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Case Report

Localized Tenosynovial Giant Cell Tumor: An Incidentally Found Lesion in the Shoulder

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Keywords

Tenosynovial giant cell tumors · Localized tenosynovial giant cell tumors · Pigmented villonodular synovitis · Shoulder

Abstract

The concept of localized and diffuse tenosynovial giant cell tumors (TSGCT) is recent and may still cause some confusion among surgeons. This disorder constitutes a family of proliferative lesions characterized by their origin in the articular synovium, tendon sheaths, or bursa. It is not always easy to diagnose this pathology, especially due to its multitude of presentation. We describe an incidentally found localized TSGCT intra-articularly located in the shoulder. The localized lesions are considered a more benign form that usually present not with specific symptoms but rather a manifestation of the disturbance in the affected joint or the surrounding soft tissues. MRI is indispensable for its diagnosis, and the standard treatment is complete surgical resection. Due to its low incidence, it is difficult to find literature that goes beyond clinical reports or small case series. With this report, we intend to call readers' attention for the variable presentations, the different diagnostic and treatment strategies, and the expected outcomes.

Introduction

Throughout history, many definitions were used to label tenosynovial giant cell tumors (TSGCT). These lesions were first described in 1941 by Jaffe and colleagues and were grouped as "pigmented villonodular synovitis, bursitis, and tenosynovitis" [1]. In 2013, the WHO system for the classification of soft tissue tumors adopted the term TSGCT to describe these

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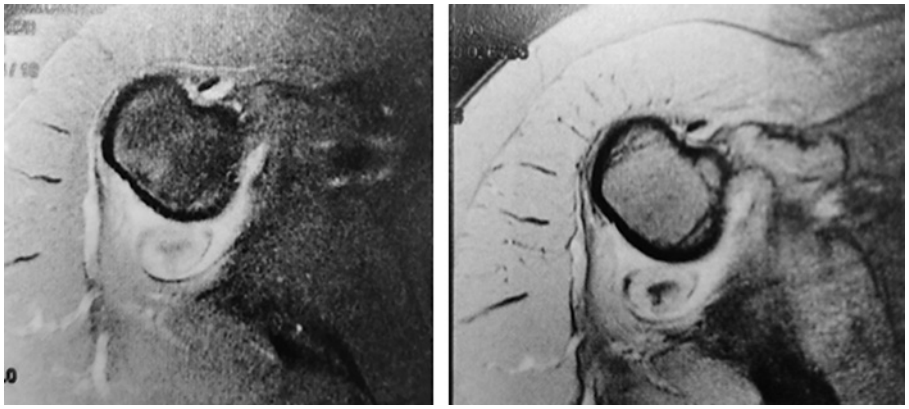

Fig. 1. MRI imaging of the intra-articular localized TSGCT. TSGCT, tenosynovial giant cell tumors.

lesions, dividing them into 2 main groups, *localized* and *diffuse* [2, 3]. They are, nowadays, considered a family of proliferative lesions characterized by their origin in the articular synovium, tendon sheaths, or bursa and their benign behavior [1–3]. Despite this nomenclature overlapping, the natural course of the disease still differs substantially between localized and diffuse tumors [3].

TSGCT are rare entities with an estimated incidence of 1.8 patients per 1 million [4, 5] affecting, predominantly, the female gender, between the fourth and fifth decade of life in cases of localized TSGCT and a little earlier for the diffuse variant [1, 3]. The localized form is more often encountered in digits (85%), near interphalangeal joints and synovial sheaths, especially on the palmar side [1]. Other locations include the knee, foot, and ankle (second most frequent location [2]) and rarely the hip or the elbow [6].

Contrasting with the diffuse variants, the localized lesions are considered a more benign form, easily diagnosed and treated without the need for aggressive surgery [7]. In this case report, we describe an incidentally found, intra-articular localized TSGCT in the shoulder in a woman with a supraspinatus tendon partial tear (Fig. 1).

