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Case Report,

Glomus Tumor of Rare Localization in the Thigh; Case Report and Literature Review

Suheyla Karaduman[1/*] Sarvinoz Albalushi[1]; Alesia Talpeka[1]; Nishtha Nigam[1]; Renu Bhargavi Boyapati[1]; Dilisha Chandanala[1], Ratesh Khillan, MD[1]; Bahadir Simsek, MD[1]; Aditya Vikram Maheshwari, MD[2]; Daniel Levitan, MD[3]

[1]Brooklyn Cancer Care, Brooklyn, NY, USA
[2]SUNY Downstate Medical Center, Department of Orthopedic Surgery, Brooklyn, NY, USA
[3]SUNY Downstate Medical Center, Department of Pathology, Brooklyn, NY, USA

Abstract:

Glomus tumors are rare tumors primarily found on the nail bed of the fingers and rarely occur in other parts of the body. We present a case of a 71-year-old male who came in with a 5-year history of glomus tumor (GT) of the right lateral thigh associated with progressively increasing intermittent sharp pain. He denied any numbness or paresthesias of the extremity. Physical examination revealed a tender mass on the lateral side of the right upper leg associated with edema and erythema. Surgical excision and immunohistochemistry were performed. A histopathological diagnosis of glomus tumor was made. The postoperative recovery was successful and the patient reported resolution of the pain.

We performed a literature review of reported cases of GTs of the thigh. The review showed that all patients had a long period of symptomatic disease before an accurate diagnosis could be made and were easily treated with surgical excision. No evidence of recurrence was noted after surgical removal. GTs of the thigh rarely recur due to their solid and encapsulated structure. This also makes them easy to recognize and surgically remove.

This study highlights the unusual location of the GT, its impact on our patient, and reviews relevant academic literature. Tumors in these areas are frequently misdiagnosed due to vague symptoms. GTs should be included in the differential diagnosis of patients who experience pain with nonspecific symptoms that are not limited to specific body locations.

Introduction:

Glomus tumors(GT) are benign mesenchymal neoplasms of vessels and glomocytes. They typically present as a bluish, painful lesion primarily found on the nail bed of the fingers and rarely occur in other parts of the body [1]. An unusual location often leads to missed or delayed diagnosis and management. In this study, we present a new case of GT located in the right lateral thigh of a 71-year-old male associated with intermittent sharp pain in his right thigh for 5 years. The pain resolved completely after the resection surgery. We study the unusual location of the GT, its effects on our patient, and review the academic literature pertaining to the topic.

Case report

A 71-year-old male presented with a history of painful mass in his right thigh for 5 years. The mass did not change in size and was associated with sharp intermittent pain radiating to the knee. The pain had been progressively increasing in intensity for the past 1-2 years and had acutely worsened in the last 3 months. It was characterized as 9/10 in intensity, persistent at night, and worse with walking or activity. The patient did not report any numbness or tingling of the extremity.

Physical examination revealed a tender mass on the lateral side of the right upper leg associated with edema and erythema.

