Jharkhand Surgical Chronicles
https://jhsurgicalchronicles.com

Case Report

A case report of a massive swelling of the jaw

Rajesh Kumar Singh[1], Eesha Ashok[2]*

1 Consultant at Devki Hospital, devkihospital@gmail.com
2 Junior Resident, Department of General Surgery, RIMS, Ranchi, eesha.ashok20@gmail.com

Received: 17 May 2024
Accepted: 17 June 2024

***Correspondence:**
Dr. Eesha Ashok
eesha.ashok20@gmail.com

ABSTRACT

The facial appearance of any person is the most important for his confidence and mental well-being, distortion of which can have repercussions on the mental status of anyone.

Background: We hereby present a case of long-standing swelling of the jaw. The growth was massive enough that it restricted side-to-side mobility of the neck and made jaw movement difficult for the patient.

Methods: patients with a growth over their jaw for the last 8 years presented with complaints of difficulty in moving their neck and chewing. His physical appearance made it difficult for him to attend social gatherings and caused a lot of damage to his mental well-being for which he consulted a surgeon. He underwent preoperative FNAC which showed a nonmalignant tumor.

Results: the case was provisionally diagnosed as adamantinoma of the jaw and the patient underwent surgery. The patient has been followed up for the last 8 years and there are no signs and symptoms suggestive of recurrence.

Conclusions: with India being a developing country there is still a lot of social stigma related to such swellings. People belonging to the socially backward category still find it difficult to approach a medical facility. The amount of mental damage met to the person is unmatched and with this report, we wish to emphasize the need to make aware to people about the facilities that are available in our part of the country and the quality of care that is provided so that no other patient needs to suffer for 8 years of his life.

KEYWORDS: adamantinoma, jaw tumor

INTRODUCTION

Adamantinoma is a rare tumor comprising less than 0.5% of all primary skeletal tumors. The term was derived from the presence of adamantine epithelium in the tibia and the intraoral enamel.[1] It is a biphasic tumor with a low potential for malignancy. Thus treatment comprises mostly of en bloc resection of the tumor. There are four varieties of adamantinoma namely: basaloid, squamous, spindle cell, and tubular. There are epithelial structures embedded in a mesenchymal stroma. This can be supported by the keratin positivity(presence of 14 and 19 and absence of 8, 18) observed in these tumors. The stromal component stains positive for vimentin.[1]The adamantinomas could be divided into three variants,i.e.

1) Osteofibrous dysplasia like adamantinoma: this comprises of meagre amount of epithelial cells embedded in the fibro-osseous stroma.

2) Classical adamantinoma: obvious epithelial elements embedded in fibro-osseous stroma.

3) Dedifferentiated adamantinoma: there is a loss of epithelial differentiation in this variety and the histology comprises mostly sarcomatoid change. [1]

In 85-90% of cases, the tumor is localized to the middle third of the tibia thus making the jaw a very rare site of occurrence of the tumor.[2]rarely it is found in the humerus, radius, ulna, metacarpals ribs, etc. [1]Progression to aggressive type is associated with epithelial to mesenchymal transition.[3] It is suggested that the epithelial component of the tumor is the primary proliferating tumor cell population capable of stimulating reactive fibrous growth.[1]

Hypothesis for the origin of adamantinoma:

1)congenital epithelial cell implantation

2)traumatic implantation

3)articular origin[1]

The tumor is associated with a pathological fracture in 16-23% of cases. On x-ray, these lesions appear as eccentric or central, lobular lytic lesions with sclerotic margins. The difference between adamantinomas and dysplasias radiologically is that adamantinomas are located in the diaphysis and extend towards the bone marrow whereas dysplasias are characterized by involvement of the cortex without extension to the canal. Treatment is usually en bloc resection with wide margins including suspicious lymph nodes, and limb reconstruction. The defects can be filled with allografts, vascularised fibular autografts, and prostheses.[1]metastasis develops in 15% of cases, mostly to the lungs and lymph nodes. Risk factors for recurrence or metastasis include a history of less than 5 years, males, younger age, pain at presentation, lack of squamous differentiation, and increased epithelium-to-stroma ratio.[1]

CASE REPORT:

