

Adenomatoid Odontogenic Tumor of the Mandible: A Rare Case Report

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Abstract

Adenomatoid odontogenic tumor is a rare tumor accounting for only 2.2-7% of the tumors originating in the oral cavity. The origins of this rare, slow growing tumor has often been the topic of debate amongst oral diagnosticians and its limited literature can often pose a challenge to diagnosis. The following case report is of a 17 year old diagnosed with adenomatoid odontogenic tumor of the mandible. Surgical enucleation of the entire lesion was done followed by a histopathologic diagnosis and the patient was followed up with for 6 months and then referred for aesthetic rehabilitation. Through this article we attempt to report an exceptional occurrence of a this rare tumor in the mandible which is otherwise commonly seen in the maxilla along with the diagnostic and surgical challenges it may pose for clinicians that may help outline the importance of a holistic approach towards diagnosis and treatment planning.

Keywords :

Adenomatoid
Tumor
Odontogenic
Diagnosis
Progressive
Surgical
Enucleation
Swelling
Benign
Adeno-ameloblastoma
Rossette

Date of Submission: 28-11-2020

Date of Acceptance: 13-12-2020

I. Introduction

Since its mention in 1915 by Habitz to its WHO histological typing in 2005 there have been various case reports stating the varied clinical and microscopic features of the Adenomatoid Odontogenic Tumours.¹ What was once believed to be a class of ameloblastomas, referred to as adeno-ameloblastoma with or without cystic variation was later adapted as a tumour on recommendation by Philipsen and Brin.^{1,2} Adenomatoid Odontogenic tumour is one of the most uncommon benign tumours of the oral cavity debated to be odontogenic in origin with a frequency of occurrence as low as 2.2% to 7%.¹ It is commonly referred to as 2/3rd tumour due to the common occurrence in about 2/3rd cases in the maxilla, with 2/3rd of cases arising in young females and

generally associated with unerupted teeth and canines in 2/3rd of the cases.³ Another frequent location for occurrence of this tumour is found to be the periapical region of teeth which can frequently lead to a misdiagnosis of periapical cyst.⁴

The exact histogenesis is still unknown but due to the increased frequency of occurrence associated with the crown of the unerupted teeth it is believed to have an odontogenic origin and demonstrates close association with dental lamina or odontogenic epithelium.⁵ The epidemiologic data available, shows an increased prevalence of Odontogenic tumours amongst African races as compared to that in Caucasians.⁶ Keeping in mind the above stated epidemiologic data it has been hypothesized that there may also be a frequent occurrence of cases of Adenomatoid Odontogenic tumours in a similar fashion.⁵ Further epidemiologic data has also shown that large number of the cases have occurred in South East Asian populations with 45.5% of the cases having being reported in Asian countries with a female to male ratio of 2:1.⁵

Adenomatoid Odontogenic tumours occur in three variants mainly, of which two are intraosseous in nature.⁵ The three most common variants have been explained below:

A. Central :

1. Follicular type: It is the most common variant generally associated with an unerupted tooth and is seen located intraosseously. It is commonly referred to as the “Dentigerous” variant and most often gives of a provisional impression similar to that of a dentigerous cyst.⁵

2. Extra Follicular type: It is generally not associated with the crown of an unerupted tooth. It is frequently provisionally misdiagnosed as any other non-developmental odontogenic cyst.⁷

B. Peripheral:

They are located extraosseously and may resemble a fibroma or an epulis.⁵ The most common location of occurrence is the gingival tissue.⁵ From all the different variants of Adenomatoid Odontogenic tumour this particular variant is seen the least and at times may be the simplest to treat.^{2,5}

All the clinical variants irrespective of their location are characterized by gradual progressive growth with cortical expansion.⁵ Displacement of the roots of the teeth involved is often noted more commonly than resorption.⁵ As mentioned earlier 2/3rd of the cases reported in literature have mentioned an occurrence of AOT in the maxilla but the occurrence of AOT in the mandible is yet regarded as a rare occurrence.⁸ This rare occurrence should be noted with the earlier mentioned data that AOT comprises of less than 10% of the odontogenic tumours which makes the occurrence of this tumour in the mandible noteworthy.⁸

