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Extradigital Glomus Tumor: A Rare Case Report

Ekstradijital Glomus Tümörü: Nadir Bir Olgu Sunumu

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Özet

Amaç: Bu çalışmada, elin hipotenar bölgesinde yerleşimli nadir bir ekstradijital glomus tümörü (GT) olgusunun sunulması ve persistan üst ekstremite nodüler lezyonlarında GT'nin ayırıcı tanıda değerlendirilmesinin tanısal güçlükler ve klinik önem açısından vurgulanması amaçlanmıştır.

Olgu: Elli dokuz yaşında kadın hasta, sağ el hipotenar bölgesinde bir yıldır devam eden ve medikal tedaviye yanıt vermeyen şişlik ve hassasiyet şikayeti ile başvurdu. Fizik muayenede, ciltte renk değişikliği veya soğuk intoleransı olmaksızın belirgin hassasiyet gösteren palpabl subkutan nodüler lezyon saptandı. Hildreth testi pozitif olarak değerlendirildi. Direkt grafilerde kemik patolojisi izlenmedi. Doppler ultrasonografide hipervasküler hipoekoik kitle saptanırken, manyetik rezonans görüntülemede lezyonun T1 ağırlıklı sekanslarda hipointens, T2 ağırlıklı sekanslarda ise hiperintens özellik gösterdiği belirlendi. Lezyon, turnike kontrolü altında rejyonel anestezi ile cerrahi olarak eksize edildi. Histopatolojik ve immünohistokimyasal bulgular GT ile uyumlu bulundu. Cerrahi sonrası hastanın semptomları tamamen geriledi ve takip sürecinde nüks izlenmedi.

Sonuç: Ekstradijital GT'leri, renk değişikliği veya soğuk hassasiyeti gibi klasik klinik bulgular olmaksızın seyredebilir ve bu durum tanıyı güçleştirebilir. Konservatif tedaviye yanıt vermeyen persistan ağrı önemli bir klinik ipucu olabilir. GT'leri, üst ekstremitenin hem ağrılı hem de ağrısız nodüler lezyonlarının ayırıcı tanısında göz önünde bulundurulmalı; tam cerrahi eksizyon ise semptomların giderilmesi ve nüks riskinin azaltılması açısından etkili bir tedavi yöntemi olarak değerlendirilmelidir.

Anahtar kelimeler: Glomus tümörü; el; hipotenar bölge; ağrı

Abstract

Aim: To present a rare case of extradigital glomus tumor (GT) located in the hypothenar region of the hand and to highlight the diagnostic challenges and clinical importance of considering GT in persistent nodular lesions of the upper extremity.

Case: A 59-year-old female presented with a one-year history of swelling and tenderness in the hypothenar region of her right hand that was unresponsive to medical treatment. Physical examination revealed a palpable subcutaneous nodular lesion with marked tenderness, without skin discoloration or cold intolerance. The Hildreth test was positive. Plain radiographs demonstrated no bony pathology. Doppler ultrasonography showed a hypervascular hypoechoic mass, while magnetic resonance imaging revealed a lesion hypointense on T1-weighted and hyperintense on T2-weighted sequences. The lesion was surgically excised under regional anesthesia with tourniquet control. Histopathological and immunohistochemical findings were consistent with a GT. The patient's symptoms resolved completely following surgery, and no recurrence was observed during follow-up.

Conclusion: Extradigital GTs may lack classic clinical features such as discoloration or cold sensitivity, which can delay diagnosis. Persistent pain unresponsive to conservative treatment should raise suspicion. GTs should be considered in the differential diagnosis of both painful and painless nodular lesions of the upper extremities, and complete surgical excision remains the treatment of choice to achieve symptom relief and prevent recurrence.

Keywords: glomus tumor; hand; hypothenar region; pain

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INTRODUCTION

The glomus body is an arteriovenous capillary shunt structure surrounded by glomus cells, which are modified smooth muscle cells innervated by autonomic nerve fibers. It is also called the neuromyoarterial body. Its function is to regulate capillary diameter and thus blood flow to the skin in response to thermal or mechanical pressure changes. Glomus tumors (GTs) develop as a result of cell proliferation in the smooth muscle component of the glomus body. They are generally well-circumscribed, encapsulated, and benign tumors. Their color can vary from pearly white to light brown (1).