A 30-year-old male presented with a swelling over his left jaw over the last 8 years which gradually increased in size. There was no history of trauma to the jaw. The patient complained of difficulty in moving his jaw and there was restricted mobility of his neck. On inspection, there was a 27x18 cm swelling extending from the lower end of the left ear to the angle of the right jaw. Vertically it extended from 4 cm below the left eye to 5 cm above the level of the nipples. The mouth was deviated to right. There was an ulcer of 8x7cm visible over the lower half of the swelling. The edge of the ulcer was sloping, There was necrotic tissue present at the base. There was no discharge present over the ulcer. There were venous prominence present over the swelling. The lower part of the swelling seemed engorged with color changes. There were no visible pulsations or scar marks over the swelling. On palpation, there was no local rise in temperature over the swelling. The swelling was non-tender. It extended from 2 cm below the zygomatic arch to 12 cm below the clavicle. It was firm in consistency and had restricted mobility. No bruit was heard over the swelling on auscultation.. There was no involvement of muscles of the tongue or muscle of mastication. There was loss of teeth on the left lower jaw namely the central incisors on both sides and the lateral incisor, canine, and premolar tooth on the left side.

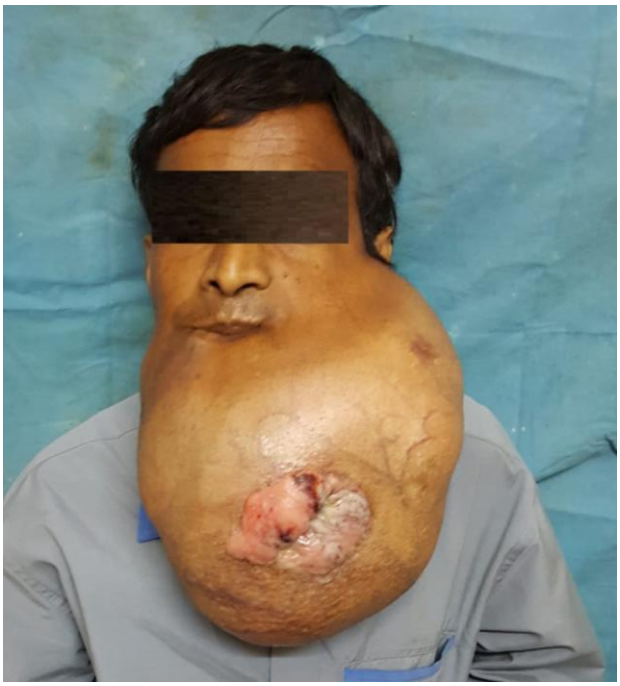

Figure 1: preoperative picture of the swelling.

The patient was subjected to Xray and CECT of the face and neck.

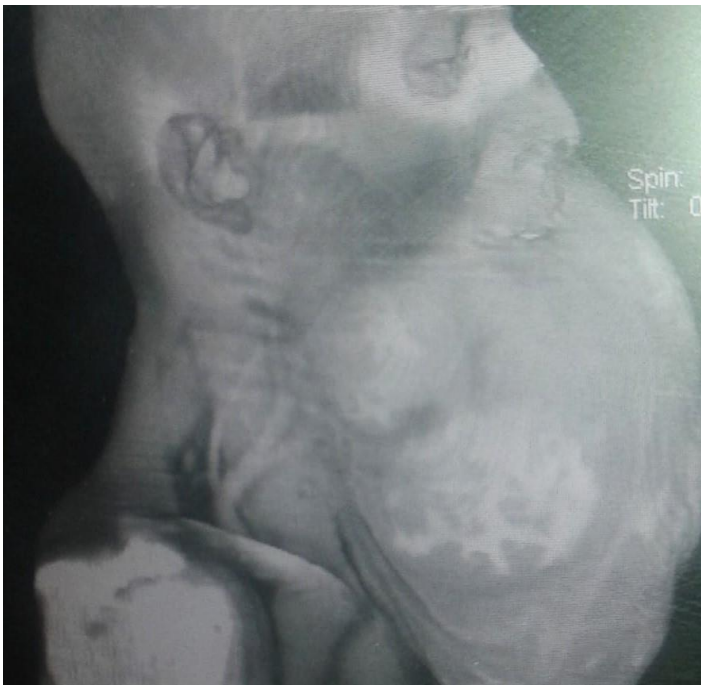

Figure 2: lateral view of the Xray of the patient

The x-ray showed a soap bubble appearance with focal areas of calcification.

The CECT face and neck were suggestive of a multilobulated heterogeneously enhancing soft tissue lesion which caused complete destruction of the mandible. bilateral eyeballs and pterygopalatine fossa were normal. the oral cavity and nasopharynx were normal. There was no involvement of the cervical vessels and no lymph node involvement was seen. Features were suggestive of adamantinoma.

