Aggressive Adenomatoid Odontogenic Tumor of Maxilla involving Impacted Third Molar: A Rare Case Report

Subrat Kumar Padhiary[1], Gunjan Srivastava[2], Satyabrat Patnaik MDS[3], Neeta Mohanty[4], Sourav Chandra B Bal[5]

[1]Professor, Department of Oral and Maxillofacial Surgery, [2]Professor, Department of Prosthodontics, [3]Reader, Department of Oral and Maxillofacial Surgery, [4]Professor and Head, Department of Oral and Maxillofacial Pathology, [5]Senior Lecturer, Department of Public Health Dentistry, Institute of Dental Sciences, Siksha 'O' Anusandhan Deemed to be University, Bhubaneswar, India

Abstract

Adenomatoid odontogenic tumor (AOT) is a rare tumor comprising about 2.2% to 7.1% of all odontogenic tumors. It is a neoplasm of odontogenic origin having slow-growth rate, noninvasive and benign in nature. Here, we report a case of a young female patient who presented with an aggressive AOT in maxilla involving the impacted third molar. Usually, the treatment for AOT comprises of enucleation and curettage, whereas due to the fast growth rate and aggressiveness of the lesion, subtotal maxillectomy was performed in this case. The authors have described about the clinical, radiographic, histopathological and surgical aspect of the case.

Keywords*: Adenomatoid odontogenic tumor, Maxilla, subtotal maxillectomy*

Introduction

Adenomatoid odontogenic tumor (AOT) is a neoplasm of odontogenic origin having slow-growth rate, is noninvasive and benign in nature[1]. AOT usually presents with an impacted tooth and contributes about 2.2% to 7.1% of all odontogenic tumors[2]. It has high female predilection in the age range of 20–30 years. The most common site of occurrence is anterior maxilla, usually associated with the impacted canine.

Stafne was the first to report AOT as a separate entity but the term AOT was not coined by him[3]. Nomenclature of AOT was not possible due to its similar histologic features with other odontogenic tumors until 1971, when WHO adopted the term "adenomatoid odontogenic tumor" proposed by Philipsen and Birn[2,3]. There is a lack of consensus on pathogenesis of the tumor, but according to the developmental theory, some authors believe it to develop from the epithelium of a dentigerous cyst[4].

Here we report a case of AOT of the maxilla. The case presents certain atypical features such as enormous size and aggressive behavior contrary to its smaller size and slow growing nature. The involvement of maxillary third molar is also not so frequently seen associated with AOT, maxillary canine being the favored site. Considering the aggressive nature of the lesion, treatment was done with subtotal maxillectomy under general anesthesia.

Case Report

A 22-year-old female patient reported to the Department of Oral and Maxillofacial Surgery complaining of painless swelling in the left side mid-facial region and upper left posterior teeth region with surface ulceration since last two months. The swelling was insidious in origin, but its rate of growth was very fast. There was deviation of regional teeth and ulceration

Corresponding author:
Dr. Subrat Kumar Padhiary
Professor, Department of Oral and Maxillofacial Surgery, Institute of Dental Sciences
Siksha 'O' Anusandhan Deemed to be University, Bhubaneswar, India – 751003
Email: subratpadhiary@soa.ac.in

over the swelling due to occlusal trauma from mandibular teeth. Due to difficulty in chewing food and marked extra-oral swelling, she consulted a local physician and was subsequently referred to our institute.

On clinical examination, there was marked mid facial diffuse swelling on the left side of the face with normal-appearing overlying skin. (Fig.1) Intraorally the swelling was evident both on the buccal and palatal aspect with obliteration of vestibular sulcus from maxillary canine to the third molar region. Palatally, the swelling extended up to the mid palatal raphe. There was marked displacement of maxillary molars and premolars with missing maxillary third molar. The swelling on the maxillary tuberosity region was impinging on the mandibular molars resulting in ulceration of the overlying mucosa. (Fig.2) The swelling was soft in consistency indicating cortical bone resorption both buccally and palatally. On aspiration, scant mucoid fluid was noted.

