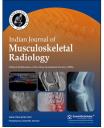
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Case Report

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Pair of glomus tumors in thigh – both periosteal and intramuscular: Rare tumor in atypical location

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ABSTRACT

Glomus tumors are benign neoplasms that arise from neuromyoarterial glomus bodies accounting for <2% of soft-tissue tumors. Glomus tumors represent around 1–5% of all soft-tissue tumors and 1–5% of all hand tumors. About 75% of these tumors occur in hand with 75–90% of these occurring in characteristic subungual location. Extradigital location of glomus tumor is very rare. We report an exceptional case of extradigital glomus tumor causing thigh pain, where there were a pair of lesions both in periosteum and intramuscular location. A 47-year-old male with complaints of pain over medial aspect of the left lower thigh for 2 years on ultrasound revealed well-defined solid hypoechoic lesions within the left vastus medialis muscle and cortical-based lesion abutting the periosteum of distal diaphysis of the left femur on medial aspect. These lesions were hypointense on T1W, hyperintense on T2W and showed early homogenous enhancement on MRI. Histopathology revealed glomus tumors, which commonly occur in digits. Glomus tumors located in both periosteal and intramuscular locations in thigh were in very uncommon. Non-specific presentation with pain in such cases remains a diagnostic dilemma. Role of multimodality imaging and histopathology correlation is important in such situation.

Keywords: Glomus tumor, Extradigital location, Ultrasound, MRI, Core biopsy

INTRODUCTION

Glomus tumors are benign neoplasms that arise from neuromyoarterial glomus bodies^[1] accounting for <2% of soft-tissue tumors.^[2] Glomus tumors represent around 1–5% of all soft-tissue tumors and 1–5% of all hand tumors.^[3] About 75% of these tumors occur in hand with 75–90 % of these occurring in characteristic subungual location.^[4] Typical glomus tumors are relatively easy to diagnose due to the tumor's characteristic solitary lesion and classic triad of symptoms: Pain, pinpoint tenderness, and hypersensitivity to cold. Extradigital glomus tumors are much more difficult to diagnose because of their atypical location and symptoms. They are usually misdiagnosed and unproperly treated, due to the absence of the typical symptoms.^[5] We report an exceptional case of extradigital glomus tumor causing thigh pain, where there were a pair of lesions both in periosteum and intramuscular locations.

CASE REPORT

Glomus tumor is a benign soft tissue tumor, commonly occur in hands. Such a tumor in extradigital location is very rare. We hereby present a case of extradigital glomus tumor. A 47-year-old male with no history of previous surgery presented with pain over medial aspect of the left lower thigh for 2 years. On local examination, there was tenderness in the medial aspect of the left lower thigh. No mass

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was clinically palpable. No significant muscle atrophy was there. Nor there was history of hypersensitivity to cold. Clinically, these features were very non-specific. Musculoskeletal ultrasound was advised as primary imaging modality.

On ultrasound with high-frequency transducer, a well-defined solid hypoechoic lesion measuring 1.8×1.0 cm was seen in medial aspect of the left thigh within left vastus medialis muscle [Figure 1a]. It showed internal vascularity on color Doppler imaging. Another similar morphology hypoechoic solid cortical-based lesion was seen abutting the periosteum of distal diaphysis of the left femur on medial aspect [Figure 1b]. Provisional diagnosis was soft-tissue tumor at this stage. MRI was done to further characterize these lesions. Three Tesla multiphasic MRI revealed a well-defined oval lesion within the left vastus medialis muscle [Figure 2 a, c and e]. It measured 1.8×1.0 cm in maximum axial dimensions. Another similar morphology lesion was seen abutting the periosteum of distal

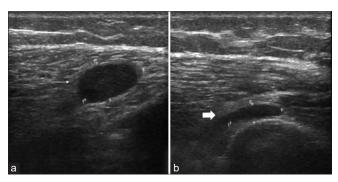


Figure 1: Gray scale ultrasound image showing well-defined oval hypoechoic lesions in vastus medialis (thin arrow in a) and based on periosteum of diaphysis of the left femur (thick arrow in b).

