bursa, which could correspond to pannus. As the medical history and physical examination were not compatible to JIA, a second MRI was performed, which showed an increase of the synovitis. The patient was submitted to an arthroscopy*

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artroscopia para biópsia, que revelou SVNP. Durante o procedimento, foi realizada sinovectomia, e uma nova RM feita após nove meses mostrou ausência de sinovite. A SVNP do ombro é incomum e a sinovectomia foi curativa nesse caso.

Palavras–chave: monoartrite, sinovite vilonodular pigmentada, ombro, sinovite.

with biopsy and the histopathological examination showed PVNS. A complete synovectomy was performed and a new MRI, nine months later, showed no synovitis. PVNS of the shoulder is uncommon, and synovectomy was curative in this case.

Keywords: *monoarthritis, pigmented villonodular synovitis, shoulder, synovitis.*

INTRODUÇÃO

O envolvimento monoarticular crônico pode ocorrer em doenças inflamatórias como a artrite reumatoide (AR) e doenças infecciosas como a tuberculose. Outras entidades podem comprometer uma única articulação, como as patologias de origem mecânica e aquelas que cursam com derrame articular e espessamento sinovial, como a sinovite vilonodular pigmentada (SVNP), o hemangioma sinovial, a osteocondromatose sinovial e o lipoma arborescente sinovial.[1-3]

Nesses casos, a história e o exame físico do paciente são fundamentais para nortear o diagnóstico. Nos casos de AR, o envolvimento do ombro como única articulação acometida é muito incomum.

Relatamos o caso de uma paciente de 15 anos de idade com quadro de monoartrite em ombro e FAN positivo cuja investigação diagnóstica por imagem e biópsia através de artroscopia revelou tratar-se de sinovite vilonodular. A sinovite vilonodular foi descrita pela primeira vez em 1852, por Chassaignac,[4] como uma lesão nodular com origem em bainhas tendinosas flexoras dos dedos, e em 1864, Simon[5] observou a ocorrência da forma localizada dessa doença no joelho, tendo, em 1909, sido Moser[6] o primeiro a relatar a sua forma difusa. Só em 1941, Jaffe *et al*.[7] propuseram o termo sinovite vilonodular pigmentada em estudo de 20 casos; atualmente, a nomenclatura proposta por Granowitz *et al*. é a mais usada.[8]

RELATO DO CASO

J.C.M., 15 anos, sexo feminino, branca, estudante, apresentava há um ano história de dor no ombro direito (D), ritmo inflamatório, que melhorava com o uso de anti-inflamatório não-esteroidal (AINE) e que tinha duração de dois a três dias. Teve, nesse período, de cinco a seis episódios de dores na mesma articulação, sempre melhorando com o uso de AINE. Negava outras queixas articulares ou sistêmicas. Referia lesões de pele, eritematosas, por todo o tronco e braços. Fora encaminhada ao dermatologista, que diagnosticou farmacodermia por AINE. Trazia exames normais ou negativos: hemograma, VHS, proteína C reativa, fator reumatoide, com exceção do FAN, positivo 1/80 pontilhado fino. Trazia ressonância magnética (RM) do ombro evidenciando sinovite glenoumeral com derrame articular com conteúdo expansivo de tecidos moles na bursa subescapular e no recesso axilar, segundo laudo radiológico, podendo corresponder a *pannus* e sugerindo a possibilidade de doença autoimune (artrite idiopática juvenil, AIJ) (Figuras 1A e 1B).

Com esta hipótese de AIJ foi encaminhada ao reumatologista. No entanto, o exame físico não revelava artrite em outras articulações e os movimentos do ombro D eram normais com um pouco de dor à movimentação. Como a história e o exame físico não eram compatíveis com AIJ e a paciente mantinha a mesma queixa, foi realizada outra RM, que mostrou piora da sinovite já descrita em ombro D (Figuras 2A e 2B).

Foi então encaminhada ao ortopedista para realização de artroscopia com biópsia sinovial, que revelou SVNP (Figuras 3A, 3B, 3C).

Durante o procedimento de artroscopia, a paciente foi submetida a sinovectomia, e o controle da RM feito após nove meses mostrava ausência de sinovite (Figura 4).

