Glomus Tumor - A Delayed Diagnosis, Yet Simple Cure: Case Reports

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Abstract

Glomus tumors are rare soft-tissue neoplasms that constitute 2% of all soft-tissue tumors in the extremities. They are painful lesions that can impair quality of life. Diagnosis is often delayed as they can easily be confused with more common pathologies. They are best diagnosed through MRI scan or ultrasonography of the concerned digit. We present two cases in this article, one of a subungual glomus tumor of the great toe (Hallux) and one of the index finger. Both tumors had a delay in diagnosis as this tumor cannot easily be diagnosed through simple radiographs. They showed classical clinical symptoms. Both were eventually diagnosed on MRI scan. Surgical excision is the gold standard in treatment of subungual glomus tumors and is also curative. We used a transungual approach for the hallux and a lateral subperiosteal approach for the index finger. In conclusion, glomus tumors are rare, benign tumors that commonly occur in the sub-ungual region. High clinical suspicion is important for clinical diagnosis. MRI scan and ultrasound can diagnose the tumor and yield information about local spread. Surgical excision is usually curative with approximately 10% chance of recurrence and rare chance of malignant transformation.

Keywords: Glomus Tumor, Sub-ungual, Love test, Transungual, Benign tumor, Hallux tumor, Glomus body

Introduction

lomus tumor is a benign mesenchymal neoplasm comprising less than 2% of soft tissue tumors. Glomus tumors, in general, show no sex predilection; however, solitary subungual lesions are more commonly observed in women and multiple lesions are slightly more common in men. While previously thought to occur predominantly in young adults (ages 20-40 years), they have been reported to be frequent in older adults (ages 40-70 years) and recently across all ages.^[1]

The tumor is composed of cells resembling modified smooth muscle cells of the normal glomus body. The glomus body, a thermo-regulator, is a specialized form of arteriovenous anastomosis localized in dermal and precoccygeal soft tissue. Glomus tumors are typi-

cally composed of 3 components: glomus cells, vasculature, and smooth muscle cells (Figure 1).

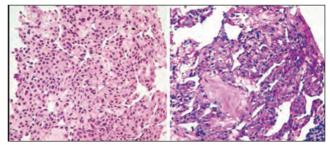


Figure 1: H&E and Periodic acid-Schiff stained sections of glomus tumor cell. The tumor cell has a sharply punched-out rounded nucleus with amphophilic or pale eosinophilic cytoplasm. The lacework of basement membrane around the tumor cells is accentuated with a periodic acid-Schiff stain.

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Glomus tumor usually occurs in areas rich in glomus bodies such as the subungual regions of digits or the deep dermis of the palm, wrist, forearm, and the foot. Patients typically present with subcutaneous or subungual nodules that may vary from red to purple to blue, depending on the depth of the lesion. Apart from these, solitary glomus tumors have been described in many locations like deep soft tissues, mediastinum, penis, nerve, bone, or soft viscera like stomach, small bowel and lungs.^[2]

These tumors are characteristically painful, often causing paroxysmal pain in response to temperature changes (especially cold) or pressure. If these tumors are stimulated by cold, they may precipitate a temperature change of the entire extremity, which is thought to be because of reflex vasodilatation.^[2]

While most bone and joint conditions do well with a simple radiograph, plain radiography may not be sensitive in a large percentage of cases, except where subtle signs of bony erosions can be picked up.^[3]

Ultrasonography, especially color-duplex ultrasonography, has a high detection rate and is known to have very few false-negative results. It has been documented to detect glomus tumors as small as 2 mm. [4,5,6]

MRI is a highly sensitive diagnostic tool in glomus tumors. It is not only a very sensitive test, but it may also be particularly useful for the detection of multiple tumors or in cases in which the diagnosis or the specific location of the lesion is in question.^[5,6] It can help delineate the relationship of subungual tumor to surrounding structures during pre-treatment planning and also in cases where there has been a recurrence.

An overwhelming majority of glomus tumors are benign and are cured by simple local excision as we will show through the two cases. A 10% risk of local recurrence is likely because of incomplete extirpation. Etiopathology of recurrence has evolved over time and we will discuss that later in this article. Just about 1% of glomus tumors are reported to be malignant.^[7]

The prognosis for patients with glomus tumors is excellent. Excision of painful lesions, most often, results in cure, with a low recurrence rate for solitary lesions. Less commonly, complications associated with surgery can include recurrence, nail deformity and infection. Prognosis of malignant glomus tumors is also good if they are treated with wide excision; otherwise, there is a risk of local recurrence.^[8,9]

Case 1

History

A 38-year-old female, otherwise healthy, presented to us with history of pain in the right great toe since five to six weeks. The symptoms were preceded by an uncertain history of blunt trauma to the same toe. Symptomatic treatment from a family doctor had given relief only for a while.