Case Report/Case Presentation

A 42-year-old female nurse, healthy, without any usual medication, presented to us with shoulder pain, 1 month after a minor trauma. There was no limitation in range of motion despite some mild pain in internal rotation. Anterior flexion and external rotation were asymptomatic. Jobe's test, as well as the lift off test, were positive. Conservative treatment was initiated with rest and nonsteroid anti-inflammatory drugs, and an MRI was requested. The image (Fig. 1) revealed a nodular structure with a 2 cm diameter, well defined, in the inferior glenohumeral recess, which was isointense on T1-weighted images and hyperintense on T2-weighted images. It captured the contrast product but did not show calcifications, fat, or hemosiderin (Fig. 2). It also did not show a blooming artifact (images 1 and 2). A 3-mm partial-thickness tear of supraspinatus tendon was also evident. An arthroscopic approach for surgical excision of the lesion and repair of the tear was proposed and accepted by the patient. The lesion was easily identified in the inferior recess and was pedunculated and multilobulated (image 3). It was removed by sectioning the pedicle and excised as a unit. The partial tear was also confirmed and repaired. The anatomopathological results revealed a proliferative lesion with connective tissue fibroblasts, fibrin, and hemosiderin compatible with localized TSGCT (image 4). The patient is completely asymptomatic 3 months after surgery, and a close follow-up will be maintained.

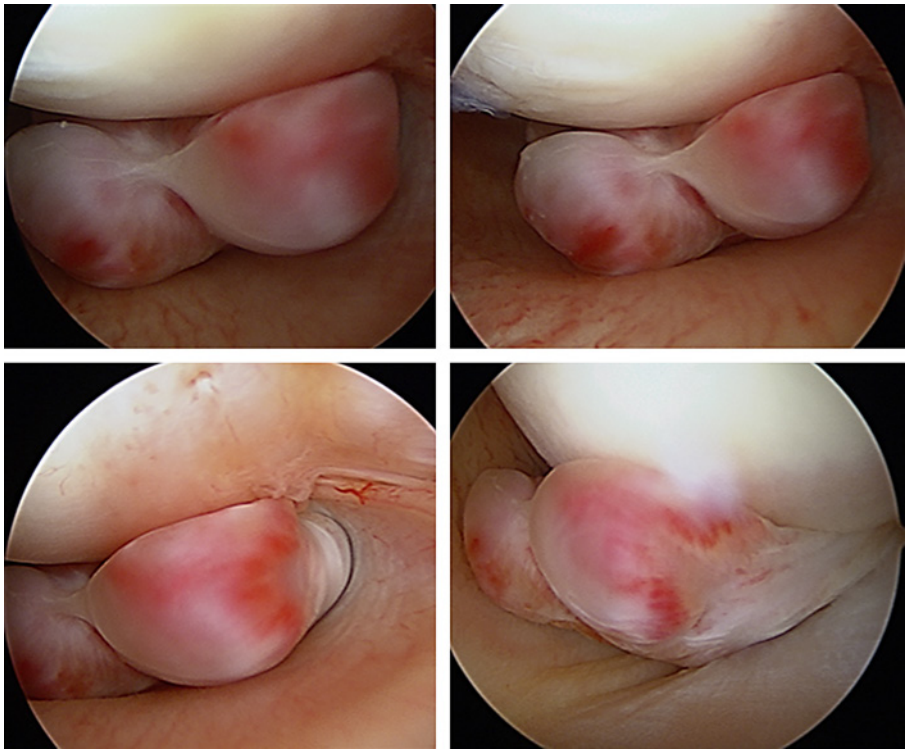

Fig. 2. Arthroscopic view of the intra-articular localized TSGCT. TSGCT, tenosynovial giant cell tumors.

Discussion/Conclusion

We described a case of a localized TSGCT in a 42-year-old female, which is in agreement with the general epidemiologic data. Localized forms predominate in the digits (85%), extra-articularly, and are rarely encountered in the shoulder [1]. The causality is a matter of controversy, but the truth is that in half of the cases, a careful interview reveals a history of trauma [1]. The reported case also referred a history of minor trauma one month before.