CT scan demonstrated a large cystic expansile radiolucent lesion with corticated borders on the left maxillary region with marked expansion of both buccal and palatal cortices. Left maxillary sinus was completely obliterated by the SOL (Space Occupying Lesion) and approximating to the orbital floor. Orbital floor was slightly elevated anteriorly by the SOL. Displaced maxillary third molar was seen intra-lesionally near to the orbital floor. There was no area of calcification inside the SOL. (Fig.3) Based on these clinical and radiological findings, a provisional diagnosis of malignant neoplasm of connective tissue origin of the left maxilla was made, and an incisional biopsy was done. Histological findings revealed the presence of solid nodules of epithelial cells forming sheets, rosettes, and whorled masses in a scanty connective tissue stroma with a fibrous capsule surrounding the tumor. (Fig.4) Histopathological diagnosis was adenomatoid odontogenic tumor (AOT).

Considering the extensive size and rate of growth of the lesion, a subtotal maxillectomy was planned. With standard Weber–Ferguson incision, sufficient access to the tumor mass was obtained, and a subtotal maxillectomy preserving the infraorbital rim and floor of the orbit was done. (Fig.5) Complete resected specimen measuring 8cmX7cm was sent for histopathological examination. (Fig.6) Biopsy report reconfirmed it to be AOT. The surgical defect was packed with a whitehead varnish-soaked gauge pack. A surgical obturator was given to hold the pack in place. Postoperative course of healing was satisfactory and uneventful. (Fig.7) Patient is under regular follow up with interim obturator prosthesis.

