Villonodular Synovitis of the Subtalar Joint: A Case Report

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Learning Point of the Article:

Villonodular synovitis (SVN) is a benign pseudotumor that is characterized by locally aggressive synovial proliferation of unknown etiology.

Abstract

Introduction: Villonodular synovitis (SVN) is a rare benign pseudotumoral proliferation of the synovial joint, of unknown etiology. In general, it reaches the big joints, especially the knee. Localization at the subtalar level is extremely rare, with only a few cases published in the literature.

Case Report: We report the case of an SVN of the subtalar in a 60-year-old patient without particular histories, who consulted for a swelling of the external border of the right foot evolving for 2 years. Biology, as well as the standard radiographs, was without anomalies. The diagnosis was evoked on magnetic resonance imaging (MRI) and confirmed by surgical biopsy and excision. The operative follow-up was simple with no recurrence after 3 years.

Conclusion: SVN of the subtalar is rare. Surgical treatment is based mainly on synovectomy, and the prognosis depends mainly on the osteocartilaginous lesion and the quality of excision.

Keywords: Villonodular synovitis, Subtalar, Synovectomy.

Introduction

Villonodular synovitis (SVN) is a benign pseudo-tumor that is characterized by locally aggressive synovial proliferation of unknown etiology. It essentially affects the knee and hip. Localization at the foot joints is extremely rare. We report the case of an SVN of the subtalar.

Observation

Patient G.J, 60 years old with no particular pathological history, was hospitalized for exploration of a swelling of the external border of her right foot that had been evolving for 2 years. The physical examination showed an oval swelling of 3 cm long, lying in the submalleolar external region, painless, mobile, and of firm consistency (Fig. 1). The biological assessment did not show any inflammatory syndrome. Standard radiography was

normal (Fig. 2). Ultrasonography showed a polylobed lesion, more than 3 cm long, hypoechogenic, heterogeneous, nonvascularized, and in communication with the subtalar joint. Magnetic resonance imaging (MRI) found a synovial, early development, clear bound, and heterogeneous signal lesion on all sequences with no significant bone abnormalities (Fig. 3) A surgical biopsy confirmed the diagnosis of SVN. The patient, therefore, had a mass resection with subtotal synovectomy through an external approach (Fig. 4). Peroperatively, we didn't observe any osteocartilaginous involvement and no bone gesture was performed. At 3 years of follow-up, there was no clinical recurrence and the functional impairment was moderate. However, a local relapse cannot be ruled out, requiring close clinical and radiological monitoring.

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Figure 1: Tumefaction of the external edge of the foot.

Figure 2: (a and b) Foot front and profile X-rays.

Discussion

SVN is a rare benign fibrohistiocytic tumor of the synovium. Myers and Masi [1] estimated its incidence at 1.8 cases per million inhabitants per year in 1980. It is also the most common tumor lesions originating in the synovium, i.e., 29.6% of cases [2, 3]. It can also affect tendinous sheaths and bursae. The etiopathogenesis of SVN is poorly known and still discussed today. Although several hypotheses have been put forward such as the occurrence of synovial proliferation linked to chronic inflammation, benign neoplasia of unknown etiology, abnormality of local lipid metabolism and repeated trauma [4]. Some cytogenetic aberrations are detected in most cases of SVN [5]. Some authors such as Perka et al. [6] suggest that the etiopathogenesis of SVN is inflammatory in localized and neoplastic in diffuse forms. All joints may be affected, but the most commonly affected are the knee (80%), hip (10%), ankle (5%), and exceptionally the joints of the foot and spine. The clinical presentation of SVN is non-specific. In both forms, diffuse and localized, the evolution is slow, and generally, the diagnosis is made at an advanced stage of the disease, about 2-3years after the beginning. In our case, the delay was 2 years. The main clinical signs of SVN are the appearance of a monoarticular swelling sometimes palpable in the form of a mass, responsible for mechanical pain, as well as functional impairment of progressive worsening, this is the case of our patient, but in a general way, the clinical symptomatology of the SVN is non-specific making the diagnosis difficult. Complementary examinations are of paramount importance in this pathology. Biologically, all authors are unanimous about the absence of an SVN-specific biological syndrome regardless of its location [3, 6, 7]. Standard radiographs are usually normal. Bone erosions are sometimes observed at an advanced stage because of the destructive character of the SVN. In localized forms, ultrasound can objectify a hypoechoic formation that

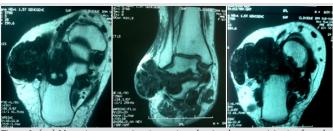


Figure 3: (a-c) Magnetic resonance imaging sections showing the mass originating from the subastragalienne.

