Bulletin of Environment, Pharmacology and Life Sciences
Bull. Env. Pharmacol. Life Sci., Spl Issue [2] 2022 : 06-11

Online ISSN 2277-1808
Journal's URL:http://www.bepls.com
CODEN: BEPLAD

ORIGINAL ARTICLE **OPEN ACCESS**

Rare Presentation of Adenomatoid Odontogenic Tumour in Anterior Mandible: Case Report

Amit B. Lall[1]*, Tanya Kusum[2], Priyanka T[3]
[1,2,3] Department of Oral & Maxillofacial Surgery, Santosh Deemed to be University, Ghaziabad, Uttar Pradesh
Email: dean.research@santosh.ac.in

ABSTRACT
AOT is a rare and distinctly described odontogenic tumor that exclusively originates from odontogenic epithelium. There are numerous reports available regarding the clinical presentation and histological spectrum of AOT. Although majority of literature reports describe it's marked predilection for site of occurrence as the maxilla. This article aims to discuss an unusual case report on a rare presentation of AOT causing swelling in the mandibular anterior region. A collaborated extensive literature review from 1990 to 2022 with the established scope of literature was carried out. This paper presents an endeavour of the authors to highlight it's unusual presentation in the anterior mandible, associated histological variant, impacted tooth, treatment and recurrence rate of AOT's.
KEYWORDS: *Aot, Adenomatoid Odontogenic Tumour, Odontogenic Tumour.*

Received 22.07.2022 Revised 23.09.2022 Accepted 21.10.2022

INTRODUCTION

The tumour that meets the present widely accepted diagnostic criteria of an adenomatoid odontogenic tumour (AOT) has been known for more than 100 years. Adenomatoid odontogenic tumor (AOT) is a distinct, relatively uncommon odontogenic neoplasm that was first highlighted in literature by Harbitz in 1915 under the name of cystic adamantoma [1]. Subsequently, a variety of terms have been used to discuss this tumor by numerous authors in their studies. Adenomatoid Odontogenic Tumour is acknowledged by various nomenclature mentioned in literature: adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma or teratomatous odontoma. In the recent 2nd edition of the WHO, histological typing of odontogenic tumours: AOT has been defined as a tumour of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumour may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst. It is believed generally that the lesion is not a neoplasm [2]. AOT is a perfect imitator of dentigerous cyst, hence, befittingly known as the 'master of disguise'. In 1999, Philipsen stalwarted extensive research in odontogenic tumours and presented a comprehensive review based on reports which showed many interesting aspects regarding epidemiology, histology, clinical features of AOT [3]. With due course of time, numerous case reports and series have been published and come into light all over the world. AOT is a relatively uncommon distinct odontogenic neoplasm and is rightfully called as 'Master of Disguise' and 'Perfect imitator of dentigerous cystIt's incidence in the anterior mandible is relatively uncommon and this article intends to emphasize on these rare occurrences in the anterior mandible reported and documented worldwide.

According to Philipsen and Reichart, AOT exists in three clinic-topographic variants: follicular, extrafollicular and peripheral. The follicular and extrafollicular variants are intrabony and account for approximately 96% of all AOTs[4]. The follicular type shows a well-defined, unilocular (round or ovoid) radiolucency associated with the crown and often part of the root of an unerupted tooth thus mimicking a dentigerous or follicular cyst. In fact, 77% of follicular type AOT are usually initially misdiagnosed as dentigerous cysts. The extrafollicular type is not associated with an unerupted tooth and a well-defined, unilocular radiolucency is found between, above or superimposed upon the roots of erupted, permanent teeth. Depending on the actual intraosseous site of the lesion, these tumours lead to the preoperative, tentative diagnosis of a residual, radicular, `globulo-maxillary' or lateral periodontal cyst. The peripheral type manifests as a soft tissue swelling, gingival fibroma or epulis attached to the gingiva labially. This type of AOT usually depicts slight erosion of the alveolar bone crest but radiographic changes are often