DISCUSSÃO

Essa jovem paciente apresentou leve dor em ombro com discreta limitação de movimento, e a primeira avaliação por imagem mostrou espessamento da sinóvia sugestivo de *pannus*, motivo pelo qual foi pesquisada a presença do fator reumatoide, que era negativo, e o FAN que era positivo 1/80, buscando confirmar o diagnóstico de AIJ. Não havia qualquer outra evidência clinica ou laboratorial de AIJ e a característica da dor não era de doença inflamatória. O FAN pode ser justificado pela frequência na população sem doença autoimune; nesses casos, o padrão mais encontrado é o pontilhado fino denso, como observado nessa paciente. Outro fato que chamava a atenção do clínico naquela ocasião era a presença de lesões eritematosas que, associadas ao FAN positivo, fez suspeitar de doença autoimune, mas a avaliação dermatológica afastou essa hipótese por tratar-se de farmacodermia.

ficados dentro da articulação. O lipoma sinovial é uma massa intra-articular isolada; tem intensidade de sinal semelhante à da gordura em todas as sequências, tal como o lipoma arborescente, mas pode distinguir-se pela sua forma redonda ou oval e pela ausência de proliferação sinovial. O lipoma arborescente é uma rara doença intra-articular, de etiologia desconhecida, que consiste em infiltração difusa da membrana sinovial por gordura. O conteúdo lipomatoso da lesão é bem caracterizado na RM ou na tomografia computadorizada. O sarcoma sinovial apresenta extensão extra-articular, a calcificação e a invasão óssea, aspectos estes não encontrados na SVNP.[36-39]

O tratamento consiste na sinovectomia, que tem muito bom resultado e pouca recorrência em se tratando da forma nodular. Já a forma difusa pode ter alta recorrência. Os pacientes não tratados adequadamente podem evoluir para destruição articular e a artroplastia pode ser a solução nesses casos.[41-45]

Essa paciente teve excelente resposta à sinovectomia, como demonstra a RM realizada nove meses após.

Villonodular synovitis of the shoulder in an adolescent: imaging and pathologic diagnosis

INTRODUCTION

Chronic monoarticular involvement can occur in inflammatory diseases such as rheumatoid arthritis (RA) and infectious diseases like tuberculosis. Other entities can compromise a single joint, like the mechanical originated disorders and those which presents with articular effusion and synovial thickening, like pigmented villonodular synovitis (PVNS), synovial hemangioma, synovial osteochondromatosis and synovial arborescence lipoma.[1-3]

In these cases, the history and the physical exam of the patient are fundamental to guide the diagnosis. In cases of RA, the involvement of the shoulder as the only involved joint is very uncommon.

We report the case of a 15 years old patient with a shoulder monoarthritis and a positive antinuclear antibody (ANA), whose diagnostic investigation by imaging and biopsy through an arthroscopy revealed to be villonodular synovitis. The PVNS was described for the first time in 1852, by Chassaignac,[4] as a nodular lesion with origin in flexor tendinous sheath of the fingers, and in 1864, Simon[5] observed the occurrence of the localized form of this disease in the knee, being Moser[6], in 1909, the first to relate its diffuse form. Only in 1941, Jaffe *et al*.[7] proposed the term pigmented villonodular synovitis, in a study of 20 cases; today the nomenclature proposed by Granowitz *et al*. is the most used.[8]

CASE REPORT

J.C.M., 15 years old, female, Caucasian, *student*. She presented, for a year, a history of pain in the right shoulder (R) with inflammatory caractheristics, which got better with the use of nonsteroidal anti-inflammatory (NSAID) and lasted for 2 to 3 days. She had in this period 5 to 6 episodes of pain in the same joint, always improving with the use of NSAID. Other complaints, articular or systemic, were denied. The patient referred erythematous skin lesions all over the trunk and arms. She was referred to the dermatologist that diagnosed pharmacodermia by NSAID. The following tests were normal or negative: complete blood count, ESR, C-reactive protein, rheumatoid factor. ANA were positive 1/80, fine speckled. The magnetic resonance imaging (MRI) of the shoulder showed glenoumeral synovitis with joint effusion and expansive contents of soft tissues in the subscapular bursa and in the axillar recess, according to radiological report, could correspond to pannus and suggesting the possibility of an autoimmune disease (juvenile idiopathic arthritis- JIA) (Figures 1A e 1B).

