Radiology Case Reports 19 (2024) 6034–6038

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

Case Report

Glomus tumor: A rare differential diagnosis for subungual lesions ☆

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ARTICLE INFO

Article history:
Received 17 May 2024
Revised 20 August 2024
Accepted 21 August 2024

Keywords:
Glomus tumor
Subungual lesion
Painful nail
Finger sensitivity

ABSTRACT

Glomus tumors are rare, benign vascular hamartomas of the glomus apparatus with unknown etiology. They can arise from anywhere in the body. However, up to 90% of them are located in the subungual region of the fingers, as in the case of our patient. These tumors typically present with the classic triad of pain, cold sensitivity, and point tenderness. Characteristic US and MRI findings aid the clinical diagnosis; nevertheless, a histopathologic examination is confirmatory. There is a well-documented mean delay in diagnosis of around 7 years, due to the rarity, benignity, small size, and lack of proper knowledge about the condition. However, we reported a case with a delay in diagnosis that reached 40 years, which is much longer than what is documented in the literature. A high index of suspicion is required for early diagnosis and management of glomus tumors to relieve the patient's long-term suffering and prevent possible secondary nail deformities. The curative treatment of glomus tumor is complete surgical excision, which is crucial to prevent recurrence and relieve the patient's symptoms.

Introduction

Glomus tumors are distinct neoplasms that resemble the Sucquet-Hoyer canal of the normal glomus body and account for 1.6% of all soft tissue tumors. It is found in the subcutaneous tissue, which controls blood pressure and body temperature. These tumors typically have a solitary appearance, and range in color from deep blue to purple [1]. Glomus tumors most frequently develop on the hand, particularly in the subungual region, which has a high concentration of glomus bodies and comes with the classic triad of pain, cold sensitivity, and point tenderness. It can also develop in the genitourinary, gastrointestinal, liver, stomach, pancreas, and lungs. On

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2024.08.116

Table 1 – Differential diagnosis of subungual lesion.

Painful subungual lesions	Painless subungual lesions
■ Glomus Tumor	■ Melanoma
■ Traumatic	■ Eccrine Spiradenoma[b]
■ Hematoma	■ Ganglion Cyst[b]
■ Neuroma	
■ Leiomyoma	
■ Exostosis	
■ CRPS[a]	
■ Gouty Arthritis	
■ Calcinosis	

[a] CRPS, Complex Regional Pain Syndrome.
[b] Usually asymptomatic but can occasionally cause pain.

rare occasions, glomus tumors can undergo malignant transformation or malignant glomus tumors can arise de novo. Glomangiosarcomas are the name for malignant glomus tumors; they have a very low rate of metastasis and a high rate of local recurrence [2].

Those that are cancerous frequently exceed 2 centimetres in size, are profound, and are visceral. They are slow-growing and only detectable by MRI years after the onset of the symptoms. Histology showing capillaries lined with glomus cells confirms the diagnosis. Given that anti-inflammatory medications have little or no effect, complete excision is necessary for the prevention of recurrence and the only way to alleviate pain. The surgical approach is determined by the lesion's location [3]. Glomus tumors are rare benign neoplasms, and their incidence is likely underestimated due to a high rate of misdiagnosis. Therefore, improving our understanding of these lesions is essential for accurate epidemiological data and better disease management. In approximately 1% of the lesions are malignant. Therefore, early diagnosis is critical. Before a diagnosis of glomus tumors is made, patients may visit several different departments and doctors. Therefore, the patient and the healthcare system may experience physical, emotional, and financial burdens when ultimately, symptoms are often completely cured by the complete removal of a benign glomus tumor [4]. This case discusses a differential (Table 1) for a subungual lesion that is rare and interesting, namely a glomus tumor. We chose to write this case study to increase awareness of the condition, as the average time from the onset of symptoms to diagnosis is about 7 years and can last up to 15 years if diagnosed at all. During this time, patients may experience severe discomfort [5].

