Multiple Adenomatoid odontogenic tumours associated with eight impacted teeth

Chané Nel [a]**:** BChD, MSc (Maxillofacial Radiology).

E-mail: chane.nel@up.ac.za

ORCID: 0000-0003-4047-6356

André Uys [a]**:** BSc, BChD, PGDipDent (Endodontics), PGDipDent (Aesthetic Dentistry), MSc (Maxillofacial Radiology), PhD (Anatomy).

E-mail: andre.uys@up.ac.za

ORCID: 0000-0001-8250-7662

Liam Robinson [a]**:** BChD, PDD (Maxillofacial Radiology), PDD (Forensic Odontology).

E-mail: liam.robinson@up.ac.za

ORCID: 0000-0002-0549-7824

Willie FP van Heerden [a]**:** BChD, MChD (Oral Path), FC Path (SA) Oral Path, PhD, DSc, MASSAf.

E-mail: willie.vanheerden@up.ac.za

ORCID: 0000-0003-2494-667X

[a] Department of Oral Pathology and Oral Biology, Faculty of Health Sciences, University of Pretoria, South Africa.

Corresponding author:

Chané Nel

Address: Room 6-17, Pretoria Oral Health Care Centre, Corner of Steve Biko and Dr Savage Road, Pretoria, 0084

Contact number: +27 12 319 2311

E-mail: chane.nel@up.ac.za

Disclosure

Conflict of Interest: The authors declare that they have no conflict of interest.

Funding: This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethics

This study was approved by the University of Pretoria, Faculty of Health Sciences Research Ethics Committee (Reference no.: 266/2020). All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation and with the Helsinki Declaration of 1964 and later versions. This article does not contain any studies with human or animal subjects performed by the any of the authors.

Word Count:

Abstract: 117

Manuscript: 2116

Number of references: 41

Number of figures: 8

Abstract

Adenomatoid odontogenic tumour (AOT) is a benign tumour that arises from odontogenic epithelial remnants. AOTs usually present as innocuous lesions with limited growth potential. Multiple AOTs are frequently reported in the literature, with reports of tumours associated with up to seven impacted teeth. Multiple AOTs have also been described in association with Schimmelpenning syndrome. This case report highlights the rare occurrence of multiple AOTs involving eight impacted teeth in a 9-year-old male patient. Radiographic examination showed features of enamel hypoplasia and prominent dilated gubernaculum dentis associated with some of the impacted teeth. The patient also presented with a linear epidermal nevus involving the left face and intraoral mucosal papillomatous growths, clinical features highly compatible with Schimmelpenning syndrome.

Keywords: Adenomatoid odontogenic tumour, Schimmelpenning Syndrome, Gubernaculum dentis, Multiple impacted teeth

Introduction

Odontogenic tumours include a spectrum of lesions with diverse biological behaviours ranging from innocuous hamartomatous lesions to aggressive neoplasms. Adenomatoid odontogenic tumour (AOT) is a benign tumour that arises from odontogenic epithelial remnants. Authors have hypothesised that the epithelial source of this tumour could be derived from the dental lamina or remnants located within the gubernacular dentis [1–4].

Adenomatoid odontogenic tumours are classified into intraosseous lesions, which includes follicular and extra-follicular variants, and a peripheral variant [1]. In the follicular variant (47-71% of cases), the lesion encircles the crown of an unerupted tooth, closely resembling a dentigerous cyst [5–7]. The extra-follicular variant (16-45% of cases) presents in the alveolar bone with no specific relation to teeth [5–7]. The peripheral variant (0.02-2% of cases) is rare, with limited information on its clinical presentation and biological behaviour [5,7,8]. Some authors argue that a tooth associated with AOT can erupt, transporting transformed cells to the alveolar bone (extra-follicular variant) or gingiva (peripheral variant). Age, sex and location similarities between the three types may support this hypothesis [1]. AOTs usually present as innocuous lesions with limited growth potential, reaching an average size of approximately 3cm [7,9]. In contrast, a South African review of 33 AOTs showed extensive growth in more than half of cases [10].