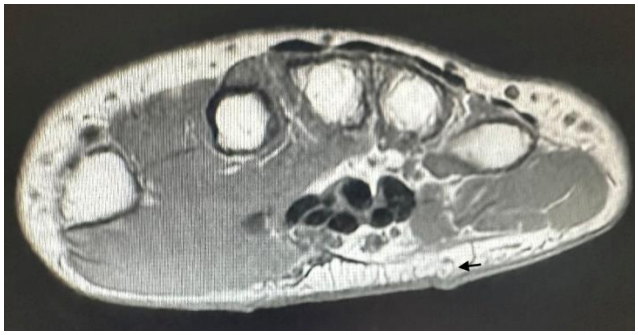
Extradigital GTs most often present as small, clinically inconspicuous, and non-palpable lesions; in some cases, pain may also be absent. This complicates the diagnostic process. In cases where pain is present, the clinical picture is often misinterpreted as idiopathic pain, and patients may be exposed to misdiagnosis and inappropriate treatment approaches for a long time (2).

CASE REPORT

A fifty-nine-year-old female patient, who had been experiencing swelling and tenderness on palpation in the hypothenar region of her right hand for approximately one year, presented to our outpatient clinic. She reported that her symptoms had not improved despite various medical treatments. Physical examination revealed a palpable, subcutaneous nodular lesion in the hypothenar region with marked tenderness on palpation. No skin discoloration was observed, and the patient did not report cold intolerance. The Hildreth test was positive, and the tenderness disappeared with tourniquet application.

Direct radiographic examination revealed no pathological findings related to bone structures. Doppler ultrasonography detected a hypoechoic mass with increased vascularity. Magnetic resonance imaging reported that the lesion showed hypointense signal characteristics on T1-weighted sequences and hyperintense signal characteristics on T2-weighted sequences (Figure 1).

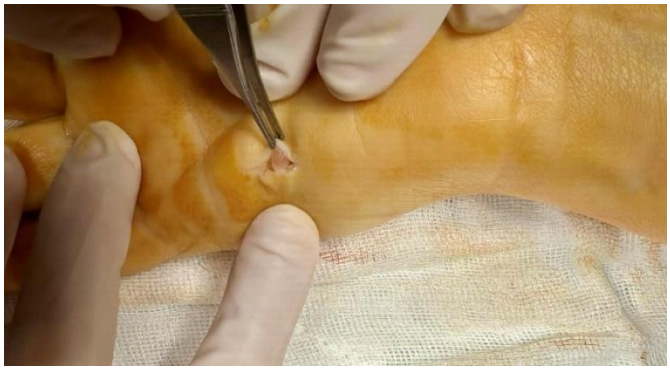
Figure 1: The appearance of glomus tumor on T1-weighted magnetic resonance imaging is indicated by an arrow.



Due to a known history of allergy to contrast agent, contrast-enhanced magnetic resonance imaging could not be performed. In light of these radiological findings, surgical excision was planned for the patient with a preliminary diagnosis of GT. It should be emphasized that imaging modalities do not possess diagnostic specificity; nevertheless, they play a crucial role in determining the precise localization of the lesion and defining its anatomical extent.

The surgical procedure was performed under regional block anesthesia with pneumatic tourniquet application. Following a longitudinal incision over the lesion, the mass was totally excised by carefully dissecting the deep tissues with blunt and sharp techniques (Figure 2).

Figure 2: The appearance of the glomus tumor exposed by dissection as viewed from the incision line.



The specimen was sent for histopathological and immunohistochemical examination. It was observed that the patient's symptoms completely resolved in the second postoperative week.

Pathological examination revealed a macroscopically smooth, white nodular lesion measuring 0.5×0.3×0.3cm. Microscopic evaluation showed

tumor cells with uniform morphology, round-oval nuclei, and eosinophilic cytoplasm, arranged in solid layers around thin-walled capillary structures (Figure 3 and 4).

Figure 3: The lesion is composed of sheets of glomus cells with focal myxoid stromal areas and intervening vascular structures (H&E ×20).

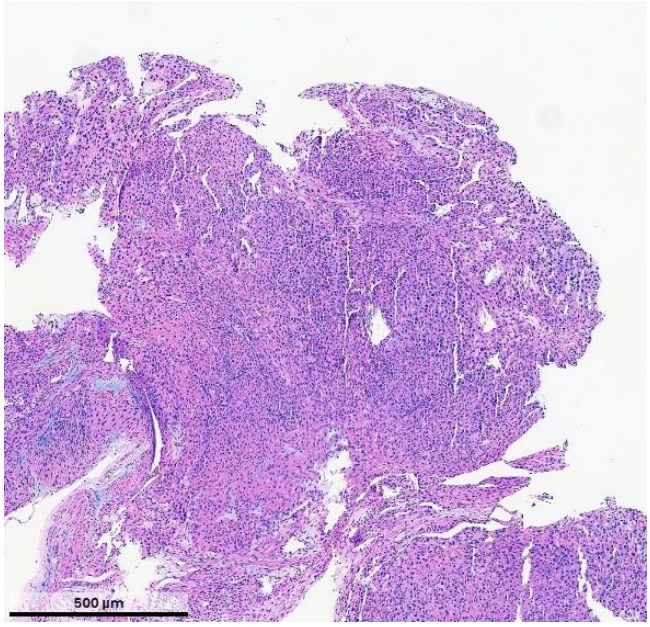
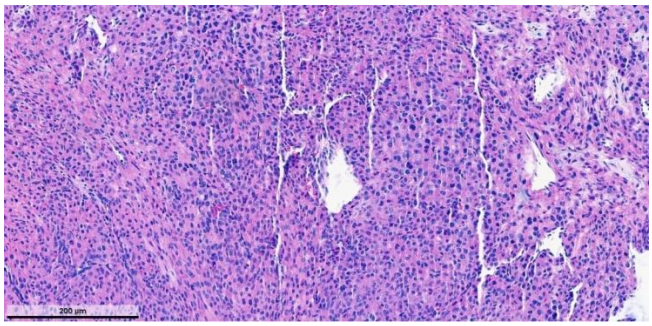


