ORAL SURGERY

Oral Surgery ISSN 1752-2471

CASE REPORT

Adenomatoid odontogenic tumour in the anterior mandible: a case report

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Key words:
adenomatoid odontogenic tumour, cone-beam computed tomography, mandible, pantomography

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Accepted: 19 April 2020

doi:10.1111/ors.12496

Abstract

Adenomatoid odontogenic tumours are hamartomatous lesions that rarely deviate from their well-recognised radiological features. However, increasing numbers with atypical radiological features have been reported in recent years. This study reports on a large extrafollicular lesion in the anterior mandible with uncommon radiological features in a 17-year-old female. Treatment included enucleation with a histopathological confirmation. Healing was uneventful.

Statement of clinical relevance

The current lesion depicted atypical radiological features. Distinctive radiological features may not always be apparent in conventional radiographs. Hence, the use of advanced imaging may improve radiographic interpretation in differentiating from radiographically similar appearing lesions.

Introduction

Adenomatoid odontogenic tumours (AOTs) are relatively rare odontogenic lesions. It is classified according to the World Health Organization (WHO) as a benign odontogenic tumour of epithelial origin[1–5], and is known to represent 2.2% to 7.1% of all odontogenic tumours[6–11]. A recent global survey revealed a prevalence of 0.6–38.5% based on more than 1500 reported cases[12]. For several years the AOT was known as 'adenoameloblastoma' for it was considered to be a histological variant of the solid/multicystic ameloblastoma. Subsequently, it has been proven to be a separate entity due to its distinctive clinical and biological features[10,13,14]. All AOT variants demonstrate identical histology which indicates a common derivation[2,9]. It is hypothesised that the lesion originates from reduced enamel epithelium, enamel organ epithelium and cell rests of Malassez. Recently, it has been suggested that the lesion may arise from remnants of the dental lamina associated with the gubernacular cord[3,10,15,16]. Conventional presentation, include clinically, a slow-growing asymptomatic intraoral swelling; demographically, occurrence in the second decade of life with females affected more frequently than males (2:1); site, marked predilection for the anterior segments of the

Table 1 Comparison of clinical and radiological features of previous studies

	Mean age (years)	Female: Male	Maxilla: Mandible	Most common site	Association with teeth	Unilocular: Multilocular	Well-defined: Ill-defined	Presence of calcifications
Reichart and Philipsen[11]	±19	1.9:1	2.2:1	Max. ant.	71.3%	NA	NA	NA
Philipsen *et al.*[29]	±19	1.9:1	1.6:1	Max. ant.	70.8%	NA	NA	NA
Mohamed *et al.*[14]	15	5.6:1	1.5:1	Max. incisor to molar	100%	1:0	1:0	0%
Becker *et al.*[6]	18.4	1.4:1	1.7:1	Max. ant.	69%	10:1	5.7:1	77%
Jiang *et al.*[7]	17	7:1	3:1	Max. ant.	87.5%	3:1	3:1	100%
Madiyal *et al.*[16]	19	2.5:1	1:1.2	Mand. post.	65.7%	1.1:1	NA	40%
Sethi *et al.*[21]	19.2	1.5:1	1:3.2	Mand. ant.	NA	NA	NA	NA
Chrcanovic and Gomes[12]	19 ± 9	1.9:1	1.6:1	Max. ant.	73%	90:1	76:1	NA

NA, not available.

jaw, with incidence twice as often in the maxilla; and radiographically, a well-circumscribed homogenous unilocular radiolucent lesion with a sclerotic border[2,3,9,17] rarely exceeding 3 centimetres (cm) in maximum diameter[1,8,18]. Other features include associated tooth displacement and though rare, root resorption. Some present with fine 'snowflake' like calcifications which can aid in differentiation from other radiographically similar appearing lesions[1–3]. There are three clinical variants: (1) intraosseous follicular (pericoronal) type; the (2) intraosseous extrafollicular (extracoronal) type and the (3) peripheral (extraosseous) type[6,19,20]. The majority of the lesions arise intraosseously and are associated with the crown of an unerupted tooth, most often the canine. The less common extrafollicular type arises in the inter-radicular alveolar bone between the roots of teeth. The rare peripheral extraosseous type occurs on the buccal gingiva[3,9,10,18]. Although the majority present with the common described clinical and radiological features, more lesions with uncommon and extraordinary features are being reported (Table 1)[16,21–28].

