Scholars Journal of Applied Medical Sciences (SJAMS) ISSN 2320-6691 (Online)
Sch. J. App. Med. Sci., 2014; 2(1B):231-233 ISSN 2347-954X (Print)

(An International Publisher for Academic and Scientific Resources)
www.saspublisher.com

Case Report

A Case of Adamantinoma Involving the Entire Tibia

Dr. Siddalinga swamy MK[1], Dr. Vedashree MK[2], Dr. Maruthi CV*[2], Dr. Shiva prakash SU[2], Dr Ravi Kumar K[3]
[1]Professor, Department of Orthopaedics, MVJ MC and RH, Hoskote, Bangalore, India
[2]Assistant Professor, Department of Pathology, MVJ MC and RH, Hoskote, Bangalore, India
[3]Assosiate Professor, Department of Orthopaedics, MVJ MC and RH, Hoskote, Bangalore, India
MVJ Medical College & Research Hospital, Hoskote, Bangalore -562 114, India

***Corresponding author**
Dr. Maruthi CV
Email: cvmaruthi@sify.com

Abstract: Adamantinoma is a rare neoplasm representing less than 1% of all primary malignancies of bone. Here we are presenting a case of adamantinoma of right tibia with extra compartmental involvement. A 43 years male patient came to us with the complains of pain in the right leg since three years and was operated by prophylactic intramedullary interlocking nail one year back for the osteolytic lesion. Patient was evaluated for the lesion by X- ray, CT scan, MRI and open biopsy. The histopathological and Immunohistochemical examination confirms the diagnosis of adamantinoma. Workup for secondaries was negative. As the lesion was extra compartmental and limb salvage was not possible hence treated him with above knee amputation. Histopathological and Immunohistochemistry examination confirms the diagnosis. If the adamantinoma is extra compartmental limb salvage procedures are not useful for the better prognosis hence amputation to be considered.
Keywords: adamantinoma, histopathology, amputation.

INTRODUCTION

Adamantinoma is a rare neoplasm representing less than 1% of all primary malignancies of bone. It has been postulated that adamantinoma arises from aberrant nests of epithelial cells, which would account for the fact that this tumour primarily occurs in bone that is in a subcutaneous location. Here we are presenting a case of adamantinoma of right tibia with extra compartmental involvement.

CASE REPORT

A 43 years male patient came to the outpatient department with the complains of pain in the right leg since three years and operated by prophylactic intramedullary interlocking nail for the right tibia one year back for the osteolytic lesion in the tibia without evaluating the aetiology of the lytic lesion.

Patient was admitted and evaluated for the lesion by X- ray which show Irregular well defined osteolytic lesion with minimal periosteal reaction in the diaphysis of right tibia with intramedullary nailing in situ and minimal soft tissue swelling –the differential diagnosis of Adamantinoma / low grade chondrosarcoma/ plasmacytoma of tibia were thought (Fig. 1, 2). C.T scans of right tibia and fibula show periosteal elevation, focal lytic areas with cortical irregularity- suggestive of neoplastic / infective aetiology (Fig 3). Implant removal with open biopsy was done for the evaluation of the aetiology. The histopathology show islands of epithelial cells in a fibrous stroma and Immunohistochemistry examination show- the neoplastic cells are focal CK and EMA positive and negative for CD34 and CD31. The features are consistent with adamantinoma right tibia. We evaluated for the secondaries with C.T scan of thorax plain and contrast, which was normal (Fig 4, 5). Bone scan which was positive only for the right tibia and final diagnosis of adamantinoma of right tibia with extra compartmental involvement was made. Patient was treated by above knee amputation, as limb salvage will be having high recurrence rate in our case.