may be associated with joint effusion. MRI remains the examination of choice and can show multiple synovial lesions presenting a hypointense or intermediate signal in T1 and a hyposignal in T2 and gradient echo [8]. Small areas of hyposignal are evidenced in the synovial masses without signal enhancement by gadolinium which corresponds to hemosiderin deposits. This MRI appearance is almost pathognomonic of SVN [9]. In our case, the MRI allowed us to strongly evoke the diagnosis but cannot replace the histological study which remains essential to confirm or affirm the diagnosis. The locoregional evolution of SVN is slow with an extension to the entire synovium, bone, and adjacent soft tissues. An untreated localized SVN may evolve into a diffuse form, hence, the need for early and adequate treatment. This treatment is not yet standardized; there is no uniform therapeutic strategy because of the rarity of this condition. It rests in the first place on a careful surgical synovectomy, the most complete possible. In localized forms, like our case, a synovectomy is the rule. In diffuse forms, total synovectomy should be supplemented by bone curettage. Some authors advocate synoviortheses (isotopic or osmic) in cases where the synovectomy is incomplete. Others recommend the use of antitumor necrosis factor-α in patients who refuse usual treatments, but its effectiveness is debated. The long-term prognosis of the disease depends on the extent of the lesion at the time of diagnosis, as well as the location and quality of the excision. In general, localized forms heal after complete exeresis, with almost no recurrence risk [10, 11]. For our patient, at more than 3-year follow-up, there was no local recurrence and this confirmed the quality of synovium excision.

Conclusion

SVN is a locally aggressive synovial proliferation that can affect joints, tendon, and pouches. Localization at the subtalar level is rarely described in the literature. The clinical picture is most often non-specific. MRI remains the key examination in this pathology for both diagnosis and follow-up. The treatment is surgically based mainly on synovectomy, and the final diagnosis is made by the histological study of synovial biopsy or specimen. The prognosis depends mainly on osteocartilaginous involvement and the quality of excision.



Figure 4: (a-b-c) Intraoperative aspect of the synovial mass.



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Clinical Message

SVN of the subtalar is rare; its diagnosis is evoked by MRI and confirmed by histology. The treatment is surgical and the prognosis depends on the quality of excision.

References

- 1. Myers BW, Masi AT. Pigmented villonodular synovitis and tenosynovitis: A clinicalepidemiologic study of 166 cases and literature review. Medicine 1980; 59:223-38.
- 2. Adelani MA, Wupperman RM, Holt GE. Benign synovial disorders. J Am Acad Orthop Surg 2008;16:268-75.
- 3. Ma X, Shi G, Xia C, Liu H, He J, Jin W, et al. Pigmented villonodular synovitis: A retrospective study of seventy five cases (eighty one joints). Int Orthop2013; 37:1165-70.
- 4. Sharma H, Rana B, Mahendra A, Jane MJ, Reid R. Outcome of 17 pigmented villonodular synovitis (PVNS) of the knee at 6 years mean follow-up. Knee 2007;14:390-4.
- Murphey MD, Rhee JH, Lewis RB, Fanburg-Smith JC, Flemming DJ, Walker EA, et al. Pigmented villonodular synovitis: Radiologic-pathologic correlation. Radiographics2008; 28:1493-518.
- 6. Perka C, Labs K, Zippel H, Buttgereit F. Localized pigmented villonodular synovitis of the knee joint: Neoplasm or reactive granuloma? A review of 18 cases. Rheumatology 2000; 39:172-8.

- Nissen MJ, Boucher A, Brulhart L, Menetrey J, Gabay C. Efficacy of intra-articular bevacizumab for relapsing diffuse-type giant cell tumour. Ann Rheum Dis 2014; 73:947-8.
- 8. Carr CR, Berley FV, Davis WC. Synovitis of the hip. JBone Joint Surg 1954; 36a:1007-13.
- HernigouPH, Marichez M, Voisin MC, Delepine G, Goutallier D. Magnetic resonance imaging in the evaluation of a pseudotumoral form of villonodular synovitis of the knee. A case report. Rev ChirOrthop1990; 76:218-21.
- 10. Dale K, Smith HJ, Paus AC, Refsum SB. Dynamic MR-imaging in the diagnosis of pigmented villonodular synovitis of the knee. Scand J Rheumatol 2000; 29:336-9.
- 11. Ogilvie-Harris DJ, McLean J, Zarnett ME. Pigmented villonodular synovitis of the knee. The results of total arthroscopic synovectomy, partial, arthroscopic synovectomy, and arthroscopic local excision. J Bone Joint Surg Am 1992; 74:119-23.

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