CASE REPORT

A 17-year-old female was referred to the Department of Maxillofacial and Oral Surgery, Faculty of Dentistry, University of the Western Cape, Tygerberg Hospital, South Africa. She presented with a firm non-tender expansile lesion in the anterior mandible which was first noticed 3 months prior. Intraoral examination revealed several carious teeth, partially erupted third molars and mobile mandibular anterior incisors. The patient was otherwise healthy with no medical history or known allergies. A pantomograph (PAN) was performed revealing: several carious teeth, partially erupted third molars, a grossly carious 26 with a periapical radiolucency; and a large well-defined, corticated, ovoid, homogenous, radiolucent lesion in the anterior mandible, extending in the mediolateral plane from the mesial aspect of the 33, crossing the midline, to the mesial aspect of the 46, and superior-inferiorly from the lower border of the mandible to the alveolar ridge; with expansion, thinning of the cortex, tooth displacement and root resorption (Fig. 1). A cone-beam computed tomography (CBCT) scan was performed and the volume evaluated in all three planes. Maximum intensity projections (MIP) (Fig. 2A–C) and CBCT images (Fig. 3A–D) revealed a large circumscribed, thin-walled, expansile, cystic-like lesion in the anterior mandible. It appeared as a low-density containing multiple fine flecks of scattered high-density calcifications along the periphery; causing, root resorption, tooth displacement and multiple interruptions in the lingual and buccal cortical plates. The lesion measured 33 mm × 42 mm × 30 mm in its maximum dimensions. Thickening of the left maxillary sinus membrane was noted. All lower anterior teeth were shown to be vital on a cold test. Surgical treatment was performed under general anaesthesia. A crestal incision facilitated the extraction of teeth 32, 31, 41, 42 and 43 along with complete enucleation of the lesion. The lining separated fairly easily from the underlying bone. Bismuth iodoform iodide paraffin paste (BIPP) impregnated gauze was firmly packed in the cavity and secured using 3/0 silk sutures and wound closure was achieved with 3/0 chromic sutures. The BIPP pack was removed 4 days later and the patient was instructed to irrigate the surgical cavity with a syringe containing chlorhexidine as provisional home care. Patient follow-up and healing were uneventful. Macroscopic histopathological examination revealed a cyst-like structure with included teeth. Microscopic examination (Fig. 4A–F) showed a cyst-like lesion with nodules of odontogenic epithelium and a thick fibrovascular connective tissue

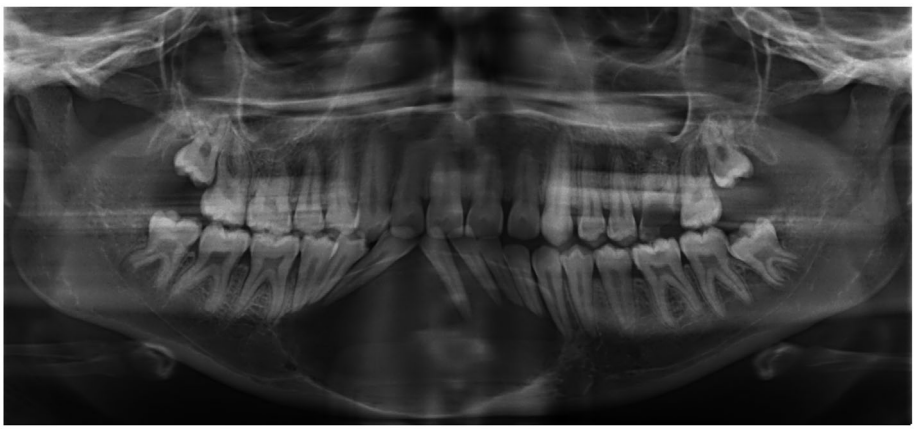

Figure 1 Pantomograph shows carious lesions, partially erupted third molars, grossly carious 26 with a periapical radiolucency and a very large well-defined, corticated, ovoid, homogenous, cystic-like radiolucent lesion in the anterior mandible. Causing expansion, thinning of the cortex, tooth displacement, and root resorption.

