

Subungual Glomangiomyoma of the Distal Phalanx: A Case Report

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AUTHORS' CONTRIBUTIONS

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DISCLOSURES

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No

HUMAN AND ANIMAL RIGHTS

This review does not include experiments on human or animal subjects

ABSTRACT

Glomus tumors typically affect the soft tissues in the hands and fingers in young to middle-aged adults, and glomangiomyoma is the least common subtype of glomus tumor. These tumors often present as painful nodules, characteristically with cold sensitivity. This report describes a 42-year-old female who presented with 18 years of pain and cold sensitivity in her right distal ring finger. Work up with plain radiographs, CT, and MRI eventually led to likely diagnosis of glomus tumor. After excision, histopathological analysis revealed the rare glomangiomyoma subtype. This report highlights the typical clinical history, the multimodality imaging appearance, and the histopathologic features of this rare and interesting entity.

CASE REPORT

BACKGROUND

Glomus tumor of the finger is an interesting entity with typical clinical features such as cold sensitivity, that is often inconspicuous or demonstrates a nonspecific appearance on initial plain film imaging. This case emphasizes the importance of detecting subtle radiographic signs such as cortical bone remodeling, prompting cross sectional imaging evaluation. We present a case of the rarest histologic subtype of glomus tumor, glomangiomyoma, where literature describing both the radiologic and histopathologic diagnostic process is minimal.

CASE REPORT

Patient presentation

A 42-year-old female presented for evaluation of right ring finger pain. She has had worsening pain in her right distal ring finger for 18 years with no known trauma to the area. Tylenol provided no relief of pain, and she noted her pain was

exacerbated by putting her hand in cold water. Physical exam revealed tenderness over the right distal ring finger, with slight discoloration noted under the nail.

Imaging findings

Right hand radiographs (Figure 1) were initially interpreted as negative, but demonstrated subtle, smooth cortical indentation on the dorsal aspect of the 4th finger distal phalanx, suggestive of a subungual soft tissue mass exerting chronic mass effect and remodeling the bone.

Right hand CT (Figure 2) confirmed smooth cortical indentation on the dorsal aspect of the 4th distal phalanx, again suggestive of an adjacent soft tissue mass (Figure 2). MRI was recommended.

Right hand MRI (Figure 3) demonstrated a T1 hypointense, T2 hyperintense, solidly enhancing mass in the subungual space

of the distal phalanx compatible with a glomus tumor, measuring 10 x 3 x 10 mm. Osseous remodeling in the adjacent distal phalanx was again visualized as on prior CT and radiographs.

Management

The lesion was surgically removed piecemeal, noted to be a friable mass located in the distal 4th finger soft tissues. The mass was approximately 4mm in diameter.

Pathology

H&E demonstrated multiple round, homogenous cells with round/ovoid nuclei suggestive of glomus cells (Figure 4). Immunohistochemical tests were positive for desmin and smooth muscle actin (SMA), both markers of myogenic differentiation indicative of the glomangiomyoma subtype (Figure 4). SOX10 (seen in tumors of neural crest origin) and p40 (seen in tumors of squamous cell differentiation) stains were negative.

Follow-up

No recurrence of tumor was noted in follow up visits, though localized pain remained in the area.

DISCUSSION

Etiology & demographics

Glomus tumor is a type of soft tissue tumor, which occurs primarily in the hands and feet due to their high predominance of glomus bodies [1]. A glomus body is a neuromyoarterial plexus composed of chemoreceptive glomus cells, smooth muscle cells, and blood vessels which form specialized arteriovenous anastomoses [2]. Glomus bodies play a role in sensing and regulating local temperature and circulation [3]. Glomus tumors are most often found in the hands, with 75% of cases involving the subungual region of the finger and otherwise affecting the deep dermal layers of the wrist and palm [4]. These tumors most frequently occur in adults aged 20 to 40 with equal distribution between males and females, though subungual tumors are more common in females [4,5]. They are uncommon, and account for 1–5% of all soft tissue tumors affecting the hand [6]. 90% of glomus tumors develop as a solitary sporadic lesion, but 10% of cases are inherited and present in multiples secondary to glomulin gene (GLMN) mutations [7,8]. There are three subtypes of glomus tumors, classified by their proportion of glomus cells, blood vessels, and smooth muscle cells: Solid subtype, which are the most common (75%), consisting mainly of glomus cells; Glomangioma (20%) consisting mainly of blood vessels; Glomangiomyoma (5%) consisting mainly of smooth muscle cells [6,9].

