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Case Report

Aggressive Plexiform Ameloblastoma in a Young Girl: A Case Report.

Dr. S. Kumar Mukul[1] ,Dr. A. Kumar[2] ,Dr. E. Ahmad Mokhtar[3],Dr. S. Pandey[4] , Dr. S. Singh

[1]Assistant Professor and HoD Department of Dentistry, AIIMS Patna,Patna.
[2]Resident Doctor, Department of Dentistry, AIIMS Patna, Patna.
[3'4]Resident Doctor, Department of Dentistry, AIIMS Patna, Patna.
[5]Ex Resident, Department of Medicine, AIIMS Patna,Patna.

correspondence Address: Department of Dentistry,Room no.223,Department of Dentistry,Ayush PMR Building, AIIMS Hospital Campus,All India Institute Of Medical Sciences Patna,Patna,Bihar,Pin-801507

ABSTRACT: Jaw tumours in children occur infrequently and diagnosing these lesions and predicting their biological behaviour are difficult. Some of them are not diagnosed correctly at the initial stages as having a neoplasm and are wrongly treated for infections by antibiotic administration. Ameloblastoma is a common and aggressive odontogenic epithelial tumour. It has an aggressive behaviour and recurrent course, and is rarely metastatic. Ameloblastoma represents 1% of all tumours and cysts that involve the maxillomandibular area and about 10% of the odontogenic tumours. We are presenting a case report of a large and aggressive tumour in a 9 year old young female patient who was diagnosed as plexiform Ameloblastoma and was managed by hemi mandibulectomy and reconstructed with condylar recon plate and free iliac crest bone graft.

Key Words: Plexiform ameloblastoma, Spontaneous bleeding, Aggressive nature.

Introduction:

Ameloblastoma is rare tumour in paediatric age group and it accounts for approximately 10-15% of all reported cases (1). Among all variants of Ameloblastoma in children and young adolescent unicystic ameloblastoma is most common type (2)(3) representing less aggressive tumour (4). Plexiform ameloblastoma is reported more frequently in 3rd decade of life(5). We present a case report about the diagnosis, management and follow up of plexiform type of multilocular ameloblastoma in a young female child.

Case Report:

A 9 year old female child reported with a huge aggressive lesion of the right side of lower jaw. The presenting complaints were spontaneous bleeding from the lesion, inability to close mouth and eat, difficult respiration due to huge size of the lesion .The lesion had aggressively grown in the last 5 months. Spontaneous bleeding from the lesion had started in last 4 days. The growth had covered almost entire mouth and pushed the tongue to the other side. The swelling had involved the right middle and lower facial region (Fig 1).

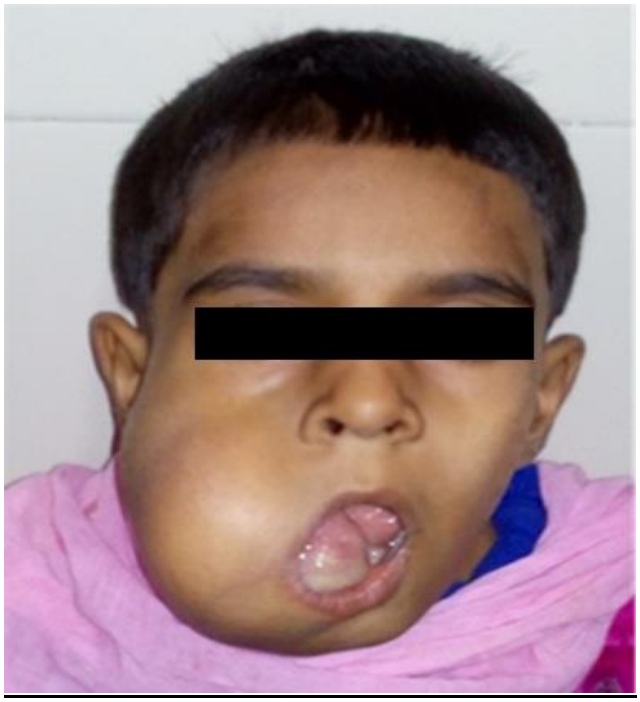

FIGURE 1:

CT scan showed radiolucent lesion involving entire half of right mandible extending up to the condyle (Fig 2).