difficult to detect. In approximately two-thirds of the intrabony variants, the radiolucency depicts discrete foci having an occulent pattern of scattered radiopacities. Growth of the intrabony variants commonly results in cortical expansion. Displacement of neighbouring teeth due to tumour expansion is much more common than root resorption. It has been estimated that the AOT accounts for 2.2 and 7.1% of all odontogenic tumours which gives this tumour a ranking of fourth or fifth among the odontogenic tumours only surpassed by odontomas, myxomas, ameloblastomas and cemento-osseous tumours or lesions. The age range of patients with AOT varies between 3 and 82 years at the time of diagnosis. About 68.6% of the tumours are diagnosed in the second decade of life and more than half of the cases (53.1%) occur within the teens (13-19 years of age). This age distribution with a very tall peak in the second decade makes the AOT unique among odontogenic tumours. The female: male ratio for all age groups and AOT variants together is around 1.9:1. Concerning the distribution of unerupted permanent teeth found in association with the follicular AOT, all four canines account for 59% and the maxillary canines alone for 40% [4-5]. Unerupted first and second molars are the teeth most rarely involved in AOTs, only four cases having been reported so far.

CASE REPORT

A 16 year old girl named Ashka presented with a chief complaint of swelling in the mouth in the right lower jaw from the past 2–3 months which had gradually increased in size to the present size. There was a history of localized dull aching pain in the lower right back tooth region 6 months back which subsided on taking antibiotics and analgesics prescribed by the local dentist. Her medical history was quite unremarkable with no apparent systemic problems. There were no significant extraoral findings and no associated lymphadenopathy. Intraorally however a solitary swelling obliterating the right lower buccal vestibule extending from the mesial aspect of the canine to the mesial aspect of the first molar was evident. There was a remarkable expansion of the buccal cortical plate (Figure 1). Swelling was tender, soft in consistency and fluctuant in areas with egg shell crackling perceptible. The lower right first premolar was missing. Mild heaviness was elicited pre-operatively over the right lower lip. Electronic pulp testing revealed 43, 45 to be vital. OPG revealed a solitary, well defined radiolucency, approximately 3*2.5cm in superio-inferior (from the thinned-out crest of the alveolar process upto 0.5cm above the inferior border of the mandible) and anteroposterior dimensions (between the roots of 43 and 45) Resorption and thinning of the alveolar process in the area of the missing premolar was evident. Displacement of the roots of 43 (mesially) and 45 (distally) was observed (Figure 2). Aspiration revealed a yellowish straw-coloured fluid (Figure 3). A provisional diagnosis of dentigerous cyst, adenomatoid odontogenic cyst involving the impacted first premolar was made. After routine haematological examination the case was considered for surgical enucleation under LA. sulcular incision was placed all along from 41 to 46 region with an anterior releasing incision in 41 region. A full thickness triangular mucoperiosteal flap was raised to expose the underlying lesion taking care not to perforate the cystic lining (Figure 4). Marked expansion with perforation of the buccal cortical plate was evident (Figure 5). The cystic lesion was enucleated in to along with the involved tooth. The impacted premolar was seen to be nestled within the bony cavity but outside the lining of the lesion. The lining was found to be attached to the CEJ of impacted tooth (Figure 6). The specimen was preserved in formalin and sent for histopathological examination (Figure 8). Peripheral ostectomy was performed and the sharp bony margins were filed (Figure 7). The bony cavity was debrided thoroughly with betadine and saline. Primary closure was done after haemostasis (Figure 8). Patient was follow-up at regular intervals and OPG X-ray was taken depicting good bone formation. The obtained specimen was grossed and measured about 2 x 3cm. The colour of the specimen was black-brown, it was firm in consistency & ovoid shaped. Upon further examination, microscopically it depicted multinodular proliferation of neoplastic cells. The neoplastic cells were predominantly spindle shaped, columnar and cuboidal. Striking feature included duct like structure which was lined by a single layer of cuboidal or columnar cells with its nuclei polarised away from the lumen (Figure 9). There were few areas which depicted presence of rosette like structure with significant centrally present eosinophilic material. Scattered foci's of calcification was also appreciated along with scanty connective tissue stroma presence.

