Contents lists available at ScienceDirect



International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



# Case report Rare glomus tumor formation following trauma to the first digit

Mohammad Saba<sup>a,b,\*</sup>, Austin Moser<sup>a,b</sup>, Joshua Rosenberg<sup>a,b</sup>, Jian-Hua Qiao<sup>b</sup>, Gary Chen<sup>b</sup>

<sup>a</sup> Ross University School of Medicine, 2300 SW 145TH AVE, SUITE 200, Miramar, FL 33027, USA
<sup>b</sup> California Hospital Medical Center, 1401 S GRAND AVE, Los Angeles, CA 90015, USA

ARTICLEIN	FO
-----------	----

# ABSTRACT

 Keywords:
 Introduction: Glomus tumors are rare and few cases are reported in the literature. They typically occur in females on the digits of the hands.

 Mass excision
 Case presentation: We report a case of a 30 year-old woman who presented with a mass that developed on the distal tip of her right thumb after traumatic injury. Magnetic resonance imaging (MRI) was conducted and mass resection was performed. Histopathology confirmed that the mass was a glomus tumor.

 Clinical discussion: Clinical presentations of glomus tumors are typically non-specific, mainly consisting of a small mass with chronic pain, with a lengthy time to diagnosis and potentially improper management. MRI is the preferred diagnostic step, followed by curative surgical excision and pathological confirmation.

 Conclusion: Glomus tumors can cause significant discomfort for patients, and clinicians should be aware of the rare diagnosis when treating painful masses on the extremities, as surgical excision is often curative.

## 1. Introduction

The anatomy and physiology of the glomus body are of interesting note. This cutaneous apparatus consists of various neuromyoarterial components that function in thermoregulation [1–3]. In addition to regular smooth muscle cells, specialized smooth muscles cells known as glomus cells, envelope the arteriovenous anastomoses seen in glomus bodies [1–4]. These structures are further surrounded by nerve endings and mast cells that play a role in pain perception as well [1,4]. Hyperplasia of the glomus body components leads to benign tumor formation. However, malignancies have also been rarely reported [5].

Glomus tumors account for approximately 2% of soft tissue tumors, and comprise 1–5% of tumors of the hand [3,4]. Two sub-classifications of glomus tumors exist: the 'solitary' form, which is more common than the 'multiple' form. Although glomus tumors can occur throughout the body, they have a predilection to the distal digits, especially the subungual area. This localized tendency has been attributed to as high as 75% of glomus tumors [6]. Females develop glomus tumors more than men; however, males more often exhibit extra-digital sites of tumor development [3].

Further categorization is based on the major histopathological component of the glomus body expressed in the tumor, which yields three distinct sub-types: the solid, glomangioma, and glomangiomyoma forms. The differentiating factor between these three sub-types is the presence of glomus cells, and varying degree of vascular and smooth muscle composition. These benign tumors tend to be painful and have a large gap in time, often multiple years, between symptom development and definitive diagnosis [1,8]. This is unfortunate, as minor surgical resection is often curative [1-2,5,9].

This case was reported in accordance with the SCARE 2020 guidelines [10].

## 2. Case presentation

Our patient was a 30 year-old female who presented to an outpatient clinic with a painful right first digit mass. Fifteen years ago, she suffered a crush injury to her right thumb, followed by subungual bleeding and bruising. Throughout the year following the crush injury a round solid mass slowly formed with progressive paroxysmal pain and tenderness at the exact site where the trauma occurred. She had been seen by multiple providers in the past without definitive treatment of her symptoms. The patient stated that her previous providers did not suspect a glomus tumor and would manage her complaints medically. They would focus on short courses of non-steroidal anti-inflammatory drugs, which did not fully address the patient's concerns. She denied changes in temperature sensation around the mass. The patient had no relevant family or

https://doi.org/10.1016/j.ijscr.2021.106324

Received 8 May 2021; Received in revised form 25 July 2021; Accepted 15 August 2021 Available online 19 August 2021

<sup>\*</sup> Corresponding author at: Ross University School of Medicine, 2300 SW 145TH AVE, SUITE 200, Miramar, FL 33027, USA.