The patient was referred to the rheumatologist with a JIA hypothesis. However, the physical examination revealed no arthritis in other joints and the movements of the right shoulder were normal with discrete pain. As the history and the physical exam were not compatible with JIA, and since the patient kept the same complaint, another MRI was performed, showing an increase of the synovitis already described in the right shoulder. (Figures 2A and 2B).

The patient was then referred to the orthopedic surgeon to perform an arthroscopy with synovial biopsy, which revealed pigmented villonodular synovitis (Figures 3A, 3B, 3C).

During the arthroscopy procedure, the patient underwent a synovectomy, and the control MRI done after nine months showed absence of synovitis (Figure 4).

DISCUSSION

This young patient presented a mild pain in the shoulder with a discrete limitation of movement, and the first evaluation by imaging showed synovial thickening suggestive of pannus. In an attempt to confirm the JIA diagnosis, the patient was checked for the presence of rheumatoid factor, which was

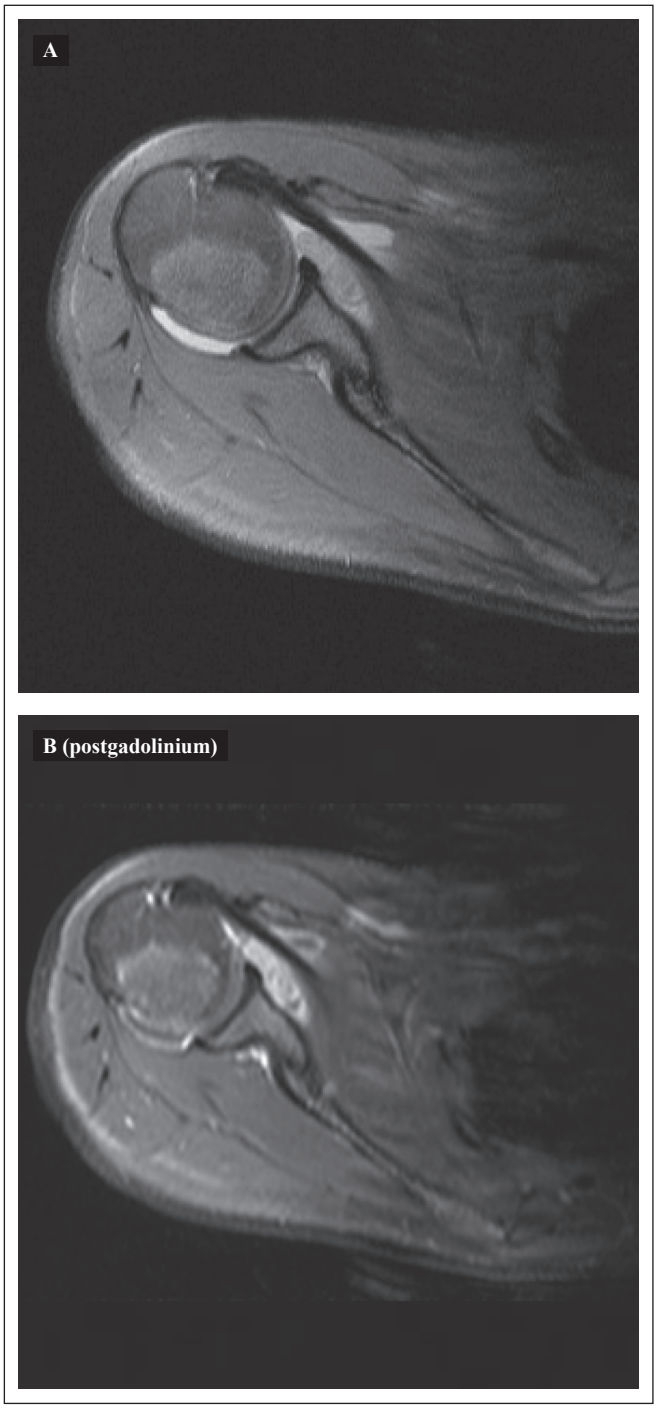


Figure 1. MRI of right shoulder demonstrate discrete diffuse synovial thickening, hypercaptant by contrast, with moderated effusion in the