Adenomatoid Odontogenic tumour shows the presence of a capsule which encircles the entire tumour, this capsule is clearly visible on radiographic scans.⁵ The capsule enclosing the tumour has a reddish brown color and is slightly thin to thick on palpation.^{1,5} The tumour exhibits marked consistency in histological findings. There are characteristic varying sized solid or spindle shaped cells that are seen arranged in nodules, which forms nests or “Rosette” like structures with minimal stromal tissue surrounding them.⁵ Eosinophilic droplets are seen at the centre of the rosette like configurations.⁵ Tubular or duct like spaces are present lined by cuboidal or columnar epithelium with nuclei that are polarized away from the luminal surfaces of the ducts or acini, with the luminal surface seen filled with either eosinophilic materials or cellular debris.⁵ The duct shaped structures that are often seen show marked variation in diameter from person to person, they may sometimes be present as convoluted cords of cuboidal or columnar cells or as bodies with complicated patterns, which may also feature invaginations.⁵ A very rare variant may show the presence of polyhedral, eosinophilic, squamous epithelial cells with well-defined boundaries and prominent intercellular bridges.⁵ The presence of islands of amyloid like material and translucent calcified substances has also been reported often leading to misdiagnosis of Adenomatoid Odontogenic tumours as Calcifying Epithelial Odontogenic tumour.⁵ The periphery of the tumour shows the presence of 1-2 layers thick epithelial cells with evidence of mitotic activity, which may explain the growth of the capsule along with the tumour.⁵ The stromal tissue of AOT is found to be very scarce and loose with congested blood vessels, which gives of an overall congested appearance to the tumour under the microscope.⁵

II. Case Report

A 17 year old reported to our department with the chief complaint of swelling on the left side of the face near the chin region since 3-4 months (refer to Figure 1). The swelling had gradually increased in size over the last 3 months. The patient gave a negative history of any pain or parasthesia associated with the same and also had taken no medication for same. On further questioning the patient mentioned that she was regular with her oral and dental hygiene and had no history of any substance abuse. A complete medical history was taken which revealed no past systemic illness and a negative history of any medical and dental treatment.

Clinical Features:

Further examination and palpation of the swelling revealed that it measured up to 2cm by 2cm approximately. The overlying skin was intact with no redness or evidence of inflammation. The patient presented with no tenderness extraorally or intraorally. The swelling was ovoid in shape, with smooth texture and had diffused edges. It was firm on palpation with no fluctuation or indentation and the swelling was not mobile. Trans illumination test for the swelling was negative. On intraoral examination the swelling was seen (refer to Figure 2) extending roughly from the distal surface of the mandibular lateral incisor in the fourth quadrant to the mesial surface of the mandibular 2nd premolar in the third quadrant. The swelling was roughly 2cms in diameter and was seen obliterating the buccal vestibule. The mucosa overlying the swelling was stretched and translucent. On palpation egg shell crackling sound was heard over the swelling which confirmed the expansion and thinning of the buccal cortical plate. Dental examination revealed no sign of carious involvement of the affected teeth (refer to Figure 2) and the teeth in the vicinity of the swelling. The permanent mandibular canine on the left side in the third quadrant was missing and bilaterally over retained deciduous mandibular canines were present in the third and fourth quadrants. The teeth involved within the swelling region in the third and fourth quadrants showed no signs of mobility, but from the lateral incisor in the fourth quadrant to the 2nd premolar in the third quadrant showed marked displacement. All the teeth involved were negative for any sign of pain on percussion and showed no signs of fracture. All other parameters of dental examination revealed no significant findings.

