

## Case report



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## Glomic tumor of the forearm associated with homolateral carpal tunnel syndrome: a case report

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## Abstract

*Glomic tumors in the extra-digital localization are extremely rare and difficult to diagnose because of their rarity. The authors report a case of a glomus tumor of the posterior surface of the forearm associated with homolateral carpal tunnel syndrome, incidentally discovered after surgical excision in a 71-year-old patient.*

## Introduction

Glomus tumors are tumors developed from a cutaneous vascular formation first described in 1924 by Masson, called glomus [1]. This corresponds to an anastomotic network between an arteriole and a venule, producing a shunt devoted to thermoregulation; richly innervated smooth muscle fibers located around the vessels help regulate the degree of vascular opening. Glomic tumors correspond to an almost always benign neuro-myo-arterial hamartomatous proliferation. Although often diagnosed with delay, they are well known in the subungual region of the fingers. Glomic tumors with extra-digital localization are much rarer; a few cases have been reported in the limbs, spine, face or viscera. We describe a case of a glomus tumor on the dorsal side of the wrist.

## Patient and observation

He is a 64-year-old manual laborer with no medical history. For the past 5 years, the patient had had slight pain in the dorsal side of the wrist, with no notion of trauma or triggering factor. Two years ago, these pains worsened, which forced the patient to consult his doctor who had concluded with neuroma. Faced with the exacerbation of pain, he again consulted an IBN SINA hospital center. These pains were exacerbated for six months, occurring at the slightest contact, with significant discomfort to perform the activities of daily life with paraesthesias in the territory of the median nerve (described as tingling, electric shocks) associated with pain radiating to the forearm, elbow or shoulder, as well as discomfort at night due to night awakenings related to pain. Later, a decrease in grip strength. The interrogation found no hypersensitivity to cold but sensitivity to climatic variations. The inspection did not find any swelling or bluish coloration of the integuments. Palpation, limited by pain, found a subcutaneous tumor opposite the ulnar border of the ulnar carpal expander, infracentimetric and adherent to the deep plane. The pain, extreme, was localized

precisely at the level of the tumor with an increase in intracanal pressure variable depending on the position of the wrist (Figure 1). The patient did not describe sensory disturbances in the territory of the radial nerve. Complementary examinations previously prescribed were carried out during the consultation. Standard front and side wrist x-rays were normal. An ultrasound, limited by pain, showed without certainty a well limited homogeneous hypoechogenic formation of 5-6 mm in large diameter (Figure 2). The scanner was not performed. In the end, EMG showed a carpal tunnel syndrome. Due to the small size of this tumor, a surgical biopsy-excision was, therefore, performed under locoregional anesthesia. The branches of the ulnar nerve were intact. A plan of cleavage was easily found, the tumor being very limited; it was in contact with the ulnar bundle, partially covered by the ulnar extensor tendon of the carpus, without innuendo among the muscle masses (Figure 3), followed by a section of the anterior annular ligament of the carpus at the level of the palm of the hand. Histopathological examination of the whole part, Histology found a glomus tumor (actin +, desmin, CKAE1/AE3, CD34) (Figure 4, Figure 5, Figure 6). The postoperative operations were perfectly simple and the pains disappeared completely, immediately and after one year.

## Discussion

The usual sites of glomus tumors are most often found at the digital extremities, and descriptions of extra-digital locations are few. Glomus tumors can in fact develop everywhere where there are glomus, but also in tissues which do not contain them in the normal state [1-3] in patients aged on average from 20 to 40 years, with a sex ratio 1 [1,4]. A common point in most of these tumors is the generally long delay between the onset of symptoms and diagnosis, a certain number of patients had even been oriented to a psychiatric consultation. The diagnosis can be evoked in front of the clinical triad associating pain, a trigger zone and hypersensitivity to cold [5]. Often, the surface lesions have a slightly purplish hue. The presence of

paroxysmal pain with hypersensitivity to palpation and trigger zone should suggest the diagnosis, particularly in patients with trailing symptoms. On the radiological level, even if they do not provide diagnostic certainty, allow to make it suspect. Standard radiographs are normal apart from the rare bone forms [3-6] and subungual. On ultrasound images we generally find a well-defined, more or less encapsulated, homogeneous and hypoechoic lesion [7-9], although Abela *et al.* [2] find a hyperechoic lesion. MRI most often reveals an intermediate signal lesion in T1 which increases after Gadolinium injection and hyperintense in T2 [8-10]. In our case, the rounded, homogeneous hypoechoic mass, well limited with a Doppler spot on the periphery, hyperalgesic when the probe passes, not very compressible, unrelated to the underlying muscle fascias, continuity with a nervous branch is not found.