*E-mail addresses:* mohammadsaba@mail.rossmed.edu (M. Saba), austinmoser@mail.rossmed.edu (A. Moser), joshuarosenberg@mail.rossmed.edu (J. Rosenberg), jian-hua.qiao@dignityhealth.org (J.-H. Qiao), drgary@aitmed.com (G. Chen).

<sup>2210-2612/© 2021</sup> The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

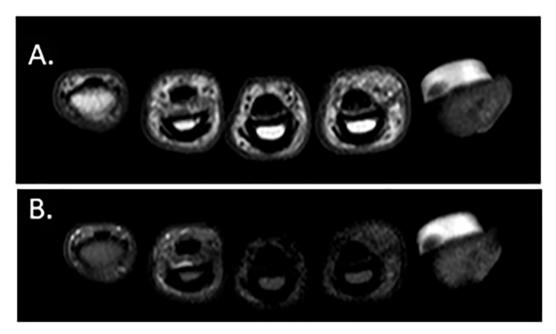


Fig. 1. MRI of patient's right five digits in the transverse plane. T1 (A.) & Proton Density (B.) weighted MRI of patient's right hand in the transverse plane showing a zone of hyper-intensity at the distal phalanx of first digit consistent with a soft-tissue mass.

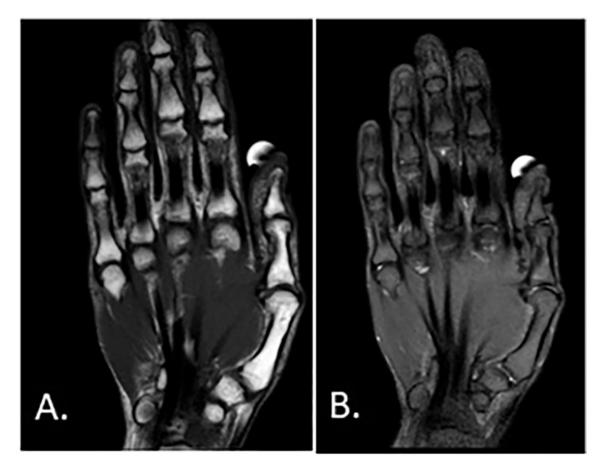


Fig. 2. MRI of patient's right hand in the coronal plane. T1 (A.) & Proton Density (B.) weighted MRI of patient's right hand in the coronal plane showing a zone of hyper-intensity at the distal phalanx of first digit consistent with a soft-tissue mass.

psychosocial history. She is a non-smoker who does not use recreational drugs, and rarely drinks alcohol.

A well-rounded 3mmx4mm bead-like lump without discoloration

was visualized on the distal phalanx of the right first digit, along with a slight nail deformity. The projecting mass was firm, slightly mobile, and exquisitely tender to palpation. Multiple views of T1-weighted and

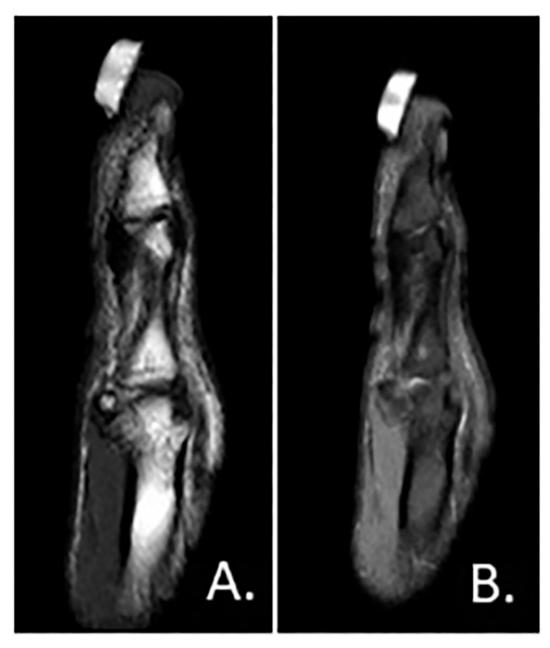


Fig. 3. MRI of patient's right first digit in the sagittal plane. T1 (A.) & Proton Density (B.) weighted MRI of patient's right first digit in the sagittal plane showing a zone of hyper-intensity at the distal phalanx of first digit consistent with a soft-tissue mass.