Radiographic features:

An orthopantomogram was advised for the patient, which showed the presence of a well-defined radiolucent lesion extending from the mandibular lateral incisor in the 4th quadrant to the mesial surface of the mandibular 1st premolar in the 3rd quadrant (refer to figure 3). The roots of the mandibular second premolar and the mandibular central incisor showed displacement due to enlargement of the cystic cavity in the third quadrant. The radiolucent lesion seen in the OPG was closely associated with the crown of the unerupted permanent mandibular canine. The unerupted canine was also noted to be displaced towards the inferior border of the mandible in the O.P.G. A CBCT was also advised to the patient to confirm the findings of the O.P.G. (refer to Figure 4,5,6)

A fine needle aspiration was carried out for the lesion intraorally following the radiographs and the liquid on aspiration was found to be straw colored. All routine hematological investigations were done for the patient and were within normal parameters. At this point the provisional diagnoses given to the patient were those on the lines of a dentigerous cyst, adenomatoid odontogenic tumour, odontogenic keratocyst etc.

Surgical procedure:

The patient was taken up for an excisional surgical biopsy under general anesthesia after taking an informed consent from the patient, having explained all the due risks involved in the same. An excisional biopsy was agreed upon due to the extent of the lesion and the provisional diagnosis that were taken into consideration. The patient was scrubbed in routine surgical pattern and saline with Adrenaline 1:100000 was infiltrated from the right mandibular second premolar up till the left mandibular first molar around the region of the lesion buccally and lingually. An incision was taken from the mesial surface of the mandibular first premolar in the 4th quadrant to the mesial surface of the first molar in the 3rd quadrant. A full thickness mucoperiosteal trapezoidal flap was elevated and the underlying bone was exposed. The bone was paper thin on exposure and the buccal cortex of the mandible was expanded from the mandibular central incisor of the 4th quadrant up to the mandibular first premolar in the 3rd quadrant. This paper thin bone overlying the lesion was removed using rongeurs. The underlying lesion present was well encapsulated with the involvement of the crown of the permanent mandibular canine (refer to Figure 7). With the use of periosteal elevators the capsulated lesion was separated from the underlying healthy bone and dissection was continued in the similar plane until the entire lesion was separated from the mandible. The cystic lining was enucleated in totality along with the contents of the cystic cavity and the impacted permanent mandibular canine (refer to Figure 8). On enucleation the surrounding bone and the walls of the cavity were curetted and flushed to remove any remainders of the cystic lining. On enucleation grade-3 mobility was observed with respect to the central and lateral incisors which were then extracted. The over retained deciduous mandibular canine in the third quadrant showed a resorbed root and hence was extracted (refer Figure 9). Haemostasis was achieved with pressure packs and the incision was closed with 3-0 vicryl sutures. Simple interrupted sutures were used throughout the incision (refer to Figure 10). Extensive pressure pack was given to the patient to apply pressure at the site of the surgery for two hours.

Histopathology:

The cystic lesion measuring 2.4 x 1.2 x 1 cms along with the extracted teeth were sent for histopathological investigation. The external capsule showed a brownish discoloration and the internal surface was soft to firm with whitish areas of congestion. Further investigation revealed a fibro collagenous cystic cavity wall that was well lined by attenuated squamous epithelial lining. Proliferation of epithelium was observed forming solid masses composed of whorled nodules of spindle and columnar shaped cells with

polarized nuclei. The cells were arranged in a duct like pattern and showed proliferation into tubular structures. The overall impression and histopathological diagnosis confirmed adenomatoid odontogenic tumour.

Post excision the patient was kept under observation in post-operative recovery for 24 hours and discharged with instructions to maintain oral hygiene and follow up with the department on an outpatient basis weekly for one month and then monthly for 6 months. The patient reported no further complaints and was referred for prosthetic rehabilitation of the extracted teeth.

