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Adenomatoid Odontogenic Tumor of Maxilla

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ABSTRACT

An Adenomatoid Odontogenic Tumour (AOT) is an uncommon, benign, odontogenic lesion that is characterized by slow, non-invasive growth. AOT, accounting for approximately 3% of all odontogenic tumours, is mostly found in the maxilla of young females during their second decade. Often, AOT is associated with the impacted canine tooth, typically encapsulated and with a low rate of recurrence following treatment. The case report details an atypical case of a large AOT in a 26-year-old-male presented to Moi Teaching and Referral Hospital (MTRH) with a gradual growing-swelling on the right side of his face. The growth had been developing for over four years resulting in marked facial asymmetry. The examination revealed a 10cm by 5cm, firm, non-tender mass on the right maxilla which was fixed on the underlying bone. The Computed Tomography (CT) Scan showed a well-defined, and encapsulated lesion while the incisional biopsy confirmed the AOT diagnosis. The patient underwent successful surgical excision of the tumour which was then subjected to subsequent histopathological analysis reaffirming the AOT diagnosis. This case is significant because of the AOT occurrence in a 26-year-old male which falls outside the typical AOT demographic profile. Thus, it highlights the necessity of including AOT within the differential diagnosis of maxillary swellings for an atypical patient population. The case also underscores the importance of employing a combination of clinical, radiographical, and histopathological evaluations to achieve a definitive AOT diagnosis.

Keywords: Adenomatoid Odontogenic Tumour, Surgical Enucleation, Hamartomatous Lesion.

INTRODUCTION

An Adenomatoid Odontogenic Tumour (AOT) is a rare, benign odontogenic lesion that typically presents with slow growth and is non-invasive. It is characterized by a low recurrence rate, absence of invasion, and often encapsulated by connective tissue (Matos et al., 2012). AOT accounts for approximately 3% of all odontogenic tumours and is more commonly found in the maxilla, particularly associated with impacted teeth, most frequently canines (John & John, 2010; Seo et al., 2015). This tumour is known for its distinct features, such as duct-like structures formed by the epithelial component of the lesion (Bulut et al., 2001). AOT is considered a hamartomatous lesion of odontogenic origin, first described in 1907, and has been associated with various names before being recognized as a separate entity (Motamedi et al., 2005).

The clinical behaviour of odontogenic tumours, including AOT, varies widely, ranging from benign hamartomatous lesions

to malignancies (Wright & Tekkeşın, 2017). AOT predominantly affects young patients, often in the second decade of life, with a higher prevalence in females (Erdur et al., 2016). The tumour can lead to facial asymmetry, tooth displacement, and bone expansion, particularly when multiple teeth are involved (Narayanan et al., 2013). Radiographically, AOT typically appears as a unilocular, pericoronal radiolucency, commonly seen in the maxillary anterior region in adolescent females.

AOT is a distinct odontogenic lesion with unique characteristics, such as slow growth, encapsulation, and association with impacted teeth, particularly canines. Understanding its clinical behaviour, radiographic features, and prevalence in specific demographics is crucial for accurate diagnosis and appropriate management of this rare odontogenic tumour.

LITERATURE REVIEW

AOT is a "hamartomatous lesion rather than truly neoplastic and one among the rare tumours of the oral cavity" (Krishnan, 2024). This uncommon tumour of odontogenic origin "constitutes only 0.1% of tumours and cysts of the jaw and 3% of all odontogenic tumours" (Kumar et al., 2014). Studying AOT in the maxilla is significant as this lesion is "more common in females than males" and is "most commonly located in the anterior maxilla, usually associated with an impacted canine tooth" (Gopalakrishnan et al., 2023).

AOT in the maxilla is a rare occurrence, with the tumour representing "3–7% of all odontogenic tumours" (Soorya Rao & Ravi, 2016). The condition demonstrates a "female predilection" (Gopalakrishnan et al., 2023) and typically affects "young individuals in the second decade" (Gopalakrishnan et al., 2023). A case report by Kumar et al. (2014) involved a 14-year-old girl with AOT in the maxilla, further highlighting the age distribution of this lesion.