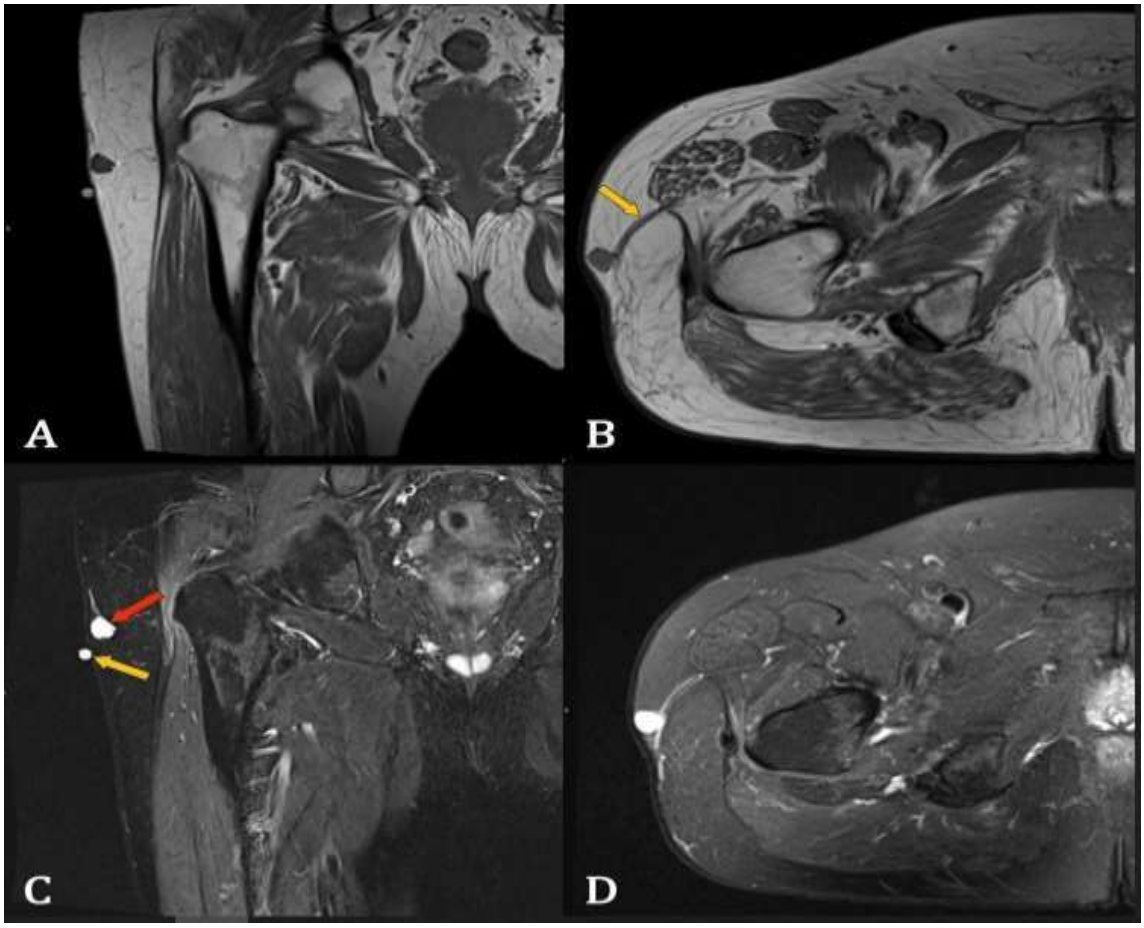

Figure 1:
Magnetic Resonance Imaging (MRI) revealed a 11x11x12 mm (AP x ML x CC) subcutaneous soft tissue mass peripheral to the caudal margin of the right greater trochanter.
(A) and (B): Iso-Intensity on Coronal, Axial T1 weighted images with a vascular feeder (B, Yellow Arrow)
(C): Coronal STIR MRI showing a mass(Red Arrow) with MRI marker(Yellow Arrow)
(D): Uniform hyperintensity on fat suppressed T2 weighted images.

Magnetic Resonance Imaging (MRI), revealed a 11x11x12 mm (AP x ML x CC) subcutaneous soft tissue mass peripheral to the caudal margin of the right greater trochanter with a vascular feed (**Fig. 1A and B, yellow arrow**). Coronal STIR MRI showed a mass (**Fig. 1C, red arrow**) with MRI marker (**Fig. 1C, yellow arrow**). Isointensity on T1 weighted images, and uniform hyperintensity on fat suppressed T2 weighted images were also noted (**Fig. 1D**). The lesion was located within 3mm of the skin surface.

Surgical excision and immunohistochemistry was performed. The tumor was well-circumscribed with a thin capsule-shaped rim of fibrous tissue, surrounded by native adipose tissue (**Fig. 2A**). The bland round cells typical of a glomus tumor were observed at high power magnification (**Fig. 2B**). Blood vessels were seen throughout (**Fig. 2B, yellow arrows**) and were quite prominent – a finding seen in overtly vascular glomus tumors, sometimes called glomangiomas. Nerves are also present and may partially explain the painful nature of these tumors (**Fig. 2B, blue arrow**). The mass was SMA + (smooth muscle actin) (**Fig. 2C**), calponin + (highlights myoepithelial cells), CD34 + (faint blush) (**Fig. 2D**), CD31 + (highlights blood vessels), and S100 + (highlights nerves) while negative for synaptophysin, chromogranin, and pancytokeratin. Ki-67 was <1%. A histopathological diagnosis of glomus tumor was made.