III. Discussion

The potential for malignant transformation is very less for AOT hence treatment by excision of the lesion in totality rarely shows any remissions.^{4,5} Patients with adenomatoid odontogenic tumour show a high recovery rate and are able to resume to their daily activities.⁵ Conservative surgical excision is the treatment of choice for AOT lesions, newer methods such as guided tissue regeneration may show promising results in rebuilding the large bony defects.⁸ The rehabilitation of the patient including recovery post-surgical procedures is highly uneventful unless complicated by secondary factors.^{4,6} In this case the patient's post-surgical follow up was as expected for AOT cases and due to the younger age of the patient, healing was faster and recovery was completed within a span of 2-3 months. The patient was then referred for further rehabilitation. An important factor to be considered when dealing with patients suffering from such pathologies is taking into account the timely referral for further rehabilitation following recovery from surgery. A histopathological diagnosis should always follow the surgery to rule out any risk of malignant transformation that may be present.^{2,4,5} The origin of AOT still remains a topic that is highly debated amongst different schools of thought.⁸ The ongoing immunohistological studies have indicated the expression of cytokeratins CK 15, CK 17, CK 19 similar to follicular cysts, which draws focus towards an odontogenic epithelial origin.⁸

IV. Conclusion

The occurrence of AOT in the mandible is extremely rare and very few cases have been reported.⁸ In this instance except an uncommon location the tumour exhibited not much variation from previously documented cases. A notable factor was the presence of bilaterally over retained deciduous mandibular canines which were undiagnosed on account of negligence of the patient in seeking professional dental care. The overall progression of Adenomatoid Odontogenic tumours irrespective of its variants appear to be gradual in progression and pain free, which makes the management along with the prognosis excellent.⁷ Although more focus should be on developing better and faster diagnostic criteria for benign lesions which would assist in better management and faster recovery.

References

- [1]. Garg D, Palaskar S, Shetty VP, Bhushan A. Adenomatoid Odontogenic tumor-hamartoma or true neoplasm: A case report. *J Oral Sci.* 2009; 51: 155-9.
- [2]. Phillipsen HP, Brin H. The adenomatoidodontogenic tumor, ameloblasticadenomatoid tumor or adeno-ameloblastoma. *ActaPatholMicrobiol Scand.* 1969; 75: 375-98.
- [3]. Marx RE, Stern D. *Oral and Maxillofacial pathology: A rationale for diagnosis and treatment.* Quintessence Pub Co, Hanover Park, 2012.
- [4]. Philipsen HP, Srisuwan T, Reichart PA. Adenomatoid odontogenic tumor mimicking a periapical (radicular) cyst: a case report. *Oral Surg Oral Med Oral Pathol Oral RadiolEndod.* 2002 Aug;94(2):246-8.
- [5]. Phillipsen HP, Reichart PA, Zhang KH, Nikai H, Yu Ox. Adenomatoid Odontogenic tumor: biologic profile on 499 cases. *J. Oral Pathol Med.* 1991; 20: 149-58.
- [6]. Mosadami A. Odontogenic tumors in an African population. *Oral Surg Oral Med Oral Pathol.* 1975; 40: 502-9.
- [7]. N Yilmaz, AAcikgoz, N Celebi, AZ Zengin, O Gunhan. Extrafollicular Adenomatoid Odontogenic Tumor of the Mandible: Report of a Case. *Eur J Dent.* 2009;3(1):71-74.
- [8]. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumor of the mandible: review of the literature and report of a rare case. *Head Face Med.* 2005;1:3.