Case presentation

A 63-year-old male, medically free of chronic diseases presented to the clinic with a 40-year history of chronic pain at the tip of right middle finger which was a sequel of an old trauma. The pain is severe, constant, sharp, throbbing in nature, and comes in attacks. Severity was constant throughout the previous years. Attacks are typically triggered by minor trauma or exposure to cold. Once an attack starts, it lasts for 15 minutes approximately; before subsiding gradually and spontaneously. In previous years, the patient was unsure about his diagnosis and believed on his own that there was inflammation in his nail for an unknown reason. According to him, this self-belief kept him from seeking formal medical advice or getting any treatment for his condition. He didn't even try any painkillers or local anesthetics. Eventually, a friend who is a doctor advised him that his beliefs might be wrong and that something could be done to relieve his suffering. This advice prompted him to seek medical help. On examination, the right middle fingernail was swelled and bluish in color showing slight ulnar deviation with a normal range of motion (Fig. 1). There was exquisite tenderness on palpation over the nail. Muscular and neurological functions were preserved. The patient has no family history of a similar condition.

Initially, plain radiography revealed normal bony texture (Fig. 2). Further investigation with noncontrast magnetic resonance imaging (MRI) demonstrated a focal oval-shaped mass lesion at the dorsal aspect of the third distal phalanx deep into the nail bed. It measures 1.3 cm in the craniocaudal dimension and × 0.8 cm × 0.7 cm in the cross-sectional dimension. It has high T2 and low T1 signal intensities. Pressure erosion on the posterior cortex of the distal phalanx with remodeling is seen as well. The characteristic high signal central dot surrounded by a zone of lower signal intensity, known as the "central dot sign," is visible on the STIR sequence. The flexor and extensor tendons are intact (Fig. 3).

Surgical excision of the lesion was performed. The mass was well-circumscribed and removed. Histopathologic examination revealed groups of epithelioid cells surrounding dilated and thick-walled blood vessels embedded within the fibrous stroma. Tumor cells were uniform with indistinct borders, eosinophilic cytoplasm, and round nuclei with minimal mitotic activity (Fig. 4).

The patient was followed for 9 months after surgical excision and showed proper healing (Fig. 5). Moreover, no complications or recurrence of symptoms were noted.

Discussion

Glomus tumors are rare, small, benign neoplasms that originate from the glomus bodies in the reticular dermis. The neuromyoarterial cells that make up the glomus bodies contain a thin network of arteriovenous anastomosis to regulate body temperature and blood pressure by controlling the cutaneous blood flow. Glomus tumors, which are identified histologically as benign vascular hamartomas, can arise from anywhere in the body. However, up to 90% of them are located in the subungual region of the fingers due to their high concentration of glomus bodies. The most common site for the development of glomus tumors is the distal phalanx of the dominant hand's index finger, followed by the middle finger. According to several published studies, female patients with glomus tumors outnumbered male individuals. Synchronous multiple glomus tumors occur at a rate of 10% and may be found in other body parts. Rarely, these may manifest as autosomal-dominant familial diseases and are usually painless [6].