Clinically, AOT in the maxilla manifests as a "right maxillary anterior swelling, facial asymmetry, continuously increase in size of swelling without any symptoms, and delayed eruption of permanent teeth" (Kumar et al., 2014). This tumour is commonly "associated with an impacted tooth, usually canine" (R. & S., 2016). Differentiating AOT from other lesions can be challenging, as Bali et al. (2023) noted that "differentiating this benign tumour from other lesions is difficult before surgical management and histopathological examination is important in accurate diagnosis."

Microscopically, AOT in the maxilla is characterized by the "formation of duct-like structures with amyloid-like deposits" (Yadav et al., 2011). This unique histological feature is a key diagnostic criterion for this tumour, as described in the case report by Yadav et al. (2011).

The primary treatment approach for AOT in the maxilla is "surgical enucleation along with the involved tooth" (Krishnan, 2024). This conservative treatment method is preferred as "this tumour has the least chance for recurrence and hence it does not require radical excision" (Krishnan, 2024). Genno et al. (2017) reported a case of AOT associated with an impacted maxillary lateral incisor, which was managed with surgical excision and followed up for 36 months and 24 months with no recurrence.

Several case reports have documented the occurrence of AOT in the maxilla. Krishnan (2024) presented a case of the "follicular variant of AOT of anterior maxilla associated with impacted canine and its surgical enucleation in a young male patient." Soorya Rao and Ravi (2016) reported a case of AOT in the "left upper 1st molar" region of the maxilla in a 17-year-old boy, highlighting the rare presentation of this tumour in an uncommon site and a male patient. Yadav et al. (2011) described a case of AOT originating in the "maxillary sinus" of an 18-year-old male, emphasizing the importance of considering this tumour in the differential diagnosis of maxillary sinus lesions.

While the majority of AOT cases occur in the maxilla, the tumour can also be found in the mandibular region, accounting for "35% of cases" (Bali et al., 2023). Bali et al. (2023) discussed a case of AOT in the anterior aspect of the mandible in a 16-year-old girl, contrasting the characteristics and management approaches between maxillary and mandibular AOT.

The primary treatment for AOT in the maxilla is "surgical enucleation" (Krishnan, 2024), which involves the "complete excision of the tumour along with the involved tooth" (Krishnan, 2024). This approach is preferred due to the "least chance for recurrence" (Krishnan, 2024) and the benign nature of the tumour, negating the need for "radical excision" (Krishnan, 2024).

The importance of regular follow-up examinations after surgical excision of AOT in the maxilla is highlighted in the case report by Genno et al. (2017), who followed up a patient for 36 months and 24 months without any recurrence. This emphasizes the need for close monitoring to ensure the long-term success of the treatment. Careful clinical, radiographic, and histopathological evaluation is essential for accurate diagnosis, as differentiating AOT from other lesions can be challenging. The primary treatment approach is surgical enucleation, which has a low recurrence rate. Further research is needed to better understand the etiology, optimal management strategies, and long-term outcomes of this uncommon maxillary lesion.

CASE REPORT

A 26-year-old male patient from the Western Region of Kenya, presented with a Swelling on the right side of the face which had been there for 4 years. He reported a slow growing swelling which progressively got larger ending in marked facial asymmetry. The patient reported with a non-remarkable medical history and had been to the local doctors who referred him to Moi Teaching and Referral Hospital (MTRH), Eldoret Kenya. On examination we noted that the patient had an obvious swelling on the right side of the midface with normal signs and normal vitals. The swelling was on the right side of the maxilla, no scar noted, the size was 10cm by 5cm, the colour over the mucosa was normal and it was non tender. The lesion was firm, and fixed to the underlying bone. A CT scan along with an incisional biopsy was ordered which revealed an AOT with well-defined capsulated margins.

