

Glomangioma of the Tibia- A rare presentation

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Abstract

A 35yr old female patient presented with a pulsatile swelling of right proximal tibia present for 2 years. There was a rapid increase in size and pain in the 6 weeks preceding presentation. Radiographs showed a lytic lesion of about 8cm x 8cm in the anteromedial aspect of the proximal tibia with MRI showing a highly vascular lesion. A needle biopsy and histopathological examination confirmed the diagnosis of Glomangioma. It was treated with marginal resection of the tumour and fibular grafting of the defect.

Introduction

The glomus tumor is a rare, benign, but painful vascular neoplasm arising from glomus cells, which are modified perivascular smooth muscle cells. As per reports of the Mayo clinic it contributes about 1.6% of consecutive 500 cases of soft tissue tumours. (1) The usual site of a glomus tumour is the sub-ungal region. It contributes for 1-5% of all hand tumours. (2) Although said to arise from glomus cells, these have also been reported to be arising in extracutaneous regions, which doesn't have glomus cells. Primary intraosseous glomus tumor is even rarer, with only about 20 cases reported in the literature so far, 5 of which involved the spine (3-18) Most of them involve the distal phalanges of the fingers or thumb, and only three cases have been previously reported in a long bone of which 2 were in ulna and one in fibula. Nose, eyelid, middle ear, lung, mediastinum, stomach, rectum, cervix and penis are the other reported areas of occurrence. Surgical resection is currently considered the treatment of choice. An uncommon case of primary intraosseous glomus tumour of the tibia treated with surgical resection is presented here. To our knowledge no case of proximal tibia

glomangioma has been reported so far.

Case Report

A 35yr old female patient presented with a solid swelling of right proximal tibia present for 2 years. There was no previous history of infection or trauma to the region. The swelling had increased in size rapidly in the last 6 weeks preceding the presentation and was associated with a significant increase in pain. The patient had taken some indigenous treatment with topical agents but there was no relief of symptoms. On examination, there was a globular swelling of the proximal tibia with visible pulsations and multiple hyperpigmented patches on the overlying skin. The swelling was firm to hard in consistency, tender and pulsatile with local increase in temperature (Fig 1). The knee range of motion was full range and pain free with no distal neurovascular deficits. Plain radiograph of the right tibia showed an expansile lytic lesion 8cm x 8cm over metaphyseal region proximal tibia with thinning of anterior cortex (Fig 2). There was no evidence of a cortical breach. Needle biopsy from the lesion showed multiple cores of a benign angiomatous lesion composed of peritheliomatous pattern and cords of tumor cells with monomorphic punched out nuclei. The diagnosis of glomangioma was suggested. (Fig 4 A&B) With this background and the absence of any signs of a malignancy in the biopsy, marginal excision of the tumour Fig 5 was done and augmented by non vascularized fibular strut graft and 8 holed

proximal tibial LCP. Fig 6

Histopathological examination

Grossly the resection specimen of right tibial lesion showed a tan brown hemorrhagic intraosseous mass measuring 7x4.5x4cm which is soft to firm, solid with central hemorrhagic areas. The tumor was seen invading the bone cortex focally. There were no areas of necrosis grossly. (Fig 7A) Microscopy shows an intraosseous cellular neoplasm composed of multiple thin walled vascular channels lined by thin endothelial cells and surrounded by prominent peritheliomatous pattern (Fig 7B, C) of monotonous round to oval and focally spindle cells with bland central nuclei, many with punched out nuclei and moderate amount of eosinophilic cytoplasm. (Fig 7D) The tumor showed focal trabecular, reticular pattern and focal solid areas. There was no evidence of cytologic atypia, intracytoplasmic neolumina formation or nuclear hobnailing. The tumor showed peritheliomatous reticulin poor stromal network unlike hemangiopericytoma. (Fig 7E) The differential diagnosis of glomangioma was considered. The immunohistochemistry showed diffuse strong vimentin (Fig 7F), focal strong cytoplasmic smooth muscle actin SMA (Fig 7G) however the tumor cells were negative for endothelial marker, CD34. (Fig 7H). The diagnosis of glomangioma was confirmed based on expression of vimentin and smooth muscle actin and negativity for endothelial marker Cd34.

