Volume 9, Issue 12, December – 2024 International Journal of Innovative Science and Research Technology
ISSN No:-2456-2165 https://doi.org/10.5281/zenodo.14576713

Unusual Presentation of a Maxillary Ameloblastoma: A Clinical and Histopathological Review

Julius Nyang'ombe[1,2]; Peter J. Ngala[2]; Lisa O. Massawe[2]; Sales Nicco[1]; Bundala Ramadhani[1]
[1]Department of Dentistry, Bugando Medical Centre, Mwanza Tanzania
[2]Department of Dentistry, Faculty of Medicine, Catholic University of Health and Allied Sciences - Bugando, Mwanza, Tanzania

Abstract:- Ameloblastoma is the most prevalent non-cancerous tumor originating from the dental epithelium. It arises in both the mandible and maxilla but with a higher occurrence in the posterior region of mandible. Ameloblastoma is characterized by its benign but locally aggressive features of a slow growing tumor ultimately causing expansion of the bone with displacement and resorption of surrounding structures. Herein, the authors describe a case report of a 22-years-old female who presented with an ulcerated and easy bleeding swelling on the left side of the midface which was clinically similar to a malignant tumor, however first histopathological diagnosis following incisional biopsy established the diagnosis of odontogenic myxoma, second phase incisional biopsy established the diagnosis of ameloblastoma all of which are benign lesions. The tumor was surgically removed and the defect was repaired by a rotational cheek flap to achieve satisfactory functional and cosmetic results. Clinical, radiological and pathological characteristics as well as surgical treatment approaches are further discussed. This was one of a kind unusual presentation of a maxillary ameloblastoma encountered in our setting.

Keywords:- *Benign, Maxilla, Ameloblastoma, Locally Aggressive, Odontogenic Tumor.*

I. INTRODUCTION

The World Health Organization classifies ameloblastoma as a benign intraosseous epithelial odontogenic tumor, marked by gradual growth that damages nearby tissues and a propensity for local recurrence if it is not adequately removed[1] . It is a locally aggressive neoplasm that arises from rest cells of dental lamina, developing enamel organ, epithelial lining of odontogenic cysts, basal cells of oral mucosa[2] .The projected frequency of ameloblastoma is around 0.5 cases per million people annually, making up 11% of all odontogenic tumors with most cases being diagnosed between 20 and 40 years of age. Maxillary ameloblastoma develops 12 years later than its mandibular counterpart. Approximately 15-20% of ameloblastomas have been reported to originate in the maxilla, with 2% arising anterior to the premolars[2], [3]. Although rare, involvement of the maxillary sinus and nasal cavity regions has been documented[1]. Ameloblastoma is characterized by slow growth, with microscopic projections called pseudopods extending to the medullary spaces of the bone, this renders the tumor to have poorly defined radiological and surgical margins making the tumor have high recurrence following inadequate surgical removal, exhibiting a locally malignant pattern[4]. The conventional radiographic presentation of ameloblastoma ranges from a unilocular or multilocular radiolucency resembling a cyst. The lesion may remain asymptomatic before the development of a facial swelling. Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) can be useful for determining the extent of the lesion, especially when it is located in the maxilla[2]. Ameloblastoma is categorized into four subtypes namely conventional ameloblastoma, unicystic ameloblastoma, extraosseous ameloblastoma and metastasizing ameloblastoma[5]. The histological variants of ameloblastoma include; follicular, plexiform, acanthomatous, granular cell, desmoplastic and basal cell types[6]. The aim of this article is to illustrate an atypical correlation of the clinical presentation and histological findings of a maxillary tumor in a 22-year-old female.

II. CASE PRESENTATION

A 22-year-old female patient reported to our facility at 13th June 2024 with a chief complaint of a painless, slow growing mass on the left side of the mid-face for 4 years. The swelling started as a small nodule that progressively increased in size with time initially associated with toothache and mobility of the teeth around the swelling. As the swelling progressed it started to ulcerate accompanied with episodes of spontaneous bleeding around the ulcer. On examination patient was cachexic. There was a facial asymmetry due to a swelling on the left side of the midface, extending from the infra-orbital rim to the corner of the mouth superior-inferiorly and from the right naso-labial fold to the zygoma area crossing the midline whose measurements are about 18 by 16 cm in antero-posterior dimension. The swelling was firm, non-tender to palpation and fixed to the underlying structures. The skin overlying the swelling was ulcerated, the ulcer measured about 10 by 8 cm in dimensions, the ulcer was elliptical with everted edges and pale pink in color, the floor of the ulcer had a greyish-yellow slough discharging serosanguineous fluid. Base of the ulcer had no signs of induration (Fig 1A-1B). On examination of the oral cavity, the swelling extended from the labial sulcus involving much of the alveolar process and the junction between hard and soft palate antero-posteriorly and from the maxillary tuberosity on

