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BRIEF REPORT - OPEN ACCESS





Glomus Tumors of the Foot and Ankle: Case Series

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ABSTRACT

Glomus tumors are rare, slow-growing vascular tumors that are uncommon in the foot or ankle. This case series describes 2 glomus tumors located in the hallux, 1 in the foot, and 3 in the ankle. One case was misdiagnosed and initially mistreated at another facility, radiographs were not diagnostic in any of the cases, and magnetic resonance imaging was diagnostic in only 2 cases. Surgical treatment was ultimately successful in all cases. It is hoped that this series may help to raise awareness of glomus tumors among foot and ankle surgeons.

Keywords: Ankle, foot, glomus, neuromyoarterial receptor, tumor

INTRODUCTION

A glomus body is a neuromyoarterial receptor found in the reticular dermis, which regulates blood pressure and temperature by adjusting blood flow (1). Glomus tumors occurring in glomus bodies are rare, usually benign, neoplasms. Although glomus tumors are most commonly found in the subungual areas of the hand, they can originate in various locations on the body (1). Glomus tumors in the hands have female dominance, while glomus tumors in other locations have been reported to have male dominance (2, 3).

Most of the available information about foot and ankle glomus tumors is based on published case reports and series (2–5). In addition, in the largest case series about extradigital glomus tumors, only 4 tumors of 56 patients were located in the foot and ankle region (6). The low incidence of foot and ankle glomus tumors can lead to mis-diagnosis and mistreatment. Cases have been reported of patients who have been misdiagnosed with Morton neuroma or an ingrown toenail, and have even subsequently undergone above-the-knee amputation due to persistent pain (7–9). The objective of this case series is to raise the awareness of glomus tumors, particularly among foot and ankle surgeons with a report of 6 glomus tumors of the foot or ankle treated surgically over a 10-year period.

METHODS

A retrospective study was conducted by reviewing the cases of foot or ankle glomus tumors managed at a single hospital between January 2007 and December 2017. The clinical, radiological, and pathological details were obtained from digital hospital charts. Patients with a pathological diagnosis of glomus tumor were enrolled. A total of 55 glomus tumors were reviewed and 49 cases were excluded from the study: 47 with glomus tumors in locations other than the foot or ankle, and 2 patients with incomplete medical history data.

RESULTS

Six patients with a glomus tumor of the foot or ankle were included: 4 males and 2 females with an average age of 48.5 ± 12.2 years (range: 29–60 years) and a mean follow-up of 7.4 ± 4.1 years (range: 2.5-12 years) (Table 1). The main presenting symptom of all patients was pain that was described as spontaneous and excruciating. The mean duration of symptoms was 4 years (range: 0.5-11 years). Of the 6 patients, 1 had cold sensitivity and 4 had tenderness with palpation of the lesion. The triad of classic symptoms (pain, localized tenderness, and cold sensitivity) was only seen in Patient #2. Patient #5 had a history of wedge toenail resection for an ingrown toenail at another hospital, but this diagnosis could not be confirmed.

Radiography and magnetic resonance imaging (MRI) were used as diagnostic tools. All of the foot radiographs were normal. Of the MRI results, only 2 resulted in an accurate pre-diagnosis of a glomus tumor located in the distal phalanx of the hallux (Fig. 1). In the other 4 patients, there were 2 pre-diagnoses of pilomatricoma, 1 of an epidermoid cyst, and 1 of a sebaceous cyst.

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Table 1. Characteristics of the patients									
	Age (years)	Gender	Location of tumor	Follow-up after excision (years)	Duration of symptoms (years)	Tumor dimensions (cm)	aSMA	S-100	CD34
1	60	Male	Dorsum of foot	12	1	1.3x1x1	+	-	-
2	59	Female	Subungual region of $1^{\rm st}$ toe	11	3	0.6x0.6x0.3	+	+	+
3	51	Male	Anterior surface of ankle	10	2	1x1x0.6	+	+	-
4	39	Male	Medial surface of ankle	6	0.5	1.1x0.5x0.5	+	+	-
5	29	Female	Subungual region of $1^{\rm st}$ toe	3	11	0.7x0.3x0.3	-	+	-
6	53	Male	Anterior surface of ankle	2.5	3	1.6x1.2x0.7	+	+	+

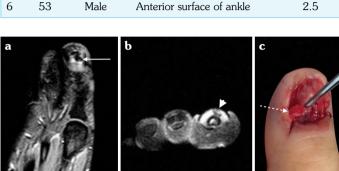


Figure 1. (a) Coronal and (b) axial section magnetic resonance images of a foot illustrating a glomus tumor in the hallux. (c) Intraoperative photograph of the excision of the hallux tumor

A transungual approach was preferred for surgical excision of subungual tumors. The nail bed was longitudinally incised after nail plate removal followed by tumor excision and repairing the nail bed with an absorbable suture. A marginal excision was used for locations other than the subungual area. The diagnosis of glomus tumor was confirmed histopathologically in all 6 cases. There was no malignant transformation.