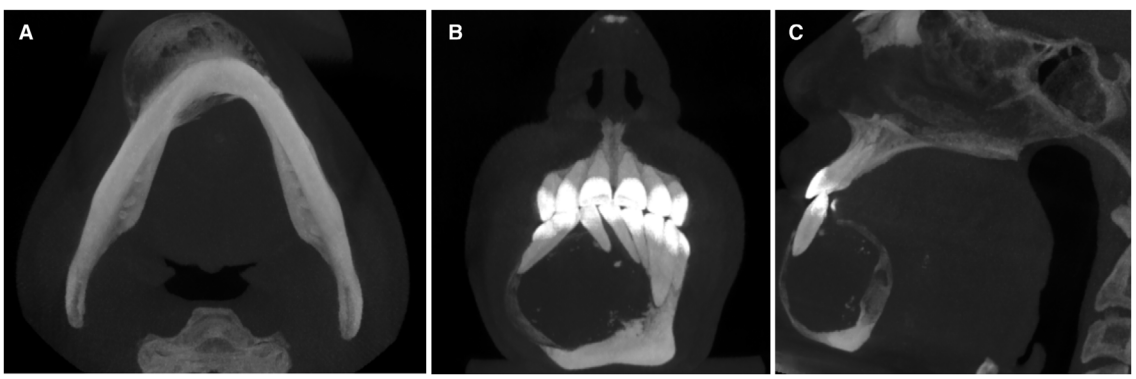

Figure 2 MIP images show a large expansile lesion in the anterior mandible appearing of low-density causing thinning of the cortex, tooth displacement, containing multiple fine flecks of scattered high-density calcifications. (A) Axial view, bucco-lingual expansion. (B) Coronal view, tooth displacement and high-densities along the periphery. (C) Sagittal view, thinning of the cortex and high-densities along the periphery.

capsule. There were characteristic duct-like structures within the epithelium containing amorphous eosinophilic material. The epithelium consisted of whorls and cords of spindle-shaped epithelial cells in a loose myxoid stroma with numerous calcifications. A histological diagnosis of an AOT was made.

DISCUSSION

The first comprehensive case reports were published by Harbitz from Norway in 1915 and then by Wohl from the United States of America[10,13,14]. However, recently it has been shown that in 1903 a Japanese general surgeon, Nakayama, reported cases with diagnostic evidence based on clinical and pathological findings[15]. AOT had several designations in the past but in 1971 the WHO adopted the term 'AOT' to describe this specific entity as it has been proposed by Philipsen and Birn[6,9].

In general, it had been accepted that AOTs comprise 2.2–7.1% of all odontogenic tumours[6–10]. Several epidemiological studies of the AOT in different parts of the world reveal variable frequency rates. According to geographic location, the reported incidence rates are Europe 1–4%, Middle East 2–4%, North America 2–7%, South America 4–7%, Asia 1–16% and Africa 1–39%.[14] Philipsen *et al.*[29] conducted a comprehensive worldwide survey of the literature that concluded a global relative frequency of 0.6–38.5% of odontogenic tumours. Recently similar findings were reported by Chrcanovic and Gomez[9,12].

Unanimous agreement regarding the peak incidence for a time of diagnosis is the second decade of life[3,9]. Studies show occurrence in this decade to be from 62.8% to 85% of all reported cases[9,14,16,19]. The mean age ranges between 15 and 24 years[6,14,18] with a global mean of 19 ± 9years[12]. Occurrence beyond