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Figure 1: Tender, pulsatile firm swelling proximal Tibia with skin changes following local indigenous therapy



Figure 2: Shows an expansile lytic lesion of the proximal tibia measuring 10x10cm in the metaphyseal region with thinning of the anterior cortex with no evidence of any cortical breach.

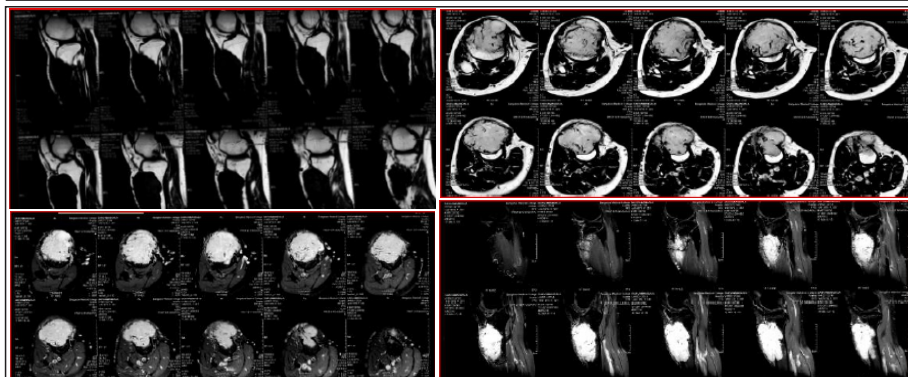


Figure 3: T1 and T2 weighted MR images of the lesion showing a highly vascular lesion with fluid level.

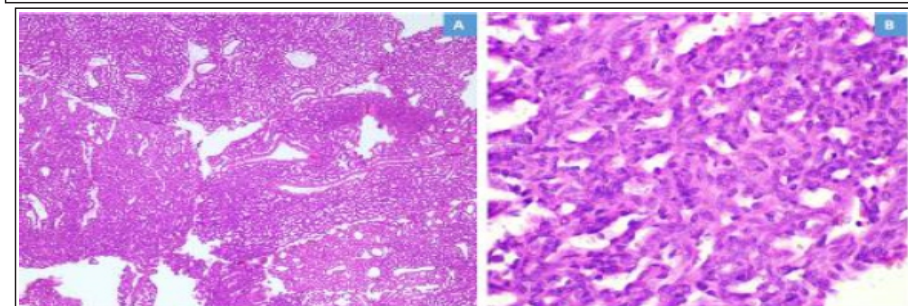


Figure 4: Fig. 1 A Needle core biopsy with peritheliomatous pattern and cords of tumor cells. Fig 1 B showing the tumor cells with monomorphic punched out nuclei.



Figure 5: Gross examination: A brown hemorrhagic intraosseous mass measuring 7 x 4.5 x 4cm which is soft to firm, solid with central hemorrhagic areas. The tumor is seen invading the bone cortex focally.



Figure 6: Shows the post op X ray marginal excision followed by fibular strut grafting and plating

Discussion

Glomus tumors are rare hamartomatous lesions that are derived from the glomus cells which are modified perivascular smooth muscle cells. A glomus tumor is characterized by excessive proliferation of vascular structure with distinctive large cells, the glomus cells, associated with thickened endothelium (1-3). The triad of pain, tenderness, and sensitivity to cold is typical of a glomus tumor [1,2]. The tumor may occur anywhere in the body, although the most common site is the subungual region of the fingers. Glomus tumors arising primarily within bone is quite rare [4,5]. A 3:1 female predisposition is observed in subungual lesions, although overall there is no gender-based predisposition. Adults between 20 and 40 years of age are usually affected. (7) The chest wall, stomach, colon, nerve, face, trachea, and possibly the mediastinum (8) are the unusual sites affected. In 1987, Rozmaryn et al. reported an intraosseous glomus tumor of the ulna (9). It was located within the medial cortex, and there was no sclerotic border. Radiographs showed an area of bone destruction in the anteromedial region of the proximal ulna. In a case reported by Bahk et al. in 2000 an intraosseous glomus tumor of the fibula, Xrays showed an ovoid, eccentric, expansive lytic lesion of the midshaft of the fibula with an outer complete, thin shell of bone (5). In a case reported by Urakawa et al the radiographs of