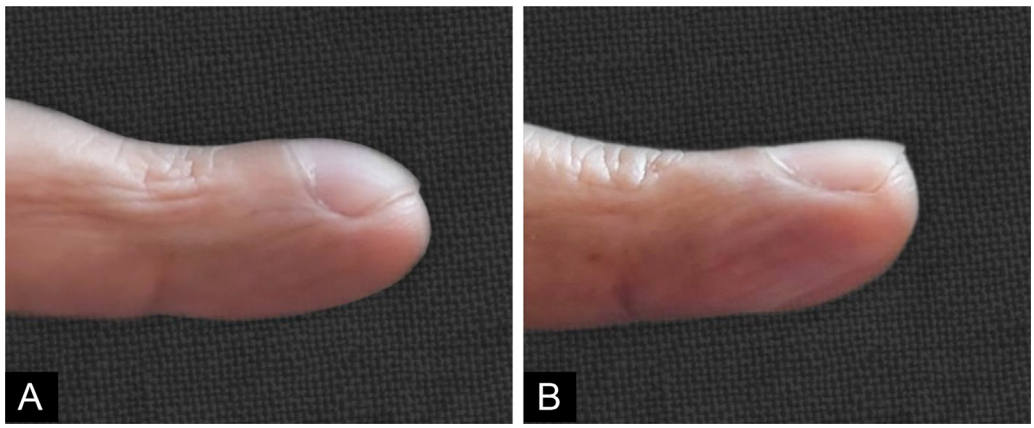

Fig. 1 – Comparison between the 2 middle fingers of (A) the right hand and (B) the left hand illustrates the swelling in the proximal nail fold and nail plate of the right one.

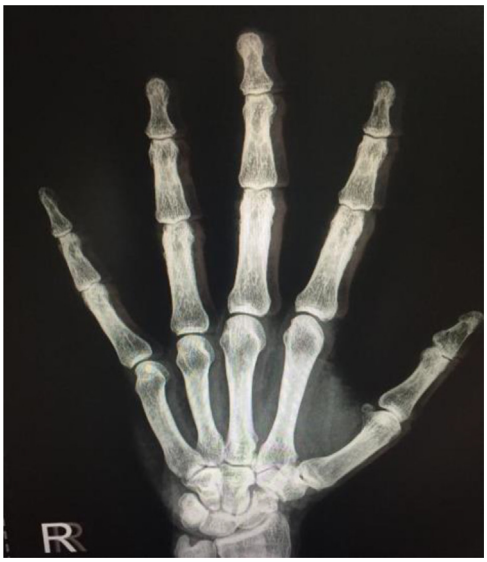

Fig. 2 – Normal plain X-ray of the right hand.

The diagnosis of glomus tumors is sometimes challenging because they are difficult to detect owing to their small size, slow growth rate, and late detection. Moreover, the following list of differential diagnoses should be kept in mind while evaluating a subungual lesion. This includes Benign solid tumors (such as glomus tumor, subungual exostosis, soft-tissue chondroma, keratoacanthoma, hemangioma, and lobular capillary hemangioma), along with benign cystic lesions (epidermal and mucoid cysts) and malignant tumors (squamous cell carcinoma and malignant melanoma), subungual angioleiomyoma, hyperplastic Pacinian corpuscles, blue nevi, blue rubber bleb nevus syndrome, eccrine spiradenoma, Kaposi sarcoma, Maffucci syndrome, neurilemmoma, and venous malformations [8]. The classic triad of symptoms that suggest the diagnosis of glomus tumor include severe pain, pinpoint tenderness, and cold sensitivity [8]. Several clinical tests such as the Love test (applying pressure to a suspected area using a pinhead, resulting in intense localized pain.), Hildreth test (reduces pain and tenderness by inducing transient ischemia with a tourniquet.), Cold test (provokes pain by applying cold water or an ice cube to the affected digit), and