ISSN No:-2456-2165 https://doi.org/10.5281/zenodo.14576713

the left to the distal of tooth 11 crossing the midline in medial-laterally direction. There was no regional lymphadenopathy, speech and swallowing functions were unaffected. Computerized tomography showed a large lobulated heterogenous enhancing soft tissue mass involving the left side of the maxilla, extending to involve the left maxillary sinus, left nasal cavity and the mouth, it is associated with bone erosion and destruction of the maxilla and nasal bone and opacification of the left nasal cavity and left maxillary sinus [Figure 2]. The first preoperative histological diagnosis showed the evidence of an odontogenic myxoma at the facility where she was referred from, the second preoperative histopathology of an incisional biopsy which was done at our facility demonstrated a multicystic tumor with predominant follicular growth pattern of columnar shaped odontogenic cells of uniform morphology accompanied by hyperchromatic nuclei with peripheral palisading cells, areas of stellate reticulum formation were evident. A diagnosis of a conventional ameloblastoma of the maxilla was confirmed and treatment was scheduled.

A. Pre-Operative Photographs

Fig 1: [1A-1B]: Shows an Ulcerated Swelling on the Left Side of the Face [18*16 cm]

B. Pre-Operative Radiographs

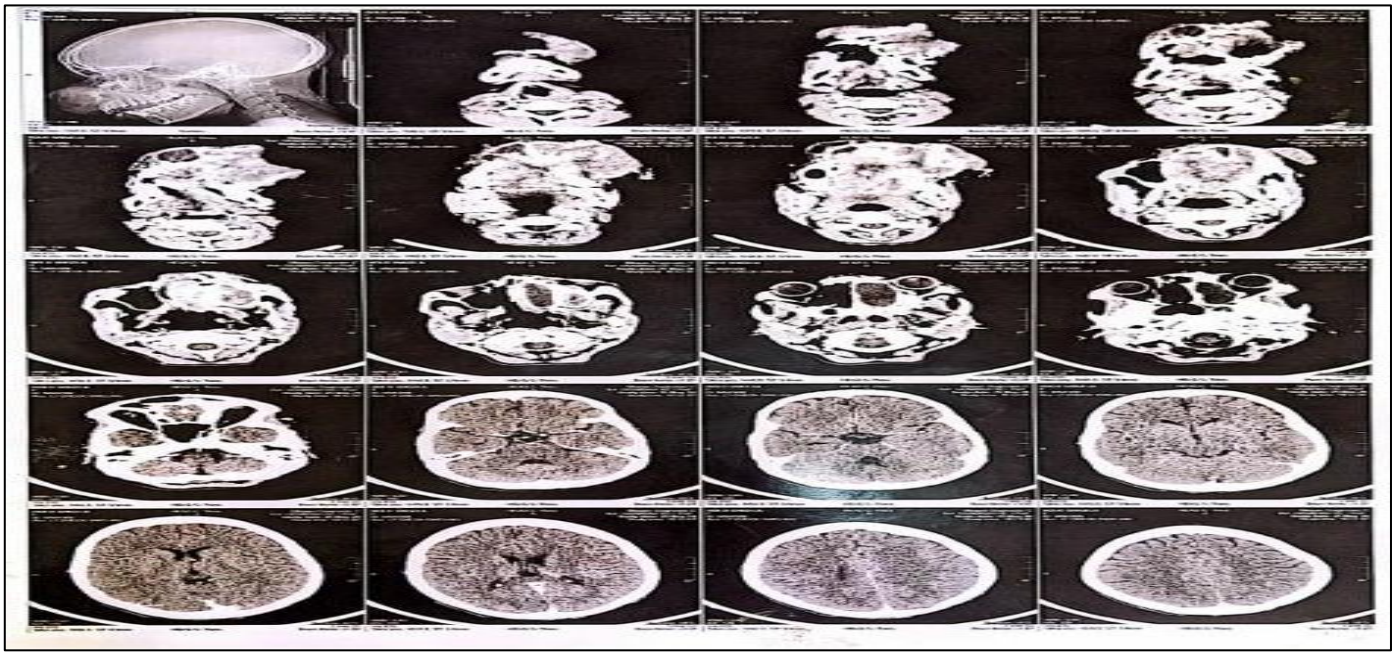

Fig 2: Multiple Axial Cuts of the CT-Scan Showing Lobulated Soft Tissue Mass Involving the Left Maxilla, Obliterating the Left Maxillary Sinus and Nasal Cavity with Bone Destruction