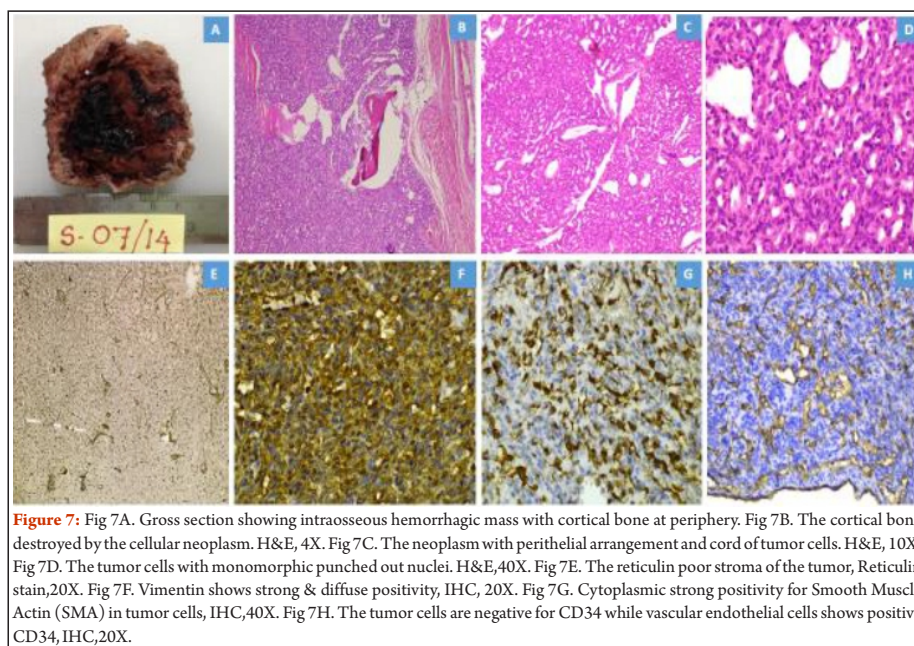


Figure 7: Fig 7A. Gross section showing intraosseous hemorrhagic mass with cortical bone at periphery. Fig 7B. The cortical bone destroyed by the cellular neoplasm. H&E, 4X. Fig 7C. The neoplasm with perithelial arrangement and cord of tumor cells. H&E, 10X. Fig 7D. The tumor cells with monomorphic punched out nuclei. H&E, 40X. Fig 7E. The reticulin poor stroma of the tumor, Reticulin stain, 20X. Fig 7F. Vimentin shows strong & diffuse positivity, IHC, 20X. Fig 7G. Cytoplasmic strong positivity for Smooth Muscle Actin (SMA) in tumor cells, IHC, 40X. Fig 7H. The tumor cells are negative for CD34 while vascular endothelial cells shows positive CD34, IHC, 20X.

the right elbow revealed an osteolytic area in the proximal bone shaft of the ulna, with the lesion measuring 1.0 cm in length. It was located within the medial cortex thickening. There was a shell-like bone formation outside of the osteolytic area (10). Our patient was 35 year old female, whose

imaging studies are comparable to the previously reported cases. They showed a lytic lesion with no cortical breach. She was treated with excision of the tumor and fibular grafting to bridge the resulting bony defect. There was no evidence of cortical discontinuity intraoperatively. The non

vascularized fibular graft was augmented using an eight holed LCP. The diagnosis of a glomus tumor was made after histopathologic examination, which was backed up by immunohistochemistry studies based on expression of Vimentin and smooth muscle actin (SMA) and negativity for endothelial marker CD34. Post op rehabilitation, and counselling was started immediately. The patient had full range of movements in the knee post operatively. She is to be followed up on a three monthly basis for the next two years. To our knowledge four glomangiomas of long bones have been reported previously. However this is the first case of glomangioma to be reported in tibia.

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