The diagnosis of certainty is based on a bundle of arguments because only histology is formal. Three types of glomus tumors exist: solid or solid glomus tumors, glomangioma and glomangiomyoma. The solid glomus tumors, the most frequent, are well limited and the glomus cells are organized in nests and in epithelioid spans around the vascular lights which are not very visible, erased by the confrontation of their endothelium [4,11]. This interstitial sector is inhabited by numerous nerve fibers. This form can pose problems of differential diagnosis with solid epithelial tumors, in particular with solid epithelial tumors, such as nodular hidradenoma or spiradenoma. A pseudoangiomatous naevocellular nevus can also be evoked [12]. Immunohistochemically, all types of glomus tumors express smooth muscle actin and have an abundant production of type IV collagen. The negativity of epithelial markers (EMA, CEA), desmin, CD34, cytokeratin and protein S100 correct the diagnosis [13]. The glomangioma is more readily located by hand and forearm. It is less painful and is associated with multiple clinical and/or familial forms. Histologically, the vascular component is evident with more or less dilated lumens giving the tumor the appearance of a cavernous hemangioma. Glomic cells form single or

bi-layered perivascular sleeves. The tumor is less well limited than the solid form. For glomangiomyoma is rare and occurs in both upper and lower extremities. It takes up the architecture of a solid glomus tumor or a glomangioma, but there is a smooth muscle component entangled in glomic proliferation, with all the forms of passage from the glomic epithelioid cell to the mature smooth muscle cell [12].

For digital forms, the diagnostic time varies from three to seven years depending on the studies [5,14,15]. Concerning extra-digital forms, for which the clinical picture is often incomplete, the diagnostic delay can go up to ten years [13]. Often for these extra-digital forms, the diagnosis is based on surgical excision and anatomopathological examination. Our patient presented two signs of the triad: the trigger zone causing dazzling pain and the palpable nodule 6 mm in diameter. In front of the small size, the localization of this tumor and before the long delay of the symptomatology, an excision was carried out in order to have the histological diagnosis and at the same time to carry out the curative treatment. We point out that this diagnosis of glomus tumor of the posterior aspect of the forearm has been ignored. Indeed extra-digital forms are very rare and can have various localizations [1,4,13]. In all cases, the treatment is surgical and is based on complete excision, which allows, as in our case, the total disappearance of the pain. Recurrences have been reported concerning glomus digital tumors [16], but the small number of cases of extra-digital forms makes it impossible to be able to give an estimate of the risk for them. Multiple [17,18] or malignant [19,20] forms have been described but are exceptional; the latter must be particularly evoked in front of a large deep tumor.

## Conclusion

Extra-digital glomus tumors of the limbs are very rare lesions especially associated with carpal tunnel syndrome are exceptional, the diagnosis of which must be evoked in the face of the clinical triad of paroxysmal pain, especially if there is a trigger zone, hypersensitivity to cold or a purplish coloration of the integuments opposite without confusion with the symptomatologies of carpal tunnel syndrome. Knowledge of the possibility of this clinical form can allow treatment and help regain indolence.

## Competing interests

The authors declare no competing interests.

## Authors' contributions

All the authors have read and agreed to the final manuscript.

## Figures

**Figure 1:** posterior aspect of the forearm showing the incision marking

**Figure 2:** surgical sight

**Figure 3:** macroscopic aspect of the lesion

**Figure 4:** HES (standard staining) with increasing magnification: single-stranded proliferation of small round or polygonal cells with abundant and poorly limited cytoplasm and round core without atypia, vented by vascular lumens. No mitosis. a: magnification 4

**Figure 5:** HES (standard staining) with increasing magnification: single-stranded proliferation of small round or polygonal cells with abundant and poorly limited cytoplasm and round core without atypia, vented by vascular lumens. No mitosis. b: magnification 10

**Figure 6:** HES (standard staining) with increasing magnification: single-stranded proliferation of

small round or polygonal cells with abundant and poorly limited cytoplasm and round core without atypia, vented by vascular lumens. No mitosis. c: magnification 20

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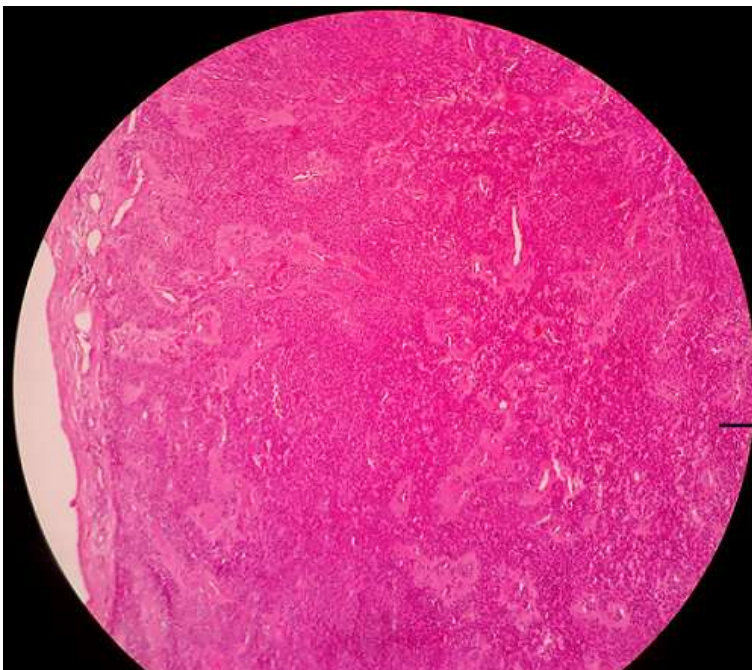
**Figure 1:** posterior aspect of the forearm showing the incision marking