glenohumeral articular space. Presence of soft tissues mass, of oval aspect, compromising the subscapular bursa and the axillar recess regions. Images in T2-weighted sequences with fat saturation with low intensity signal, with small granules in between. Signal indicative of glenohumeral synovitis with joint effusion. Expansive content of soft tissues in the subscapular bursa and in the axillar recess, which could correspond to tissual hypertrophy of the synovia (pannus).

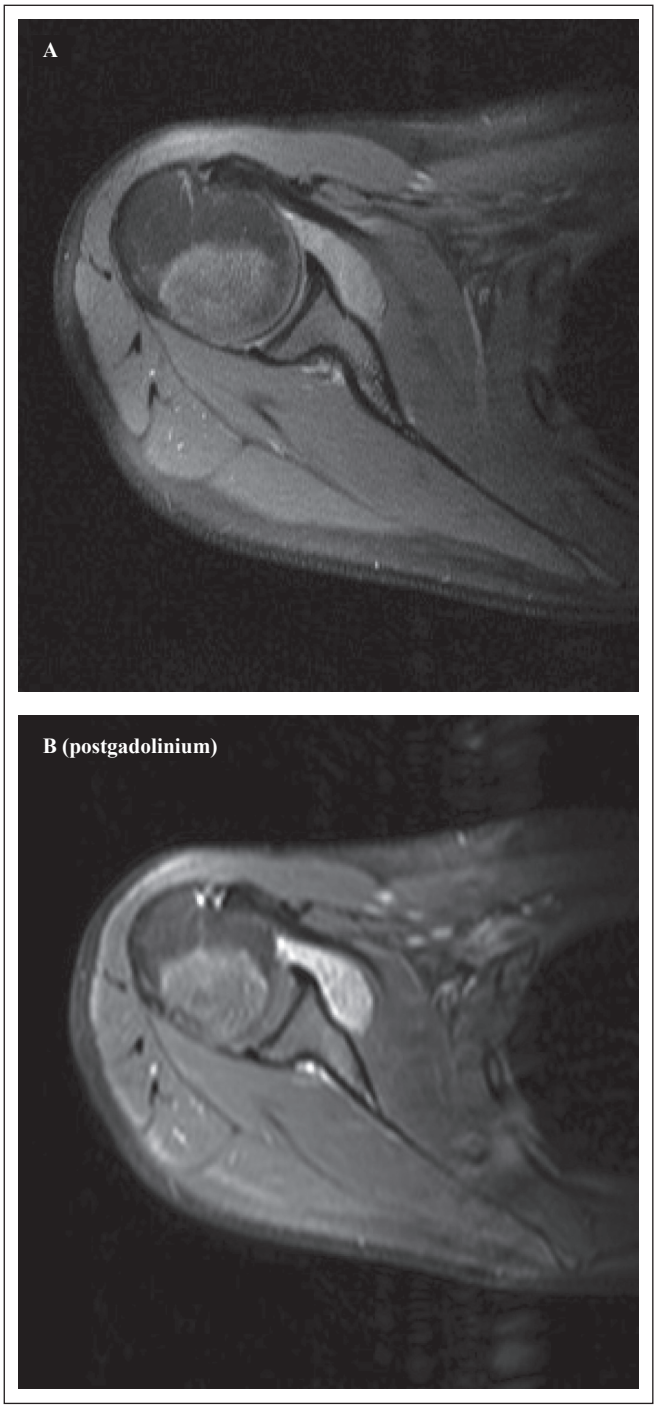
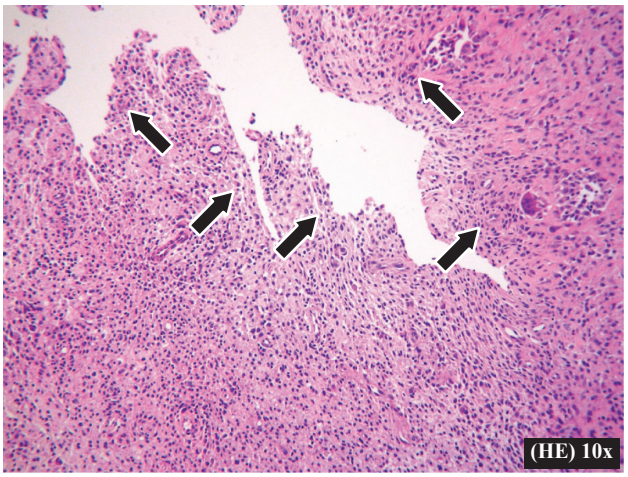
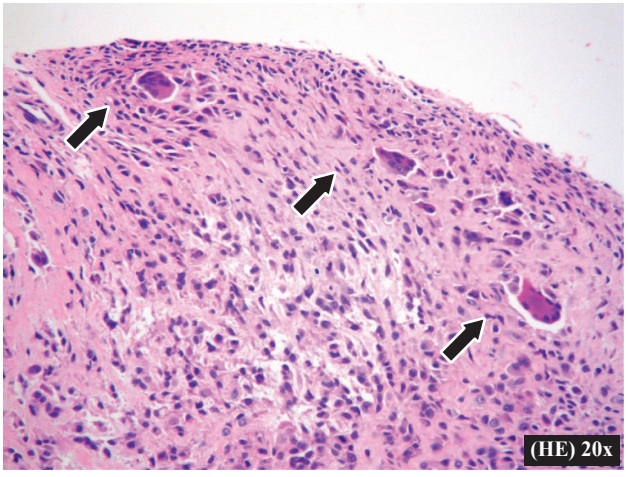
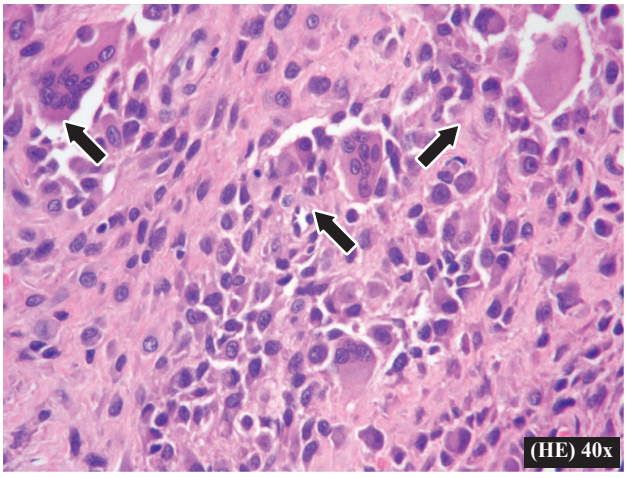