Figure:1



Figure: 2



Figure: 3

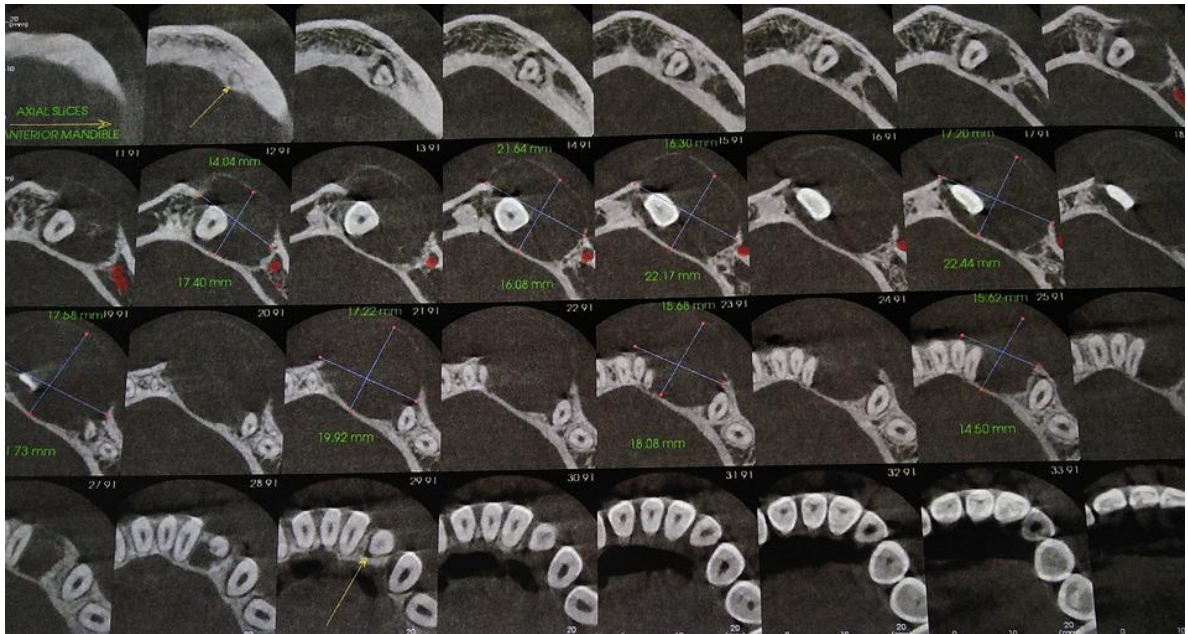


Figure: 4