On examining further history, she experienced worsening of symptoms after morning walks which she took every day and also they were often at their worst when she woke up certain mornings.

There was no history of fever, sudden loss of weight, clear and perceived trauma, previous similar history or any swelling or discharge from the nail fold.

On examination, there was no swelling of the toe, the metatarso-phalangeal joint was not tender and there was no discharge, or change in the colour of skin. Capillary refill was normal. Range of movement at the IP and MTP joint were normal. There was no deformity at either of these.

There was grade III tenderness on deep palpation of the nail and nail bed, but no axial tenderness.

Simple radiographs and routine blood tests were requested. Radiographs didn't show a fracture or arthritis or any bony lesion. It was reported normal as well. Blood parameters including serum uric acid levels were within normal limits. Inflammatory markers were normal as well.^[10]

Rest from exercise, change of footwear, contrast bath and simple anti-inflammatory medications were prescribed.

The patient followed up with us after 2 months with worsening of symptoms though she felt momentary relief with previous treatment. A rheumatoid panel was requested, which turned out to be normal as well.

Considering a rare but possible diagnosis of a glomus tumor in the hallux, an MRI was requested. MRI showed, an intensely enhancing T1W hypointense lesion and STIR hyperintense areas in subungual region of 1st toe measuring 9mm*12mm and 2 mm in vertical thickness. No bony erosion was seen. There was high likelihood of it being a subungual glomus tumor more than anything else. No other noteworthy findings were observed in the rest of the foot (Figure 2a, 2b, 2c, 2d).

On re-examining the patient with a specific diagnosis in mind, we found that the morning pain was probably due to the use of air-conditioning at home. Pain increased when the patient washed her feet with cold water, which pointed towards cold sensitivity.



Figure 2a: MRI coronal section: Right great toe

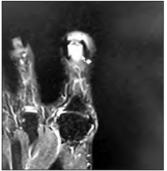


Figure 2b: MRI contrast enhanced coronal view



Figure 4a: Rubber tourniquet Figure 4b: Localisation of tumor with removal of nail plate and incisions along nail fold



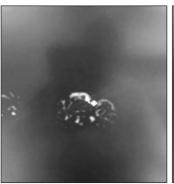


Figure 2c: MRI contrast enhanced axial view.

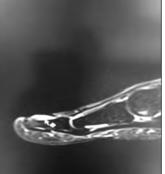


Figure 2d: MRI contrast enhanced saggital view



Figure 4c: Incision of nail bed with excision of tumor



Figure 4d: Suturing of nail fold and repositioning of trimmed nail plate



Figure 3: Clinical picture of great toe with dull discolouration

On removing nail polish we could see an area of dull blue discolouration, which was very tender to pinpoint palpation as popularised by Love *et al.*^[11] (Figure 3).

Management

All pre-operative investigations being normal, the patient underwent a transungual excision of the glomus tu-

mor under ring block anaesthesia with sedation.^[12] A single shot of IV cephalosporin was given.

Under thorough aseptic precautions, a slip of No. 7 sterile gloves was rolled onto the great toe and was used as a tourniquet. It was ensured that there was enough place for a haemostat to pass through.

The nail plate was removed gently by undermining it with a Freer's elevator. Care was taken not to injure the nail bed. The nail plate was gently released from the proximal nail fold. The nail bed was inspected and the glomus mass was visualised and palpated.

The proximal nail fold was incised and the incision extended on both sides of the nail fold for better exposure. A No. 15 blade was used to make longitudinal incision in the nail bed. The glomus mass was found right underneath the incision. Circumferential dissection of the mass was performed till cortex of distal phalanx was reached. Micro-instruments from the ophthalmology and ENT set were utilised. The tumor was sub-periosteally elevated and excised. Mild curettage of periosteum was done to prevent recurrence.

After saline lavage, the proximal germinal nail matrix was repaired with 5-0 monocryl and the nail fold was repaired with 4-0 monocryl. The nail plate

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Figure 5a: Glomus tumor specimen- Great toe

was trimmed, cleaned and then replaced so it would stay as a biological cover for the nail bed. It was secured with 4-0 monocryl (Figure 4a, 4b, 4c, 4d).