This case report highlights the rare occurrence of multiple AOTs with extensive growth involving numerous impacted teeth. In addition, the patient presented with a linear epidermal nevus involving the left face and intraoral mucosal papillomatous growths, clinical features highly compatible with Schimmelpenning syndrome.

Case Report

A 9-year-old male patient presented with a slow-growing tumour of unknown duration involving the left maxilla. The patient's mother reported no co-morbidities. A previous neurological examination was normal with no history of seizures. Extraoral examination revealed multiple raised pigmented lesions arranged in a linear pattern involving the left side of the face and upper lip (Fig.1). On intraoral examination, the left maxillary alveolus showed intrabony expansion with unilateral exophytic papillomatous growths involving the palate and gingival mucosa (Fig.2). There were numerous absent teeth on the left side, with only a displaced permanent central incisor and canine visible on examination. The deciduous central incisor was over-retained in a labial

position to the erupted successor. On questioning, the mother of the patient reported no previous history of extractions.

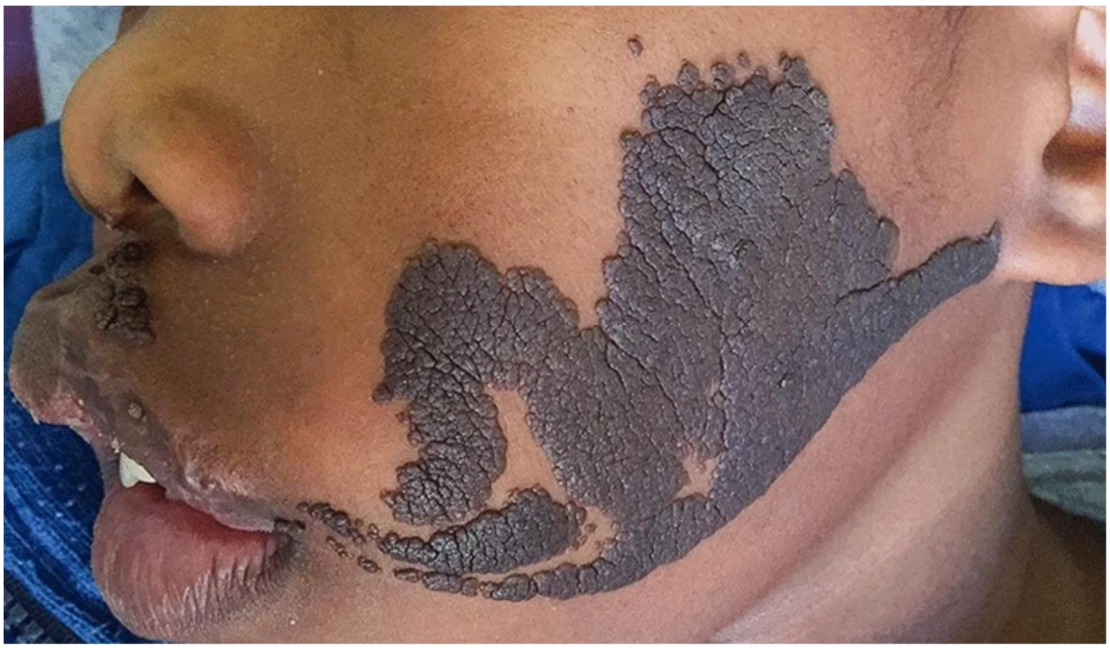

Figure 1. Clinical image showing multiple raised pigmented lesions arranged in a linear pattern involving the left side of the face and upper lip

