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CASE REPORT

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Giant Cell Tumor of the Tendon Sheath in the Anatomical Snuffbox – Report of an Unusual Location

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ABSTRACT

The aim of this study is to present a case of giant cell tumor of the tendon sheath in the anatomical snuffbox, an extremely unusual location of such neoplasms. A 54-year-old male came to our hospital with a mass in his left hand that had appeared over the past two years, demonstrating a recent rapid increase in size. The physical examination revealed that the mass did not cause any pain or restriction in the range of motion. The neurovascular function remained intact. The magnetic resonance imaging (MRI) detected a 2.5 x 1.5 cm mass in the anatomical snuffbox but without a clear diagnosis. A meticulous marginal surgical excision was performed and biopsy sample was sent for histopathologic examination. Intraoperatively, it was observed that the tumor extended in-between the tendons of the first and third dorsal compartments, reaching the radial artery at the depth of the anatomical snuffbox. Macroscopically, the tumor resembled a giant cell tumor, which was confirmed by histological examination. The patient was discharged from the hospital on the same day. He fully resumed his daily activities three weeks after surgery. Three years later, he remained free of symptoms without any recurrence of the tumor. Giant cell tumor of the tendon sheath is a benign neoplasm that demonstrates local aggressiveness. In several cases, nerves and blood vessels might be entrapped within the tumor. This complication along with its high recurrence rate renders surgical excision of the tumor a particularly demanding procedure.

Keywords: giant cell tumor, tendon sheath, anatomical snuffbox, neoplasm, excision.

INTRODUCTION

Giant cell tumors of the tendon sheath (GCTTS) are benign proliferative masses of synovial origin that are commonly slow growing and originate from the synovial cells of the tendon sheaths. They might be nodular or diffuse and are classified as types 1 and 2. Type 1 displays a single round or multilobulated tumor, while in type 2, two or more separated distinct tumors are typically observed (1-3). These well-circumscribed firm neoplasms are mostly painless, unless they are near a joint. In this case, pain and decreased range of motion might be observed (1-3).

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These lesions are usually seen in the third to fifth decade of life, with a slightly higher predominance in women (4-6). Despite their non-malignant characteristics, GCTTS demonstrate local aggressiveness and a propensity for recurrence (7). In the hand and wrist, they are the second most common mass following the ganglion cyst and the most frequent mesenchymal tumor found in this location (5, 6, 8). In fact, they are found almost exclusively in a digit of the hand. Rare occurrences have been reported in other parts of the body, including palm, wrist, foot, knee, ankle and elbow (4, 6, 8-12). When GCTTS concern a joint, they commonly grow outside with an extension into or near the joint (11).

The purpose of the current study was to present a case of a giant cell tumor of the tendon sheath that was found in an unusual place, and more specifically, in the anatomical snuffbox. The diagnostic process, surgical approach and postoperative management that were implemented have been also emphasized. ❑

CASE PRESENTATION

A 54-year-old left-handed male farmer visited our hospital reporting an asymptomatic palpable mass in his left hand that was first noticed two years prior to presentation but had a considerable and rapid increase in size over the last three months. The physical examination revealed the existence of a tumor in the anatomical position of the snuffbox, between the tendon of the extensor pollicis longus and the tendons of the extensor pollicis brevis and abductor pollicis longus. The mass did not cause any pain, tenderness, numbness or tingling. No limitation in the range of motion of radiocarpal or any other adjacent joint was found. Regarding his medical report, the patient displayed no trauma history or evidence of systemic or degenerative joint diseases. In parallel, it was remarked that the neurovascular function remained unaffected.

Radiographic assessments that included standard anteroposterior and lateral views were performed without showing any pathognomonic sign (Figure 1). The MRI revealed a 2.5 x 1.5 cm well-defined mass into the anatomical snuffbox in close association with the adjacent tendons without any bony erosion (Figure 2). No clear diagnosis could be given.