RESULT AND DISCUSSION

AOT is also a unique lesion that presents in a fairly consistent manner, hence, famously regarded as a "tumor of two-thirds," two-thirds occur in female patients, two-thirds occur in the second decade of life, two-thirds develop in the anterior sextant of the maxilla, two-thirds are superimposed on dentigerous cysts, and two-thirds of associated unerupted teeth are permanent canines. In addition, two-thirds of cases show scattered dot-like (snowflake) opacities within the unilocular radiolucency [6]. However, in a worldwide collaborative retrospective study, the relative frequency of AOT ranged from 0.6% to 38.5%

out of all tumours [7]. Numerous retrospective studies with large case series revealed a female predominance for AOT, with global female-to-male ratio of 1.9:1. A comprehensive Sri Lankan study revealed female-to-male ratio as 2:1. Toida *et al*. in Japan had reported in their study that the female-to-male ratio was approximately 3.0:1 [8]. However contrastingly, Swasdison *et al*. in their retrospective review of 67 cases conducted in Thai population showed female-to-male ratio as 1.8:1 [9]. Arotiba *et al*. in a previous study from Nigeria and de Matos *et al*. reviewed 15 cases from Brazil showed a female-to-male ratio of 1.4:1 [10]. Considering age distribution of AOT's, it was reported that more than two-thirds of AOT's were diagnosed in young female patients, especially higher preponderance in second decade of life and more than 86% were found before 30 years of age. However, interestingly Swasdison *et al*. in a retrospective review of 67 cases from Thailand indicated that 21.1 years was the mean age [11]. Furthermore, Ochsenius *et al*. in a retrospective review from Chile revealed a similar mean age of 21.03 years. Leon *et al*. in a clinicopathological and immunohistochemical study of 39 cases of AOT showed lower mean ages of 16 years [12]. Arotiba *et al*. in their retrospective review of 57 cases for Black African population reported the mean age as 17 years [13]. AOT occured predominantly in the maxilla, with maxilla-to-mandible ratio of 2.1:1 with greater likelihood of occurring in the anterior maxilla. Another interesting noteworthy finding included greater incidence of the right side being affected. The maxilla-to-mandible ratio of AOT for Sri Lanka previously reported as 2.3:1. Comprehensive studies from Nigeria by Arotiba *et al*. also concluded with an anterior maxillary preponderance (Table 1). Literature reports of AOT in the anterior mandible are few and hence intruiging cases in the anterior mandible were usually discussed as case reports. It has been the endeavour of the authors to highlight the incidence of AOT occurring in the mandible through published literature from 1990 to present alongwith its histological variant, impacted tooth, treatment and recurrence rate [14]. Philipsen *et al* in his landmark article reviewed 499 cases to study the biological profile of AOT's. Their noteworthy finding included that mandibular involvement was higher in age group of 30 years and above. Matos *et al* reiterated the marked predilection of AOT in the maxilla and reported only 1 (6.7 %) case of follicular AOT in the premolar region of mandible out of a total 15 cases. Muhammed *et al* in their study reviewed 33 cases, in which the incidence of AOT's in the mandible included 13 cases (39%). These AOT's were further described according to their site: 6 cases were large extending from the entire area from the incisors to the molars (18%), and 5 cases (15%) affected the anterior incisor to the premolar area [15]. Only 2 reported cases (6.1%) affected the incisor area of the mandible. A striking feature of this study in South African population was the large size of the AOT's extending beyond the midline in half of the cases. Adisa *et al* conducted a retrospective review of 61 cases of adenomatoid odontogenic tumour which reported the total mandibular presentation of 44.3%. The incidence in anterior mandible was 20 (32.8%) cases whilst the posterior mandible included 7 (11.5%) cases. The increased percentage of extra-follicular variant of AOT (32.8%) obtained in this study partly attributed to this finding. Arotiba *et al* in their study comprising of 57 cases reported mandibular involvement in 35.7% (20 cases) with increased predilection of extrafollicular tumours in the mandible (69.2%). Fernandes *et al* reported the incidence of mandibular AOT's in 7 out of 54 cases (17%) they had reviewed [16]. Philipsen *et al* in their recent study (2007) reported the incidence of mandibular follicular and extrafollicular AOT's which accounted for 35.7% and 35.3% respectively. They also indicated that the peripheral variant was rarely encountered in the mandible (0.3%). Swasdison *et al* conducted a comprehensive study in Thai population and reviewed 67 AOT cases and the reported incidence in the mandible was total of 22 (32.8%) out of which 14 (20.9%) were in the anterior region and 8 (11.9%) were in the posterior region. Clinical features of AOT generally pivoted around complaints associated with a missing tooth. AOT was described as a gradually slow growing swelling which asymptomatic in nature. The extent of the lesion is approximately 2 to 7 cm, gradually increasing in size over time, resulting in a painless swelling of the jaws. Radiologically, an unilocular cystic mass enclosing the unerupted tooth was appreciated. Immunohistochemical investigations can also be carried out for the diagnosis of AOT [17]. It involves the identification of monoclonal and polyclonal antibodies which are used to detect specific antigens in the sections of the tissues [18-20]. Several immunohistochemical markers of the tumor include keratin, vimentin, amelogenin, enamelin, and matrix metalloproteinase (MMP-7 and MMP-26). The treatment of choice for AOT is usually enucleation of the tumor along with the removal of teeth associated with the lesion. For intra-bony defects secondary to AOT, guided tissue regeneration along with the placement of a membrane is advisable for filling of large defects.