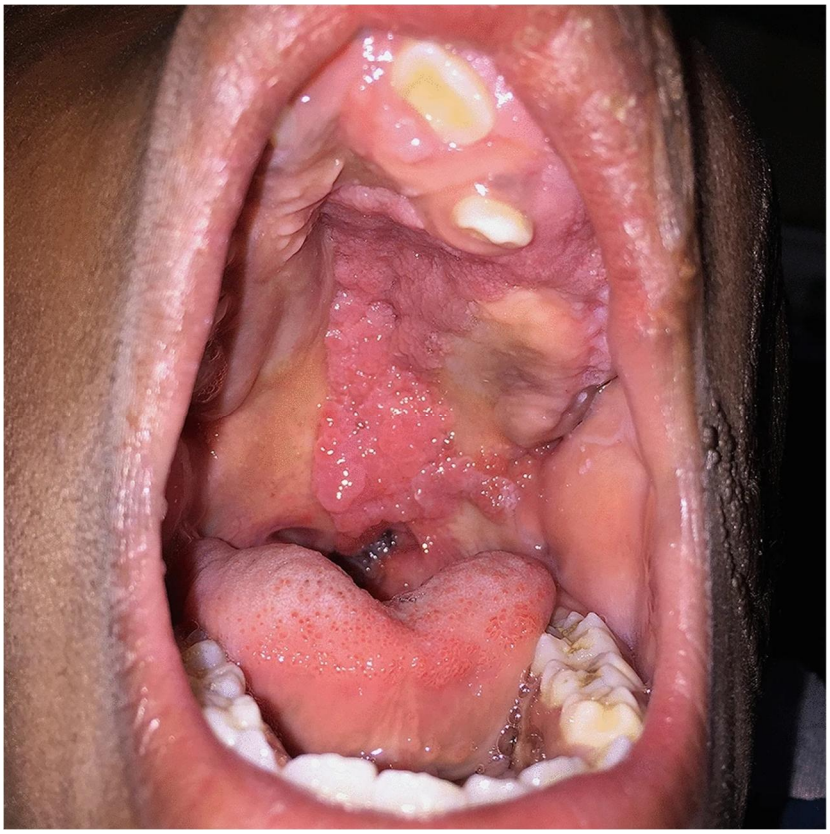

Figure 2. Intraoral image showing expansion of the left maxilla with numerous clinically missing teeth. Unilateral exophytic papillomatous growths involving the palate and gingival mucosa were also visualised

A panoramic radiograph showed multiple expansile radiolucent lesions with internal calcifications associated with numerous impacted teeth (Fig.3). Unfortunately, due to superimposition, the individual impacted teeth could not be delineated on the panoramic radiograph. Cone-beam computerised tomographic (CBCT) imaging was performed to examine the extent of the lesions. The lesions resulted in significant expansion and displacement of the left maxillary sinus floor with associated mucosal thickening (Fig.4). The deciduous teeth initially suspected to have undergone spontaneous exfoliation, were impacted and contained within the expansive lesions (Fig.5A-E). A radiolucent lesion with varying amounts of internal calcifications encased each impacted tooth. The left maxillary permanent lateral incisor exhibited areas of enamel hypoplasia, with a canal-like structure connecting the pericoronal lesion to the alveolar ridge (Fig.5B). The authors speculated that this structure represented a dilated gubernacular dentis. A similar phenomenon was seen involving the second deciduous molar and second premolar (Fig.6). Interestingly, the deciduous molars were impacted superiorly in relation to their permanent successors. In addition, the first permanent molar was also impacted superiorly in comparison with the developing second permanent molar.