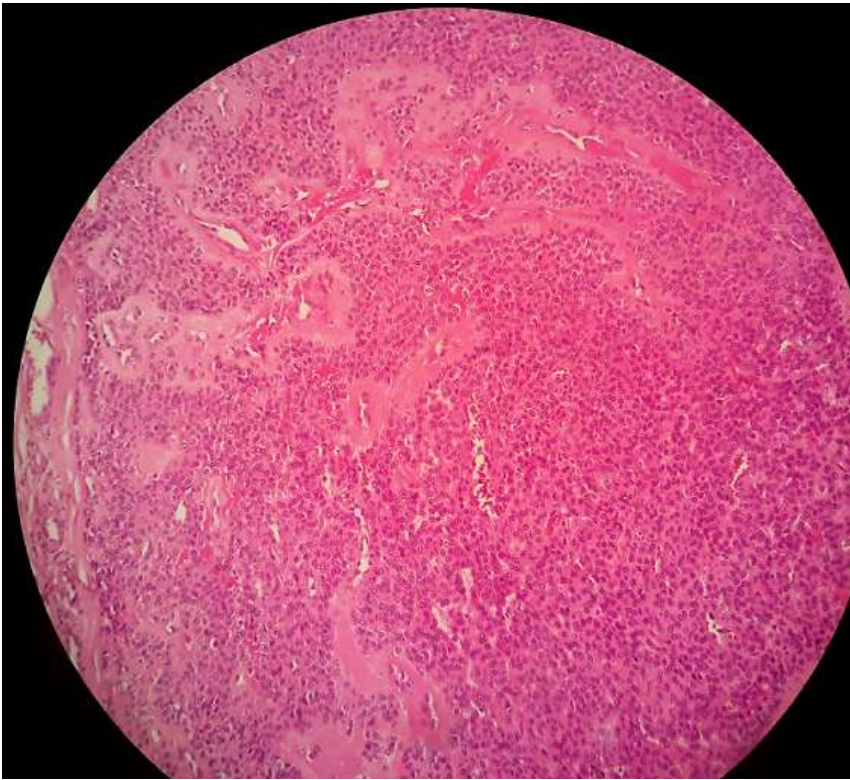
**Figure 2:** surgical sight



**Figure 3:** macroscopic aspect of the lesion

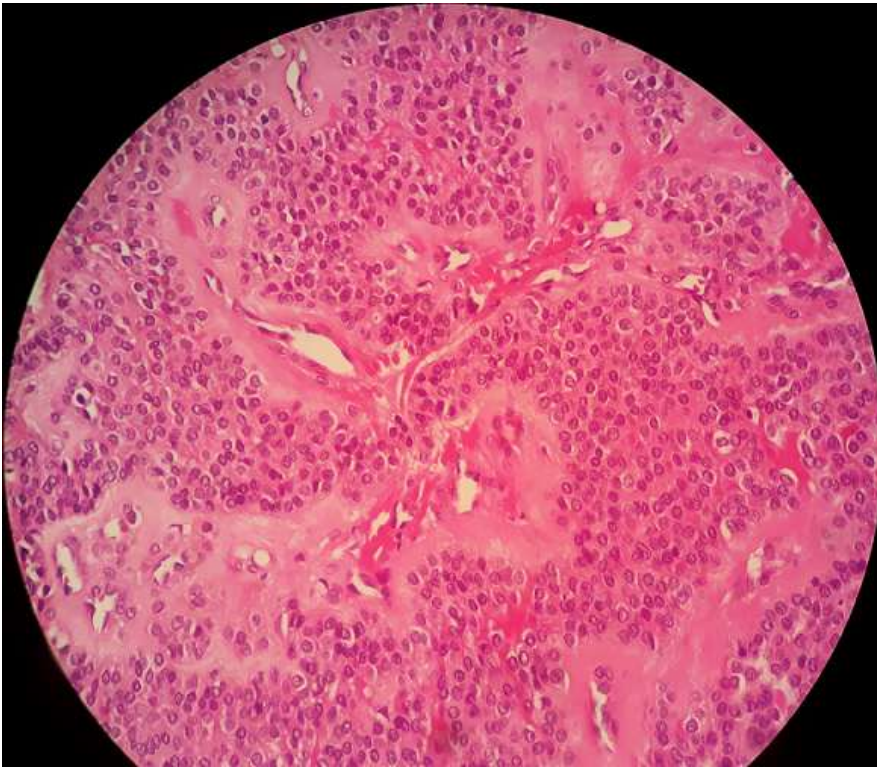


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