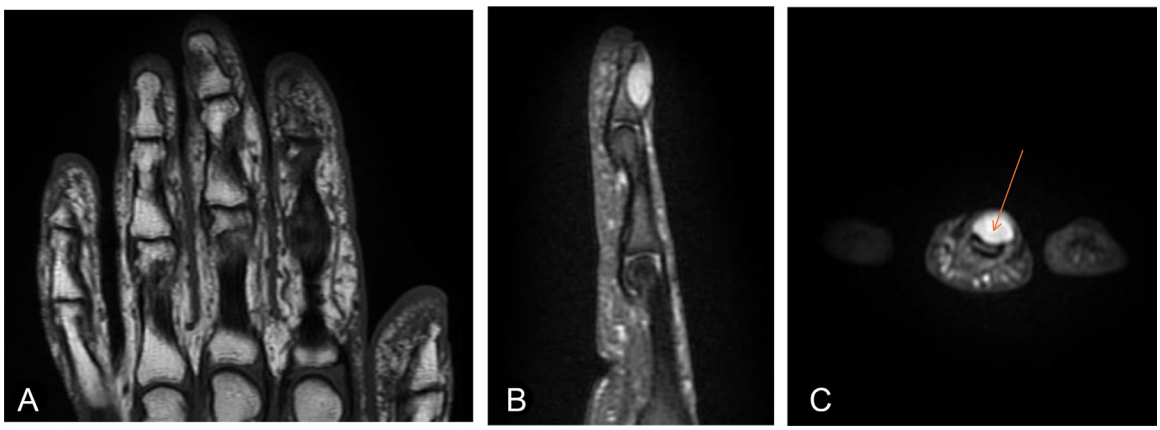

Fig. 3 – MRI appearance of the tumor: The mass has low signal intensity on T1 (A) and marked hyperintensity on T2 (B). The characteristic "central dot sign," is visible on the STIR sequence (C, orange arrow).

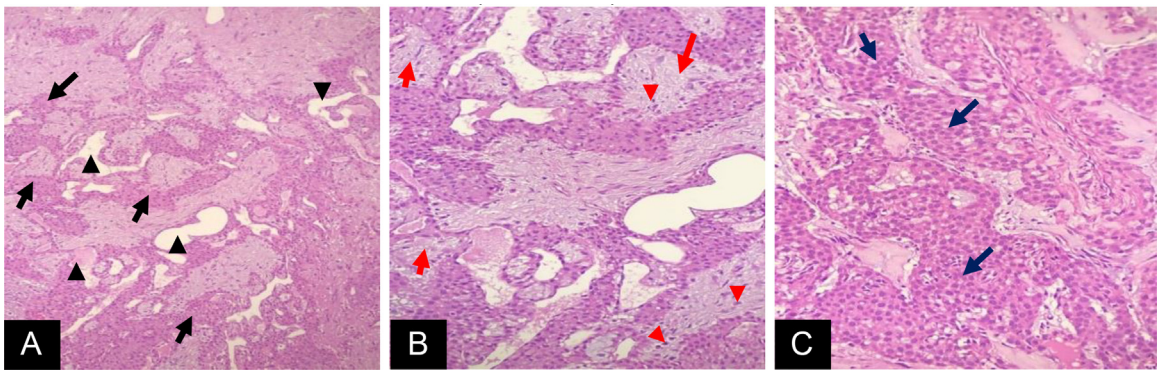

Fig. 4 – (A) Tumor consists of collars, nests, and groups of epithelioid cells (Black arrows) surrounding dilated and thick-walled blood vessels (Black arrowheads) embedded within fibrous stroma (H&E, 10x). (B) There are focal myxoid changes in the stroma (Red arrow) with mild mononuclear inflammatory infiltrate including mast cells (Red arrowheads) (H&E, 20x). (C) Tumor cells (Blue arrows) are uniform with indistinct borders, eosinophilic cytoplasm, round nuclei, and bland chromatin with minimal mitotic activity. No atypia, mitoses or necrosis were identified (H&E, 40x).