Figure 4: Cells with round nuclei and abundant eosinophilic cytoplasm with indistinct cell borders (H&E ×100).



No significant nuclear pleomorphism was observed in the cells, and mitotic activity was assessed at a low level. No necrosis was observed. Immunohistochemical analysis revealed smooth muscle actin (SMA) and CD34 positivity, and the Ki-67 proliferation index was reported to be below 2% (Figure 5 and 6).

Figure 5: Diffuse cytoplasmic staining with smooth muscle actin (DAB×10).

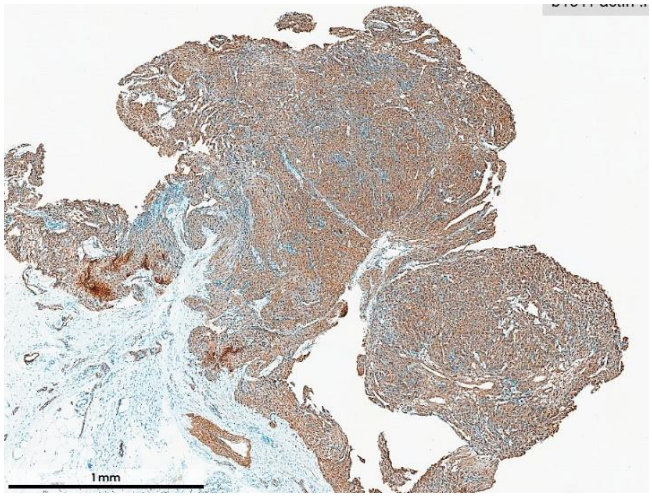
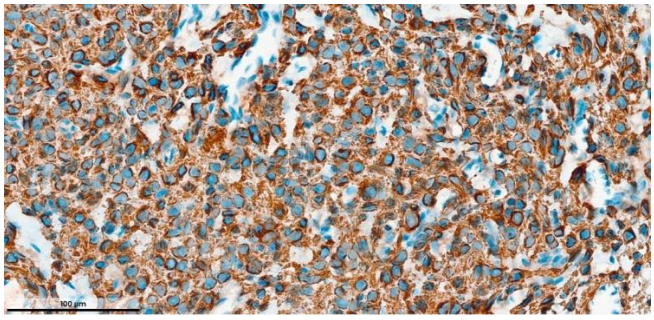


Figure 6: Cytoplasmic staining for smooth muscle actin (DAB×200).



Postoperatively, regular wound dressings were recommended at scheduled intervals. The patient was invited for follow-up two weeks after surgery for suture removal and evaluation of the histopathological report. At the end of the second postoperative week, the patient's pain and tenderness had completely resolved. The sutures were removed, scar massage was advised, and the patient was informed that she could return to her daily activities without restriction. Subsequently, outpatient follow-up visits were conducted at the 1st, 3rd, 6th, and 12th postoperative months. No recurrence of symptoms was reported during these follow-up evaluations.

DISCUSSION

GT is a rare, benign, mesenchymal neoplasm arising from the modified smooth muscle cells of the normal glomus body (3). Classic symptoms of solid GT include spontaneous paroxysmal pain, trigger point tenderness increased by local trauma, and sensitivity to cold. Classic clinical tests used in diagnosis include

the Love needle test (4), the cold sensitivity test, and the Hildreth test (5). The Hildreth test is a clinical evaluation method with high sensitivity and specificity.

In imaging methods, bone erosion in the affected phalanx can be seen in direct radiographs (6). High-resolution Doppler ultrasonography can show the size, location, and increased vascularity of the lesion (7). In magnetic resonance imaging, it has been typically reported that the lesion shows hypointense signal characteristics in T1-weighted sequences and hyperintense signals in T2-weighted sequences; and exhibits significant contrast enhancement after gadolinium administration (8).