ISSN No:-2456-2165 https://doi.org/10.5281/zenodo.14576713

➢ *Surgical Procedure*

Under general anesthesia, patient was orally intubated. The incision site was marked around the tumor through preauricular with retromandibular and temporal extension. Under aseptic technique an incision was made then tissues were deflected using blunt dissection. The tumor was then exposed. The tumor was a multilobulated mass with nodules of rubbery consistency, bleeding and discharging brownish fluid. Periosteal elevator was used to detach the tumor intraorally, from distal of tooth number 11 which was also resected together with the involved maxillary tuberosity. The remaining masses were removed using periosteal elevator, the defect left was significant, exposing the nasopharynx, the nasal septum and maxillary sinus. Therefore, the defect was repaired by firstly undermining the tissues under the incision to loosen the skin and subcutaneous tissues and then sutured using vicryl 3-0 on the mucosa, muscular and subcutaneous layers, then using nylon 4-0 on the skin[Figures 3A-3F]. Hemostasis was achieved using dry gauze compression, clamping with hemostat, suturing and cold cauterization. Sterility was successfully achieved throughout the surgery.

C. Intra-Operative Photograph 3(A, B,C,D,E,F)

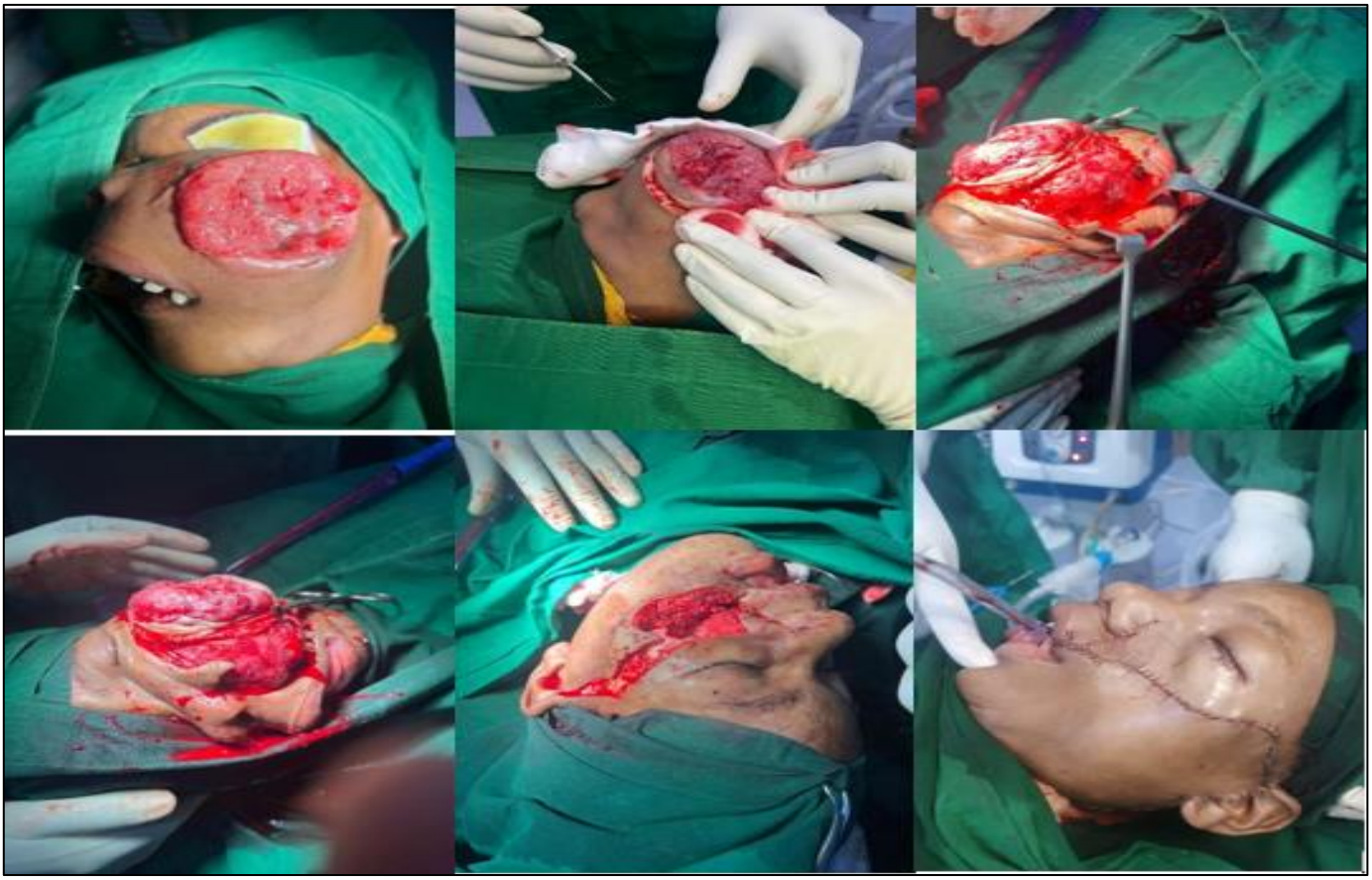

Fig 3: [3A-3F]: Surgical Approach used to Obtain Satisfactory Esthetic and Functional Outcome

