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Case Report

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Subungual glomus tumor: a clinical case and a review of the literature

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ABSTRACT

Glomus tumors are rare vascular lesions, located in the dermis, very difficult to diagnose. A 55 years old female patient with pain at first finger of her right hand, which eradiated to the forearm, with several years of evolution. The pain was worst with local compression and cold exposure. The magnetic resonance imaging was compatible with a subungual glomus tumor. Surgical resection of the lesion by a lateral approach was done. The histopathological analysis confirmed the diagnosis and the patient complaints disappeared after the procedure. The surgical treatment is usually curative and the lateral approach, although not the most commonly used, could be a good alternative.

Keywords: Glomus tumor, Nail, Surgical procedure, Lateral approach

INTRODUCTION

Glomus tumors are rare vascular lesions, difficult to diagnose.[1,2] These tumors originate from glomus body, present in reticular layer of dermis.[3] These bodies are neuromyoarterial structures responsible for regulate blood flow, interfering with arterial pressure and body temperature.[4,5] Disease was described by Wood, in 1812, and histopathology by Masson, in 1924.[6,7] Despite that, etiology remains unclear, however factors such as age, sex, trauma and heredity may be related with the development of the disease.[8]

These neoplasms result from hyperplasia of part of these structures and typically affects people between 30 and 50 years old, especially women and are located, most frequently, in subungual zone of fingers, although can appear anywhere in human body.[9] 75% of lesions are located in hands, with 65% located in distal phalanx zone.[10] The classic triad of symptoms includes pain (the most frequently reported complaint), point tenderness, and cold sensitivity.[2-5,11] However, other clinical presentations are possible, such as nail deformities, nodular lesions, or nail discoloration.[3,8] Magnetic resonance imaging (MRI) is the exam of choice to help in differential diagnosis and is especially useful for detecting tumors as small as 2 mm in size.[1,2] Surgical treatment is often described as the only effective and typically curative treatment, when the entire lesion is removed.[5]

CASE REPORT

We present a case of a 55-year-old woman (with no significant medical history) referred to orthopedic appointment due to pain in the 1st ray of the right hand, radiating to the forearm, with years of evolution and no improvement over time. The patient reported that the pain was worse when riding a motorcycle, especially during winter.

Non-steroidal anti-inflammatory drugs previously prescribed by her family doctor were ineffective in treating the pain. On physical examination, there was intense pain touching the nail of the right hand's 1st finger (without any evident deformity). An MRI was requested (suspecting a glomus tumor) which revealed a lesion in the subungual region of the 1st finger, consistent with the proposed diagnosis (Figures 1 and 2). The patient underwent surgical treatment with a lateral approach to the nail, where a rounded lesion was identified, dissected, isolated, and

removed without any complications during the surgical procedure (Figures 3, 4, and 5). Histopathological study revealed a lesion measuring 0.4×0.3×0.3 centimeters. Microscopically, it was a well-defined lesion composed of round cells (with poorly defined borders) organized in nests and trabeculae surrounding small thin-walled vessels in a myxoid stroma.

The surgical margin contained neoplastic cells that were positive for actin but negative for the presence of the S100 protein, confirming the diagnosis of a glomus tumor (solid subtype). The postoperative period was uneventful, and at 6 months postoperative, the patient was pain-free, with a very discreet surgical scar (Figure 6), and able to perform daily activities without any limitation, including riding a motorcycle without any complaints.

Figure 1: Axial T2-weighted MRI showing the lesion.

Figure 2: Sagittal T2-weighted MRI showing the lesion.

Figure 3: Lateral approach.

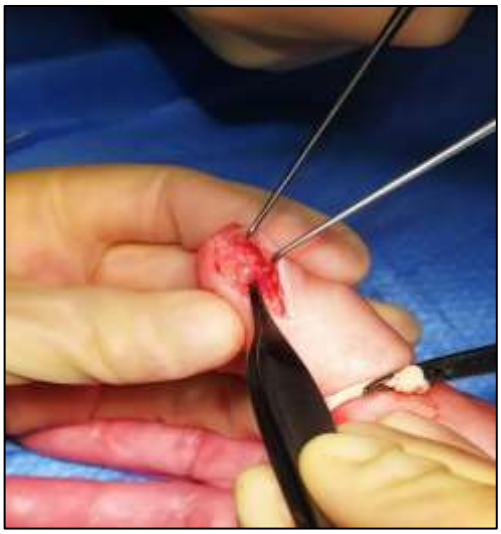

Figure 4: Macroscopic aspect of the tumor,

Figure 5: Bone sulcus made by the tumor.

Figure 6: Surgical scar 1year after surgery.