The pathogenesis of pain, a common symptom in GTs, has not yet been fully elucidated. Various mechanisms are thought to contribute to the development of pain, including pressure sensitivity of the tumor capsule, mediators such as histamine and heparin released from mast cells, substance P and TRPV1 expression in glomus cells, and increased nerve fiber density within the tumor tissue (2,8). As in most cases, in the presented case, pain was the primary reason for the patient's visit to the healthcare facility.

Extradigital GTs are generally painless and typically present as painless cutaneous nodules or lesions with color change (9). In line with the presented case, cases of extradigital GTs characterized by palpation-induced pain, nocturnal pain, or progressive pain have also been reported in the literatüre (10-12). Pain may persist despite prolonged medical therapy and may not show significant improvement. Similarly, cases of extradigital GTs in which diagnosis could not be established despite four years of medical treatment have been documented (13). Clinical courses that do not conform to classic diagnostic criteria may complicate the diagnostic process and contribute to delays in diagnosis. The literature reports that only approximately 20% of extradigital GTs are correctly diagnosed by clinicians (14).

In extradigital GTs, color change on the lesion is commonly observed. Although studies in the literature have reported clinical presentations such as bluish nodules or erythematous plaques, cases of extradigital GTs without any associated color change, similar to the present case, have also been documented (10,12,13,15,16).

Cold intolerance, as also not observed in our case, has been reported at a lower rate in extradigital GTs compared with digital GTs (16). While pronounced sensitivity to cold is considered a key component of the classic clinical triad in subungual GTs, this finding may be milder or entirely absent in extradigital localizations. Therefore, the presence of cold intolerance (11,13) represents a supportive clinical feature, but its absence does not exclude the diagnosis of extradigital GT. Diagnostic evaluation based solely on this symptom may lead to misleading conclusions.

The differential diagnosis of extradigital GTs can be challenging due to nonspecific clinical findings and variable anatomical localization. Extradigital GTs may clinically and radiologically mimic various benign and malignant lesions, including hemangioma, leiomyoma, neurogenic tumors, epidermal inclusion cysts, and soft tissue sarcomas. Additionally, studies in the literature have included lesions such as foreign body granuloma, granular cell tumor, and dermatofibroma in the differential diagnosis (12,15,16). Therefore, a definitive diagnosis is most often established through histopathological examination and, when necessary, immunohistochemical evaluation.

In immunohistochemical evaluation, positivity for smooth muscle actin (SMA), vimentin, and CD34, a vascular endothelial cell marker, is commonly investigated in accordance with the literature (10,12). In our case, immunohistochemical analysis was performed using SMA and CD34, and this approach is consistent with the immunophenotypic characteristics described in previous reports. The diagnosis was primarily established on the basis of the characteristic histomorphological features observed on microscopic examination, while immunohistochemistry was employed for supportive purposes. The demonstration of the vascular component with CD34 and the confirmation of perivascular smooth muscle differentiation with SMA were considered sufficient to support the diagnosis in the presence of typical morphological findings. As no diagnostic uncertainty was encountered, an expanded immunohistochemical panel was not deemed necessary.

GT is histologically divided into three subtypes: solid type GT consisting mainly of glomus cells (70%), glomangioma with predominantly vascular structures (25%), and glomangiomyoma with prominent smooth muscle cells (5%) (17). Although GT is mostly benign, it can rarely exhibit malignant features. High mitotic activity, tumor diameter greater than 2 cm, presence of necrosis, and prominent nuclear pleomorphism are findings that can be evaluated in favor of malignancy (18).

The primary approach in the treatment of GT is complete surgical excision. The main goal of surgical

intervention is the complete elimination of symptoms, especially pain. In cases of inadequate resection, the risk of recurrence increases, with recurrence rates reported to range between 12% and 33% in the literature (19). Therefore, adequate surgical resection within anatomical limits is of critical importance. In the presented case report, no recurrence was observed during the one-year follow-up period. This finding suggests that the surgical intervention achieved adequate resection margins.

In conclusion, this case demonstrates that extradigital GTs may present without all components of the classic clinical triad and without significant color change. It also highlights that the most characteristic clinical symptoms of GTs, such as cold intolerance and pain, may not always be present. By emphasizing that extradigital GTs should be considered in the differential diagnosis in the presence of subcutaneous, long-standing, localized pain unresponsive to medical therapy, it contributes to clinical awareness. Furthermore, it provides a practical clinical contribution by showing that diagnosis can be established with typical histomorphological features and limited immunohistochemical evaluation, and that appropriate surgical excision may result in complete resolution of symptoms, thereby supporting both diagnostic and therapeutic approaches.

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