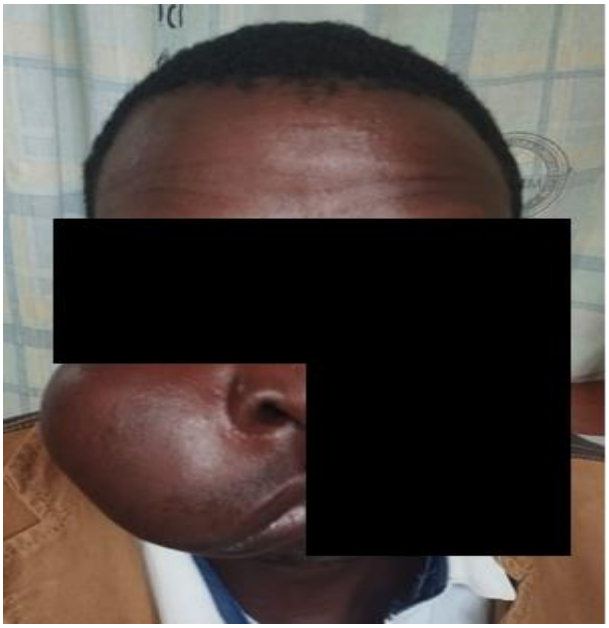

Figure 1: 26-year-old male patient from the Western Region of Kenya, presenting with a Swelling on the right side of the face which had been there for 4 years

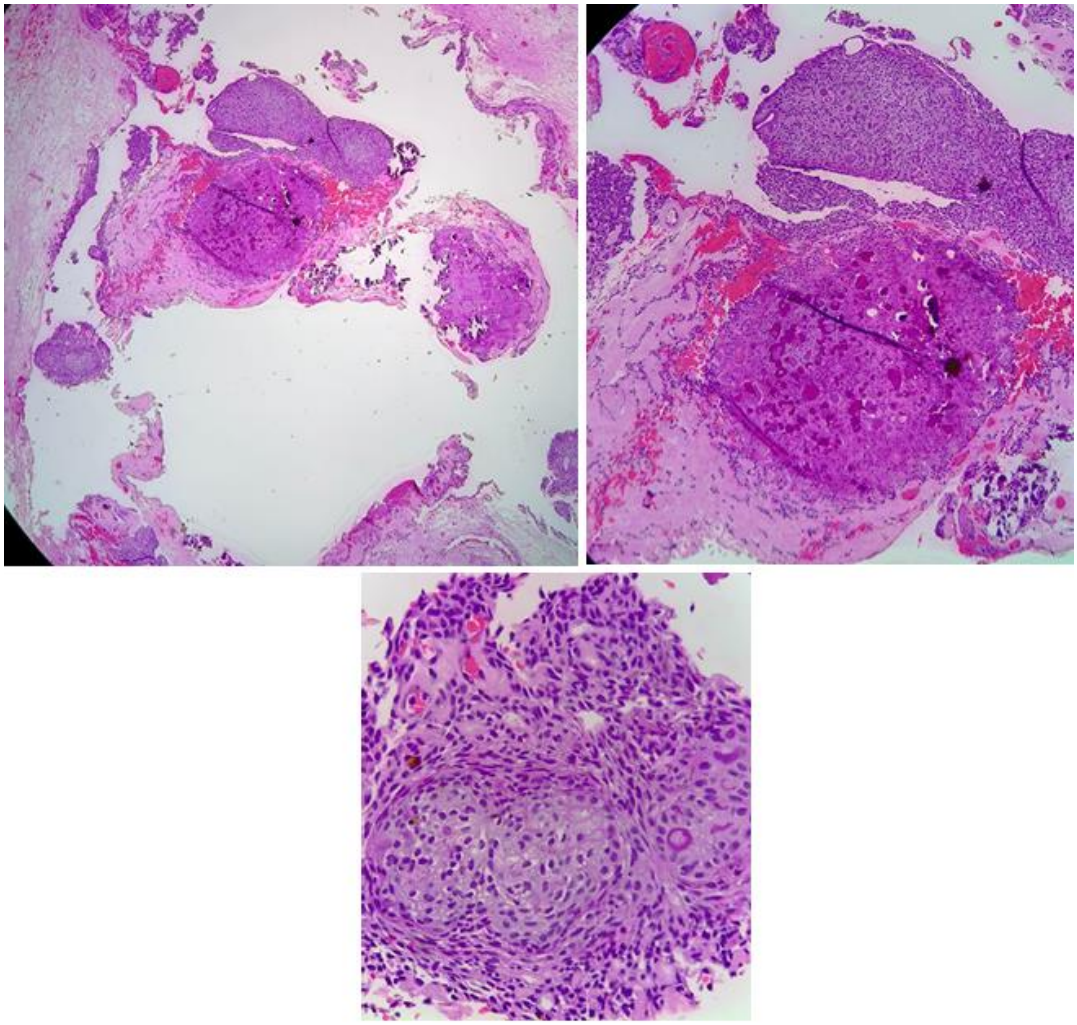

Figure 2: Histological appearance of the Incisional Biopsy done for Histopathology Report