The sample was sent for histopathological diagnosis which confirmed that it was a glomus tumor (Figure 5a, 5b).

Nature of Material:

Biopsy from a lesion in the subungual region of the right great toe.

Gross Description:

Received three glistening whitish tissue bits aggregating to 0.8x0.6x0.2 cm. Submitted entirely.

Microscopic Description:

The biopsy reveals a lesion composed of haphazardly arranged small vessels. Some of these vessels show perivascular proliferation of homogenous round cells with round to ovoid nuclei arranged in multicellular layers.

These perivascular cells express SMA and CD34.

Impression:

Glomus tumour.

Figure 5b: Histopathology report

Post-operative Care

The patient was ambulatory as soon as she had recovered from sedation.

A simple sterile gauze and bandage dressing was done and the toe was strapped to the 2nd toe with roller bandage.

She was discharged the next morning after surgery after checking vascularity with standard instructions of care for the bandaged toe and wound.

Sutures were removed at 2 weeks from surgery. The nail fold incisions had healed well. In our patient we had to remove the nail plate at 4 weeks as the patient had discomfort from it. The nail bed underneath had healed very well.

Patient was allowed to wear protective foot wear and resume morning walks at 6 weeks from surgery when the nail bed had healed completely.

At the time of writing this article, there was no pain and tenderness and the nail had started growing but hadn't grown fully.

Case 2

History

A 47-year-old male, otherwise healthy, presented to us with a history of pain in the left index finger for six months. The symptoms were of sudden onset and progressive over 6 months. They were not preceded by any specific trauma. The patient had visited a few specialists and had persistently been given conservative symptomatic treatment.

On further history, the symptoms particularly worsened on holding cold glasses, and often, fine movements that involved the index finger were very painful. There was no history of fever, sudden loss of



Figure 6a: Clinical picture of index finger showing no discolouration or deformity of nail plate



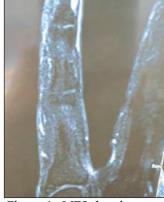


Figure 6b: MRI showing Contrast enhanced coronal view

Figure 6c: MRI showing contrast enhanced saggital view

weight, clear and perceived trauma, previous similar history or any swelling or discharge from the nail fold.

On examination, there was specific tenderness at 1 particular point in the nail bed and no other findings, just like the previous case. There was no deformity of the nail. Blood parameters and radiographs were normal.

An MRI was instantly advised considering a possibility of glomus tumor and it confirmed the clinical diagnosis (Figure 6a, 6b, 6c).

Management

All pre-operative and surgical protocols being same, we chose to use a modified lateral sub-periosteal approach for excision of this glomus tumor of the index finger, as it was pre-operatively located to be slightly lateral in the coronal views.^[13]

Under rubber tourniquet and thorough aseptic precautions, a high mid-lateral incision was taken from just proximal to the tip of the pulp of the finger till a few millimetres proximal to the nail fold.

A subperiosteal flap was raised to reach the subungual region without invading the germinal nail matrix or nail bed.

The dull blue glomus tumor was identified by bunt



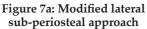




Figure 7b: Flap raised deep to nail bed and tumor localised



Figure 7c: Tumor circumferentially dissected and excised with capsule



Figure 7d: Void left by resected tumor with undersurface of nail and nail bed visible in flap

dissection and was slowly and carefully dissected off the soft tissue attachments and the underlying periosteum.

The tumor was excised en-masse and sent for histopathological confirmation.

The periosteum was curettaged and after thorough lavage the wound was closed with 4-0 monocryl in a single layer. A repair of the nail bed was not necessary as a sub-periosteal approach was used. The patients nail plate had also been conserved via this approach. (Figure 7a, 7b, 7c)



Figure 8: Glomus tumor specimen- Index finger

The excised tissue was sent for histopathological diagnosis and glomus tumor was confirmed (Figure 8).

Post-operative Care

The patient was ambulatory as soon as he had recovered from sedation.

A simple sterile gauze and bandage dressing was done and

the index finger was strapped to the middle finger with roller bandage.

He was discharged the next morning after confirming vascularity with standard instructions of care for the bandaged finger and wound.

Sutures were removed at 2 weeks from surgery. The incision had healed well.