Fig.1 Preoperative View (A) Frontal (B) Profile

Fig.2 Preoperative Intraoral View

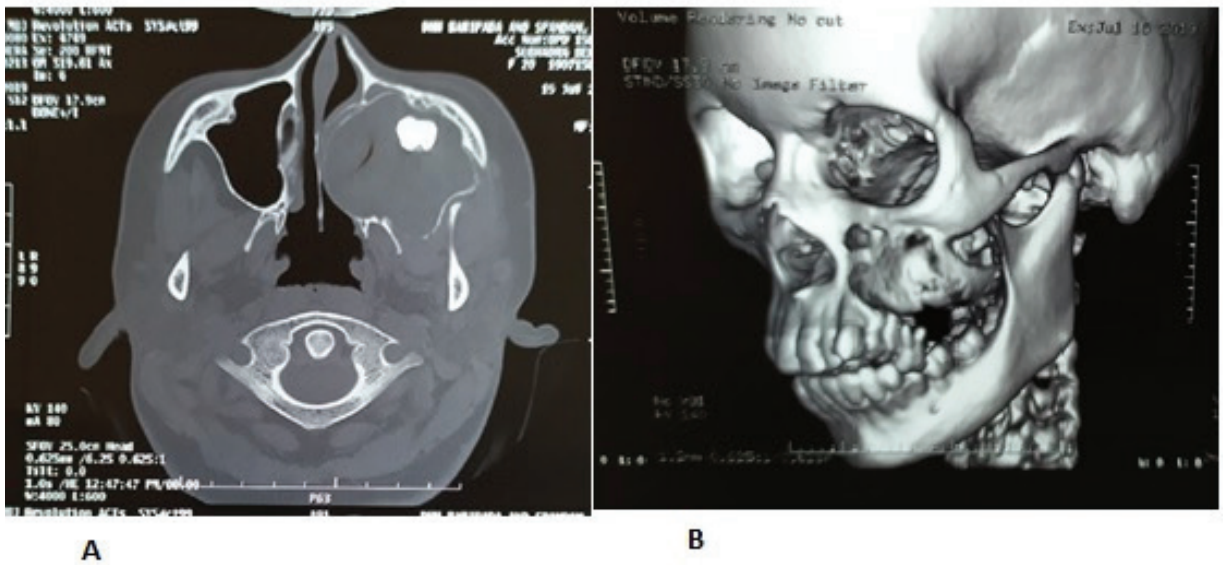

Fig.3 CT Scan showing the enormous size of the tumor. (A) Axial View (B) 3D View

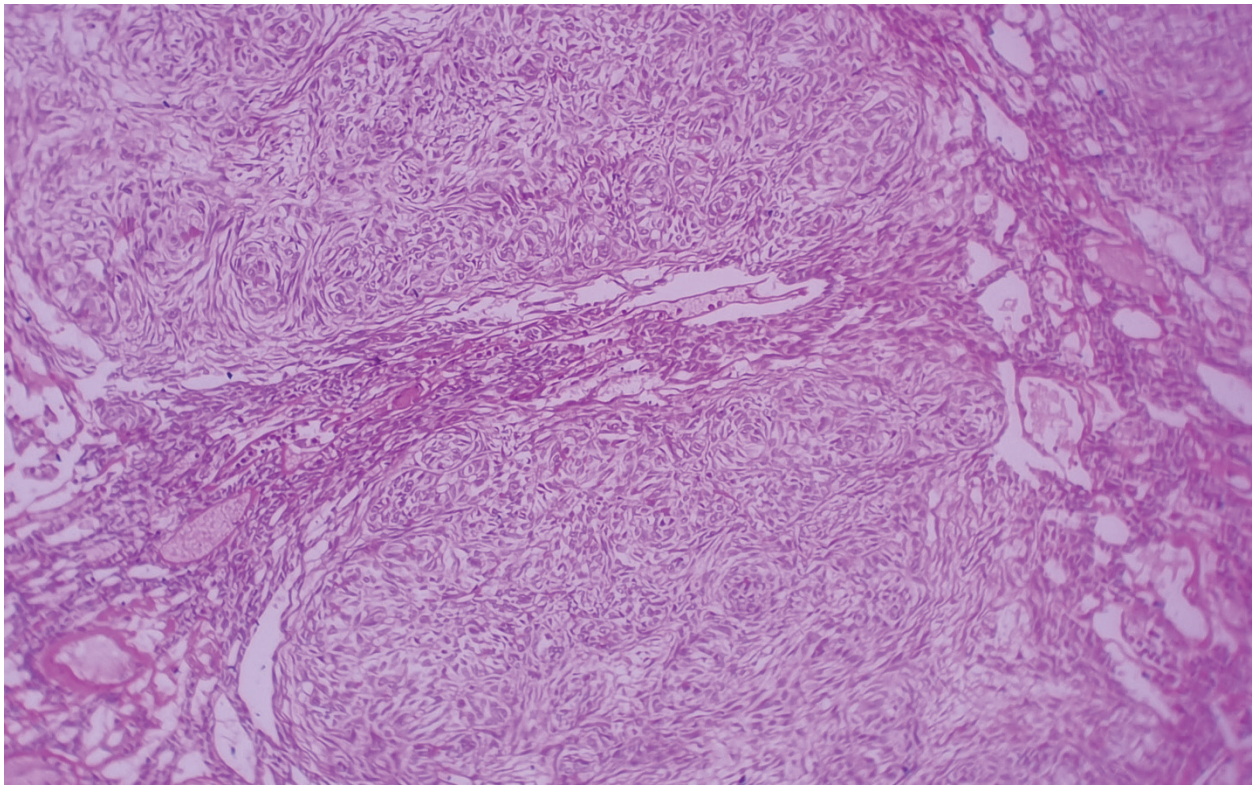

Fig.4 Histopathological photomicrograph.