Clinical & imaging findings

Clinically, glomus tumors typically present as a small nodule at the distal aspect of the finger, commonly subungual, and are most often tender with pain out of proportion to tumor size, and often sensitive to cold temperatures [9]. A sensitive

and specific test for glomus tumor is Hildreth's sign, where there is pain relief upon inflation of a sphygmomanometer and sudden worsening of pain when pressure is released [10].

Radiograph: Glomus tumors arising from soft tissues are usually imperceptible on plain radiographs. However, as many develop in the subungual region, limited space leads the tumor to expand into deeper tissues and become apposed to adjacent bone, where it may cause cortical remodeling and scalloping [6]. Though rare, glomus tumors may occur in the intraosseous location, and in that case would appear as a radiolucent lesion [11].

Ultrasound (US): Glomus tumors appear as a hypoechoic nodule with vascularity on doppler images [4].

MRI: Glomus tumors show low to intermediate signal on T1, high signal on T2, and avid enhancement on post contrast sequences [4].

Histology: Glomus tumors are composed of glomus cells, blood vessels, and smooth muscle cells, with a higher relative proportion of one component identifying the tumor as solid, glomangioma, or glomangiomyoma respectively. Glomus cells are identified as round/ovoid slightly eosinophilic cells with central nuclei [4]. In the glomangiomyoma subtype, tissue samples will show positive stains for Smooth Muscle Actin (SMA), Muscle Specific Actin (MSA), and/or h-Caldesmon, indicating the presence of smooth muscle cells [9]. Positive staining for desmin may also be seen [9].

Treatment & prognosis

The mainstay of treatment is surgical excision. Recurrence related to incomplete excision happens in approximately 20% of cases and is treated with repeat excision [1].

Differential Diagnoses

Differentials to consider related to glomus tumors, particularly in the subungual location, include epidermoid cysts, mucoid cysts, tenosynovial giant cell tumors, foreign body granulomas, hemangiomas, soft-tissue chondromas, and malignancies such as squamous cell carcinoma (SCC) and melanoma.

Epidermoid cysts, also known as epidermal inclusion cysts, are keratin-filled structures lined by epidermis located in the soft tissues or intraosseous. They are thought to develop secondary to penetrating trauma, which causes epidermal fragments to become entrapped in the dermis and proliferate [5]. Intraosseous lesions are proposed to develop secondary to soft tissue epidermoid cysts eroding into nearby bone [5]. Radiographs may be unrevealing, or display radiolucency within bone if intraosseous [12]. Ultrasonography will demonstrate a nonspecific hypoechoic mass. On CT, epidermoid cysts appear iso or hypodense and heterogenous, without contrast

enhancement [13]. T1 weighted MRI these appear isointense, and T2 weighted appear hyperintense [13]. Pre-contrast imaging findings of epidermoid cysts and glomus tumors are similar, but post-contrast imaging is helpful in their differentiation, since glomus tumors demonstrate strong diffuse enhancement, while epidermoid cysts are non-enhancing or demonstrate thin rim enhancement only. History of penetrating trauma is also an important consideration for favoring epidermoid cysts, and epidermoid cysts are also less likely to involve the nailbed compared to glomus tumors.

