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Review Article

Glomangiomyoma Vs Fibrous Histiocytoma, Complex Differential Diagnosis in An Unusual Location: Case Report and Literature Review

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Abstract

Introduction: Glomus tumors account for 1.6% of soft tissue tumors, most frequently in the subungual region, and fibrohistiocytoma is a rare mesenchymal tumor that can affect the dermis and soft tissues of any part of the body.

Clinical case: A 65-year-old female patient with a palpable nodule in the right axillary region, which progressively increases in size. Physical examination revealed a palpable mass measuring 8 cm x 8 cm with irregular edges, a hard, mobile, and painful consistency. Surgical excision was performed, and immunohistochemical studies established a differential diagnosis between glomangiomyoma and fibrohistiocytoma.

Discussion: The clinical presentation of glomus tumors is characterized by a triad of localized tenderness, paroxysmal pain of great intensity, and hypersensitivity to cold. Immunohistochemically, they have smooth muscle characteristics, they are positive for smooth muscle actin markers and CD34, whereas fibrohistiocytoma clinically manifests as a slow-growing, painless, ovoid nodular mass, which is negative for CD34 and S100 protein; in view of the findings obtained, the definitive diagnosis is “glomus cell tumor: Glomangiomyoma”.

Conclusion: The definitive treatment for both pathologies is surgical excision and subsequent anatomopathological diagnosis, allowing the nature of the tumor to be determined by immunohistochemistry.

Kew Words: tumor; biopsy; fibrohistiocytoma; immunohistochemistry; glomus tumor

Introduction

The glomus apparatus is located in the reticular dermis and soft tissues. It is composed of a contractile neuromyoarterial receptor, whose function is to regulate blood flow in the arteriovenous anastomoses, thus controlling blood pressure and body temperature [1-3]. Glomus tumors are rare, mostly benign hamartomas, comprising 1.6% of all soft tissue tumors (2, 3). They are generally solitary in adults, being more common in the third and fourth decades of life, and only 10% are multiple, especially in children and adolescents. Their etiology is still unknown; however, they are associated with neurofibromatosis type 1. 60% of patients have a family history of glomus tumors, and in some cases, there was prior trauma at the lesion site. They originate in the glomus apparatus and are made up of cells similar to the modified smooth muscle cells of this structure. They are mainly located on the fingertips, with the most common location being the subungual region [1, 2]. Glomangiomyomas are composed primarily of a transition of glomus cells to smooth muscle-like spindle cells surrounding blood vessels [4]. They constitute 2–8% of glomus tumors [1, 2, 5], and less than 1% are malignant [6]. They are usually located in areas with a high density of glomus bodies, such as the subungual region, the deep dermis of the palms and soles, the wrists, and the forearm. However, there are reports of glomangiomyomas in anatomical regions where glomus bodies are absent, presumably due to the differentiation of pluripotent mesenchymal cells or smooth muscle cells [2]. On the other hand, benign fibrohistiocytoma, also called dermatofibroma, is a rare mesenchymal tumor that manifests in the dermis and soft tissues of any part of the body [7, 8]. It comprises approximately 3% of skin lesions and is usually associated with epithelial hyperplasia in response to an injury with tissue repair [9]. It primarily

affects women between the second and fourth decades of life [10]. Its etiology is unknown; however, most patients present lesions in sun-exposed areas with prior trauma, recurrent infections, or chronic irritation [8]. This case report presents a female patient with a differential diagnosis of glomangiomyoma vs. axillary fibrohistiocytoma, possibly being the first report in the literature with this location.

Clinical case A 65-year-old female patient was referred by Medical Oncology to the Surgery Department of the University Hospital of Caracas. She reported having a palpable nodule in the right axillary region since May 2022, which had been progressively increasing in size. She consulted a physician who ordered a skin and soft tissue ultrasound that showed a hypoechoic, Doppler-positive, space-occupying lesion (SOL) in the axilla, with increased lymphatic drainage measuring 5x4.5 cm in the right axillary region. An MRI showed isointense T1 images and hyperintense T2 images, showing cleavage planes with muscular structures in the axillary fossa measuring 5.4x4.2 cm (Figure 1). A core needle biopsy was subsequently performed, which revealed a glomangiomyoma in the right axillary region and is planned for surgical resolution. Physical examination revealed a palpable axillary mass measuring 8 cm x 8 cm with irregular borders, mobile, and painful the patient was taken to the operating table, where dissection, clamping, and ligation of the vascular pedicle of the axillary EOL were performed (Figure 2), followed by excision (Figure 3). The surgical findings were: 1) an intact axillary neurovascular bundle adhered to the axillary EOL; 2) an axillary EOL in the right axilla measuring 5.6 cm x 4 cm x 2.5 cm, adhered to deep planes. A biopsy was performed with millimeter sections and stained with hematoxylin and eosin, identifying a proliferation of spindle-shaped cells with a central ovoid nucleus with clear chromatin and eosinophilic cytoplasm, concluding that the tumor was a mesenchymal tumor: glomangiomyoma. An immunohistochemical study of the tissue material also revealed: AML: positive only in the blood vessel walls. Calponin: intense cytoplasmic positivity. S100 protein: nuclear and cytoplasmic positivity (focal). CD34: positive only in the blood vessel walls (Figure 4). The immunohistochemical diagnosis was made as "benign mesenchymal tumor: fibrohistiocytoma." The sample was sent to another center for a second pathological examination, which reported a mesenchymal tumor characterized by a proliferation of spindle cells with a central ovoid nucleus with chromatin clearing, occasional inconspicuous nucleoli, and enhancement of the nuclear contour with eosinophilic cytoplasm and ill-defined cellular boundaries; proliferation of thin-walled blood vessels with focal branching and no atypia, most of them hyperemic. The pathological diagnosis was: glomus cell tumor: glomangiomyoma. Immunohistochemistry was also performed, showing immunoreactions such as AML: positive, CD34: positive, desmin negative, and S100 protein negative. The conclusion was that the tumor was glomangiomyoma. The patient progressed satisfactorily during her hospital stay and was discharged.