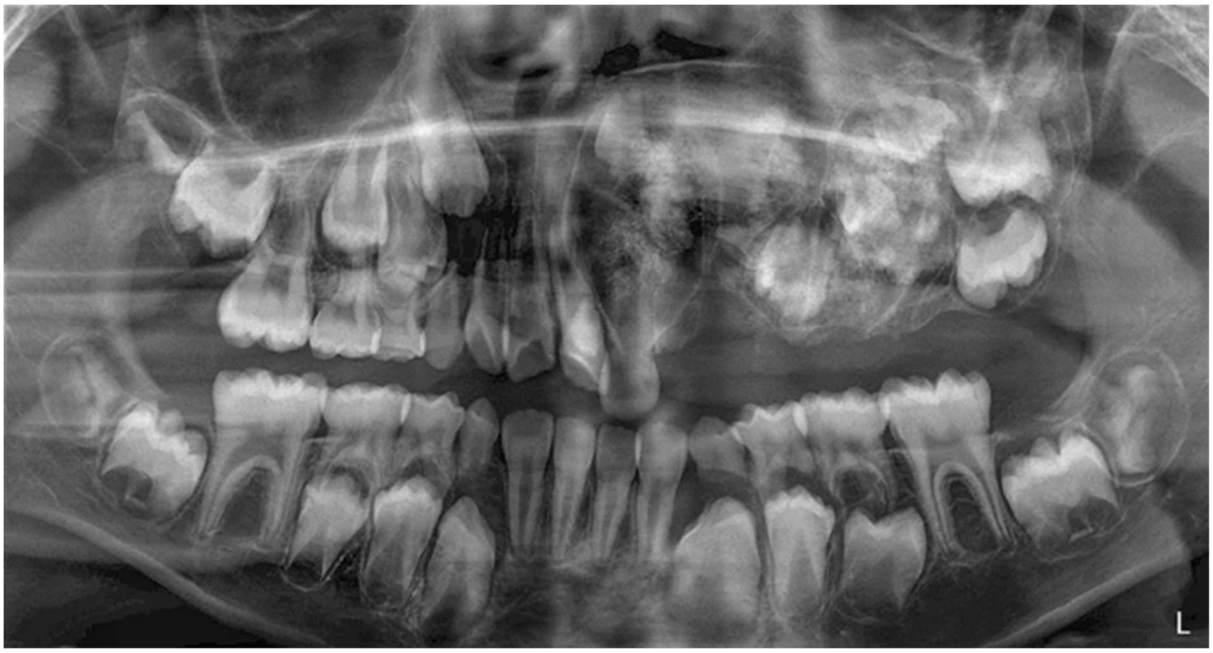

Figure 3. Panoramic radiograph illustrating multiple mixed radiolucent–radiopaque expansile lesions involving the left maxilla associated with numerous impacted teeth

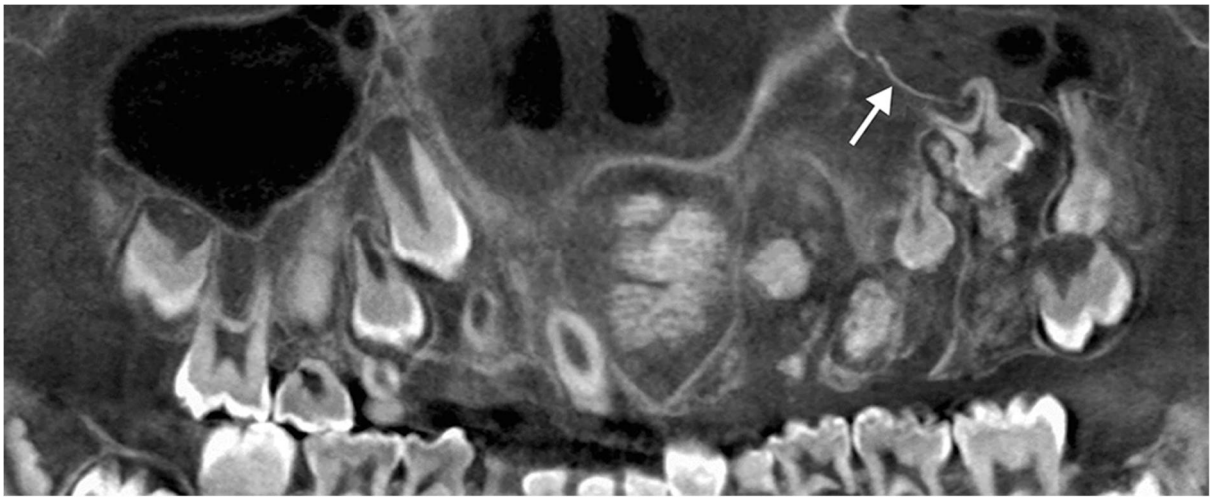

Figure 4. Panoramic reconstruction of the cone-beam computerised tomography (CBCT) scan showing superior displacement of the left maxillary sinus floor with associated mucosal thickening (arrow)