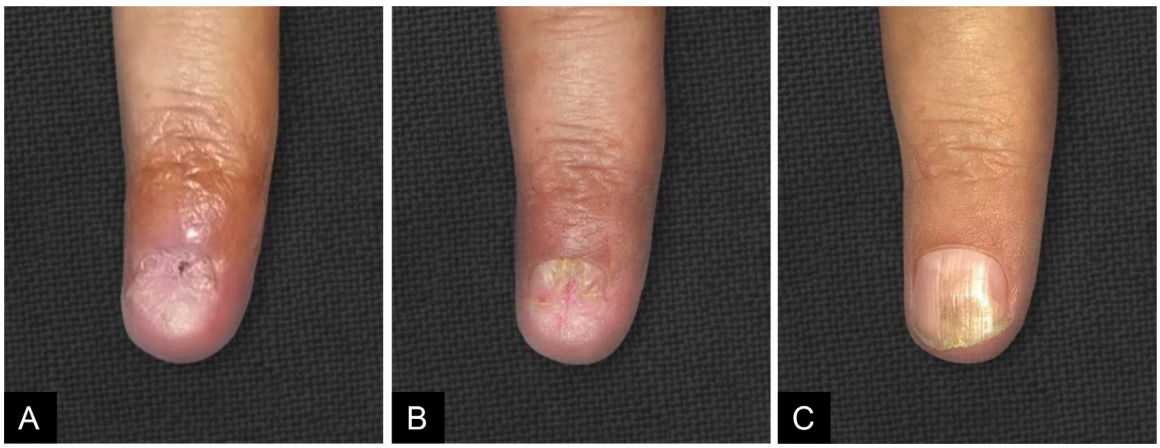

Fig. 5 – The patient showed uneventful healing and nail growth at (A) 2 months, (B) 3 months, and (C) 9 months.

Joseph-Posner test (uses ethyl alcohol spray to demonstrate cold-induced pain) have been described to provoke the symptoms of glomus tumor and aid in the diagnosis.

Radiological tests can detect a glomus tumor and high-resolution magnetic resonance imaging (HR-MRI) is currently considered the gold standard diagnostic investigation and is extremely effective in locating the tumor and delineating its extent preoperatively. In our case due to the patient's refusal to undergo contrast-enhanced imaging, we relied on a combination of clinical and radiological findings to diagnose a glomus tumor. Fortunately, the patient's history and examination findings were characteristic, facilitating this diagnosis. Typically, both subungual glomangiomas and cysts exhibit isointense to hypointense signals on T1-weighted images and hyperintense signals on T2-weighted images [13]. However, a high signal central dot surrounded by a zone of lower signal intensity, known as the "central dot sign," is indicative of a glomus tumor and was observed in our case. Additionally, in contrast-enhanced MRI studies, cysts demonstrate peripheral rim enhancement, whereas glomus tumors show hyperenhancement with increasing tumor blush on multiphasic acquisition [13]. Other suggestive radiological findings of glomus tumor include bone erosions on plain radiographs, and low signals within the hypoechogenic mass on Doppler US [7]. The confirmatory test to diagnose glomus tumor is based on the histopathology report after tumor resection [3]. The average duration of symptoms before establishing the correct diagnosis was reported by many authors to range from 18 months up to 15 years! Moreover, our patient has been suffering from fingertip pain for 40 years! Hence, early detection and prompt diagnosis of glomus tumors are of paramount importance because a delay in diagnosis might worsen the patient's symptoms, cause secondary nail deformities, and prolong their suffering [5,6,8–12].

The curative treatment of glomus tumor is complete surgical excision, which is crucial to prevent recurrence and relieve the patient's symptoms. The transungual and lateral subperiosteal approaches have been described with the latter being particularly more suitable for more proximal subungual lesions, as suggested by some studies. Additionally, it has been described that the lateral approach reduces the incidence of germinal matrix injury and postoperative nail deformities [7].

Possible postoperative complications of glomus tumor resection include recurrence and nail deformities. The recurrence rate is variably reported by authors to range between 4% and 50% [6]. It is advised that repeat imaging and re-exploration be seriously considered if symptoms persist for longer than 3 months following excision because of the high recurrence rate and the high occurrence of synchronous multiple lesions [7].

The relatively short follow-up period is the main limitation of this case report as it was limited to 9 months. For future studies, we recommend that longer follow-up periods of up to 10 years should be targeted to detect delayed recurrence or new lesions arising over the subsequent years [6].

Conclusion

Glomus tumors are rare, benign neoplasms that can be difficult to diagnose. A high index of suspicion is required for diagnosis and early detection. This would allow for timely management to be provided with subsequent reduction in patients morbidity and tumor malignant potential. This can be achieved by creating awareness among doctors and the general population to identify an early presentation. Suspicion should be raised if a patient presents with a triad of punctate-tenderness, severe paroxysmal pain, and cold sensitivity, particularly in the tips of the digits. Management requires radical surgical removal.

Patient consent

Written informed consent was obtained for experimentation with human subjects from the patient and for publication of their details.

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