The patient was booked in for Surgical Excision and the specimen presented for histopathology which confirmed the diagnosis above

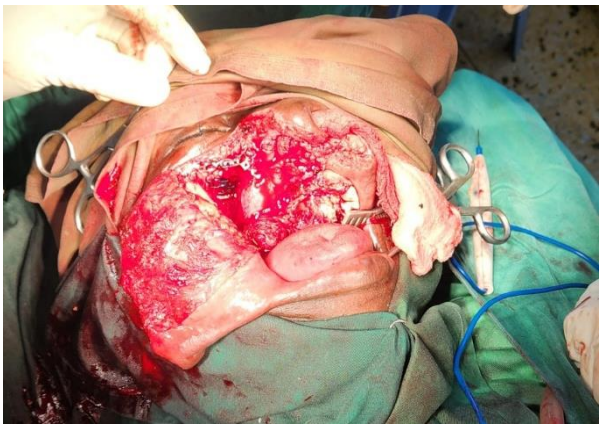

Figure 3: Weber Fergusson Incision used to approach the tumor from the Right side

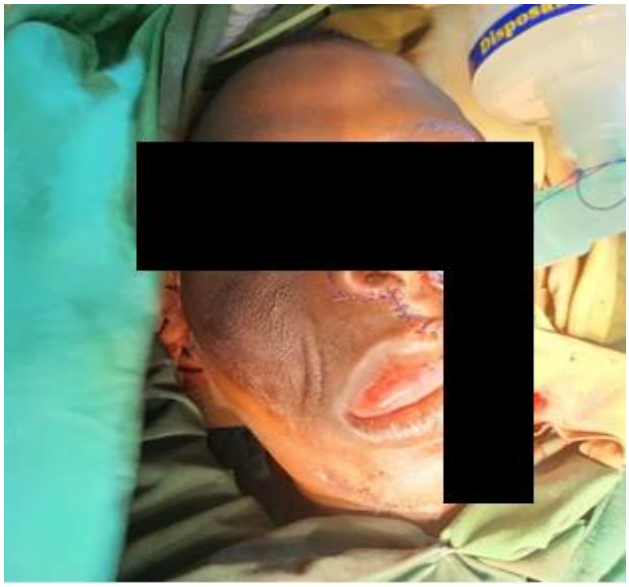

Figure 4: Suturing post resection

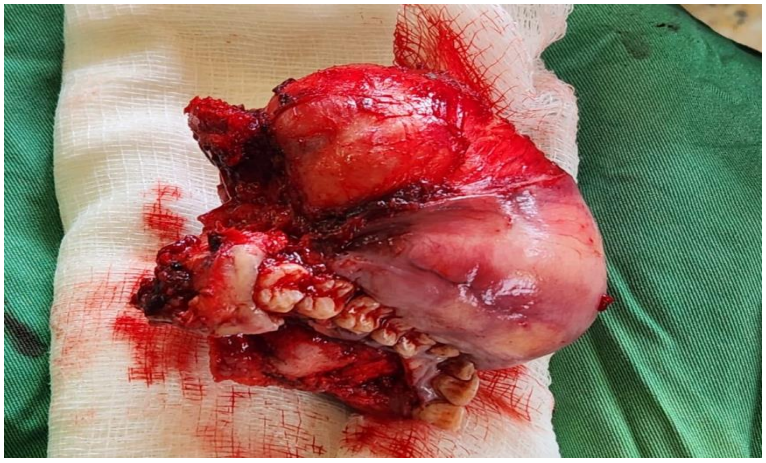

Figure 5: Excisional Biopsy: Full extent of the tumor extending from the 21, 11 to 18

DISCUSSION

The presented case report of an AOT in the maxilla contributes significantly to the understanding of this rare and benign odontogenic lesion. AOTs are known for their slow growth, non-invasive nature, and low recurrence rates. They are often encapsulated by connective tissue, accounting for a small percentage of all odontogenic tumours, and typically affect young females, particularly in the anterior maxilla associated with impacted canines (Matos et al., 2012; John & John, 2010; Seo et al., 2015). This case, involving a 26-year-old male patient, provides valuable insights into the clinical presentation, diagnostic process, and treatment of AOTs.