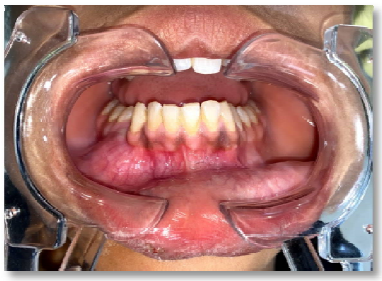
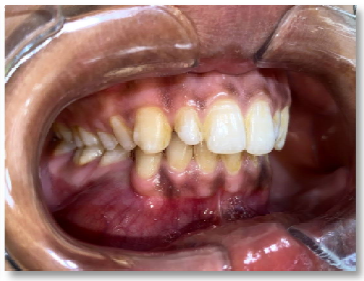



Figure 1: swelling in the Right buccal vestibule Figure 2: Pre-operative OPG

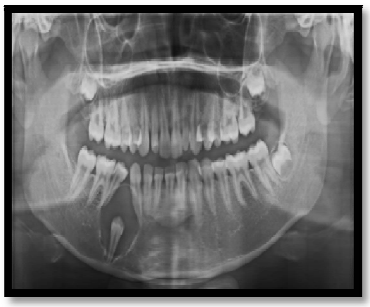
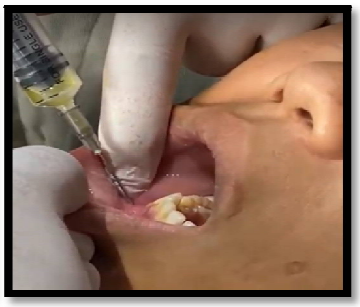



Figure 3: Aspiration revealed a yellowish straw-radiolucency with 44 in situ and coloured fluids

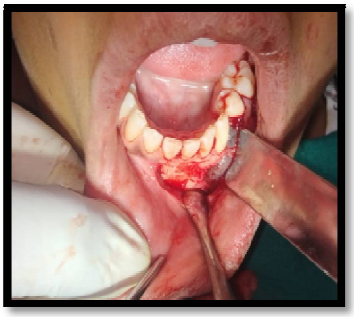
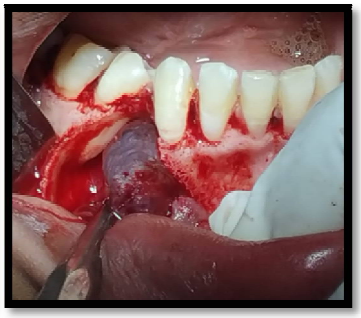



Figure 4: exposing the underlying lesion. Figure 5: Buccal cortical plate.

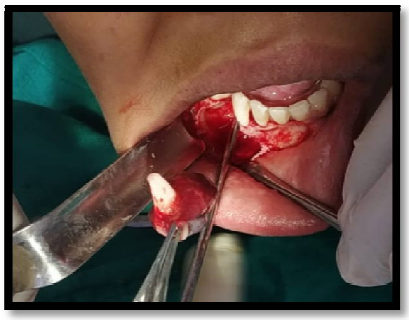
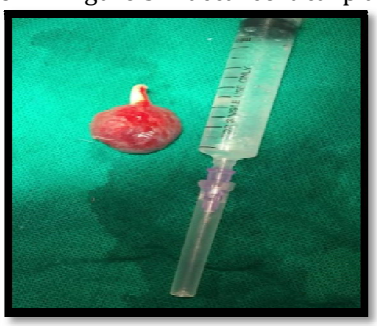



Figure 6: The cystic lesion was enucleated Figure 7: specimen (2*3 cm)