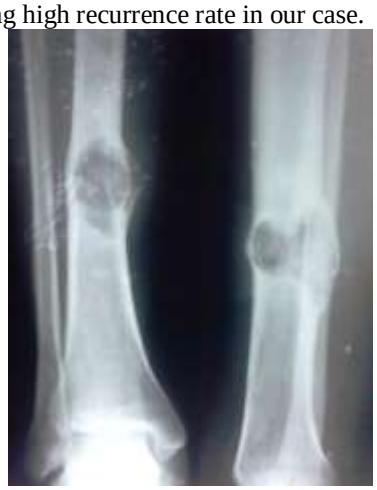

Fig. 1: Osteolytic lesion in the diaphysis of tibia 2 years back

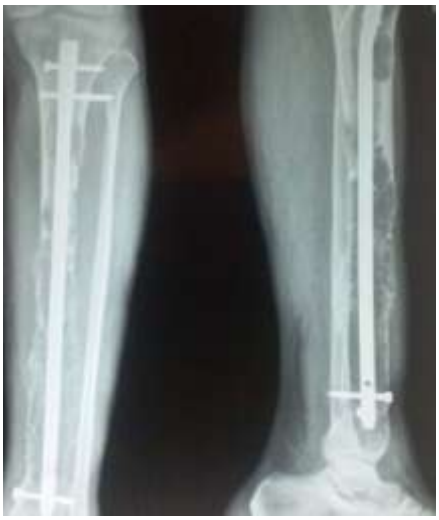

Fig. 2: Adamantinoma involving the entire tibia with nail *insitu*

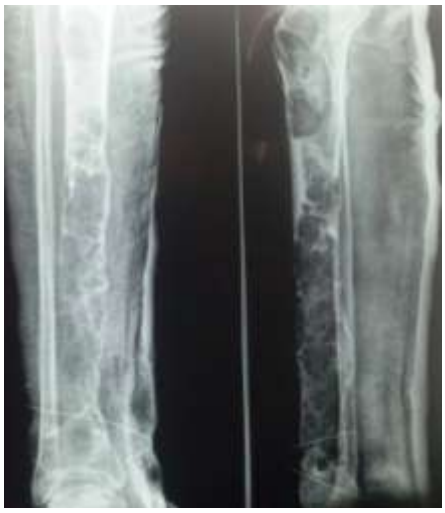

Fig. 3: Adamantinoma of tibia after nail extraction

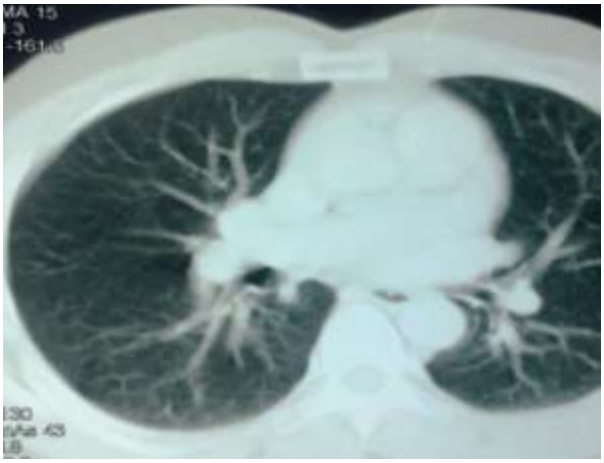

Fig. 4: CT thorax normal

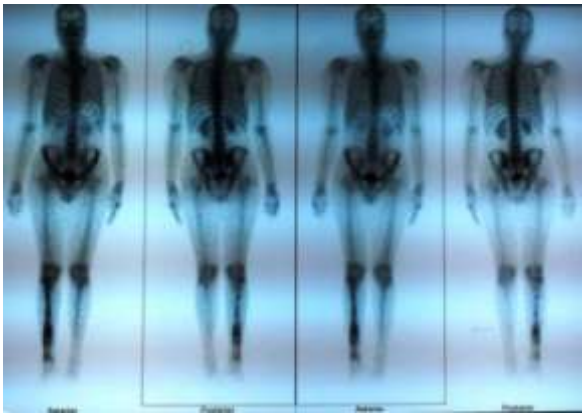

Fig. 5: Bone scans hot spot in right tibia

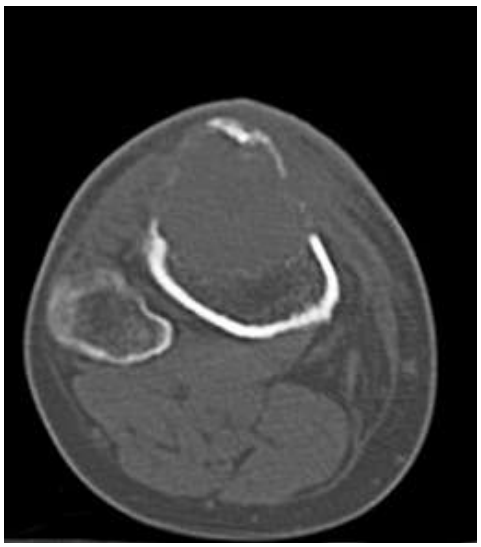

Fig. 6: CT showing breach in the cortical continuity

CONCLUSION

Any lytic lesion is to be evaluated for the aetiology. Prophylactic fixation to be done once the aetiology is known. Histopathological and Immunohistochemistry examination confirms the diagnosis of adamantinoma. Evaluation for the secondaries is must for the treatment and prognosis. If the adamantinoma is extra compartmental limb salvage procedures are not useful for the better prognosis hence amputation to be considered.

DISCUSSION

Adamantinoma was first described by Fischer in 1913[1]. Adamantinoma has a wide age distribution, but most patients are in the second or third decade at the time of diagnosis. Adamantinoma are classified into 2 distinct types: classic and differentiated. Classic adamantinoma usually occur in patients older than 20 years, whereas differentiated adamantinoma occur almost exclusively in patients younger than 20 years. It has an 85% predilection for occurring in the tibia. Pain is the most common symptom. Because the lesion is typically slow growing, the pain can be present for many years before the patient seeks medical attention.

Because the lesion usually occurs in a subcutaneous location, a palpable mass may be present. Approximately 20% of patients have a pathological fracture. The most common radiographic appearance is that of multiple, sharply demarcated radiolucent lesions in the tibial diaphysis. The radiolucent lesions are separated by areas of dense, sclerotic bone. A large portion or even the entire tibia can be involved. Frequently, the fibula also is involved by direct extension of the tumour [2].