Mucoid cysts develop near the distal interphalangeal (DIP) joint, most often secondary to underlying osteoarthritis of the joint [5]. They present as fluctuant skin colored or translucent masses on the dorsal finger between the DIP and proximal nail fold, and may distort the nailbed [14]. These soft tissue lesions may not be visualized on XR and CT, however degenerative changes indicating osteoarthritis of the DIP are often present [14]. US findings are nonspecific, displaying an avascular hypoechoic or anechoic cystic mass [5]. On MRI mucoid cysts can appear identical to epidermoid cysts, in that they are T1 hypointense, T2 hyperintense, and non-enhancing [5]. The most distinguishing feature that allows confident diagnosis of mucoid cyst is presence of a pedicle extending to the DIP joint, which was found in 83% of mucoid cysts in one series [14].

Tenosynovial giant cell tumors are soft tissue tumors common in the fingers, but not typically subungual like glomus tumors. They originate in the synovial tissues of tendon sheaths, joints, and bursae, therefore continuity to these structures is key in suggesting this diagnosis [1]. Radiographs and CT often only show nonspecific mass, though lysis of adjacent bone may be seen [1]. US may visualize a heterogeneous mass with vascularity [15]. MRI appearance is similar to glomus tumors though more heterogeneous on T2-weighted sequences, with diffuse enhancement [1].

Foreign body granulomas are an inflammatory reaction to exogenous material remaining in the tissue after a penetrating skin injury [16]. If a patient is unaware this trauma occurred, symptoms present as nonspecific pain and swelling. Depending on the material of the foreign body, radiographs and CT may be negative. US may suggest a foreign body when a non-anatomic echogenic structure with acoustic shadowing is identified. MRI may show the foreign body as hypointensity on both T1 and T2 weighted images, with the surrounding ring of the granuloma appearing as a low to isointense area with strong contrast enhancement [16].

Hemangiomas are highly vascular masses which can affect any soft tissues and can also present with similar pain. They are often visibly reddish-pink, and exhibit a pattern of growth in infancy followed by regression in later childhood [5, 1]. Radiographs and CT may show only nonspecific soft tissue mass, but sometimes demonstrate characteristic phleboliths [17]. Ultrasonography may show a hypoechoic mass, and phleboliths may be suggested by echogenic foci with acoustic

shadowing within the mass [17]. Hemangiomas on MR imaging show a high signal on both T1- and T2-weighted images, often with regions of fat interspersed between vascular channels [1]. Without phleboliths and intralesional fat, hemangiomas can be indistinguishable from glomus tumors because they both enhance strongly.

Soft tissue chondromas are benign cartilaginous tumors primarily affecting the fingers [5]. They are painful, and grow slowly [5]. Radiographs and CT may show a soft tissue mass with areas of calcifications [5]. US appearance is hypoechoic and heterogeneous with marked vascularity [5]. T1 weighted MRI will show isointensity, T2 shows hyperintensity, and these masses show variable contrast enhancement [5]. Notably, if calcifications are present, these will appear as hypointensities on MRI and may serve as a distinguishing factor from glomus tumors [5].

Subungual malignancies such as squamous cell carcinoma or melanoma may present with nonspecific symptoms of nail discoloration, drainage, bleeding, or pain [18]. On XR and CT, both may show a soft tissue mass and with possible erosion into nearby bone, and present as hypoechoic on US [5]. For MRI, SCC is hypointense on T1-weighted images with intermediate signal on T2, while melanoma is hyperintense on T1 and hypointense on T2, with both showing diffuse solid enhancement [5]. Histological analysis, however, is the most definitive for distinguishing between malignancies. Of note, subungual melanoma most commonly occurs on the thumb [18].

TEACHING POINT

Glomus tumors usually arise in soft tissues of the fingers and are not directly visible on plain films and CT, but because they commonly occur in the tight subungual space, they often result in indentations on the adjacent bone. A patient presenting with characteristic features of a tender and temperature sensitive mass in the finger should prompt the radiologist to consider glomus tumor, and familiarity with this entity's imaging features on all modalities will aid in the diagnosis.

QUESTIONS

Question 1: Which of the following describes the typical clinical presentation of a glomus tumor?