proton density weighted magnetic resonance imaging (MRI) showed a zone of hyper-intensity at the peri-ungual distal pulp of the right first digit [see Figs. 1–3]. The imaging demonstrated a soft tissue mass continuous with subcutaneous tissue of the pulp of the digit and not an extension of the distal phalanx bone.

We performed a mass excision under general anesthesia using a less extensive version of the lateral sub-periosteal approach. A tourniquet was placed over the right upper extremity and an additional local block was placed at the base of the digit, consisting of a 50:50 solution of 0.25% bupivacaine with 1% lidocaine. After exsanguination of the right thumb, a 1 cm incision was made at the tip of the mass and extended to the lateral edge of the nail bed, avoiding insult to the nail [see Fig. 4]. Careful dissection through the subcutaneous tissue and fascia was made until the mass was identified. A 5 mm  $\times$  4 mm white mass with a faint bluish hue was excised [see Fig. 4]. A 5 mm  $\times$  2 mm pedicle that was associated with the mass [see Fig. 4] was also carefully resected to ensure no margins remained. The distal portion of the digit nail fold was reconstructed and primary closure was obtained using 5–0 nonabsorbable sutures in a simple interrupted fashion. The wound site was further dressed with a petrolatum gauze dressing and additional gauze bandages wrapped with a soft roll. A short arm splint was applied to maintain the right hand in a functional position.

The resected mass and pedicle were sent for histopathological analysis, which showed patchy proliferation of glomus tumor cells in fibrotic stroma and perivascular regions [see Fig. 5]. Immunohistochemistry staining for a vascular tumor marker, friend leukemia integration-1,was performed as well [see Fig. 5]. Given the single lesion and the major composition of the tissue sample being glomus cells with limited vasculature and smooth muscle, the glomus tumor was deemed to be of solitary solid type.

The patient followed up in clinic two days post-procedure with a well-healing wound and no complications. She continued to adhere to post-intervention wound care and sutures were removed on postoperative day ten, with no signs of wound infection and full



Fig. 4. Image showing patient's right first digit after mass resection with a 1 cm incision (A.). Image showing excised mass superficially and the mass' excised pedicle below it (B.).

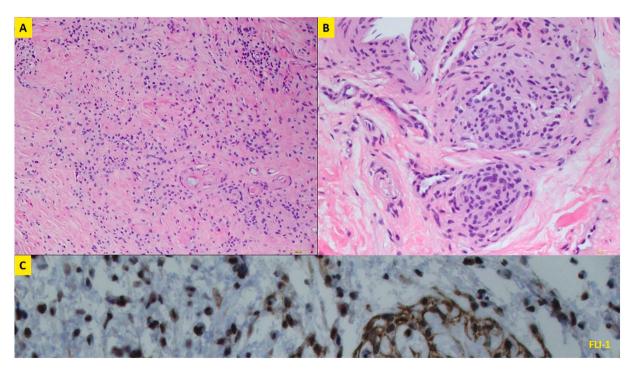


Fig. 5. Perivascular sheets and nodular proliferation of glomus tumor cells. The tumor cells are relatively uniform with moderate amount of pale cytoplasm and round to oval-shaped nuclei, H& E stain,  $200 \times$  (A.) and  $400 \times$  (B.). Nuclei of tumor cells are diffusely positive for FLI-1. IHC staining,  $400 \times$  (C.).

functionality intact.