Results

The clinical presentation of glomus tumors is characterized by a triad of localized tenderness, intense paroxysmal pain, and hypersensitivity to cold. They usually present as nodular lesions less than 1 cm in diameter, with an irregular, dark red surface. Ultrasonography reveals solid, hypoechoic masses with a positive Doppler response, findings consistent with our case [1, 2]. Magnetic resonance imaging is the most sensitive imaging study for establishing the diagnosis, showing a capsulated lesion that is isointense or slightly hyperintense on T1-weighted images and hyperintense on T2-weighted images when compared to the muscle, findings similar to those reported for the patient in the study. Immunohistochemical studies show smooth muscle characteristics, with positive results for smooth muscle actin (SMA) and CD34 markers, which are highly specific markers for the diagnosis of glomus tumor [11]. They are usually negative for desmin and S100 protein [3, 6]. These markers are consistent with the immunohistochemical markers from the patient case study, which are conclusive for the diagnosis of glomangiomyoma. Differential diagnosis should be established by immunohistochemistry due to the complexity of its pathological study with other vascular lesions such as intradermal nevus and hemangioma, and with soft tissue lesions such as leiomyomas, leiomyosarcomas, and rhabdomyosarcoma [6]. Definitive treatment is surgical excision; recurrence is a rare event that is usually associated with incomplete excision of the lesion [1, 2].

Conclusions

Benign fibrohistiocytoma clinically manifests as a slowly growing, painless, ovoid, nodular mass with no skin discoloration, usually measuring less than 1 cm in diameter. Symptoms are related to invasion of the surrounding anatomy, usually affecting the upper or lower limbs [8-10], characteristics that were mostly absent clinically in the present case. Ultrasonography reveals a hypoechoic area with regular borders, predominantly located in the dermis, mostly with limited blood flow [12]. MRI shows hypointense or isointense lesions on T1-weighted images, and T2-weighted images may be hypointense. Diagnosis is primarily based on histopathological and immunohistochemical analysis. Histologically, a proliferation of spindle-shaped fibrohistiocytic cells with a storiform or whirlpool pattern can be observed, and multinucleated giant cells and foam cells may be present. Immunohistochemical studies revealed positivity for CD68, vimentin, α-1-antitrypsin, and α-1-antichymotrypsin, and negativity for CD34, S100 protein, cytokeratin, and epithelial membrane antigen [13]. Differential diagnosis should include neurofibroma, leiomyosarcoma, and atypical forms of dermatofibroma. Definitive treatment is excision of the lesion (7), and recurrence is very rare, even in cases of incomplete excision. In this case, extensive resection is recommended because the patient may be a fibrohistiocytoma with a high risk of malignancy [12].

In the present case, as previously described, two immunohistochemical studies of the tissue material were performed, in which the relevant markers were AML, S100 protein, and CD34. The differential diagnoses for glomangiomyoma and fibrohistiocytoma are established. However, according to reports in the literature, the markers AML and CD34 are highly specific for glomus tumors [11]. Furthermore, in the case of fibrohistiocytomas, these are usually negative for CD34 and S100 protein [12], which allows a definitive diagnosis of axillary glomangiomyoma. It is interesting to highlight that this type of diagnosis requires several elements, such as those described, to reach a conclusion (diagnosis). It should be noted that the described location (axilla) was not found in the studies analyzed here, and may correspond to the first case report in this location.

Compliance with Ethical Standards

Informed Consent: The authors declared that written informed consent was obtained from the patient for the publication of this case report and the images included within.

Conflicts of Interest: The authors declared no conflicts of interest.

Use of Artificial Intelligence: The authors declared that no artificial intelligence (AI)-assisted technologies (such as large language models, chatbots, or image generators) were used in the preparation of this work.

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