The patient underwent surgical excision of the tumor under local anesthesia with the use of tourniquet control and magnifying loupes (3.5 x magnification) for better visualization. In addition, a subsequent biopsy sample from the tumor was sent for histopathologic examination. Intraoperatively, it was noted that the mass extended in-between the tendons of the first and third dorsal compartments into the anatomical snuffbox, approaching the radial artery at the depth of the snuffbox. Macroscopically, the well-encapsulated gray-brown to yellow lobulated soft-tissue mass strongly resembled a giant cell tumor of the tendon sheath and thus a meticulous marginal excision was conducted (Figure 3). As the

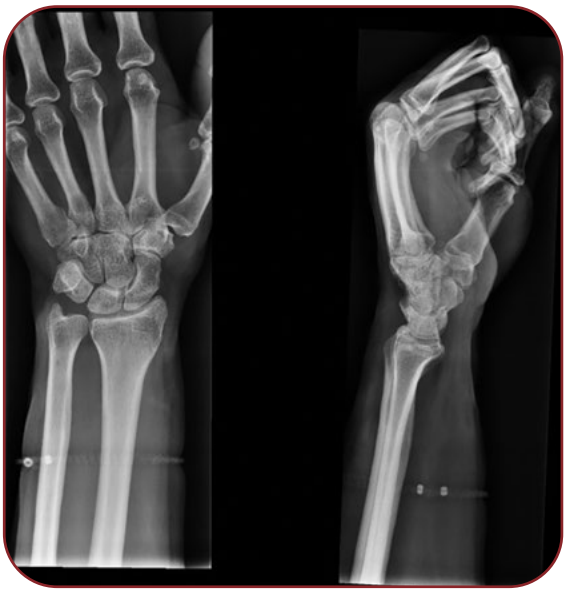

FIGURE 1. Preoperative anteroposterior and lateral radiographs of the left radiocarpal joint

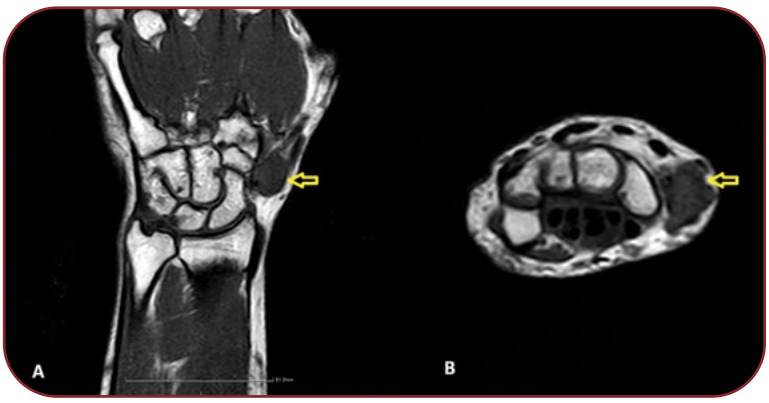

FIGURE 2. Preoperative magnetic resonance imaging depicting the tumor into the anatomical snuffbox: (A) coronal view; (B) transverse view.

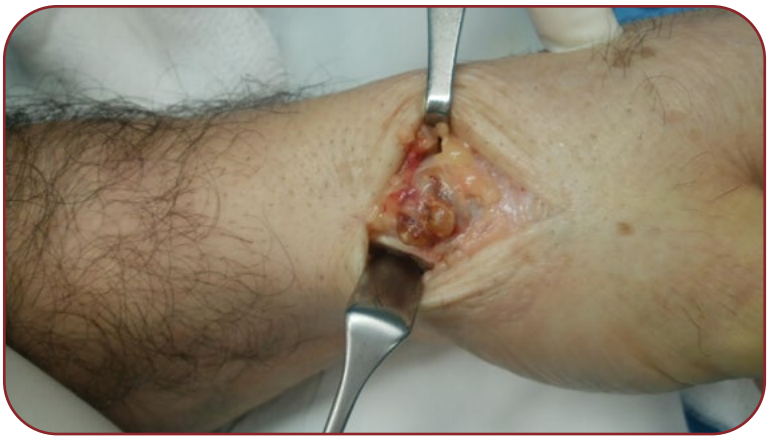

FIGURE 3. Intraoperative image displaying the lobulated soft-tissue tumor

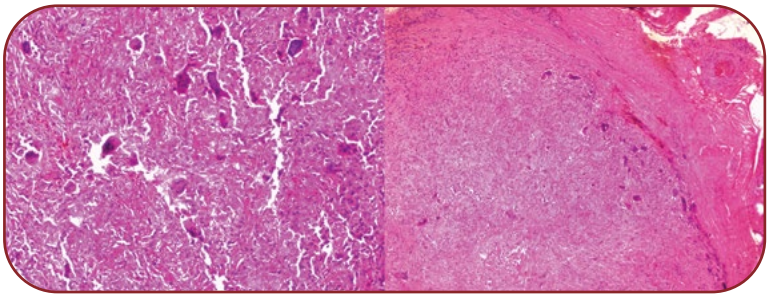

FIGURE 4. Histopathology of the neoplasm resected showing polynuclear giant cell surrounded by mononuclear stromal cells [H&E, (A) × 100, (B) × 40, scale bar = 100 μm]

tumor was originated from the synovial sheaths, care was taken to completely remove the neoplasm without injuring the adjacent tissues such as the tendons or neurovascular structures. No erosion of the nearby bones was observed.