diaphysis of the left tibia on medial aspect [Figure 2a, b and d]. It did not scallop the periosteum or outer cortex. It measured 2.3×1.0 cm in anteroposterior \times transverse dimensions. No cortical breach or periosteal reaction was seen. These lesions were hypointense on T1W, hyperintense on T2W and showed early homogenous enhancement on dynamic post-contrast images. After clinical examination, ultrasound and MRI examination possible differential diagnoses included softtissue tumors such as myxomas and neurogenic tumors. The patient underwent ultrasound-guided core needle biopsy with 18-gauge coaxial needle. On histopathology, the left vastus and left distal femoral periosteal-based lesion biopsy showed skeletal muscle bundles and adjacent sheets of oval to round cells with moderate cytoplasm, bland-looking nuclei with many of them showing intranuclear inclusions [Fig 3 a, b and c]. There were interspersed thin capillaries and foci of epithelial looking cells [Figure 3a and b]. No mitosis or necrosis was seen. Morphological features were compatible with diagnosis of glomus tumor. On immunohistochemistry, tumor cells were negative for AE1/AE3, CD31, S100, and EMA. Tumor cells were [Figure 3h] diffusely positive for SMA [Figure 3g], focally positive for calponin while they were negative for p63, HMWCK. CD31 highlighted the vessels [Figure 3d and e]. Ki67 proliferation index was very low approximately 2% [Figure 3i]. The patient underwent surgery for the lesions with complete recovery from symptoms after surgery. Final histopathology was also glomus tumor.

DISCUSSION

Glomus tumor may be observed at any age. In most instances, it occurs in the fourth or fifth decades of life.^[6] In our case,

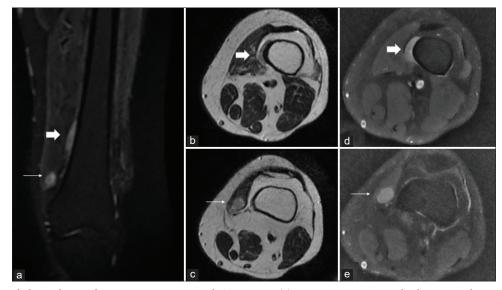


Figure 2: MRI revealed, two lesions hyperintense on Coronal STIR image (a). Lesion in intramuscular location is hyperintense on T2 axial image (thin arrow in c) and showed homogenous enhancement (thin arrow in e). Another similar intensity lesion based on periosteum of left femur (thick arrow in a , b , d)

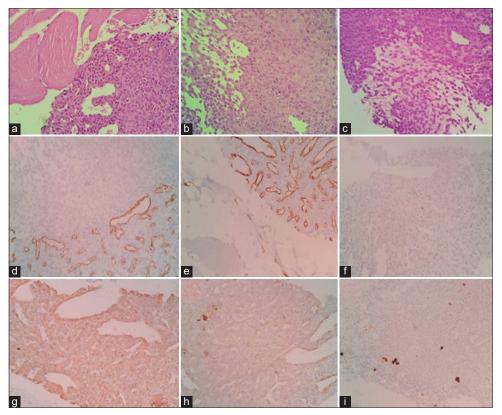


Figure 3: (a) Microsection shows skeletal muscle bundles and adjacent sheets of bland oval to round cells with interspersed thin capillaries, (b) foci of epithelial looking cells are seen which resemble synovial sarcoma, (c) focal myxoid stroma was noted, (d and e) CD31 and CD34 highlight the capillary endothelial cells, while tumor cells are negative, (f) PanCK was negative, (g) SMA was positive, (h) calponin was positive, (i) Ki67 proliferative index was very low (approximately 2%).

two glomus tumors in the left thigh were detected. One was located in intramuscular compartment within the left vastus medialis muscle. Another one was abutting diaphysis of the left femur. These unusual locations are very atypical for a glomus tumor which commonly originates from subungual bed of hand. Besides, there was no clinically palpable lump Location of glomus tumor in thigh is rare. Periosteal location of glomus tumor is extremely rare. In a 36 year old lady, Hermann et al^[7] reported a glomus tumor in thigh . In this case , the lesion was solitary and confluent with periosteum of left femur base . There was no adjacent intramucular similar lesion ,unlike our case. Masazumi et al.[8] reviewed 63 patients with glomus tumor of the soft tissues. Fifteen occurred in the lower extremity, among them seven around the knee. The tumors were found most often in the dermis and subcutaneous tissue. Heys et al.^[9] in 1992 in analysis of 43 patients with glomus tumor, only 6 occurred in the thigh and lower leg. Abou Jaoude et al.[10] found out of 11 cases of glomus tumor, 2 were in the soft tissue of the thigh. None of them extended to the bony cortex.

CONCLUSION

Glomus tumors predominantly involve subungual location in digits of upper extremity. The presence of these lesions in lower extremity particularly in thigh in both periosteal and intramuscular locations is extremely rare. None of previous published literature reported extradigital presentation of pair of such lesions in both periosteal and intramuscular locations in thigh. Non-specific presentation with pain in such cases remains a diagnostic dilemma. Our case undermines the importance of multimodality imaging and histopathology correlation in such cases.

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Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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