1. A mass arising near the distal interphalangeal (DIP) joint which is skin colored and fluctuant.
2. A spot of discoloration noted beneath the nailbed of the thumb which intermittently bleeds.
3. An exquisitely tender nodule beneath the nail plate which is more painful when exposed to the cold. (applies)
4. A painful swelling at the fingertip which develops months after a splinter was lodged in the skin and removed.
5. A painful pink mass which appears to initially grow and then regresses over the course of infancy to early childhood.

Explanation:

1. [Mucoid cysts develop near the distal interphalangeal (DIP) joint most often secondary to underlying osteoarthritis of

the joint. They present as fluctuant skin colored or translucent masses on the dorsal finger between the DIP and proximal nail fold, and may distort the nailbed.]

2. The symptom of intermittent bleeding should raise suspicion of a malignant lesion such as squamous cell hematoma and melanoma [Subungual malignancies such as squamous cell carcinoma or melanoma may present with nonspecific symptoms of nail discoloration, drainage, bleeding, or pain.]

3. [Clinically, glomus tumors typically present as a small nodule at the distal aspect of the finger, commonly subungual, and are most often tender with pain out of proportion to tumor size, and often sensitive to cold temperatures.]

4. [Foreign body granulomas are an inflammatory reaction to exogenous material remaining in the tissue after a penetrating skin injury. If a patient is unaware this trauma occurred, symptoms present as nonspecific pain and swelling.]

5. [Hemangiomas are highly vascular masses which can affect any soft tissues and can also present with similar pain. They are often visibly reddish-pink, and exhibit a pattern of growth in infancy followed by regression in later childhood]

Question 2: Which of the following statements is true regarding the imaging appearance of glomus tumors?

1. Glomus tumors appear hyperintense on T1 and hypointense on T2 weighted MRI sequences.
2. Glomus tumors appear hypointense on T1, hyperintense on T2 weighted sequences, and enhance diffusely on MRI (applies).
3. Glomus tumors demonstrate thin rim enhancement and central nonenhancement.
4. Glomus tumors show no contrast enhancement at all.
5. Glomus tumors appear hyperechoic on ultrasound.

Explanation:

[Glomus tumors show low to intermediate signal on T1, high signal on T2, and avid enhancement on post contrast sequences.] [Glomus tumors appear as a hypoechoic nodule with vascularity on doppler images.]

[Epidermoid cysts are non-enhancing or demonstrate thin rim enhancement only.]

Question 3: Which of the following statements is true?

1. Genetic conditions involving mutations in the glomulin gene (GLMN) account for 90% of all glomus tumors.
2. The treatment of glomus tumor is surgical excision, with a 20% chance of recurrence requiring additional excision. (applies)
3. Glomus tumors typically develop in adults over 65 years of age, with a predominance in males.
4. The reason that glomus tumors predominantly occur in the hands is due to the increased density of sensory nerves.
5. Glomus tumors are always nearly completely composed of glomus cells.

Explanation:

1. [90% of glomus tumors develop as a solitary sporadic lesion, but 10% of cases are inherited and present in multiples secondary to glomulin gene (GLMN) mutations.]

2. [The mainstay of treatment is surgical excision. Recurrence related to incomplete excision happens in approximately 20% of cases and is treated with repeat excision.]

3. [These tumors most frequently occur in adults aged 20 to 40 with equal distribution between males and females, though subungual tumors are more common in females.]

4. [Glomus tumors are a type of soft tissue tumor, which occur primarily in the hands and feet due to their high predominance of glomus bodies. A glomus body is a neuromyoarterial plexus composed of chemoreceptive glomus cells, smooth muscle cells, and blood vessels which form specialized arteriovenous anastomoses.]

5. Glomus tumors may be classified as solid (mostly glomus cells), glomangioma (mostly blood vessels), or glomangiomyoma (mainly smooth muscle cells). [There are three subtypes of glomus tumors, classified by their proportion of glomus cells, blood vessels, and smooth muscle cells]

Question 4: Which of the following statements is false?