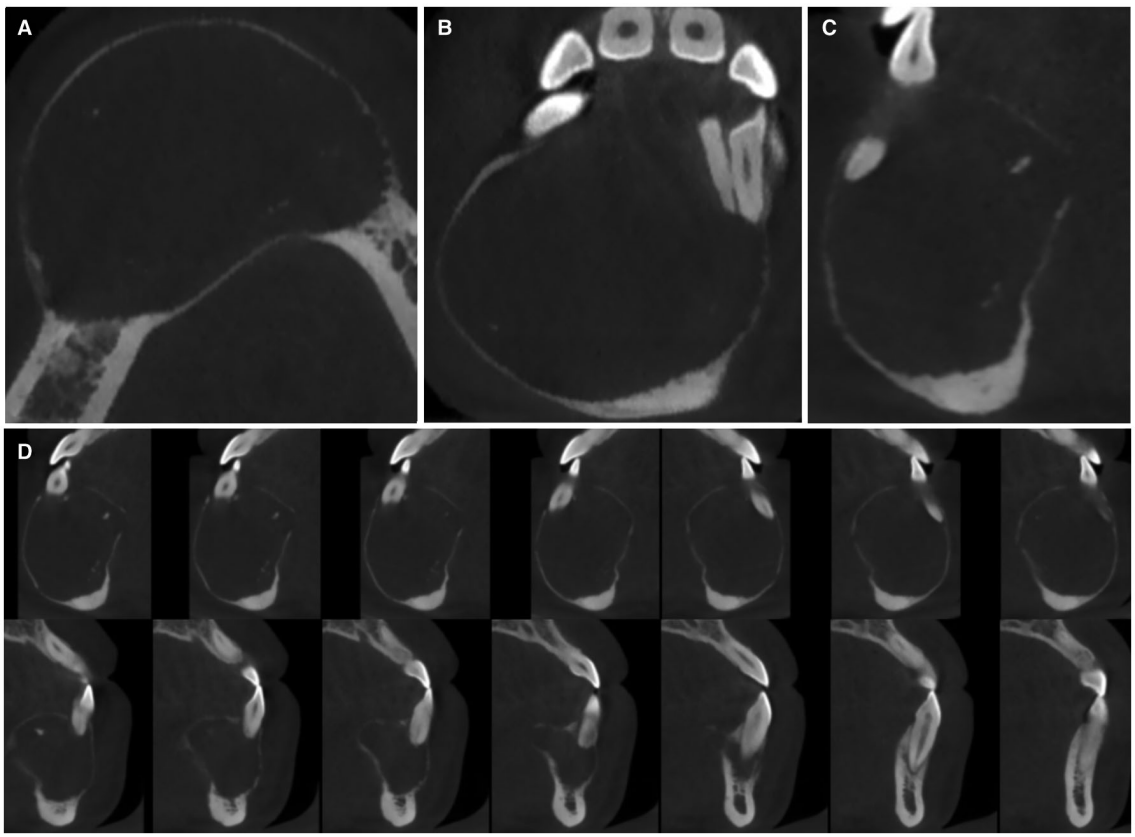

Figure 3 CBCT images show a large circumscribed, thin walled, expansile, cystic-like lesion in the anterior mandible. Appearing of low-density containing multiple fine flecks of scattered high-density calcifications. Causing root resorption, tooth displacement, and interruptions of the buccal and lingual cortices. (A) Cropped CBCT axial view, bucco-lingual expansion, and thinning of the cortex. (B) Cropped CBCT coronal view, root resorption. (C) Cropped CBCT sagittal view, discontinuity of the lingual cortical plate. (D) CBCT cross-sectional slices with 1.5 mm spacing, high-densities along the periphery.

the age of 30 years is considered to be uncommon[6,8]. Various age ranges of occurrence are reported from 5 to 50 years, 2 to 44 years with global reviews suggesting 1 to 82 years[9,12]. There is a definitive gender predilection towards females[3,9,12]. With 65.2% of lesions occurring in females according to a recent global review[12]. Studies reported female-to-male ratios ranging from 1.4:1 among Africans and non-Asians, 2.3:1 in Asians, 3:1 among the Japanese population, a study from South Africa concluded 5.6:1 and the global average 1.9:1[9,11,12,14,29]. Predilection for the maxilla and the anterior regions of the jaw is unanimous. With reported maxilla-to-mandible ratios ranging from 1.4:1 to 2:1[6,9,14] and a global average of 1.6:1[12]. Occurrence in the maxilla is recorded from various studies ranging from 45.7% to 76.3% with reported global average 61.8%. Subsequently, 67.9% of all cases involve a canine of which 40–83.3% and a global average of 66% involve the maxillary canine. The anterior maxilla is affected in 40–83.3% of reported cases with a global average of 66%[6,7,12,14,16,18–21,29]. Lesions in the mandible attribute to 27–54.8% of occurrences and globally 38.2% of which the anterior segments are affected in 28–58% of cases[12,14,16,18,21].

Intraosseous lesions consist of up to 97% of all AOTs[10,12,19]. With the majority of these the follicular type known to be from 65% to 97.2%[6,16,21] with a global average from 70% to 73%[7,8,11,18–20]. The extrafollicular type attributes from 24.3% to 27%[6,8,12,20] and the rare peripheral lesion is seen in 2.3–5% of the total reported cases[6,8,10–12,18,20]. Most often these lesions are asymptomatic but up to 13% reports symptoms such as pain[6,8].