DISCUSSION

Patients can remain symptomatic for years before a diagnosis is made. Clinical experience and awareness of this pathology allow for much earlier diagnosis.[3] Early recognition and diagnosis are extremely important, as delayed diagnosis and recognition can exacerbate the patient's complaints.[10] The diagnosis is essentially clinical and besides the classic presentation, there are three tests that can be performed during the physical examination which are quite useful, which are Love's pin test, Hildreth's test, and the cold sensitivity test.[5,8,12] The Love's pin test involves using the tip of a clip to press the painful area, which will exacerbate the pain and sometimes cause patients to withdraw their hand.[13] Using a pressure tourniquet, the pain disappears (when this occurs, it is known as Hildreth's sign).[11] The cold sensitivity test involves pain exacerbation upon the application of cold water or alcohol. There are also another tests, such as the transillumination test, where a light is passed through the finger in a dark room, showing a reddish, opaque image, which helps estimate the lesion's size.[10]

The differential diagnosis includes various pathologies such as neuroma, schwannoma, foreign body, gout, osteoarthritis, subungual exostosis, hemangioma, hematoma, pyogenic granuloma, mucoid cyst, epidermoid cyst, nodular hidradenoma, keratoacanthoma, pigmented nevus, soft tissue chondroma, squamous cell carcinoma, melanoma.[1,11,14,15] Imaging exams available and useful in clinical practice include radiography (which may reveal cortical thinning or erosive bony changes), ultrasound with or without Doppler (which is a more cost-effective exam than MRI and can detect lesions equal to or greater than 2 mm), and MRI (which classically shows a hyperintense central point with a less intense surrounding halo).[5,16] This imaging modality shows a hyperintense lesion on T2 and low signal intensity on T1, which enhances with gadolinium administration, but unfortunately, it is not always available.[15] Both MRI and ultrasound can be dispensable for diagnosing this pathology if the clinical history and physical examination are very suggestive.[12,17] However, they are extremely valuable for preoperative planning, assessing the location, size, and shape, and being useful in choosing the surgical approach.[1,17,18] These tumors can be classified as solitary or multiple.[3] The first ones are usually encapsulated. Multiple tumors can be synchronous lesions, representing about 10% of cases, usually asymptomatic and generally located in areas other than the subungual region.[11,14] There are three histological types described, type I (glomangiomyoma), type II (solid type) the most frequent and type III (glomangioma).[14] The vast majority of these tumors are benign, with malignant ones (glomangiosarcoma) occurring in only 1% of cases, more frequently located in the lower limb.[3] Although other treatment options are described, such as sclerotherapy and laser, surgical treatment is the gold-standard treatment, usually effective and curative when the lesion is completely removed.[5,14,19] The subungual region is difficult to approach due to the unique nail anatomy. The best surgical approach combines the least risk of nail deformity and maximizes the surgeon's field of vision, with no consensus in the literature.[8]

Several surgical approaches are described, such as transungual-classic transungual, modified transungual (with nail plate preservation) lateral-lateral subperiosteal, modified lateral subperiosteal, Keyser-Litter, nail bed margin approach, shark-mouth flap approach, latero-ungual and volar.[2,13,20-27] The classic surgical approach is the transungual, allowing adequate exposure of the subungual region. Although some authors use this approach only for central subungual lesions.[25] Technically, this approach involves nail removal, matrix incision, and meticulous repair of matrix after tumor excision.[3] The major complication associated with this approach is the resulting nail deformities.

To avoid this complication, some authors have described this approach with minor modifications.[13,21,28] Roan et al compared 29 patients undergoing the classic approach with the approach with a single incision over the nail and matrix in 17 patients, reducing the recurrence rate from 13.8% to 0% and the nail injury rate from 24.1% to 0%.[13] Lee et al described the modified transungual approach in 34 patients, with nail preservation, where none developed nail deformity and only 2 patients had disease recurrence.[21] More recently, Bae et al, described a series of 17 cases where they performed an eponychium flap elevation, a good alternative for central subungual lesions, but limited in cases where patients have preoperative nail deformities.[28] The key technical point to avoid nail deformity is not injuring the nail matrix during tumor excision.[21] Thus, the main advantage of lateral approaches is the aesthetic result, achieved by the lower risk of nail deformity.[22] Some authors advocate the use of lateral approaches mainly for periungual lesions or lesions

located in the fingertip pulp, but also for more lateral subungual lesions, being inadequate for more centrally located lesions.[4,27] The Keyser-Litter approach uses an incision below the perionychium fold, identifying and retracting the distal phalanx ligament, then elevating the matrix to identify the tumor.[24] The lateral subperiosteal approach is very similar to the approach described by Keyser-Litter, but a flap of skin, nail, and matrix is obtained, avoiding retraction of the distal phalanx ligament.[22] It is mainly useful for proximal subungual lesions. Later, Garg et al described a modified lateral subperiosteal approach in 30 patients, avoiding injury to the nail matrix and the interosseous ligament, with no cases of recurrence or nail deformity.[23] The shark-mouth flap approach described in a series of 24 patients also presented excellent results in central subungual lesions, with only 1 case of recurrence 2 years after surgery.[26] The use of a microscope in recent years also seems to contribute to a lower incidence of iatrogenic nail bed injury, as well as a lower disease recurrence rate, due to more precise excision of the entire tumor.[27]

Recurrence of the disease after surgery is rare and is usually related to incomplete tumor resection.[4,5,10] However, recurrence of this pathology can be due to the presence of synchronous lesions. Generally, when it occurs early (i.e., in the weeks following surgery), it may be due to incomplete excision or undetected synchronous lesions, while late onset (years later) is associated with the development of a new lesion.[11,22] It is postulated that these late recurrences occur from satellite lesions already existing in the index surgery, but only detectable with the use of a microscope.[4]

CONCLUSION

Despite the good surgical exposure in the presented case, the lateral approach is likely associated with a higher recurrence rate due to less exposure of the lesion, especially in central lesions or in small subungual lesions. Thus, we believe that the choice of the surgical approach to be adopted should be individualized given the lesion's location and the surgeon's experience performing the procedure. The use of a microscope, whenever possible, also seems to offer advantages concerning surgical outcomes.

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