1. Glomus tumors, epidermoid cysts, and mucoid cysts appear similar on MRI sequences, but glomus tumors may be distinguished by a pattern of diffuse contrast enhancement.
2. Soft tissue chondromas may be distinguished from glomus tumors on MRI by the presence of calcifications.
3. Squamous cell carcinoma (SCC) is hyperintense on T1-weighted MRI images and hypointense on T2, while melanoma is hypointense on T1 with intermediate signal on T2 (applies).
4. Foreign body granulomas may appear on MRI as a hypointense object on T1 and T2 weighted images with a surrounding ring of contrast enhancing granulomatous tissue.
5. Hemangiomas and glomus tumors may be indistinguishable from one another on imaging unless phleboliths are present to indicate hemangioma, which manifest on ultrasound as echogenic foci with acoustic shadowing.

Explanation:

1. Glomus tumors, epidermoid cysts, and mucoid cysts show similar T1 hypointensity and T2 hyperintensity, however epidermoid cysts and mucoid cysts do not uniformly contrast enhance as glomus tumors do. [Pre-contrast imaging findings of epidermoid cysts and glomus tumors are similar, but post-contrast imaging is helpful in their differentiation, since glomus tumors demonstrate strong diffuse enhancement, while epidermoid cysts are non-enhancing or demonstrate thin rim enhancement only.]

2. [On MRI mucoid cysts can appear identical to epidermoid cysts, in that they are T1 hypointense, T2 hyperintense, and non-enhancing.] [Notably, if calcifications are present, these will appear as hypointensities on MRI and may serve as a distinguishing factor from glomus tumors.]

3. [SCC is hypointense on T1-weighted images with intermediate signal on T2, while melanoma is hyperintense on T1 and hypointense on T2, with both showing diffuse solid enhancement.]

4. [MRI may show the foreign body as hypointensity on both T1 and T2 weighted images, with the surrounding ring of the granuloma appearing as a low to isointense area with strong contrast enhancement.]

5. [Without phleboliths and intralesional fat, hemangiomas can be indistinguishable from glomus tumors because they both enhance strongly.] [Ultrasonography may show a hypoechoic mass, and phleboliths may be suggested by echogenic foci with acoustic shadowing within the mass.]

Question 5: Which of the following accurately describes the histological appearance of glomus cells?

1. Ovoid cells, peripheral nuclei, slightly eosinophilic.
2. Oblong cells, peripheral nuclei, slightly basophilic.
3. Round cells, central nuclei, slightly eosinophilic. (applies)
4. Round cells, central nuclei, slightly basophilic.
5. Oblong cells, central nuclei, slightly eosinophilic.

Explanation:

[Glomus cells are identified as round/ovoid slightly eosinophilic cells with central nuclei.]