Radiographically the AOT usually presents as a well-defined, unilocular, radiolucent lesion with curved, sclerotic borders giving it a cystic-like appearance. More often than not it is associated with an unerupted tooth. Tooth displacement with jaw expansion may be observed. The predilection for the

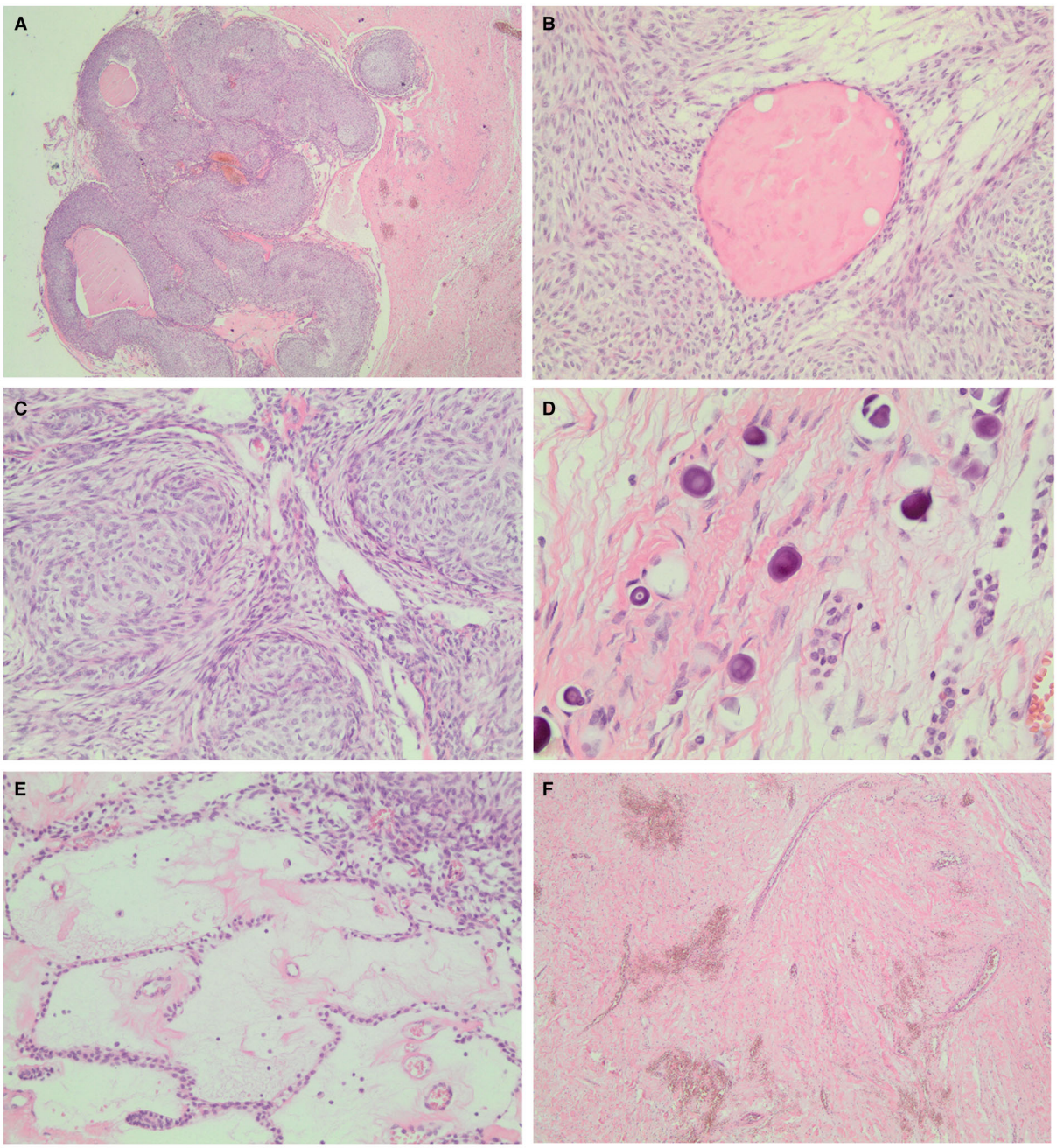

Figure 4 Photomicrographs of the present case's histological sections. (A) Showing nodules of odontogenic epithelium and a thick capsule. (B) Duct-like structures within epithelium containing amorphous eosinophilic material. (C) Whorls of spindle shaped epithelial cells in scant stroma. (D) Presence of multiple calcifications. (E) Epithelial cords in loose myxoid stroma. (F) The cyst-like wall comprising of fibro-vascular connective tissue.