The postoperative recovery was successful. The patient had no complaints regarding his surgical excision and the pain subsided immediately.

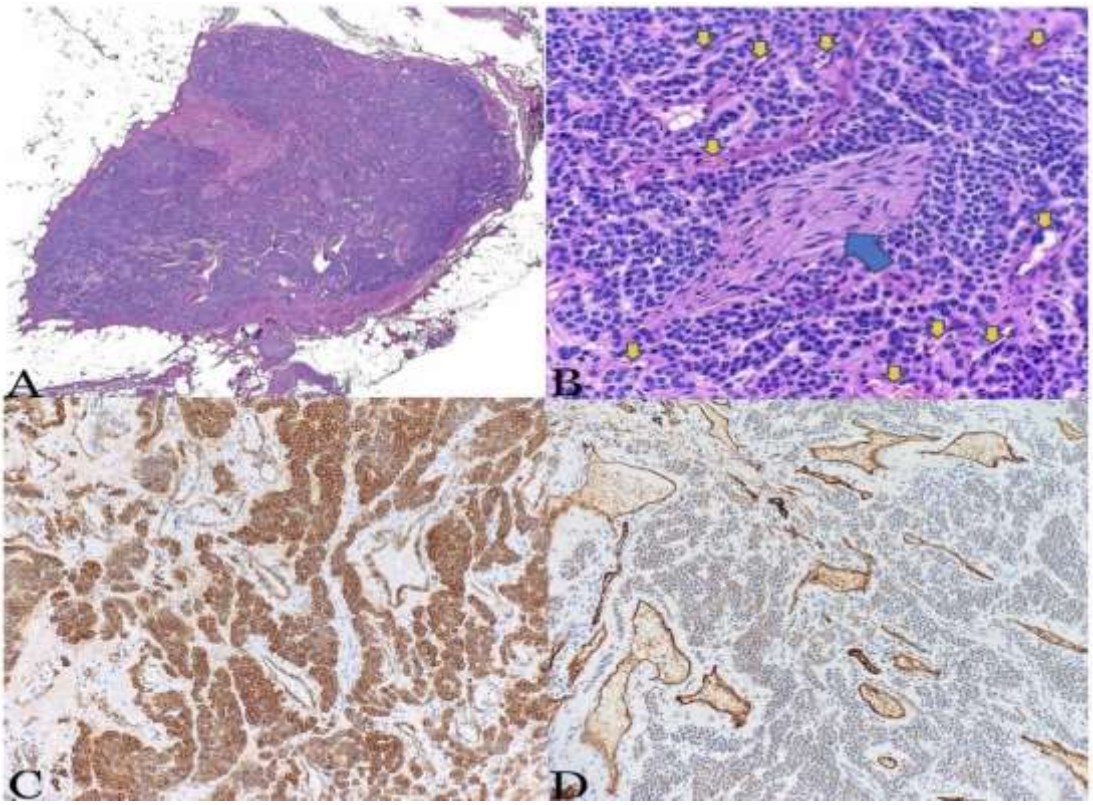

Figure 2: **(A)**Low power magnification reveals a well-circumscribed tumor with a thin capsule-like rim of fibrous tissue, surrounded by native adipose tissue.
(B) High power magnification reveals bland, round cells, typical of a glomus tumor. Blood vessels are seen throughout**(yellow arrows)** and are quite prominent—a finding seen in overtly vascular glomus tumors, sometimes called glomangiomas. Nerves are also present and may partially explain the painful nature of these tumors. One nerve can be seen in the middle of this image **(blue arrow).**
(C) Immunohistochemical staining for smooth muscle actin is positive (brown) in glomus tumors/glomangiomas.
(D) Immunohistochemical staining for CD34 highlights the vascular component.