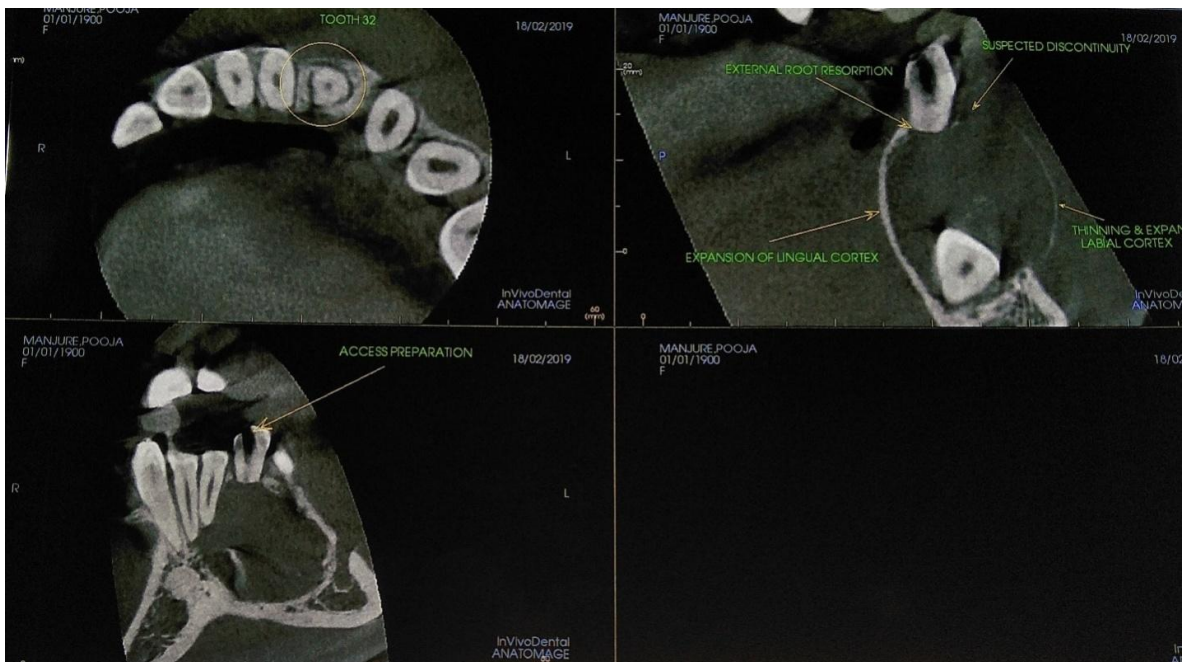


Figure: 5

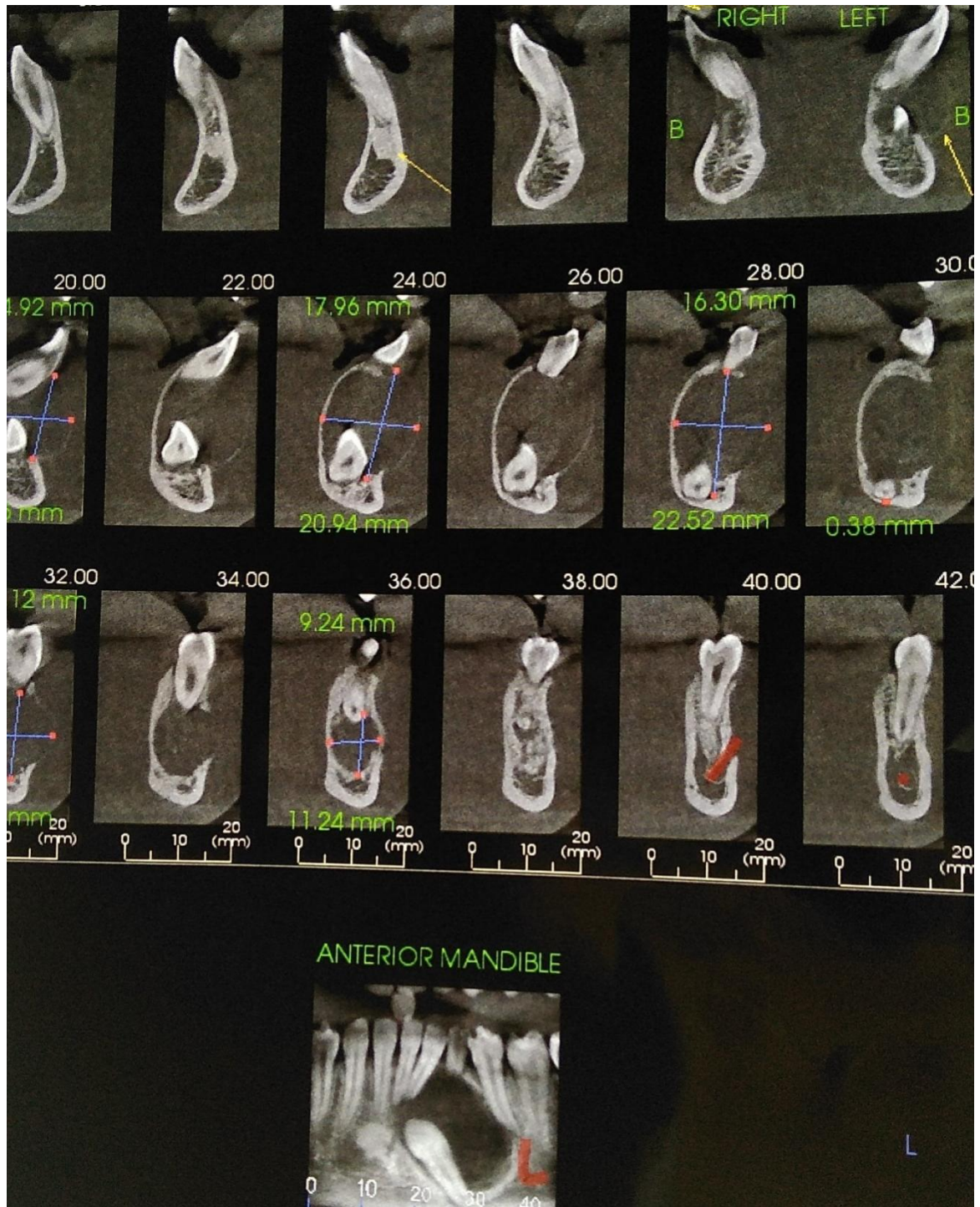