Figure 2. MRI of right shoulder demonstrate persistence of soft tissues mass, of oval aspect, compromising the subscapular bursa and the axillar recess, with volumetric increase when compared to a previous exam. T2-weighted image with fat saturation shows low signal intensity with small granules in between (of riziform aspect), and with isosignal to musculature in T1-weighted image. After contrast administration shows marked enhancement.







Figures 3. Pigmented villonodular synovitis: Nodular and villous proliferation (arrowheads) which projects itself in the synovial space. Constituted by round and oval shaped cells without nuclear atypicalities, having in between, giant multinuclear cells, sparse or forming small groupings (arrows) over fibroconnective stroma with more colagenated areas.
HE = Hematoxilin-Eosin.

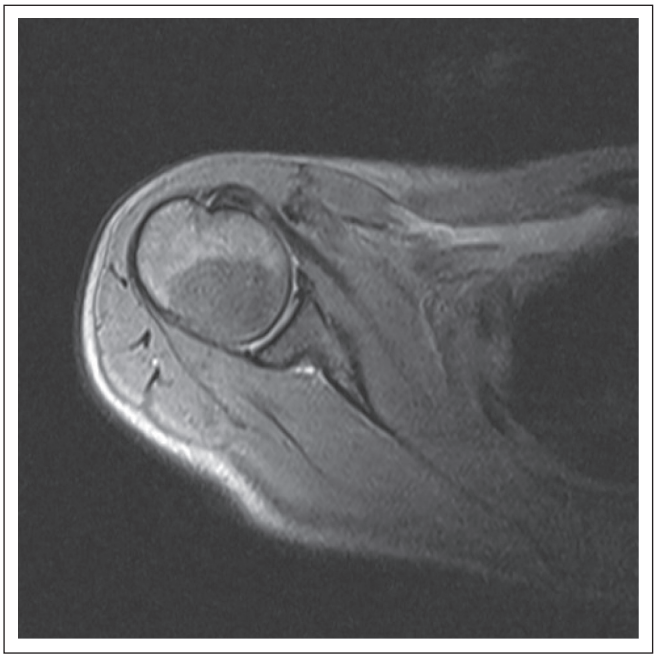


Figure 4: MRI of right shoulder with no evidence of intra-articular effusion or in the synovial prolongment, as well as in the shoulder bursa. There is no evidence of synovitis or joint effusion.

negative, and ANA, which was positive (1/80). There wasn't any other clinical or laboratorial evidence of JIA and the characteristic of the pain wasn't that of an inflammatory disease. The ANA can be justified by the frequency in the population without autoimmune disease; in these cases the most frequent pattern is the dense fine speckled, as observed in this patient. Another fact that drew the attention of the clinician at that occasion was the presence of an erythematous lesion, which associated with the positive ANA raised the suspicion of an autoimmune disease, but the dermatologic evaluation rejected this hypothesis for being a drug eruption.

A second MRI revealed that there was a deterioration of the synovitis after seven months, but there wasn't any suggestive evidence, either clinical or laboratorial, of other disorder. An arthroscopy was then performed for diagnostic elucidation and a complete synovectomy was performed during the procedure. The histopathology revealed pigmented villonodular synovitis (PVNS).

The PVNS is a rare disease, estimated in 1,8 cases/year by million of people. The intra-articular lesions appears in two forms: localized and diffuse. Giant cell tumors of the tendon sheath and villonodular bursitis are related lesions, generally extra-articular, and can be considered as an extra-articular extension of the villonodular synovitis. The PVNS is of an

unknown etiology and is characterized by hyperplasia of synovial villosities in joints and tendinous sheaths, with accentuated proliferation of the stroma cells, with great quantity of hemosiderotic pigment intra and extracellular and multinucleated giant cells, as observed in this patient. The disease is observed in patients between 20 and 50 years old, more in the male gender, its beginning is gradual, with intermittent pain, associated with hemorrhagic joint effusions and limitation of the articular amplitude. In this patient, the symptoms had lasted around a year, but there wasn't limitation of movement and the pain was fairly discrete.[9-19]

PVNS is a monoarticular and progressive disease, affecting large joints, with the knee compromised in 80% of the cases in the diffuse form, followed by the coxofemoral and ankles and, more rarely in other, joints. The shoulder, as occurred in the present case, is not frequently involved; there are a few more than 30 cases of villonodular synovitis in the shoulder published in English and French literatures, with the same age distribution already referred.[20-35]

The pathogenesis of this lesion is unknown, being attributed to the reaction to chronic trauma with repeated hemorrhages, or of inflammatory origin, to a local alteration in the metabolism of the lipids, leading to an accumulation of them in the interior of the phagocytes and traumatic secondary alteration or even to a benign neoplastic process, being this last the most probable hypothesis, even though, despite of its aggressive local characteristic, it has not been documented a case of malign transformation or metastasis. In the case of this young patient, the only factor to be considered is the many years of ballet practice, which could act as a traumatic factor. The invasive nature of the diffuse PVNS form, with destruction of the joint cartilage and bone is well documented, some believe that a similar infiltrating process as the one observed in the pannus of rheumatoid arthritis can occur.