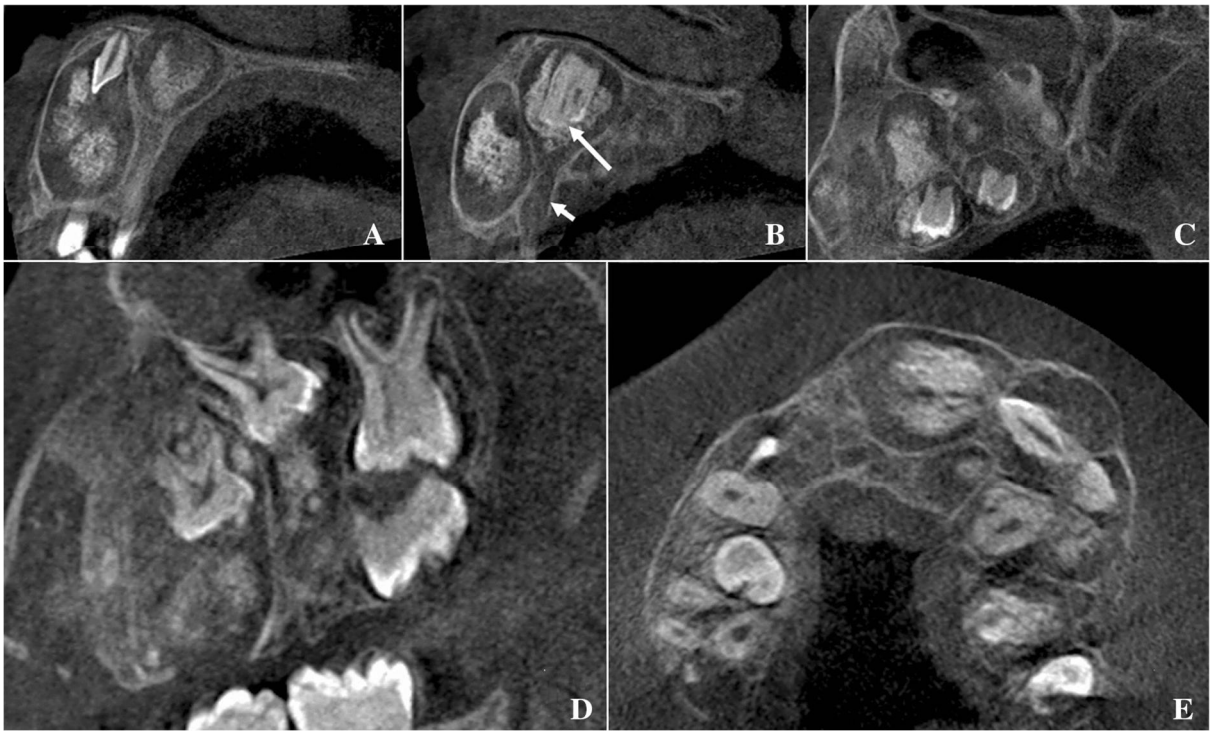

Figure 5. CBCT images illustrating the impacted teeth. **a** Sagittal slices of CBCT scan illustrating the impacted left maxillary deciduous canine and **b** permanent lateral incisor. Note the enamel hypoplasia (long arrow) and canal-like structure (short arrow) associated with the permanent impacted lateral incisor. **c** Sagittal slices also illustrating the impacted left maxillary permanent premolars and **d** primary molars with first and developing second permanent molars. **e** Axial slice illustrating a horizontally impacted left deciduous lateral incisor