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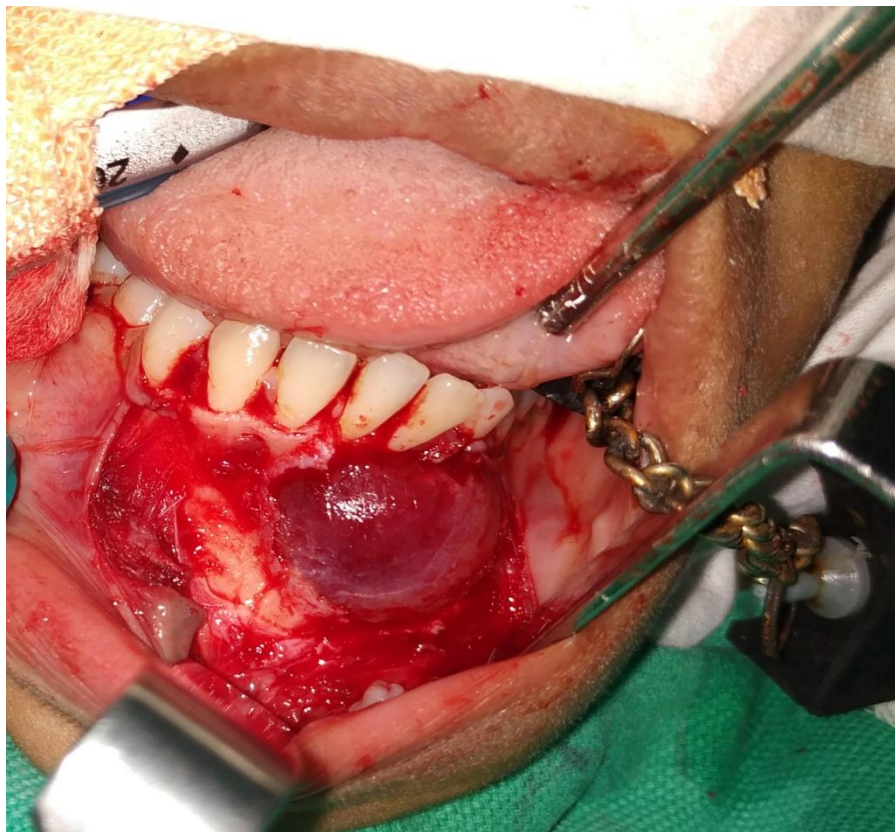