Microscopically, adamantinoma consists of islands of epithelial cells in a fibrous stroma. Some areas of the tumour resemble fibrous dysplasia or osteofibrous dysplasia. Nuclear atypia is minimal, and mitotic figures are rare. Immunohistochemical staining usually is positive for cytokeratins and vimentin. It generally is a low-grade lesion, and histological features are not predictive of behaviour [3, 4]. The optimal treatment of adamantinoma is wide resection or amputation [5, 6]. The tumour generally is radio resistant and chemo resistant. Local recurrence occurs in approximately 25% of patients, and amputation should be considered for these patients. Metastases are rare at presentation, but may occur later in 30% of patients. Overall survival is approximately 85% at 10 years. Prognosis depends most on the adequacy of the surgical margin. Compared with patients who have marginal or intralesional surgical procedures, patients who have wide or radical procedures have significantly reduced rates of local recurrence and metastases (<10%). Because of the slow-growing nature of this lesion, local recurrence or metastasis may occur very late—reportedly 19 years after the initial treatment. The importance of long-term follow-up must be stressed.

REFERENCES

1. Fischer B; Uber ein primares Adamantinom der Tibia. Frankfurt: Zeitschr F Path., 1913:422-441.
2. Bethapudi S, Ritchie DA, Macduff E, Straiton J; Imaging in osteofibrous dysplasia, osteofibrous dysplasia-like adamantinoma, and classic adamantinoma. Clin Radiol., 2013.
3. Resnick D; Diagnosis of Bone and Joint Disorders. Volume 6, Philadelphia, PA: W. B. Saunders Co., 1995: 3882-3884.
4. Taveras JM; Radiology, Diagnosis, Imaging Intervention. Volume 5, Philadelphia, PA: Lippincott Williams & Wilkins, 1993.
5. Kitsoulis P, Charchanti A, Paraskevas G, Marini A, Karatzias G; Adamantinoma. Acta Orthop Belg., 2007; 73(4): 425-431.
6. Gleason BC, Liegl-Atzwanger B, Kozakewich HP, Connolly S, Gebhardt MC, Fletcher JA et al.; Osteofibrous Dysplasia and Adamantinoma in Children and Adolescents: A Clinicopathologic Reappraisal. Am J Surg Pathol., 2008; 32(3): 363-376.