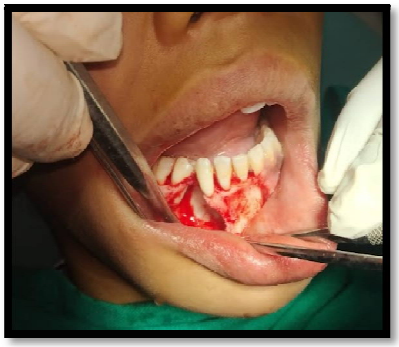
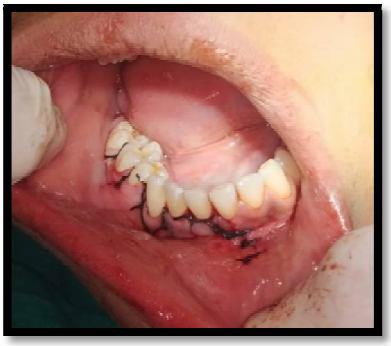
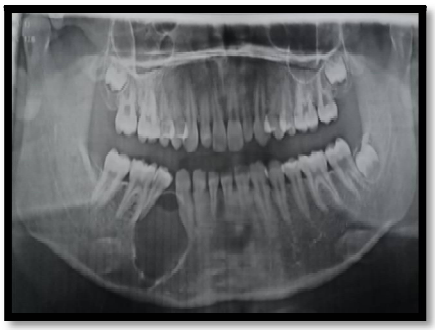



Figure 8: Residual bony defect in peripheral ostectomy Figure 9: Primary closure done



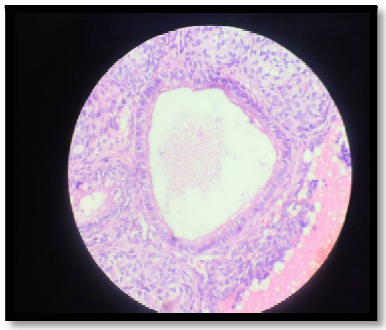
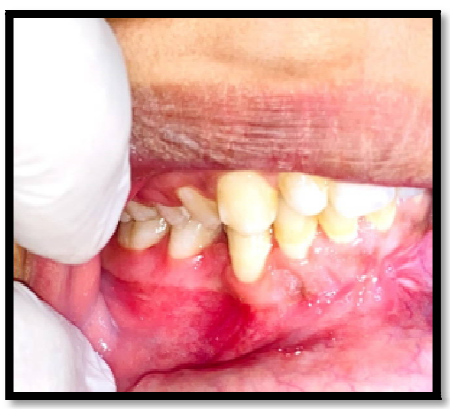


Figure 10: Immediate post-operative OPG Figure 11: histopathological examination



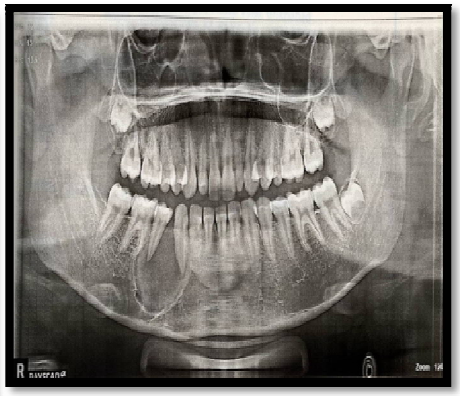


Figure 12: Post-operative depicting good soft tissue healing. Figure 15: Post-operative OPG 3 months

CONCLUSION

The results from the present review of articles concluded that AOT occurred most frequently in the second decade of life, it was more prevalent in females and oftenly associated with maxilla, predominantly affecting anterior region. and presenting mostly in the right side. However, mandible as a site of occurrence is reported in various case reports and case series, and we intended to highlight the rare incidences and review them in detail within the established scope of reported literature. Immaculate documentation of these rare cases is required with longer follow-ups discussing the treatment and recurrence in detail in further studies.