The clinical presentation of the patient, who experienced a slow-growing swelling on the right side of his face over four years, is consistent with typical manifestations of AOT. The lesion led to marked facial asymmetry, which is a common consequence of the tumour's growth. The patient's medical history was unremarkable, and physical examination revealed a firm, non-tender swelling fixed to the underlying bone with normal mucosal coloration (Wright & Tekkeşın, 2017; Erdur et al., 2016). This presentation aligns with the known characteristics of AOT, which often leads to facial asymmetry, tooth displacement, and bone expansion (Narayanan et al., 2013; Bulut et al., 2001).

Diagnostic imaging and histopathological examination are crucial in confirming the diagnosis of AOT. In this case, a CT scan revealed well-defined capsulated margins,

and an incisional biopsy confirmed the presence of duct-like structures formed by the epithelial component of the lesion (Bulut et al., 2001). These features are hallmark indicators of AOT, distinguishing it from other odontogenic tumours. The presence of amyloid-like deposits and the formation of duct-like structures are key histological characteristics that aid in the diagnosis (Yadav et al., 2011).

Surgical enucleation remains the primary treatment approach for AOT, involving the complete excision of the tumour along with the involved tooth. This method is preferred due to its low recurrence rate and the benign nature of the tumour, which negates the need for radical excision (Krishnan, 2024). In this case, the patient underwent surgical excision, and the specimen was submitted for histopathological examination, which confirmed the diagnosis of AOT (Kumar et al., 2014).

The literature review supports this treatment approach, highlighting the importance of conservative management for AOT. Studies have shown that AOT has a very low recurrence rate following surgical enucleation, and regular follow-up examinations are essential to monitor for any signs of recurrence. The case report by Genno et al. (2017) emphasized the importance of long-term follow-up, with no recurrence observed after 36 and 24 months, respectively. This underscores the need for close monitoring to ensure the long-term success of the treatment and to detect any potential recurrence early.

Differentiating AOT from other odontogenic lesions can be challenging due to overlapping clinical and radiographic features. Accurate diagnosis requires a combination of clinical, radiographic, and histopathological evaluations. Radiographically, AOT typically appears as a unilocular pericoronal radiolucency, commonly seen in the maxillary anterior region in adolescent females (Seo et al., 2015). However, other odontogenic tumours and cysts can present with similar features, making histopathological examination crucial for definitive diagnosis. Bali et al. (2023) noted the difficulty in differentiating AOT from other lesions before surgical management, emphasizing the importance of histopathological confirmation.

The literature review highlights the epidemiology and clinical features of AOT, noting its higher prevalence in females and its typical occurrence in the second decade of life (Gopalakrishnan et al., 2023). The tumour is most commonly associated with impacted canines in the anterior maxilla, but it can also occur in other regions, including the mandible. The female predilection and age distribution are important considerations for clinicians when evaluating patients with similar presentations.

Several case reports document the occurrence of AOT in the maxilla, further supporting the demographic and clinical trends observed. For instance, Krishnan (2024) presented a case of the follicular variant of AOT in a young male patient, while Soorya Rao and Ravi (2016) reported a case in the left upper first molar region of a 17-year-old boy, highlighting the variability in presentation sites and patient demographics.

This case report adds to the growing body of knowledge on AOTs, reinforcing the characteristics, diagnostic approaches, and treatment modalities for this rare tumour. The successful surgical management and emphasis on follow-up care demonstrate the effectiveness of current treatment strategies. However, the need for further research remains to better understand the etiology, optimal management strategies, and long-term outcomes of AOTs, particularly in the maxillary region (Soorya Rao & Ravi, 2016; Wright & Tekkeşın, 2017).

CONCLUSION

In conclusion, AOT in the maxilla is a rare benign lesion of odontogenic origin that predominantly affects young females. Accurate diagnosis relies on a combination of clinical, radiographic, and histopathological evaluations. Surgical enucleation is the preferred treatment

approach due to its low recurrence rate, and regular follow-up is essential to monitor for recurrence. This case report contributes valuable insights into the clinical presentation, diagnostic process, and management of AOT, highlighting the need for continued research to improve patient outcomes and understanding of this uncommon tumour.

Declaration by Authors

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