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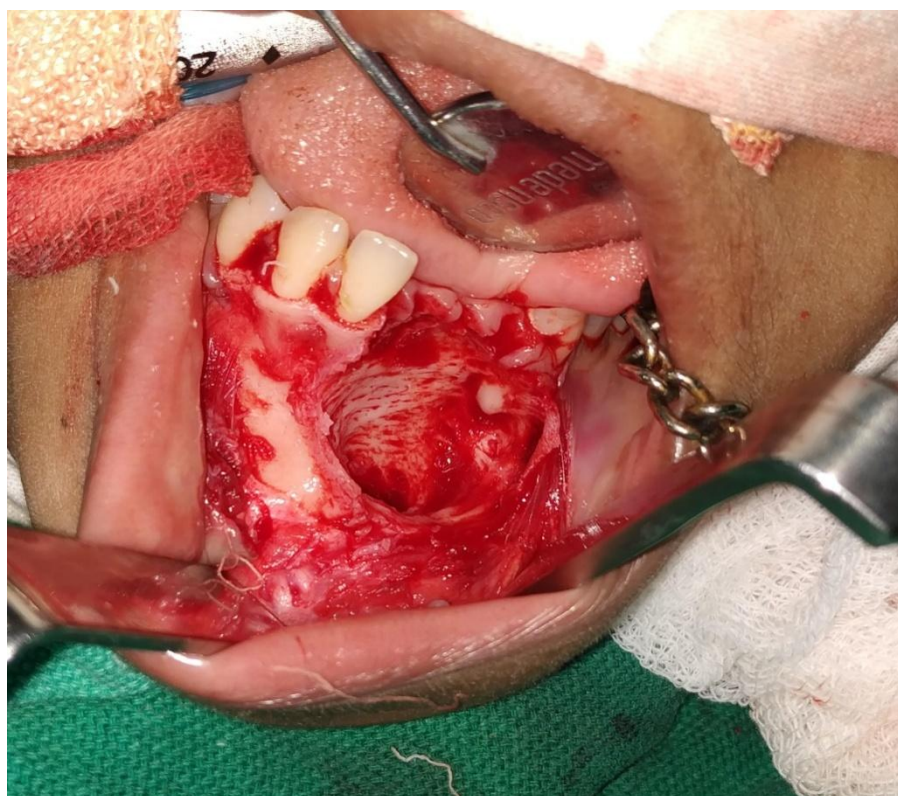


Figure: 8



Figure: 9

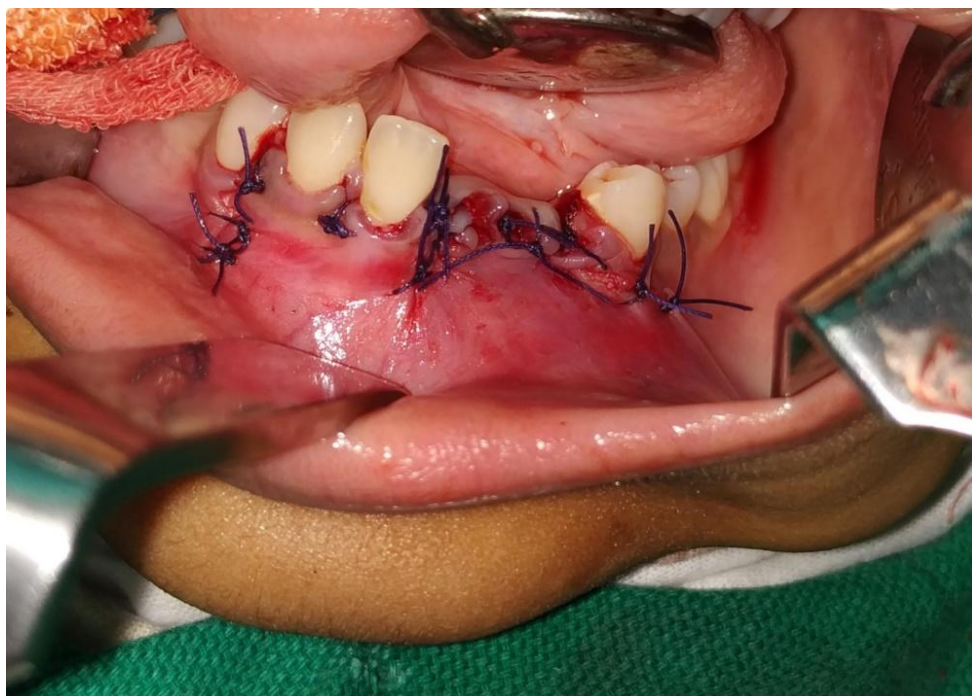


Figure: 10

Dr. Aumiyo Das, et. al. "Adenomatoid Odontogenic Tumor of the Mandible: A Rare Case Report." *IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)*, 19(12), 2020, pp. 05-13.