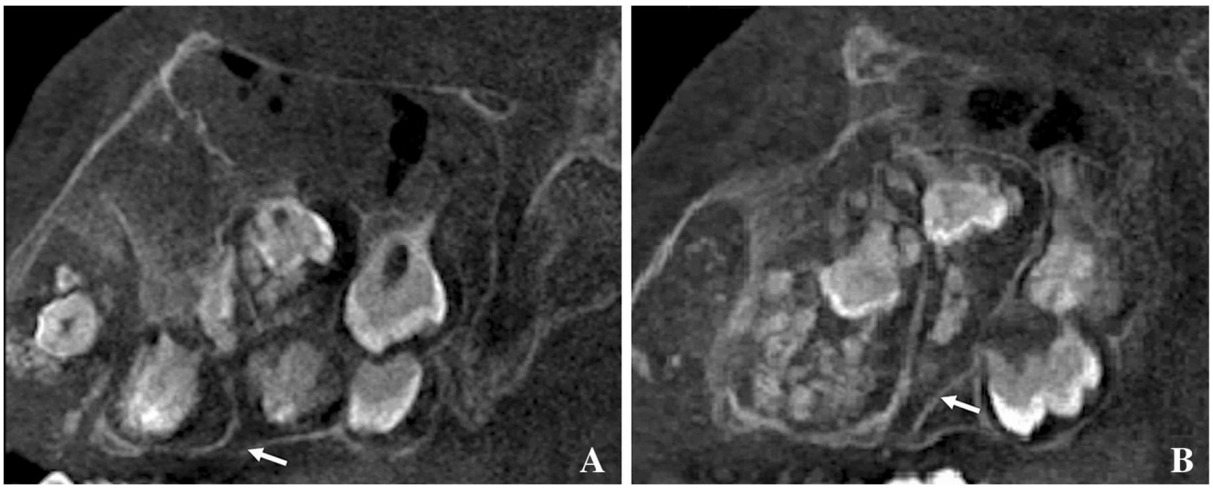

Figure 6. Sagittal slices of the CBCT scan showing additional canal-like structures (arrows) associated with **a** the second premolar and **b** the second deciduous molar

An incision biopsy was performed from the left upper lip for histological assessment. The specimen consisted of a papillomatous skin fragment measuring 8x5x4mm. Histological evaluation showed skin represented by epidermis and dermis only. The epidermis showed uniform acanthosis with hyperkeratosis and broad low papillomatosis (Fig.7). The basal cell layer showed a slight increase in melanin pigment. The dermis consisted of fibrous connective tissue with scattered skeletal muscle bundles and a mild, non-specific chronic inflammatory infiltrate. The clinical presentation and histological features were those of a linear epidermal nevus.

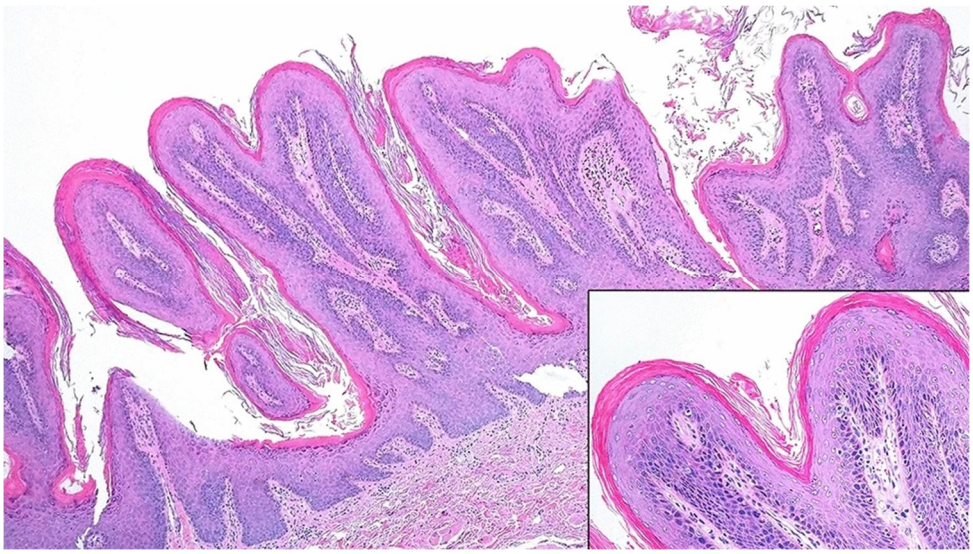

Figure 7. H&E-stained sections showing a papillomatous skin fragment with uniform epidermal acanthosis with hyperkeratosis (original magnification × 4). Insert: higher magnification of the papillomatous epidermis (original magnification × 20)

Two of the intrabony cystic lesions, in the canine and the premolar regions, were enucleated and submitted for histological assessment. The first specimen (canine region) consisted of an unopened cystic mass measuring 21 x 20mm, with a wall thickness of 4 mm. On sectioning, the cyst was attached to the neck of the tooth. The second specimen (premolar region) also consisted of a cystic mass measuring 20x15mm, with a wall thickness of 5 mm. On sectioning, the cyst was partially attached to the neck of the tooth. Histological evaluation of both specimens showed similar features. Sections showed multiple cystic tumours surrounded by thick fibrous capsules. The lumens of the cysts contained a nodular proliferation of nondescript odontogenic epithelium with minimal intervening stroma. These nodules contained variably sized duct-like structures lined by columnar and cuboidal epithelium, with nuclei showing reverse polarisation. Small foci of round calcifications were frequently seen within the nodular epithelium (Fig.8). A final diagnosis of adenomatoid odontogenic tumour was made in both specimens.