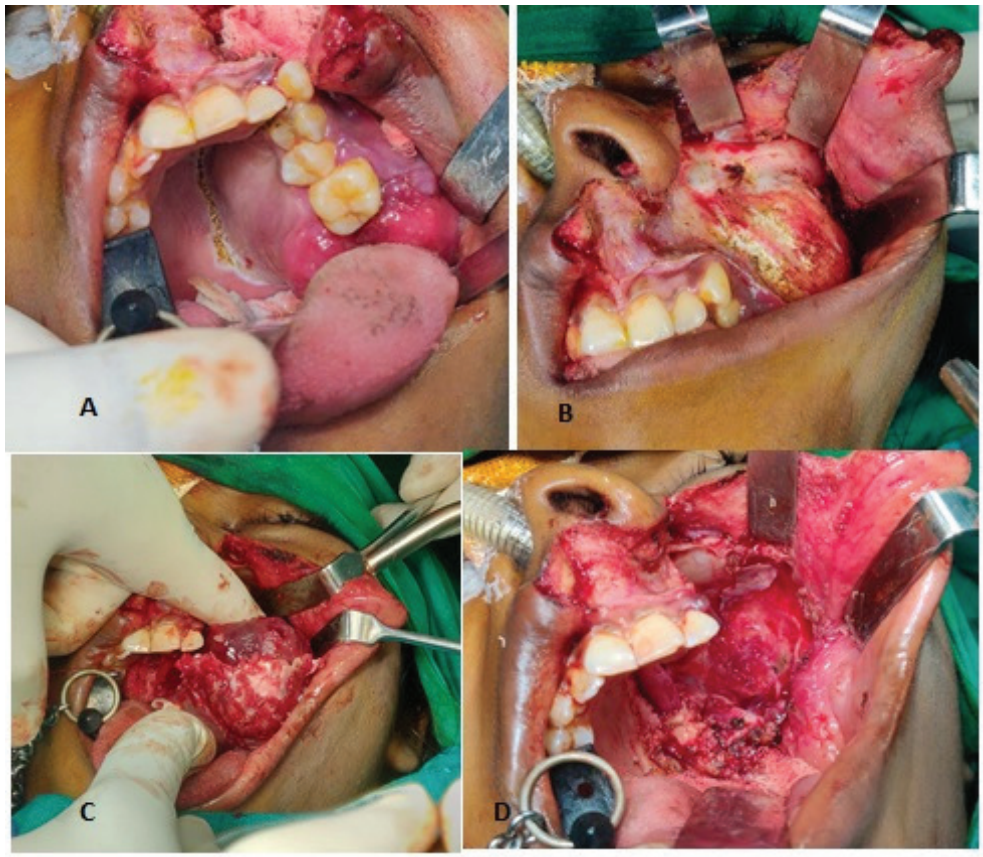

Fig.5 (A-D) Intraoperative photographs

Fig.6 Surgical Specimen after resection.

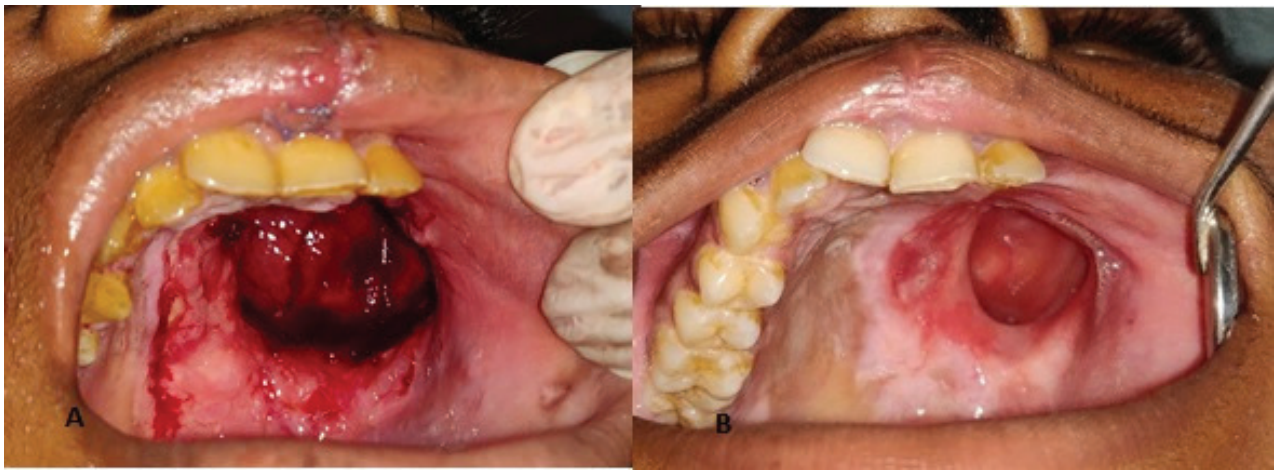

Fig.7 Postoperative intraoral view (A) 7 days postoperative photograph (B) 6-months postoperative photograph showing satisfactory healing.

Discussion

Histologically three different variants of AOT are described. The follicular variant accounts for 73% of AOT cases and is associated with an unerupted tooth. The extrafollicular variant is not associated with an impacted tooth and accounts for approximately 24% of AOT cases. The peripheral variant accounts for 3% of the cases and affects the gingival tissues[5]. In the present case, the lesion was a follicular variant as associated with impacted maxillary 3rd molar. The lesion was very fast growing contrary to its nature and attained a huge size within two months of history as reported by the patient. Due to its fast growth rate and ulceration of the overlying mucosa our first provisional diagnosis was a malignant tumor of connective tissue origin.