Patient was allowed use of finger at 3 weeks from surgery for simple activities and at 6 weeks from surgery for all activities.

At the time of writing this article, there was no pain or tenderness. The patient was completely relieved of his symptoms, was using his finger for all routine activities and there was no deformity of the nail plate.

Discussion

Glomus tumors are rare, benign, painful neoplasms which arise from hyperplasia of one or more components of the glomus body. The glomus body is a contractile neuromyoarterial receptor that regulates peripheral blood flow and temperature.^[14]

The tumor is rare enough that in literature there are only few case series reported in detail.

Glomus tumors most commonly occur in the hand and specifically the subungual region of the distal phalanx (75%–90%) given the high concentration of glomus bodies in that area. Within the finger other reported locations include the finger pulp, digital nerve and distal phalangeal bone. They are rarely multifocal and rarely malignant.^[15]

In the largest published series of extra digital (nonfinger) glomus tumour accumulated over 20 years at the Mayo Clinic, only 2 out of 56 tumors were located in the foot.^[16] Foot glomus tumors are relatively rare due to lower concentration of glomus bodies in the foot, even the subungual region.^[17] Our case report had a female with a glomus tumor of the hallux and a male with a glomus tumor of the index finger. Both were subungual presentations.

Due to rarity of this benign tumor, patients with glomus tumors frequently face delay in diagnosis, more so in the foot, where they are commonly confused with Morton's neuroma, FHL injury, gout, stress fractures and most commonly, ingrown toe nails. In one series, 4 out of 11 patients had previously undergone surgical procedures assuming an ingrowing toe nail was the cause of the pain.^[18]

Our patient with the index finger glomus was diagnosed at nearly 6 months from the onset of symptoms after much symptomatic treatment and multiple investigations and one with the glomus of the hallux was di-

agnosed at nearly 3 months after onset of symptoms.

A clinical triad of pain, point tenderness and cold sensitivity are classical findings in patients with glomus tumors. Both our patients distinctly had them. All components of the clinical triad may not be present in each case, but pain is the commonly presenting characteristic of all lesions. Pain is excruciating, paroxysmal and usually bothers the patient over a long period of time. Trehan *et al.*^[18] in their series found nearly 100% patients to have all three symptoms in a classical triad. Van Geertruyden *et al.* have previously reported in their series that pain, point tenderness and cold hypersensitivity occurred in 80%, 100% and 63% of patients respectively.^[19]

Subungual lesions can present with a bluish hue beneath the nail bed and some present with deformity of nail growth. None of our patients had a deformity but our hallux patient had a mild bluish discolouration of the toe nail. Trehan *et al.* found discolouration in about 50% patients of their series.^[18]

The Love test, which is described as using a pin point or a tooth pick to elicit point tenderness over the tumor while eliciting no pain in an immediately adjacent pinpoint area is highly suggestive of glomus tumor. [11] Hildreth's sign, describes the disappearance of pain after the placement of a tourniquet on the extremity and relates to the vascular nature of the lesion. [20]

A number of imaging techniques have been used in diagnosis of glomus tumors including plain radiographs, arteriogram, thermogram, ultrasound and MRI.

Plain radiographic images of involved digits are often unremarkable as was the case in both our patients. ^[11] Drape *et al.* concluded that plain radiography may not be sensitive in a large proportion of cases. ^[3] Trehan *et al.* in their series of 11 patients had only one positive X-ray finding where the glomus tumor was uncommonly intra-osseus. Only in rare presentations, when there are radiolucent erosions along the phalangeal border, it can be detected on radiographs, but these may not be conclusive of glomus pathology. ^[21,22]

Ultrasonography, especially color-duplex ultrasonography, had a high detection rate and no false-negative results. Chiang, Fornage and their teams used high resolution ultrasonography and could detect glomus tumors as small as 2 mm.^[23,24,25] Ultrasound may be a particularly useful tool for exploring tumors involving the nail, with glomus tumors often visualized as small, solid, hypoechoic or isoechoic nodules with hypervascularity on Doppler.^[5] Cha *et al.* excised glo-

mus tumors from a number of patients with a solitary symptom of cold sensitivity and with only positive imaging findings on ultrasound (vs no findings on CT/MRI).^[26]

Both our patients had not undergone ultrasound imaging for diagnosis and authors have no direct experience in this matter.