The preoperative FNAC of the swelling showed a non-malignant tumor. The patient was taken up for surgery wherein an incision was made in the left submandibular region extending from the midline to the left infraclavicular region. Excision of the tumor with left mandibulectomy and reconstruction was done with a recon mandibular plate.

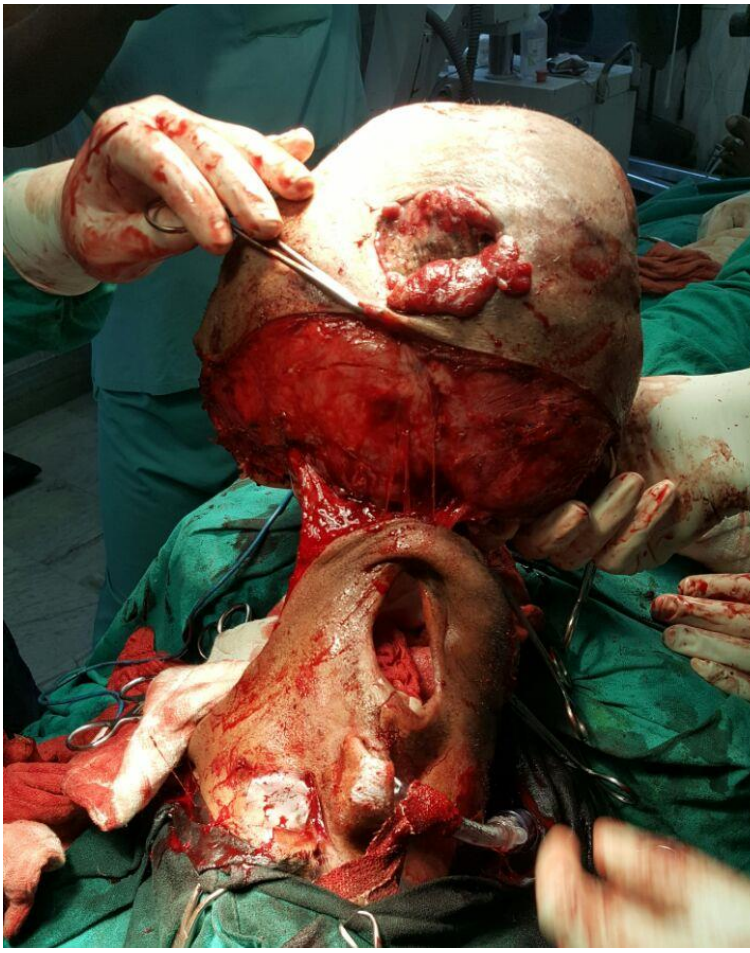

Figure 3 : intraoperative picture

The post operative histopathological examination was suggestive of adamantinoma. The patient has been followed up for 8 years with no signs of recurrence.

Discussion:

Adamantinomas are rare tumors of the bone and are of epithelial origin. The arise in the diaphysis of the tibia commonly but can occur at other sites as well. They arise usually in the adolescent or middle-aged group with slight male-to-female preponderance.[4] it is a slow-growing tumor and is usually associated with pain.

Deformity and pathological fractures are the two common reasons why patients visit a doctor.

Conclusion:

Despite the advancement in technology and the availability of surgeons at every nook and corner in India, a patient still suffers for 8 years before seeking medical help. The mental damage met to the person because of his changed physical appearance and the social stigma is unmatched. With this case report, we wish to highlight the need to make medical help more approachable to the people of India.

Annexure:

Figures and their description:

Figure	Description
Figure 1	Preoperative picture of the swelling
Figure 2	Lateral view of the X-ray of the patient
Figure 3	Intraoperative picture

References

1. Varvarousis DN, Skandalakis GP, Barbouti A, Papathanakos G, Filis P, Tepelenis K, et al. Adamantinoma: An Updated Review. In Vivo 2021;35(6):3045–52.

2. Szendroi M, Antal I, Arató G. Adamantinoma of long bones: a long-term follow-up study of 11 cases. Pathol Oncol Res POR 2009;15(2):209–16.

3. Hazelbag HM, Laforga JB, Roels HJL, Hogendoorn PCW. Dedifferentiated adamantinoma with revertant mesenchymal phenotype. Am J Surg Pathol 2003;27(12):1530–7.

4. Sahito B, Jatoi NN, Kumar D, Asad A, Hasan SS, Ali M. Understanding The Rare Bone Tumor "ADAMANTINOMA." J Ayub Med Coll Abbottabad JAMC 2021;33(Suppl 1)(4):S835–40.