Table:1

Cases/year	Age	Gender	Localisation	Size	Surgery	Followup
1*	71	M	Right thigh	1.1 x 1.1 x 1.2 cm on MRI	excision	symptom-free for 5 months of follow-up
2/2009 Hakverdi, Sibel *et al.* (6)	-	-	left hip	1.2 cm (ultrasound)	excision	symptom-free for 3 months of follow-up
3/2003 Gencosmanoglu, Rasim *et al.* (7)	68	Male	Right hip	2 cm (ultrasound)	excision	symptom-free for 2 months of follow-up
4/2008 Weiser, Jessica A *et al.* (8)	57	Male	Left lateral hip	3.1x2.2x1.5 cm (on histopathologic examination)	excision	Fully recovered at discharge
5/2023 Pena-Burgos, E M *et al.*(9)	79	Male	Thigh location	9.5cm	excision	No local relapse
6/1997 Amillo S, Arriola FJ *et al.* (10)	38	Female	Thigh	3cm (MRI)	Excision	symptom-free for 12 months of follow-up
7/1989 Inoue O, Ibaraki K *et al.*(11)	26	Female	Gluteal region	5x3x1.5cm	excision	No recurrence at 1.5 years after surgery

Cases/year	Age	Gender	Localisation	Size	Surgery	Followup
8/2005 Hermann, G., Klein, M.J., Springfield, D. *et al.* (12)	36	Female	Mid-thigh	0.9x2cm (MRI)	excision	symptom-free for 12 months of follow-up
9/2014 Ezeoke C, Xiang D *et al.* (13)	48	Female	Right thigh	3.1x2.2cm (MRI) -	Excision with re-excision in 5 months (due to the positive margin of the initial biopsy for tumor cells)	Disease free with no clinical evidence of recurrence
10/2014 So, Sang Young *et al.*(14)	65	Male	Left anterior thigh	0.8x0.6 cm2 hypoechoic cyst	excision	Pain free for six months follow-up
11/2017 Margad, Omar, and Nabile Bousselmame.(15)	40	-	Right thigh	1.5 cm	excision	Disease free for 2 years of follow-up
12/2015 Beksaç K, Dogan L *et al.*(16)	39	Male	Posterolateral side on left thigh	15x10mm (CT)	excision	Pain free after surgery
13/2016 Lee TS, Wu HT, Chan RC *et al.*(17)	66	Female	Lateral side of the right thigh	5mm	Excision	Symptom-free after surgery
14/2018 Sbai, Mohamed Ali *et al.* (18)	25	Female	Posterior side of the left thigh	3x2x2 cm (MRI)	excision	Pain-free after surgery for the last year
15/2022 Avery, Emma Claire *et al.*(19)	Mid-50s	Female	Right thigh	-	excision	Symptom-free after surgery
16/2013 Dabadie, Alexia *et al.*(20)	13	Female	Right thigh	3x5 cm highly calcified tumor	excision	No recurrence after 2 years of follow-up
17/2023 Alfattni, Ammar Abdulqader *et al.*(21)	39	Male	Left thigh	9x9 mm (MRI)	Incision	Symptom-free after surgery
18/2012 Werner, Jeff D *et al.*(22)	48	Male	Left thigh	6x6x4 mm (ultrasound)	excision	-
19/1997 Negri, G *et al.* (23)	21	Female	Left thigh	22x11x6 cm of surgical specimen	resection	Symptom-free after surgery
20/2008 Sraj SA, Khoury NJ *et al.*(24)	48	Male	Distal thigh (over the adductor canal)	1x1 cm	excision	Symptoms-free after surgery
21/2020 Kloping, Laskar Pradnyan *et al.* (25)	56	Male	Posteromedial of the distal femur	6 x 5.3mm	excision	No recurrence after 6 months of follow-up
22/2012 Lancerotto, Luca *et al.* (26)	41	Male	Lateral aspect of the intermediate third of right leg	2cm nodule	Surgically excised	Disease free for 3 years of follow-up

Dıscussıon:

GTs are benign mesenchymal neoplasms composed of various proportions of vessels and glomocytes [1], typically present with hypersensitivity to cold, paroxysmal pain and point tenderness [2].They account for less than 2% of soft tissue cancers. The cells that make up the glomus body resemble modified smooth muscle cells. They are uncommon and sometimes misinterpreted as hemangiomas or venous malformations. Glomus tumors are categorized into three types [3] based on their dominant component:Solid; dominantly glomus cells, glomangioma; dominantly blood vessels and glomangiomyoma; dominantly smooth muscle cells.