# 3. Discussion

The first clinical descriptions of glomus tumors were detailed by

Woods nearly 200 years ago and histologically elaborated on by Masson nearly a century later [1]. Despite the historical documentation of this ailment, glomus tumors remain rare and difficult to diagnose. A major unresolved issue in the management of glomus tumors is the long duration from when symptoms present and when diagnosis takes place, with a mean of approximately 7 years has been noted for this time gap [4,8]. The case series presented by Tomaket al. showed how all 14 patients were misdiagnosed through the years and prescribed non-steroidal anti-inflammatory medications [8]. Clinicians must be cognizant when presented with a differential for glomus tumors, as most patients are suffering with chronic pain and surgical management is appropriate.

This time discrepancy prior to diagnosis may be a result of the broad differentials, including neuromas, hemangiomas, neuropathic pain, arthritis, and gouty inflammation [1,9]. Furthermore, a classic symptom-triad of point tenderness, paroxysmal pain, and cold sensitivity exists, however, complete presentation of the triad is uncommon [1-2,7,11]. As with our patient, she presented with pain and tenderness, but lacked cold sensitivity. The incomplete presence of the triad should be considered by clinicians in order to maintain glomus tumors as a possible diagnosis.

Given the lack of prompt diagnosis of glomus tumors, imaging is a key element of management. Ham et al. and Al-Qattan have expressed that MRI is an important adjunct to clinical history in diagnosing glomus tumors [12,13]. MRI is specifically useful in ruling out various differentials with a reported sensitivity of 90% and establishes important structural components for clinicians, which prevent symptom recurrence due to missed residual tumor mass during excision [9,12,13].

As explained by Vasisht et al. and Garg et al. the lateral subperiosteal approach to resecting sub-ungual glomus tumors has excellent cosmetic benefit as there is no need to manipulate the nail bed. All patients in both case series reported no nail deformity postoperatively, which is unavoidable with the trans-ungual approach [5,9]. Garg et al. reported no recurrence, while Vasisht et al. reported a recurrence rate of 15.7%, which was lower than the 20–30% recurrence rate reported in literature reviewing the trans-ungual method [5,9]. Vasisht et al. reported two similar cases in which the glomus tumors were located in the distal pulps of the digits, and both cases were successfully managed using the lateral sub-periosteal approach [5]. This surgical approach seems to be optimal to avoid a nail deformity when approaching periungual glomus tumors located in the pulp of the digit.

Histopathological analysis of excised tissue is critical to properly document and confirm glomus tumor diagnosis. Friend leukemia integration-1 (FLI-1) expression has a role in cell cycle regulation and tumor formation. Our use of immunohistochemical staining for FLI-1 protein expression compound Yang et al.'s conclusions, that this stain is useful in augmenting glomus tumor diagnosis [14,15]. Despite the rarity of malignant glomus tumor, clinicians should screen for suspicious neoplasms, and features such as size (larger than 2 cm), tumor depth, and increased mitotic activity are associated with metastasis [3]. Fortunately, our patient displayed uniform bland tumor cells with no signs concerning for malignancy.

#### 4. Conclusion

Glomus tumors are rare neoplasms that give clinicians difficulty in making prompt diagnoses, leading to increased patient morbidity before symptoms are resolved. We agree with other authors that MRI is an important adjunct to diagnosis and management of glomus tumors. We demonstrate that surgical excision is curative and the lateral subperiosteal approach remains the most cosmetically appealing surgical method. We also emphasize the importance of histopathology in diagnostic confirmation of glomus tumors, specifically staining with FLI-1. Documenting rare occurrences of glomus tumors in the literature brings attention to this condition and helps reduce the lengthy time to diagnosis that currently exists today.

## **Ethics** approval

Ethical approval has been exempted because this is a case report and no new techniques were carried out.

## Funding

There was no source of funding for this work.

## Authors' contributions

Mohammad Saba assisted with the procedure, formulated the first draft, and is the guarantor. Gary Chen performed the surgery and supervised the authorship. Austin Moser and Joshua Rosenburg edited the manuscript. Jian-Hua Qiao provided pathology.

# Guarantor

Mohammad Saba is The Guarantor.

# Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Provenance and peer review

Not commissioned, externally peer-reviewed.

#### Declaration of competing interest

None of the authors have a conflict of interest to declare.