MRI has been documented to increase accuracy of detection and diagnosis of glomus tumors especially in early stages with small lesions. The lesions uniquely appear on MRI as dark, well delineated masses on T1 weighted images and as bright, well delineated masses on T2 weighted images, with strong post-contrast enhancement.^[3,6,11]

In the largest case series evaluating the utility of MRI in diagnosis of glomus tumors, Al-Quattan *et al.* found that MRI sensitivity was 90%, specificity was 50%, positive predictive value was 97% and negative predictive value was 20%.^[27]

Mundada et al. showed that an MRI can be particularly helpful in delineating the relationship of subungual tumors to surrounding structures during pretreatment planning and also in cases where a recurrence may be suspected.^[28]

Both our patients were diagnosed conclusively on MRI scans.

Multiple treatment modalities have been recommended in literature including laser ablation and sclerotherapy but the gold standard in treatment of glomus tumors especially the solitary subungual ones is total surgical excision and it is curative.^[29]

Complete removal of tumor capsule not only relieves pain but also prevents recurrence.

Two main approaches for the surgical excision of glomus tumours are - transungual and lateral (lateroungual or laterodigital Keyser-Littler approach) although many alternatives have been described recently. [30]

We used the transungual approach in our hallux patient, as it provides for a better view of the whole subungual region and makes precise and complete excision possible. The transungual approach is also useful when the exact location is not known beforehand.

Previous reports have indicated a higher rate of the nail plate deformity with transungual approach when compared to lateral approaches.^[30] This may be because of the nail bed incongruities created during the excision or formation of adhesions between matrix and

PNF.^[30] Careful handling of tissues during surgery and meticulous repair of the defect created by excision can largely prevent this outcome.

Grover *et al.* in their series of 10 patients reported only two patients who developed a post-operative longitudinal ridging of the nail plate, owing largely to the initial large size of the lesions.^[31]

Nail plate repositioning, as in our patient, has been used as a method to prevent adhesions of the eponychium to the matrix. It also favours repair of the nail bed and reduces the pain occurring when dressings are changed, as the wound is kept covered. [30]

Li *et al.* and few other authors have recommended the use of autologous fat grafting to repair the defect in nail bed with good results.^[32]

Other than the standard transungual approach, many other nail bed sparing approaches have been described by various researchers.

We used a modified lateral sub periosteal approach for our index finger patient. While the approach spared the nail bed and afforded decent access to the tumor, it also bypassed some of the possible nail related complications that can arise from a direct transungual approach.

Glomus tumors are benign tumors arising from hyperplasia of one or more components of the glomus body. Various histologic variants may be identified on histopathology, including glomangiomas, glomangiomyoma, plexiform glomus tumor or glomus tumor with classic features in an intraosseous location. Glomus tumors are composed of glomus cells, small vessels, and smooth muscle cells in various combinations. According to the relative predominance of cell components, they can be classified as classic glomus tumor (75%), glomangioma (20%), or glomangiomyoma (5%). These pathologic variants do not have known implications in terms of recurrence, aggressive local behavior, or metastatic potential. However, histologic glomus tumor variants have been reported to have atypical clinical presentations and/or radiographic findings. For example, glomangiomyoma, which shows a predominance of smooth muscle cells, is infrequently associated with point tenderness, cold sensitivity, or subungual location. On the other hand, glomangioma, which shows a prominent vascular component, is frequently multifocal and associated with an atypical clinical presentation. Therefore, there can be an association between glomus tumor histologic variants and atypical clinical presentation, which can lead to a challenging pre-operative diagnosis.[18]

Removal of subungual glomus tumors has been reported to have recurrence rates of 2%-13% (highest reported at 50%) and nail bed deformity rates of 0- 19%. [33] Recurrence can be due to incomplete excision or development of a new lesion, with the probability of recurrence of glomus tumors in general being highest for subungual glomus tumors.

Gandhi *et al.* showed that most recurrences result from pre-existing synchronous satellite lesions not seen at the time of initial surgery and which become larger and symptomatic with time.^[34] They demonstrated this in five patients of recurring glomus tumours, all of whom developed lesions in different locations of the same digit.^[34] MRI may be useful in localising the recurrent lesions.

Conclusion

Glomus tumor is a rare benign tumor commonly found in the subungual regions of the fingers and rarer in the toes. A high clinical suspicion is recommended since there is usually no preceding history. One or more features of the classical triad may be present. MRI is the most sensitive and detailed radiological diagnostic tool if cost is not a problem. Surgical excision is gold standard with few chances of recurrence and nail plate deformities.

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