The histopathological analysis exhibited a distinctive composition characterized by mononuclear cells and multinucleated giant cells with hemosiderin deposits within a densely collagenous background. The mitotic activity was low (3-4/10 high power fields (HPF), while there were no areas of necrosis. These findings were indicative of a diagnosis consistent with giant cell tumor of the tendon sheaths (Figure 4).

The surgical procedure was well tolerated by the patient, who was therefore discharged from hospital on the same day with a bulky dressing on the wound. He was followed up at weeks 1 and 2 for wound check and suture removal, respectively. Early rehabilitation exercises started from the next day to prevent joint stiffness and enhance wrist and hand function. Two weeks after surgery, the postoperative swelling had considerably subsided and the patient continued with an accelerated rehabilitation protocol in order to increase the range of joints' motion and grip strength. Three weeks after surgery, the patient was free of pain without any functional limitations or deficits and he returned to his daily routine. He was still followed up at month 6, year 1, and then yearly thereafter. Three years after surgery, the patient remained completely asymptomatic without any recurrence of the tumor. ❑

DISCUSSION

Giant cell tumors of the tendon sheath are lesions that may arise from tendon sheaths, fascial planes, joint capsules, bursae or ligaments (13). They usually represent an extra-articular localized type of pigmented villonodular synovitis, as reported by Moore et al, who used this term in order to give a more detailed description of clinical manifestation of the mass when it is developed in the hand (14). However, a large variety of names such as localized nodular tenosynovitis, pigmented villonodular proliferative synovitis, pigmented villonodular tenosynovitis, proliferative synovitis, benign synovioma, giant cell fibrohemangioma, fibroma, fibrous histiocytoma, sclerosing hemangioma, myeloxanthoma, fibrous xanthoma and xanthogranuloma, amongst others, has been referred to giant cell tumors of the tendon sheath (5, 13-16).

These multiple names might be associated with the pathogenesis of GCTTS which remains unclear. Several studies support their inflammatory origin, whereas others present evidence for their neoplastic basis (1-3, 5, 6, 12, 14, 15). However, it is also considered that factors including trauma, genetic mutations or hormone-related effects, might be related to the etiology of GCTTS (1, 13). In parallel, the connection of these tumors with rheumatoid arthritis or osteoarthritis is still under investigation (6, 16, 16).

Regarding their anatomic location, those which occur in the hand are usually extra-articular, often multiple and concern mostly the small joints of a finger (4, 13). The most frequent area of occurrence is the distal interphalangeal (DIP) joint, with the index finger being the most common digital location (4, 6, 8-15). In addition, they are predominantly located on the palmar site (13-15). Similar digit tumors can be observed in the foot with analogous characteristics but a lower incidence (4). However, rarely, there are GCTTS that can be found in larger joints like in

the wrist, ankle, foot and elbow (4, 6, 8-12). These tumors are typically intra-articular, single and relatively large (4).

To the best of our knowledge, apart from infrequent cases of GCTTS found in the palm and especially in thenar eminence, there are no studies in the literature reporting the occurrence of this mass in the anatomical snuffbox. In our case, the neoplasm was extra-articular and similar to the GCTTS found in the small joints of the fingers, and not to those noted in the larger joints such as the wrist. It was extended between the tendon of the extensor pollicis longus and the tendons of the extensor pollicis brevis and abductor pollicis longus, without causing any functional limitation in muscle contraction.

According to the existing literature, patients display an average of six months to 2.5 years after the initial presentation of symptoms, due to the slow-developing nature of the mass (13). Despite the fact that, in many cases, the lesion can cause symptoms, including pain (particularly in neurovascular involvement), gradually progressive swelling and restriction in range of motion, our patient was asymptomatic and the reason for coming to our hospital was the recent rapid increase in size of the lesion.