Immunohistochemical staining was performed in all cases. All of the glomus tumors were positive for alpha-smooth muscle actin (α SMA) and HHF-35, (muscle actin-specific monoclonal antibody) and negative for CD31, cytokeratins, and S100. CD34 was positive in 33.3% (n=2) of the cases, with few showing focal cytokeratin expression.

There was no recurrence in the follow-up period. All of the preoperative symptoms were completely resolved in all 6 patients. One patient had a postoperative toenail deformity.

DISCUSSION

The results and findings of the present series of glomus tumors of the foot or ankle were largely similar to those presented in the literature. However, Trehan et al. (5) reported female predominance in foot glomus tumors, while we observed a male predominance in this series. There was also male predominance in the largest known series of extradigital glomus tumor cases, published by Schiefer et al. (6), but it does not include information about gender distribution in foot and ankle cases. The known age distribution of patients in foot and ankle glomus tumor case reports reported in the literature is 28–61 years (3–5, 8).

It has been established that a marginal excision may be sufficient in glomus tumor surgery (5). In this case series, the absence of recurrence after marginal excision in all of the cases supports the proposal that a marginal excision may be adequate. In addition to the transungual approach, other approaches have also been defined for subungual tumors, including a nail-sparing approach (10). The purpose of the nail-sparing approach is to protect the integrity of the nail bed; however, a delicately performed transungual approach can also preserve the integrity of the nail bed.

Case reports of foot and ankle glomus tumors demonstrate that a glomus tumor has sometimes been misdiagnosed as ingrown toenail (5, 8). A patient in the current study was also diagnosed with an ingrown toenail and had previously undergone surgical treatment. In this series, the triad of classic symptoms of a glomus tumor was only seen in Patient #2. Other reports have also indicated that the triad is not always present (5, 6, 11). Since misdiagnoses of conditions other than a subungual lesion in our patients did not change the choice of treatment at our hospital, they did not adversely affect the clinical results. We know that excisional biopsy (marginal excision) is sufficient for both the histopathological evaluation and treatment of soft tissue lesions smaller than 3 cm in size (12). However, a preoperative misdiagnosis of a subungual glomus tumor as an ingrown toenail can lead to mistreatment, such as the wedge resection of the nail bed performed for Patient #5. Preoperative misdiagnosis and mistreatment caused a delay of 11 years in the diagnosis of glomus tumor in that patient. A pre-diagnosis of a glomus tumor should be considered by foot and ankle surgeons in cases of long-lasting pain in the nail bed.

Van Geertruyden et al. (13) noted that radiography is not a significant diagnostic tool in glomus tumors and is primarily used to rule out other possible pathologies (13). All of the radiographs in our patients were not diagnostic for a glomus tumor; however, MRI in 2 of the 6 cases pointed to the correct diagnosis. Al-Qattan et al. (14) reported that MRI demonstrated high sensitivity but low specificity in the diagnosis of glomus tumors.

The results of immunohistochemical analysis of the patients in this study were similar to those of previous reports (15). Although there is no specific immunohistochemical analysis of a glomus tumor, characteristic αSMA and HHF-35 expression is observed in most cases due to the pericytic/perivascular phenotype. As in the present study, the variable expression of CD34 has also been reported in previous research (15, 16).

This study has several limitations. The first and foremost is the rarity and consequent low incidence of foot and ankle glomus tumors.

Therefore, the sample size of this study is too small to perform a statistical analysis. In addition, the clinical data were evaluated retrospectively. Despite the small number of patients, the study revealed a significant number of foot or ankle glomus tumors.

In conclusion, successful diagnosis of foot or ankle glomus tumors can be made using a histopathological examination based on clinical suspicion, given the low incidence rate and broad differential diagnosis. A misdiagnosis of glomus tumors and possible unnecessary surgical interventions can be prevented with greater awareness of glomus tumors among foot and ankle surgeons. Although a clinical examination may suggest a glomus tumor, an MRI is recommended to assist in confirming the diagnosis. Finally, it is noteworthy that successful clinical results can be obtained with marginal excision of the tumor.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

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