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FIGURES

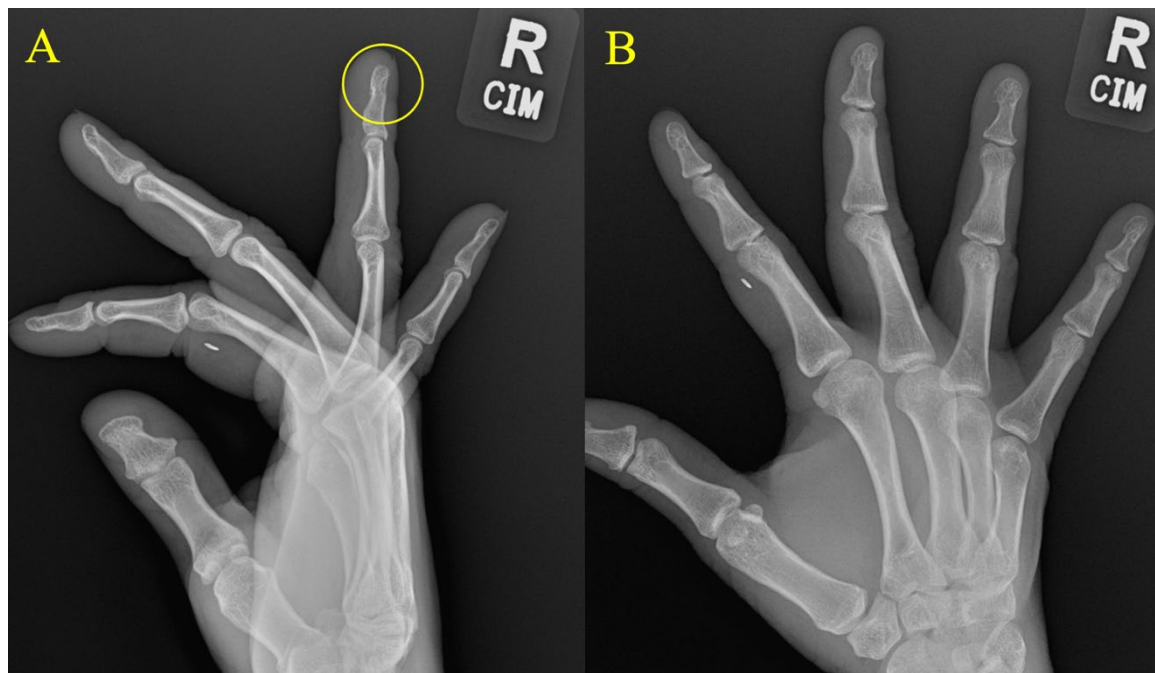


Figure 1: 42-year-old female with glomangiomyoma.

FINDINGS: (A) Lateral view plain film of the right hand demonstrates subtle indentation of the 4th finger distal phalanx's dorsal cortex (yellow circle), suggesting extrinsic soft tissue mass.

(B) Oblique view plain film of the right hand displays no distinguishable osseous or soft tissue abnormalities.



Figure 2: 42-year-old female with glomangiomyoma.

FINDINGS: Sagittal reformatted CT without IV contrast of the right hand demonstrates smooth cortical erosion of the dorsal 4th finger distal phalanx (yellow circle), again suggestive of a soft tissue mass which is not clearly visible. Images obtained on a Siemens Somatom Definition AS CT scanner with technique: mAs 136. kVp 120. Slice thickness 2 mm.