REFERENCES

1. Harbitz F. (1915). On cystic tumors of the maxilla, and especially on adamantine cystadenomas (adamantomas). *Dent Cosmos.* 57:1081–93
2. Pindborg JJ, Kramer JR, Torloni H. (1971). Histological typing of odontogenic tumours, jaw cysts and allied lesions. Geneva: WHO.
3. Philipsen HP, Reichart PA. (1999). Adenomatoid odontogenic tumour: facts and figures. Oral Oncol. 35(2):125–31
4. Philipsen HP, Reichart PA, Zhang KH, *et al*. (1991). Adenomatoid odontogenic tumor: biologic profile based on 499 cases. J Oral Pathol Med; 20: 149–58
5. Philipsen HP, Reichart PA, Siar CH, Ng KH, Lau SH, Zhang X, *et al.* (2007). An updated clinical and epidemiological profile of the adenomatoid odontogenic tumour: a collaborative retrospective study. J Oral Pathol Med. 36:383-93.
6. Ide F, Mishima K, Kikuchi K, Horie N, Yamachika S, Satomura K, Shimoyama T, Sakashita H, Saito I, Kusama K. (2011). Development and growth of adenomatoid odontogenic tumor related to formation and eruption of teeth. Head Neck Pathol.; 5(2):123-32
7. Toida M, Hyodo I, Okuda T, Tatematsu N. (1990). Adenomatoid odontogenic tumor: report of two cases and survey of 126 cases in Japan. J Oral Maxillofac Surg. 48:404-08
8. Swasdison S, Kittipong D, Jainkittivong A, Philipsen HP. (2008). Adenomatoid odontogenic tumors: an analysis of 67 cases in a Thai population. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 105:210-15
9. Arotiba GT, Arotiba JT, Olaitan AA, Ajayi OF. (1997). The adenomatoid odontogenic tumor: An analysis of 57 cases in a black African population. J Oral Maxillofac Surg; 55:14
10. Ochsenius, Germán & Ortega, Ana & Godoy, Luis & Peñafiel, Cristian & Escobar, Enrico. (2002). Odontogenic tumors in Chile: A study of 362 cases. Journal of oral pathology & Medicine. 31. 415-20.

11. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. (2005). Adenomatoid odontogenic tumor of the mandible: Review of the literature and report of a rare case. Head Face Med. 1:3.12-15
12. Sriram G, Shetty RP. (2008). Odontogenic tumors: a study of 250 cases in an Indian teaching hospital. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 105(6):14-21
13. Mohsmed A, Singh S, Raubenheimer EJ. Bouckaert MM. (2010). Adenomatoid odontogenic tumour: review and an analysis of 33 cases from South Africa. Int J Oral Maxillofac Surg. 39:843-46
14. de Matos FR, Nonaka CF, Pinto LP, de Souza LB, de Almeida Freitas R. (2012). Adenomatoid odontogenic tumor: retrospective study of 15 cases with emphasis on histopathologic features. Head and Neck Pathol. 6(4):430-437
15. Becker T, Buchner A, Kaffe I.(2012). Critical evaluation of the radiological and clinical features of adenomatoid odontogenic tumour. Dentomaxillofac Radiol. 41(7):533-40.
16. Adisa AO, Lawal AO, Effiom OA, Soyele OO, Omitola OG, Olawuyi A, *et al*. (2016). A retrospective review of 61 cases of adenomatoid odontogenic tumour seen in five tertiary health facilities in Nigeria. Pan Afr Med J. 24:102.
17. Chrcanovic BR, Gomez RS. (2019). Adenomatoid odontogenic tumor: An updated analysis of the cases reported in the literature. J Oral Pathol Med. 48:10-6
18. Siriwardena, B.S.M.S. & Udagama, Muthuranwelli & Tennakoon, Tennakoon & Athukorala, Demin & Jayasooriya, Primali & Tilakaratne, Wanninayake. (2020). Clinical and demographic characteristics of adenomatoid odontogenic tumors: analysis of 116 new cases from a single center. Brazilian Journal of Otorhinolaryngology. 88. 10-16
19. Chaabani I, Bouguila J, Kammoun R, Chebbi R, Sriha B, Khochteli H, Ben Alaya T. (2022). Radiological features of Adenomatoid odontogenic tumor: Report of a maxillary case and a mandibular one. Clin Case Rep. 10(1): 05301
20. Bansal SP, Shaikh S, Arvandekar AS, Dhanawade SS, Desai RS. (2022). Analysis of 55 cases of adenomatoid odontogenic tumor in an Indian population and review of literature. Medicina Oral, Patologia Oral y Cirugia Bucal. 27(1):85-93

CITATION OF THIS ARTICLE

Amit B. Lall, Tanya Kusum, Priyanka T. Rare Presentation of Adenomatoid Odontogenic Tumour in Anterior Mandible: Case Report. Bull. Env.Pharmacol. Life Sci., Spl Issue [2]: 2022:06-11