D. Post-Operative Radiographs

Fig 4: One week Post Operative Outcome

ISSN No:-2456-2165 https://doi.org/10.5281/zenodo.14576713

Fig 5: One Month Post Operative Outcome

Fig 6: Three Months Post Operative Outcome

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III. DISCUSSION

Literature reports that ameloblastoma develops from residual cell proliferation of enamel organ, evolution of epithelium cells of odontogenic cyst, basal layer of the oral mucosa[7]. Ameloblastoma is relatively common among young patients with a mean age in the third and fourth decade of life[4], [8], our patient's age was 22 years which falls within the third decade of life. Ameloblastoma has a relatively higher occurrence in the mandible than the maxilla[8], our present case was atypical in that the tumor's location was in the maxilla. An incidence of as low as 1% of maxillary ameloblastoma was reported to occur, among those, 47% occur in the molar region, 15% in the antrum and floor of the nose, 9% in the premolar and canine region and 2% in the palate[7][9]. Owing to the presence of abundant cancellous bone and the proximity of the maxilla to the nasal cavity, nasopharynx and the maxillary sinus the tumor has a high propensity to spread to these regions and render their delay in diagnosis[4]. Our patient's maxillary tumor involved the left maxillary sinus. The commonest symptoms of a maxillary ameloblastoma include but not limited to facial swelling, malocclusion, loosening of teeth, ulceration and nasal airway obstruction[10], similar clinical presentations were observed in our patient. However the bleeding that accompanied the tumor could not be accounted for in our patient but has been reported to occur in cases of ameloblastic carcinoma[11]. Diagnosis of maxillary ameloblastoma does not solely depend on radiographic and clinical presentations due to the overlapping characteristics from other tumors such as odontogenic myxoma, adenomatoid odontogenic tumor and ameloblastic fibroma which become their differentials. In this case histopathologic confirmation is the gold standard[12]. The author describes the histological report of ameloblastoma as odontogenic epithelial islets simulating the stellar reticulum, peripherally palisaded by cylindrical cells with inverted nuclear polarity, filled with angular cells loosely arranged[5], which were consistent with our pre-operative and post-operative histopathological diagnosis. The recommended treatment of ameloblastoma is wide resection of the jaw in attempt to prevent recurrences however this comes with undesirable effects such as facial deformities, masticatory dysfunction and abnormal jaw profile[10]. Ameloblastoma of the anterior maxilla could be treated conservatively by enucleation and curettage due to sufficient distance from vital structures whereas those of posterior maxilla extension require a more radical treatment such as partial, hemi or total maxillectomy[13]. In our case, the patient underwent partial maxillectomy due to posterior extension of the tumor with 1cm margin of clinically normal bone.

IV. CONCLUSION

Maxillary ameloblastoma as the one encountered in our setting is quite rare, let alone presenting with features of malignancy which are bleeding and ulceration that made this case so bizarre. Despite having no signs of malignancy transformation from the post-operative histological report, a close follow up for the patient is of significance for detection of signs of recurrence and possible early intervention as it is with our case [Figures 4-6]. Patients should also be informed about the possibilities of reconstruction following at least six months recurrence free so as to improve quality of life including speech and esthetics.

AUTHOR CONTRIBUTIONS

- Peter J Ngala: Conceptualization; data curation; writing – original draft.
- Julius K Nyang'ombe: Conceptualization; investigation; data curation; writing – review and editing.
- Lisa Oscar: Data curation; writing – review and editing.
- Sales Nicco: Data curation; writing – review and editing.
- Bundala Ramadhani: Data curation; writing – review and editing.
- Funding Information: None

ACKNOWLEDGEMENTS

The authors would like to thank the patient and her family for allowing us to use her medical information for academic purpose. We also acknowledge Peter Bwire, Asha Ally, Regan Mwaijumba, David Mattoke, Limbe Luhela, Fodensiana Mbele, Thomas Matatizo, Nyangwida Mangaida and the BUGANDO hospital leadership for supporting this study.

CONFLICT OF INTEREST STATEMENT

The authors declare that there are no conflicts of interest regarding the publication of this paper.

DATA AVAILABILITY STATEMENT

There is no data generated from this study.

CONSENT

Written informed consent for publication of clinical detail and images was obtained from the patient.

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ISSN No:-2456-2165 https://doi.org/10.5281/zenodo.14576713

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