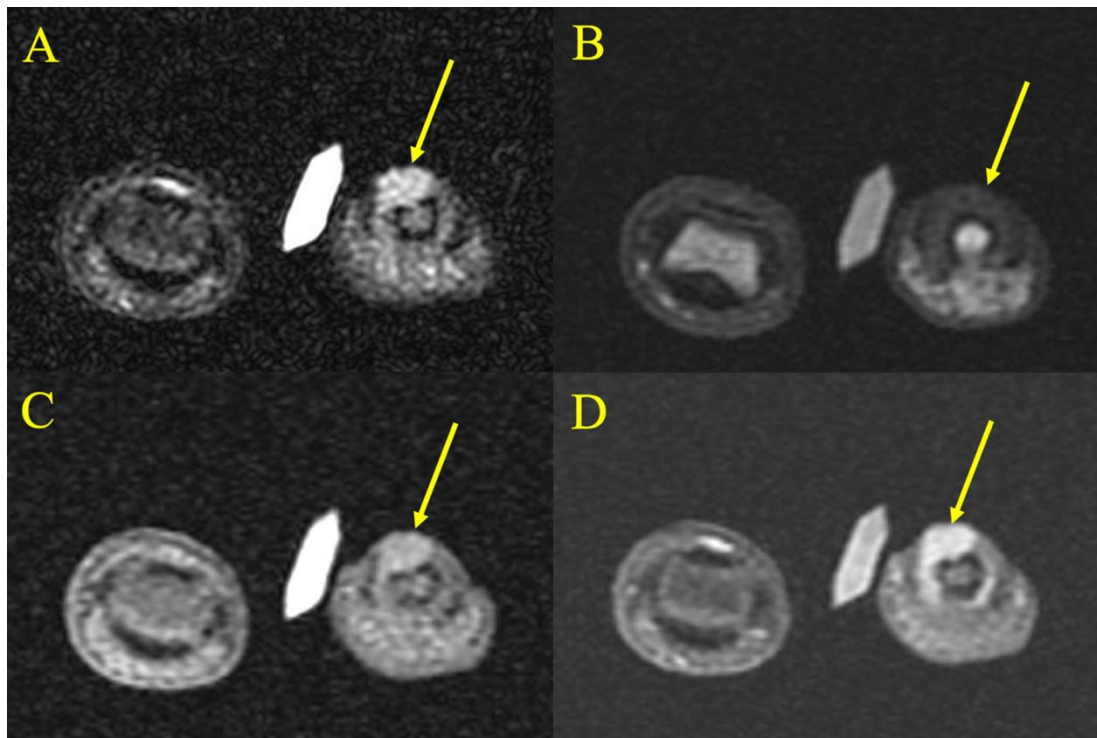


Figure 3: 42-year-old female with glomangiomyoma.

FINDINGS: (A) Axial T2 weighted fat saturated (TR 4567 ms, TE 49.5 ms) MRI of the right hand shows a soft tissue lesion of the dorsal 4th finger distal phalanx (yellow arrow) which is mildly hyperintense.

(B) The mass (yellow arrow) is hypointense on axial T1 weighted (TR 383 ms, TE 10.6 ms) MRI.

(C) Axial T1 weighted MRI with fat sat (TR 633 ms, TE 10.6 ms) pre contrast image establishes baseline signal intensity prior to contrast administration.

(D) Axial T1 weighted MRI with fat sat (TR 633 ms, TE 10.6ms) post contrast (after IV administration of 7 mL Gadobutrol) shows diffuse contrast enhancement of the soft tissue lesion (yellow arrow). These findings altogether favor glomus tumor amongst the differential diagnosis of subungual masses. Images obtained on a 1.5 T GE HDXT MRI machine.

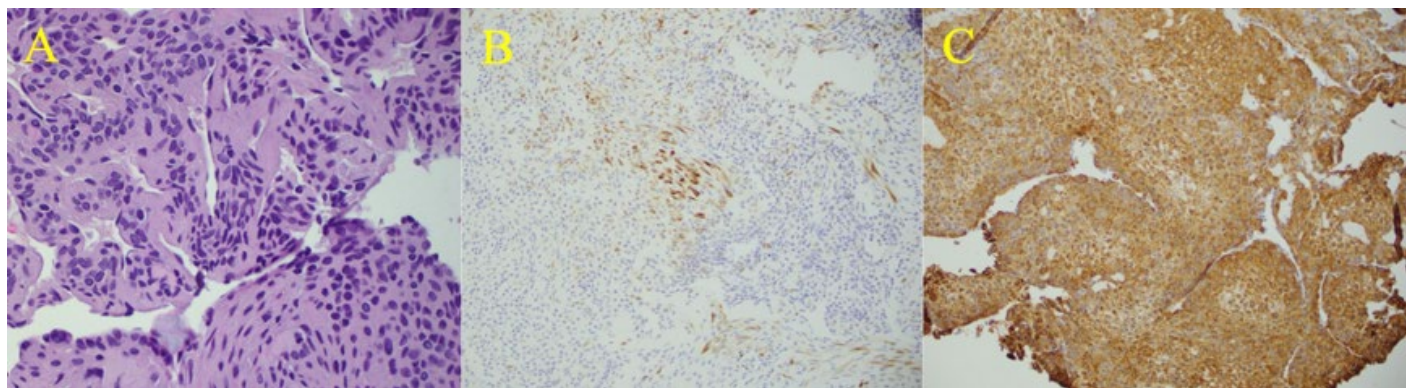


Figure 4: 42-year-old female with glomangiomyoma.

(A) H&E demonstrates round, homogenous cells with round to ovoid nuclei, indicative of glomus cells.

(B) Desmin shows patchy positivity.