Unequivocally AOT is also misdiagnosed as dentigerous cyst on clinical and radiologic examination. Roughly 77% of the follicular variant of AOT is reported to be misdiagnosed as dentigerous cyst[6]. Both AOT and dentigerous cyst present as pericoronal radiolucent area around the impacted tooth. Unlike Dentigerous cyst, AOT is most commonly seen enclosing the entire crown and root of the impacted tooth as it grows from the Hertwig's epithelial root sheath[7].The dentigerous cyst is frequently seen associated with the impacted third molar, enclosing mostly the coronal part of the tooth[8,9]. Due to this difference in presentation, the treatment options in both AOT and dentigerous cyst may vary. In AOT, a marsupialization procedure with the intention to allow the involved tooth to grow to the occlusion is less likely to be successful as there is little or no peri radicular bone available around the root of the involved tooth. In the present case, although the site of the lesion was favorable for dentigerous cyst, an aggressive course of the swelling deferred our provisional diagnosis more towards a malignant growth.

The size of AOT is usually within 1–3 cm in diameter. Rarely in literature, do we get cases of large AOTs[10,11]. The large size of this lesion has been credited to a higher growth potential in younger patients and usually delay in seeking treatment. In the present case the patient was a young female but there was no delay in seeking treatment. This suggests more aggressive nature of the tumor. The treatment of AOT usually consists of enucleation and curettage. However, in our case, the enormous size of this lesion achieved within a short period with cortical perforation and ulceration in overlying oral mucosa suggested the aggressiveness of the neoplasm. Therefore, subtotal maxillectomy was planned and carried out.

Conclusion

The clinical behavior of AOT has been widely studied in the past by many authors and it has been tagged as slow growing benign neoplasm of odontogenic origin, with favored anatomical site, age and gender of the patient. But due to some unknown factors, this seemingly docile tumor may behave aggressively masquerading as a malignant neoplasm. The present literature has very few case reports on aggressive AOTs. The cause of aggressive behavior in some cases of AOT is still to be found out to help the surgeon in formulating an adequate treatment protocol for these cases.

Conflicts of Interests: The authors declare, they do not have any conflicts of interests

Funding: None

Ethical Issues: None

References

1. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu QX. Adenomatoid odontogenic tumor: Biologic profile based on 499 cases. J Oral Pathol Med 1991; 20:149,58.
2. Reichart PA and Philipsen HP: Odontogenic tumors and allied lesions. Quintessence Publ., London, pp. 105-115, 2004.
3. Kramer IRH and Pindborg JJ: Histological Typing of Odontogenic Tumours, 2nd edition. Springer, Berlin (1992).
4. Philipsen HP, Samman N, Ormiston IW, Wu PC, Reichart PA. Variants of the adenomatoid odontogenic tumor with a note on tumor origin. J Oral Pathol Med. 1992; 21:348–52.
5. Philipsen HP, Reichart PA, Siar CH, Ng KH, Lau SH, Zhang X, et al. An updated clinical and epidemiological profile of the adenomatoid odontogenic tumour: a collaborative retrospective study. J Oral Pathol Med. 2007;36(7):383–393.
6. Dhirawani RB, Pathak S, Mallikaarjuna K, Sharma A. An adenomatoid odontogenic tumor in disguise. J Indian Soc Pedod Prev Dent 2016; 34:291-3.
7. Max RE, Stern D. Oral and maxillofacial pathology: a rationale for diagnosis and treatment. 2th ed. Quintessence Publishing: Hanover Park; 2003. pp. 609–612.
8. Lee JK, Lee KB, Hwang BN. Adenomatoid odontogenic tumor: A case report. J Oral Maxillofac Surg 2000; 58:1161-4.
9. Patel V, Sproat C, Samani M, Kwok J, McGurk M. Unerupted teeth associated with dentigerous cysts and treated with coronectomy: Mini case series. Br J Oral Maxillofac Surg 2013; 51:644-9.
10. Shetty K, Vastardis S, Giannini P. Management of an unusually large adenomatoid odontogenic tumor. Oral Oncol. 2005; 41:316–8.

11. Dhupar V, Akkara F, Khandelwal P. An unusually large aggressive adenomatoid odontogenic tumor of maxilla involving the third molar: A clinical case report Eur J Dent. 2016 Apr-Jun; 10(2): 277–280.