These lesions are normally identified for the discomfort, repeated joint swelling, and restricted range of motion [8]. We often cannot find specific symptoms but rather a manifestation of the disturbance in the affected joint or the surrounding soft tissues. Our patient presented with shoulder pain, nocturnal awakenings, and some limitation in internal rotation. A 3-mm partial-thickness tear of supraspinatus tendon was present and worked as a bias on the cause of the symptoms. The standard of care for intra-articular TSGCT is surgical excision to reduce articular symptoms and improve limb function [3]. The current literature is controversial about repairing partial-thickness tears, but some reports agree that it could have good results, especially in patients under 45 years old [9]. Therefore, we decided to repair the lesion in the same surgical approach.

A thorough imageological investigation is indispensable. Usually, a simple radiography is the first exam asked and is contributive as a way to evidence any bone abnormalities. Cystic erosions with periosteal reactions are more frequent in the hand or hip. If found outside weight-bearing areas, symmetrically and without calcifications, could be suggestive of articular TSGCT [1]. MRI is the most helpful imaging tool, allowing diagnosis, staging, and evaluation during follow-up. Localized forms tend to be well-circumscribed lesions, well demarcated by a low signal intensity capsule as a result of fibrosis or hemosiderin [10]. Macroscopically, the localized form is often familiar, with one or

multiple well-delimitated nodules, pedunculated in most cases, and yellowish brown, as a result of hemosiderin infiltration. The size varies greatly, where some lesions can go up to 13 cm [1].

The localized TSGCT are usually considered benign lesions; however, their potential for progression must be clearly understood from the beginning. The intra-articular lesions can still cause damage and discomfort as they are confined to a narrow space, so their surgical excision is desirable [1]. It can be done by an arthroscopic approach, but if the entire lesion is not visualized or is difficult to reach, the procedure should be converted to an arthrotomy [11]. Arthroscopy has the theoretical risk of joint seeding and portal contamination. Mastboom et al. [3] stated that the risk of recurrence was higher not only with arthroscopic resection (18%) compared with open resection (9%) but also with lesions >5 cm or that were already in recurrence at presentation. The lesion discussed in this report is pedunculated, one of the few characteristics that make a localized intra-articular TSGCT amenable to arthroscopic resection. Ganglion cysts, hemangiomas, and synovial sarcomas are examples of differential diagnoses; however, they normally present with high T2 signal, which differentiates from TSGCT [1]. As it was a relatively small lesion with imageological characteristics of TSGCT and amenable to complete excision (as it was pedunculated), we decided to excise the tumor completely without previous biopsy. The current literature is in favor of this strategy [11, 12]. Localized TSGCT rarely recur, and some authors reported rates of recurrence-free survival at 5 years between 73% and 88% [11].

A complete excision is sometimes difficult, more often with the diffuse variant. In these cases, some additional therapies like cryosurgery, external-beam radiotherapy, radiosynovectomy/synoviorthesis, and more recently, potent selective colony-stimulating factor-1 inhibitors can be useful to reduce functional impairment of joints, residual disease, and recurrences.

Verspoor et al. [11] recommended a new MRI 6 months after surgery as a baseline image to compare posterior evaluations, if symptoms arise. However, there are no current markers of recurrence, and many doubts persist about long-term follow-up. Clinical evaluation is of utmost importance, and timings are not defined so each physician should adapt the plan case-by-case.

TSGCT are rare benign lesions that come to our attention with relative insidious and nonspecific symptomatology. A great degree of suspicion is necessary, and MRI is indispensable for diagnosis. The standard treatment should be complete surgical resection.

Statement of Ethics

The subject included in this work has given written informed consent to publish this case (including publication of images), and therefore the study is exempt from ethics committee approval.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Jorge Gomes Lopes was involved in manuscript writing. Manuel Gutierres was involved in data collection. Luisa Vital was involved in manuscript writing. Miguel Relvas-Silva was involved in data collection. Ricardo São-Simão was involved in manuscript editing. Francisco Serdoura was involved in manuscript editing.

Data Availability Statement

The data that support the findings of this study are not openly available.

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