(C) SMA is diffusely positive. This histologic profile is diagnostic of glomangiomyoma.

Summary table:

Etiology	A soft tissue tumor affecting glomus bodies, which are specialized neuromyoarterial plexuses found primarily in the hands and function in temperature regulation
Presentation	A painful nodule which is exquisitely tender and cold-sensitive
Incidence	Account for 1–5% of all soft tissue tumors affecting the hand
Gender ratio	1:1, subungual location more common in females
Age predilection	Most common in ages 20-40
Risk factors	90% develop sporadically, 10% secondary to inherited GLMN gene mutations
Treatment	Surgical excision
Prognosis	<20% may recur
Imaging findings	XR & CT: extraosseous lesion usually not visible but may cause remodeling and scalloping of adjacent bone, if intraosseous would appear radiolucent US: hypoechoic nodule with vascularity MRI: low to intermediate signal on T1WI, hyperintense on T2WI, avid contrast enhancement

Legend: Summary table reporting epidemiologic and clinical features of glomus tumors.

Differential table

Differential	X-Ray/CT	US	Pre-contrast MRI	Post-contrast MRI
Glomus tumor	Lesion itself often inconspicuous, may cause remodeling and scalloping of adjacent bone, if intraosseous would appear as lytic lesion.	Hypoechoic nodule with vascularity.	T1 low to intermediate signal, T2 hyperintense.	Avid diffuse enhancement.
Epidermoid cyst	Similar to glomus tumor: extraosseous lesion invisible, intraosseous lesion appears as lytic lesion.	Nonspecific hypoechoic mass.	T1 isointense, T2 hyperintense.	Central non-enhancement and possible thin rim enhancement.
Mucoid cyst	Lesion itself usually invisible. Often associated with degenerative changes of the DIP joint.	Avascular hypoechoic cystic mass, may show pedicle extending to joint.	T1 hypointense, T2 hyperintense, may show pedicle extending to joint.	Do not enhance centrally but may show slight border enhancement.
Tenosynovial Giant Cell Tumor	Nonspecific soft tissue density mass, though lysis of adjacent bone may be seen.	Heterogenous soft tissue mass with vascularity.	T1 hypointense, T2 heteroeogeneous low to intermediate signal intensity.	Diffuse enhancement.
Foreign body granuloma	If foreign body density is different than soft tissues, may manifest as a radiodense or radiolucent non-anatomically shaped object.	Echogenic, nonanatomic object with posterior acoustic shadowing.	Foreign body itself usually T1 and T2 hypointense though variable. Surrounding granuloma T1 and T2 hypointense.	Granulomatous tissue demonstrates rim enhancement.
Hemangioma	Inconspicuous or nonspecific soft tissue density mass, but may demonstrate calcific density phleboliths.	Hypoechoic mass, and phleboliths may be suggested by echogenic foci with acoustic shadowing within the mass.	T1 hypointense and T2 hyperintense vascular channels, often with interspersed T1 hyperintense fat.	Marked enhancement.
Soft-tissue Chondroma	Soft tissue density mass which may ossify.	Hypoechoic mass, may be hyperechoic and show shadowing if ossified.	T1 isointense, T2 hyperintense if non-ossified. Follows bone signal intensity if ossified.	Variable patterns of enhancement.
Malignancy	Inconspicuous or nonspecific soft tissue density mass with possible erosion into nearby bone.	Hypoechoic mass.	SCC: T1 hypointense, T2 intermediate signal. Melanoma: T1 hyperintense, T2 hypointense.	Diffuse solid enhancement.

Legend: Differential table describing the imaging appearance of lesions which may develop in the subungual space.

KEYWORDS

Glomus tumor; glomus body; glomangiomyoma; subungual; neoplasms, finger mass

ABBREVIATIONS

SMA = Smooth Muscle Actin
GLMN = Glomulin
US = Ultra Sound
MSA = Muscle Specific Actin
SCC = Squamous Cell Carcinoma
DIP = Distal Inter Phalangeal

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