#### References

- E. Samaniego, A. Crespo, A. Sanz, Key diagnostic features and treatment of subungual glomus tumor, Actas Dermosifiliogr. 100 (10) (2009) 875–882, https:// doi.org/10.1016/S1578-2190(09)70558-X.
- [2] V.M. Morey, B. Garg, P.P. Kotwal, Glomus tumours of the hand: review of literature, J. Clin. Orthop. Trauma 7 (4) (2016) 286–291, https://doi.org/10.1016/ j.jcot.2016.04.006.
- [3] Z. Gombos, P.J. Zhang, Glomus tumor, Arch. Pathol. Lab. Med. 132 (9) (2008) 1448–1452, https://doi.org/10.1043/1543-2165(2008)132[1448:GT]2.0.CO;2.
- [4] R.J. Rohrich, L.M. Hochstein, R.H. Millwee, Subungual glomus tumors: an algorithmic approach, Ann. Plast. Surg. 33 (3) (1994) 300–304, https://doi.org/ 10.1097/0000637-199409000-00011.
- [5] B. Vasisht, H.K. Watson, E. Joseph, G.T. Lionelli, Digital glomus tumors: a 29-year experience with a lateral subperiosteal approach, Plast. Reconstr. Surg. 114 (6) (2004) 1486–1489, https://doi.org/10.1097/01.PRS.0000138752.36175.D5.
- [6] S.S. Kale, V.K. Rao, M.L. Bentz, Glomus tumor of the index finger, J Craniofac Surg. 17 (4) (2006) 801–804, https://doi.org/10.1097/00001665-200607000-00037.
- [7] C. Sethu, A.U. Sethu, Glomus tumour, Ann. R. Coll. Surg. Engl. 98 (1) (2016) e1–e2, https://doi.org/10.1308/rcsann.2016.0005.
- [8] Akcay I. TomakY, N. Dabak, L. Eroglu, Subungual glomus tumours of the hand: diagnosis and treatment of 14 cases, Scand. J. Plast. Reconstr. Surg. Hand Surg. 37 (2) (2003) 121–124, https://doi.org/10.1080/02844310310005676.
- [9] B. Garg, M.V. Machhindra, V. Tiwari, V. Shankar, P. Kotwal, Nail-preserving modified lateral subperiosteal approach for subungual glomus tumour: a novel surgical approach, Musculoskelet. Surg. 100 (1) (2016) 43–48, https://doi.org/ 10.1007/s12306-015-0374-z.
- [10] for the SCARE, R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, The SCARE 2020 guideline criteria: updating consensus Surgical Case Report (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
- [11] S.D. Heys, J. Brittenden, P. Atkinson, O. Eremin, Glomus tumour: an analysis of 43 patients and review of the literature, Br. J. Surg. 79 (4) (1992) 345–347, https:// doi.org/10.1002/bjs.1800790423.
- [12] K.W. Ham, I.S. Yun, K.C. Tark, Glomus tumors: symptom variations and magnetic resonance imaging for diagnosis, Arch. Plast. Surg. 40 (4) (2013) 392, https://doi. org/10.5999/aps.2013.40.4.392.
- [13] M.M. Al-Qattan, A. Al-Namla, A. Al-Thunayan, F. Al-Subhi, A.F. El-Shayeb, Magnetic resonance imaging in the diagnosis of glomus tumours of the hand, J. Hand. Surg. Br. 30 (5) (2005) 535–540, https://doi.org/10.1016/j. jhsb.2005.06.009.
- [14] B. Miao, A.S. Bauer, K. Hufnagel, et al., The transcription factor FLI1 promotes cancer progression by affecting cell cycle regulation, Int. J. Cancer 147 (1) (2020) 189–201, https://doi.org/10.1002/ijc.32831.
- [15] Y. Yang, R.L. Wang, X.U. Jin, H.B. Yin, Expression of FLi-1, h-caldesmon and collagen IV in glomus tumor: a clinicopathologic study, J. Clin. Exp. Path. 33 (12) (2017) 1337–1341, https://doi.org/10.13315/j.cnki.cjcep.2017.12.010.