According to the literature, GTs can be inherited or sporadic. The inherited type is autosomal dominant with incomplete penetrance and variable expression. The sporadic type has de-novo mutation and is present during birth. They are classically red, purple, blue or even skin-colored, which makes it even more challenging to diagnose. They are found in nail fold, hand, lower limb, middle ear, jugular bulb, and carotid artery. A differential diagnosis of the tumor includes a fibroma, hemangioma, leiomyoma, blue nevi, venous malformation, subungual exostosis, and mucoid cyst [3].

GTs clinically diagnosed, and preferred methods for diagnosing GTs are MRI and ultrasound. Ultrasonography, especially color-duplex ultrasonography can be useful in detecting small nodules that appears hypoechoic or isoechoic nodules with hypervascularity on Doppler [4-5].

Immunohistochemistry can further assist in diagnosis as they are a-smooth muscle actin (aSMA), muscle-specific actin (MSA), h-caldesmon positive, and have abundant type IV collagen [1-3].

The treatment of choice is wide local excision [6] followed by monitoring for local recurrence. Reported recurrence rate is approximately 10%[1]. Glomus tumors constitute less than 2.0% of all primary soft tissue tumors, approximately 80% of the lesions are located in the upper extremity, and over 75% are subungually located [1]. Ultrasound and MRI are used for assessment of the lesions. A complete excisional biopsy is helpful in making a definitive diagnosis.

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We performed a literature review of reported cases, to the best of our knowledge, we have found 67 published cases in lower extremity rare area glomus tumors which is mentioned briefly in Table 2.

In this study, we mostly focused on rarer area, hip and thigh glomus tumors. We reviewed 21 cases of thigh glomangiomas from 1989 to 2023. Previously reported glomus tumors in the thigh/hip area including our study: cases, age, sex, location, size, surgery, follow up are shown in Table 1.

Tumor developed in 11 men, 9 women, gender of 2 patients was not documented. Patient age ranged from 13 to 79 (median age:48) The size of the tumors varies from 0.6 cm to 9.5cm. One tumor was excised with surgical specimen, measured 22 x 11 x 6 cm, consisting in parts of musculus vastus medialis and intermedius due to infiltrative growth. One tumor was described as a round hypoechoic cyst on ultrasonography. Ultrasound and MRI were used for assessment of the lesions. A complete excisional biopsy helps to make a definitive diagnosis.

Upon reviewing the cases we can summarize that all patients had a long period of symptomatic disease before making an accurate diagnosis from 1 month to several years. No malignancies were reported in these studies. All glomangiomas were treated with surgical excision with no evidence of recurrence. Glomus tumors in the thigh region rarely recur due to their solid and encapsulated structure, which makes them easy to recognize and surgically remove.

Conclusions:

Commonly, Glomus tumors in these areas are misdiagnosed due to vague symptoms such as radiating pain and absence of sensitivity to cold and tenderness. Without a palpable mass, pain in this area may be masked as neuropathic; thus, Glomus tumor should be included in the differential diagnosis of patients who describe pain with vague symptoms without being restricted to certain body regions.

Declarations:

Ethical Approval: We affirm that the submitted work complies with all applicable ethical guidelines and regulations, including but not limited to the Declaration of Helsinki and the International Committee of Medical Journal

Editors (ICMJE) guidelines.Informed Consent and Consent to Publish Identifiable Information or Images was obtained. Privacy and confidentiality were strictly maintained.

Competing Interests: The authors declare that they have no competing interests of a financial or personal nature that could potentially influence the results or interpretation of this study.

Authors' Contributions: The contributions of each author are as follows: Corresponding Author; Suheyla Karaduman wrote the main manuscript, Sarvinoz Albalushi, Alesia Talpeka and Suheyla Karaduman reviewed literature and prepared tables. Daniel Levitan prepared pathology figures. Nishhta Nigam wrote abstract. All authors have read and approved the final version of the manuscript.

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