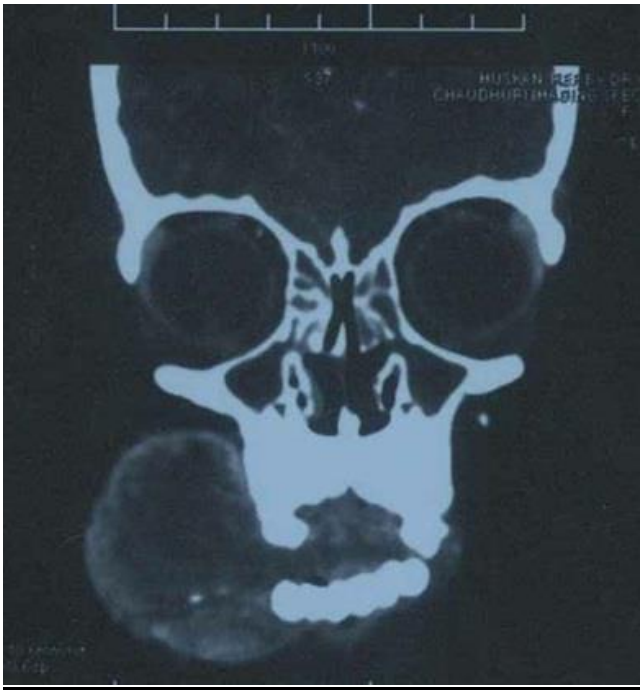

FIGURE 2:

There was a danger of an impending medical emergency due to spontaneous bleeding from the lesion and respiratory distress. Incisional biopsy was deferred as routine histopathology reporting would have further delayed the planned surgery. Right side hemi mandibulectomy with disarticulation, followed by reconstruction with free iliac crest bone graft and condylar reconstruction plate was planned. The iliac crest bone graft was harvested for mandibular body reconstruction and available stainless steel recon plate with condyle was adapted as desired for the defect after wide excision (Fig 3).

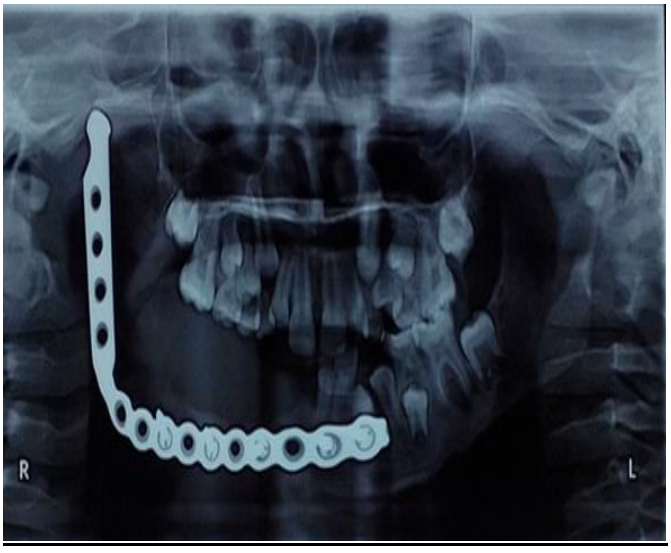

FIGURE 3:

The specimen (Fig 4) was sent for histopathological examination.

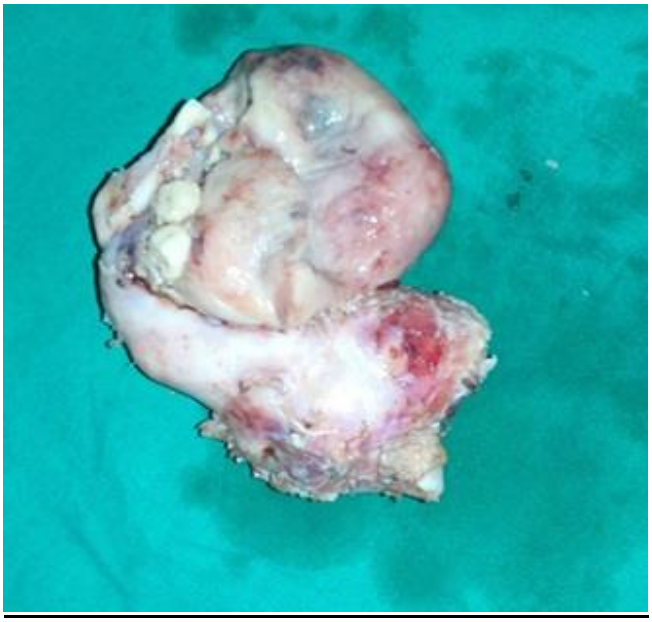

FIGURE 4:

