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RESEARCH ARTICLE

MASSIVE AMELOBLASTIC FIBROMAIN MANDIBLE: A CASE REPORT

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ABSTRACT

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Key Words:

Ameloblastic Fibroma, Odontogenic Tumors, Mesenchyme, Enucleation. Ameloblastic fibroma is an extremely rare, true benign mixed odontogenic tumor with male predilection that can occur in either mandible or maxilla but most frequently found in posterior region of mandible. It usually occurs in first two decades of life. It exhibits both epithelial and mesenchymal components. This lesion was previously considered to be benign lesion with very limited recurrence rate and malignant transformation.

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INTRODUCTION

Ameloblastic fibroma is of odontogenic origin and is characterized by proliferation of epithelial and mesenchymal tissues with absence of any calcified dental structure⁽¹⁾. It is associated with tooth enclosure causing delay in eruption or alters the dental eruption sequence. A conservative treatment strategy such as enucleation and curettage is usually sufficient however extensive lesions require radical treatment. Here, we are presenting a case of ameloblastic fibroma in a 27 years old female patient.

Case Presentation: A 27 years old female patient reported to the Department Of Dental Surgery, Mahatma Gandhi Govt Medical Hospital, Trichy, Tamil Nadu with the chief complaint of swelling in lower jaw for past 4 years. On extraoral examination prominent swelling was seen in mandibular anterior region (Fig.1). Swelling size ranged about $4.5-5 \times 6.5-7$ cm extending from left parasymphyseal region to the ramus of mandible on right side. . Skin over the swelling appeared to be pale, stretched and shiny. The lesion had a sudden onset and had increased in size gradually over time. Upon intraoral examination patient reported mobility in 31 and 41. Upon palpation bony hard, firm and non tender swelling with buccal and lingual cortical plate expansion was found. No lymph nodes were palpable in submandibular and submental region. There was obliteration of labial vestibule (Fig.2). and occlusion was dearranged

Investigations

- OPG was taken, which revealed multilocular radiolucency which is exteding from 35 to 45 region (Fig.3).
- CT scan of face showed sclerosis with cortical expansion of mandible at mentum and parasymphyseal region.
- FNAC was taken, smear study shows occasional neutrophils in a proteinaceous fluid background.
- Incisional biopsy done in facial surface of lesion and specimen submitted for Histo Pathological Examination. Microscopically, section studied shows lesion admixed with bony elements exhibiting bland fibroblasts with collagenous tissue and focal islands and cords of ameloblastic epithellial cells suggestive of Ameloblastic fibroma.

Differential Diagnosis: Ameloblastoma, Odontogenic myxoma, keratocystic odontogenic tumor, Central giant cell lesion.

Treatment: Patient's Ct scans were converted to stl files using Slicer 3D software and the 3D model of patients skull was made. (Fig 4 & 5) After segmentation a steriolithographic model of mandible was prited upon which preoperative planning of surgey was done. (Fig 6&7) A 2.7mm locking reconstruction plate was adapted on the model which was used later in the surgery. Under GA and nasoendotracheal intubation extraoral apron incision was given and mandible



Fig.1. Preoperative extraoral picture shows swelling in mandible



Fig. 2. Preoperative intraoral picture



Fig.3. OPG shows multilocular radiolucent lesion in anterior mandible



Fig. 4. Dicom to stl conversion - 3D model preparation



Fig 5. Segmented mandible 3d model



Fig. 6. 3d printed rapid prototyping model of the mandible



Fig. 7. Reconstruction plate adapted on the model



Fig. 8 Intraoperative view mandible with lesion exposed



Fig. 9. Resected specimen with involved teeth



Fig. 10. Intraoperative view with reconstruction plate

was exposed. Buccal and lingual facial artery were dissected, cut and ligated, 46 and 35 were extracted. The lesion was exposed fully intraorally and extraorally. The lesion was removed with the help of Gigli saw (Fig.8 & 9). The osteotomies were made in sound bone with 1cm margin from lesion. The remaining mandible was reconstructed using 2.7mm titanium unilock reconstruction plate with 11 2.5mm titanium unilock screws (Fig.10). The wound was irrigated using metrogyl and saline. The wound was closed in layers using 2-0 vicryl interupted sutures and 3-0 ethylon interupted sutures extraorally. Ryles tube was placed. Resected specimen was sent for Histo Pathological Examination which revelaed tumor mass insinuating between bony trabeculae and exhibiting islands of odontogenic epithelial cells with Peripheral palisading appearance and central stellate cells surrounded by fibromyxoid stroma suggestive of ameloblastic fibroma.

DISCUSSION

Ameloblastic fibroma is a neoplasm of odontogenic epithelium and mesenchymal tissues and is categorized as mixed odotogenic tumor. Ameloblastic fibroma is considered as tumor of childhood and adolescence and occurs almost exclusively in first and second decades of life. Ameloblastic fibroma has been known to affect men more commonly than women. Grossly ameloblastic fibroma appear as firm,lobular soft tissue mass with a smooth surface (Cohen, 2004). Ameloblastic fibroma may behave as true neoplasm or hamartomatous proliferation of odontogenic epithellium of enamel organ and odontogenic mesenchyme of primitive dental pulp (Verma, 2016). It is a painless, slow growing and expansile tumor, similar findings were reported in present case.

This pathology exhibits slower clinical growth than simple ameloblastoma. It enlarges gradually. Most common presenting symptom is swelling (Nelson, 2009). The density of collagen fibres in ameloblastic fibroma has an impact on the shape and direction of enlargement of epithelial follicle. Its growth is found to be restricted in areas of dense collagen deposits, leading to its enlargement in planes of less resistance (Hangelbroek). Radio graphically unilocular radiolucency with smooth outline is associated with asymptomatic patients while cases with jaw swelling gradually have multilocular radiolucent pattern in most cases exhibiting a sclerotic radiopaque boundary (Chen, 2005). Multilocular appearance is often seen in larger tumors and a unilocular pattern charcterises the smaller lesions. When associated with an unerrupted tooth the tumor may radiographically mimic dentigerous cyst (Cohen, 2004). Microscopically ameloblastic fibroma is composed of connective tissue background that appears to recapitulate dental papilla resembling stellate reticulum (Cohen, 2004). This tissue is composed of spindled and agular cells with little collagen, imparting myxomatous appearance. The epithelial component is made up of thin branching cords of cuboidal or columnar cells or small nests of odontogenic epithellium with little cytoplasm and basophillic nuclei. Larger nests may show central area of stellate reticulum. Ameloblastic fibroma does not show mitosis (Kruse, 1891). Aspirates from ameloblastic fibromas closely ressemble those of ameloblastoma. Histologically ameloblastic fibroma is characterized by proliferation of odontogenic epithellium supported by primitive mesenchymal connective tissue stroma. The presence of raged - edged aggregates of hypercellular stroma distinguishes between two lesions. Conservative surgical approach such as enucleation with curettage of surrounding bone along with removal of affected teeth is treatment of choice (Tomich, 1999). Recurrence rate varies among sources, but is considered to be low. Aggressive therapy may also be performed which includes modified block resection of mandible with the placement of immediate autologous bone graft for an extensive tumor and or multiple recurrences. Recurrence commonly results from incomplete removal of tumor (Munde, 2013). Complete excision with close follow up is highly recommended. As we have done in the present case of extensive ameloblastic fibroma of the mandible in a female adult patient.

Abbreviations

OPG- Ortho Pantomo Graphy, FNAC- Fine Needle Aspiration Cytology, CT- Computed Tomography, GA General Anesthesia.

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