The diagnosis is suspected clinically and confirmed by the radiological and anatomopathologic examinations. The clinical findings are not specific, the patient could be oligosymptomatic, as in the present case, and even present intermittent pain, progressive edema of the involved joint, limitation of movement and articular blockage. The definitive diagnosis can be retarded for years, after the appearance of the symptoms and clinical signs. The difficulty of the clinical diagnosis is due to its rarity and to the fact of simulating mechanical originated lesions (meniscal lesions, intra-articular free bodies etc.), or even synovial sarcoma, mainly when the affected joint is the knee. Given the difficulties of obtaining a conclusive PVNS clinical and radiographic diagnosis, other complementary imaging tests are essential for the diagnosis and planning of the appropriate treatment.[36]

Histologically, these lesions present synovial hyperplasia, hypervascularization, accumulation of histiocytes and extracellular deposition of hemosiderin. The presence of intra and extracellular hemosiderin shows in the macroscopy reddish or chocolate like color, hence the name pigmented. The following criteria are important in this evaluation: hypercellularity, infiltrated linfoplasmocitary, presence of multinucleate giant cells, phagocytes containing hemosiderotic pigment. The hypercellularity sometimes accompanied by mitosis can lead equivocally to the diagnosis of malignant neoplasia, a synovial sarcoma. Giant multinuclear cells, however, generally do not occur in synovial sarcoma besides the absence of phagocytes with hemosiderin pigment. Rheumatoid arthritis, synovial hemangioma and hemophilic arthritis can also produce synovial thickening, hemorrhage and hemosiderin deposit, which are very similar to the findings in PVNS.[36]

The radiological findings of PVNS are joint effusion and cystic erosion of the bone in 50% of the patients. The magnetic resonance imaging (MRI) is the test that better differentiates synovial pathologies and can make an early diagnosis.[37-40] Characteristic findings in the MRI are areas of low signal in the pondered sequences in T1 and T2, which correspond to a deposit of hemosiderin inside the synovia and are characteristic. The localized nodular aspect of PVNS is typical. The lesion is highlighted after contrast, delimiting its extension for later synovectomy. The evaluation of the extension of the bone involvement by MR is also important. This patient did not present this classic PVNS image, but a progressive synovitis that demanded other differential diagnosis.

The differential diagnosis of PVNS should be done with disorders that presents with joint effusion and synovial thickening, for example, rheumatoid arthritis and tuberculosis, diseases that have characteristics of systemic involvement and do not compromise exclusively the synovia and other components of the articulation. The MRI is important to differentiate some diseases which basically compromise the joint and neighboring structures like synovial hemangioma, synovial osteochondromatosis, synovial arborescence lipoma and synovial sarcoma.[36] Thus, PVNS presents a typically in all the imaging sequences a low signal intensity (created by paramagnetic effects of hemosiderin deposits and fibrous tissue). The low signal blooming effect of hemosiderin is best seen on T2-weighted gradient-echo images, besides presenting strong uptake of the endovenous contrast. The synovial hemangioma is a benign vascular tumor that usually extends outside the articulation; It is hypotense in T1 with punctuated images of linear

high-intensity signal, corresponding to the fibroadipose septum between the vascular channels. The T2-weighted images demostrates hypersignal and the septa are hypointense. In the synovial osteochondromatosis in the T1- weighted images, it is verified a low to intermediate intensity of the signal inside the synovial membrane and joint space, while in T2 the intensity of the signal is variable, due to the cartilaginous nature of the lesion and the extension of the calcified areas. In this disorder are observed ossified nodules, which can develop adipose marrow with the same intensity of signal of the arborescent lipoma. However, the simple RX allows the distinction between the two entities, by showing, in the first, multiple calcified bodies or ossificated inside the joint space. The synovial lipoma is an isolated intra-articular mass, has intensity of signal similar to that of fat in all the sequences, just like the arborescent lipoma, but can distinguish itself by its round shaped or oval form and by the absence of synovial proliferation. Arborescent lipoma is a rare intra-articular disease of unknown etiology, which consists of diffuse fat infiltration in the synovial membrane. The lipomatous fatty content of the lesion is well characterized in MRI or in the Computed Tomography. The synovial sarcoma presents extra-articular extension, calcification and bone invasion, aspects not found in PVNS.[36-39]

Treatment consists in synovectomy with very good results and little recurrence in the nodular form. In contrast, the diffuse form can have high recurrence rates. Patients not adequately treated can evolve to articular destruction and arthroplasty may be the solution in these cases.[41-45]

This patient had an excellent response to synovectomy, as demonstrated in the MRI performed nine months later.

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