Concerning radiographs as diagnostic tools, they can be normal as in our case or they may demonstrate a subtle soft-tissue mass. In more advanced cases, osseous alterations such as bony erosion or periosteal reaction might be seen (17). In several occasions, ultrasonography (US) may propose the diagnosis on the basis of lesion's close approximation to the adjacent tendon. In the majority of cases, US depicts a solid, homogeneous and hypoechoic mass with increased vascularity, closely associated with a tendon (18). A more definitive diagnosis can be provided by MRI, as the GCTTS are exhibited like circumscribed lobulated structures in relation to a tendon presenting decreased signal intensity on T1- and T2-weighted sequences as a result of the paramagnetic effect of hemosiderin and the existence of fibrous tissue (17).

However, clear and accurate diagnosis can be made only after histopathological examination of biopsy specimens from the mass, that can be taken either intraoperatively or preoperatively with the use of fine-needle aspiration biopsy (13). Histologically, GCTTS are comprised of multinucleated giant cells, histiocytes, macrophages and hemosiderin (6, 12). Typically, in the hand area, the tumor is multilobular and well-circumscribed, partially or fully encapsulated presenting extensions or satellite lesions (6, 12, 13). The coloration may vary from grey to yellow or yellow-orange, frequently with brownish regions, depending on the levels of hemosiderin and collagen and on the quality of histocytes of the tumor (6, 12, 13). In our patient, as MRI revealed that the neoplasm was benign, it was decided to avoid preoperative aspiration and to follow excision of the tumor and biopsy at the same time.

The aim of every surgeon should be the excision of the tumor with minimal damage to the healthy surrounding tissues. However, inadequate excision of the mass when attempting to fully preserve anatomical structures could lead to recurrence of the tumor (13). Therefore, a balance should be sustained between fully removing tumor margins and salvaging tissues. In cases where the tumor does not aggressively infiltrate adjacent tissues such as tendons, blood vessels, nerves or joint capsule, marginal excision of the lesion is the recommended treatment with low percentage of recurrence (19). On the contrary, when these structures are directly involved with the mass or when there are satellite lesions, marginal excision is frequently ineffective, presenting high recurrence rate, and surgeons should be more aggressive proceeding to radical surgical excision with resection of associated satellite nodules (19). If the lesion has invaded the surrounding bones, the excision may require sacrificing segments of them (14). When the mass originates from the joint, a capsulotomy should be conducted to explore the joint and debride any remnants of pigmented tissue (14).

In our patient, the tumor was well-encapsulated, localized in-between the tendons of the first and third dorsal compartments without any adhesions to them or to the contents of the anatomical snuffbox, including the radial artery, the superficial branch of the radial nerve and the cephalic vein. Moreover, no satellite lesions were found. Thus, a meticulous excision of the mass was performed, with caution to completely remove it without causing damage to the adjacent structures. The management that was followed was successful as our patient remained asymptomatic without any recurrence of the tumor at his last follow-up, three years postoperatively.

In the literature, the recurrence rate ranges from 5-50% after a primary excision of GCTTS (6, 8, 11-16). This variability among different studies could be attributed to the aforementioned incomplete excision of the neoplasm in order to maintain the adjacent healthy structures intact or to residual satellite lesions that could not be detected intraoperatively (6, 8). In addition, tumor location, tissue involvement, pseudocapsule presence, bony erosion or genetic predisposition have been reported as prognostic factors for recurrence of GCTTS (2, 9, 19).

Over the last years, the reported recurrence rates have demonstrated a decrease, potentially due to the improved preoperative workup with MRI and fine-needle aspiration cytology, intraoperative use of loupe magnification and use of meticulous surgical procedures (3, 13). Furthermore, it is remarked that postoperative radiotherapy has been advised as adjuvant therapy for prevention of recurrence of GCTTS at high-risk cases such as those presenting high mitotic activity, aggressive histologic features or history of previous recurrence (9, 20). Nevertheless, in instances of recurrent tumors, subsequent re-excision should be undertaken without hesitations to sacrifice surrounding involved tissues, in order to ensure complete excision (3, 9, 13). ❑

CONCLUSION

In conclusion, giant cell tumors of the tendon sheath in the hand may not only concern the digits but also areas where it is hard to occur such as the anatomical snuffbox. Frequently, these lesions present a complex mosaic regarding the clinical manifestation, radiologic examination, histopathological evaluation and therapeutic approach. If the tumor is well-encapsulated and not directly involved with the adjacent tissues or when there are not satellite lesions, meticulous marginal excision could be an adequate surgical procedure with low potential of recurrence. ❑

Conflicts of interest: none declared.

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Ethical statement: The present study was approved by the Institutional Review Board (14402/27-9-2024).

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