mandible, lesions crossing the midline, cortical bone perforation, root resorption, ill-defined borders and the extrafollicular type is thought to be less common and has been associated with older patients[2,6–8,18]. The average reported AOT size ranges from 1 to 3 cm[1,8,18], but can range from 0.4 to 12 cm, with a mean of 2.9 cm[8,12]. Presence of the distinctive radiopacities or calcifications inside the radiolucent lesion varies from as little as 0–7.1%[7,14] to as great as 40–78%[6,8,16,18] of cases. This may be due to the radiographic technique implemented or the stage of the progressive development of the lesion. These

have been described as fine, faint irregular, amorphous, patchy, scattered, areas of snowflake-like, flecks or spicules[6]. Often concentrated around the crown and root of an involved unerupted tooth in the follicular type[7]. Periapical radiographs are considered to be the gold standard for observing such calcified deposits. Observations by a recent study related to increased frequency of opacities present to the degree of expansion of a lesion[6]. Due to its cystic appearance in some cases, the AOT can be misdiagnosed radiographically if calcifications are not present or visible. Determining the absence of attachment to the cementoenamel junction may help distinguish from a dentigerous cyst[3,15]. It is reported that 51.4–98.9%[6,12,16] present as unilocular and 1.1–9%[6,12,16] as multilocular. Well-defined lesions are reported in 85–98.7% of cases and 1.3–15% as ill-defined. Root resorption appears in 17.1–19%, cortical expansion in 68–89.5%, cortical perforation 9–45.6%[12], tooth displacement in 80% and crossing of the midline is seen in 12% of which 79% are in the mandible[6,12]. Lesions presenting predominantly radiolucent are 98% compared to 2% which are predominantly radiopaque. On the basis of radiographic findings differentiation from other lesions can be difficult; whereas the dentigerous cyst, unicystic ameloblastoma, odontogenic keratocyst, ameloblastic fibro-odontoma and the calcifying epithelial odontogenic cyst can be placed among the differential diagnosis[2,6,17,18].

Advanced imaging such as multidetector computed tomography may demonstrate more detailed features such as finer calcified deposits, especially when displayed in the soft tissue window. Nevertheless, CBCT images may be sufficient for diagnostic purpose[17]. Advantages of CBCT when compared to CT, include decreased radiation dose, shorter scanning time and overall cost reduction. The PAN is often unable to demonstrate the distinctive fine radio-opacities when calcification is minimal[6,7]. In such instances intraoral radiographs may be essential for radiographic interpretation should advance techniques not be available[8,16]. CBCT is superior compared to conventional techniques due to the ability to navigate multiple planes, eliminate superimposition and excellent contrast resolution for mineralised tissue structures. Distinctive features of the AOT such as internal calcifications and predilection for anterior regions in the jaw make CBCT proficient for radiographic evaluation[7].

Recurrence is reported to be very low due to the lesions well-encapsulated borders and benign behaviour[8–10]. There are no apparent clinical and radiological differences between the AOTs' variants[12]. Consequently, treatment entails conservative thorough enucleation and curettage for all AOTs[8,9]. The mental- and inferior alveolar nerve should be considered when operating on lesions in the anterior mandible. Paraesthesia to the lower lip and chin may result due to intervention. Continued expansion of an untreated lesion has the potential of an ensuing pathological fracture[20]. It is well accepted that early follicular types can clinically and radiologically resemble a cystic lesion such as a dentigerous cyst[15]. The histological features are considered to be distinct and care should be taken to avoid misinterpretation. If mistaken for ameloblastoma, unnecessary radical surgery can result[3].

CONCLUSION

The presented AOT's size, location, uncommon clinical and radiological features make it notable. Distinctive radiological features may not be apparent in conventional radiographs. Hence, the use of advanced imaging such as CBCT could improve radiographic interpretation to differentiate from similar appearing lesions. More AOTs with unconventional radiological features are being reported: change in trends, publication bias, underreporting or geographical predilections may be suggestive[14,16,21]. A multidisciplinary collaboration that includes the treating clinician, radiologist and pathologist is recommended to achieve the correct diagnosis and an uncomplicated outcome.

Conflict of interest

The authors declare no conflict of interest.

Ethical approval

None required.

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