The report showed proliferated epithelial components in the form of Islands, strands and cords within oedematous connective tissue stroma and area of hyalisation with multiple cystic degeneration and fair number of stellate reticulum with squamous metaplasia. The histopathological features was consistent with plexiform ameloblastoma. Periodic follow up as per protocol for benign jaw tumour was followed. After 26 months of follow up there was no occlusal derangement or any draining sinuses over the skin (Fig. 5).

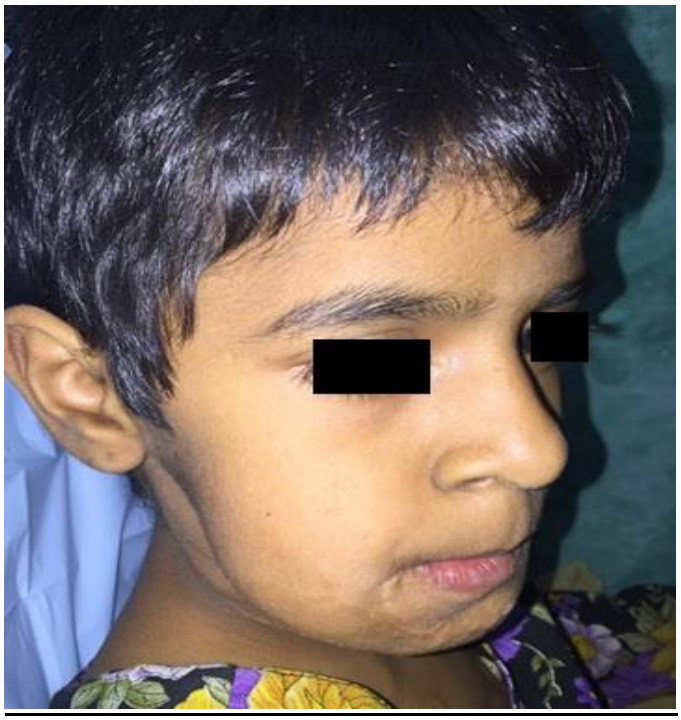

FIGURE *5:*

The iliac crest bone graft had exposed in the oral cavity in the body region. There were no associated problems of speech and mastication. Second surgery for reconstruction with free fibula flap is planned.

Discussion:

Ameloblastoma are not uncommon in Indian paediatric

population (6).Histopathologically ameloblastoma is classified into 6 subtypes :a)follicular b)plexiform c)acanthomatous d)desmoplastic e)granular and f)basal cell type(7).According to 2005 WHO classification Ameloblastoma is classified under3 major category as solid, desmoplastic and hybrid. Solid Ameloblsatopma further classified as Follicular, plexiform and acanthomatous(8). Radiologically they may be unilocular or multilocular .Plexiform type of ameloblastoma present less aggressive form (9). However we are presenting an aggressive case of plexiform type of ameloblastoma as recently reported in few literatures. The treatment of ameloblastoma is controversial in children due to continued facial growth of different bones, presence of unerupted tooth and greater percentage of cancellous bone(10). The treatment varies from conservative approach to radical resection but radical resection should be avoided in children(11) . The different treatment option include enucleation followed by chemical cauterisation with Carnoy`s solution, marsupialisation followed by enucleation, marginal resection or aggressive resection(12). Resection beyond 1.5 cm to 2 cm of radiological limit is the treatment of choice for solid or multicystic ameloblastoma(13).For solid aggressive form of ameloblastoma wide excision with suitable reconstruction is the treatment of choice.

In **conclusion** we emphasize that late presentation of tumour is a frequent problem in low and middle- income countries (LMICs). Many a times immediate surgery due to an emerging emergency (respiratory distress, bleeding) is warranted which imposes limitation in reconstructive plan. In such situations plan for excision and reconstruction with available reconstructive option should be formed. Delayed reconstructive options would depend upon the histopathological report and behaviour of the tumour in the follow up.

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