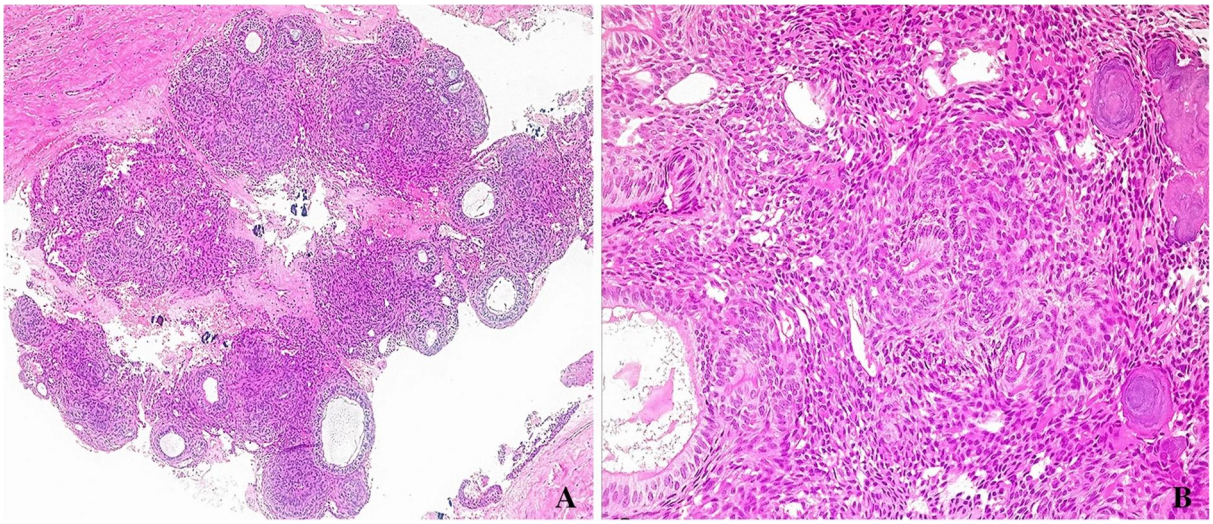

Figure 8. **a** Low-magnification H&E-stained section showing a cystic tumour surrounded by a thick fibrous capsule (original magnification × 4). **b** Higher magnification H&E-stained section showing tumour nodules consisting of variably sized duct-like structures lined by an ameloblastomatous-type odontogenic epithelium with surrounding small foci of calcification (original magnification × 20)

The presence of a linear epidermal nevus, unilateral intraoral mucosal papillomatous growths and multiple AOTs were highly suggestive of Schimmelpenning syndrome. Unfortunately, due to financial constraints, genetic testing was not performed in this case. An ultrasound study of the abdomen and a cardiac Doppler ultrasound study found no abnormalities. The patient was scheduled for surgical removal of the remaining tumours with the associated impacted teeth.

Discussion

Adenomatoid odontogenic tumour (AOT) is a common odontogenic tumour with a reported prevalence ranging from 0.6-38.5%, depending on the study population [5,6]. The variation in prevalence may be attributed to genetic diversity in the different population groups. AOTs commonly present during the second decade of life, with females affected twice as often as males [5,7,9,10]. In a South African study AOTs represented 4% of all odontogenic tumours. Interestingly, in this demographic females were affected six times more often than males [10] AOTs commonly arise in the maxilla with a predilection for the anterior maxilla over the posterior region [5–7,9]. The maxillary permanent canine is associated in about 51% of cases [6].

Whether AOT represents a neoplasm, hamartoma or cystic lesion is still contentious [11]. Some authors speculate that due to its indolent behaviour and immunohistochemical profile, AOTs may be more consistent with a hamartomatous lesion [12,13]. This argument is further supported by studies showing the limited growth potential and low recurrence rate (0.2%) of the tumour [7,11]. In contrast, large aggressive AOTs with multiple recurrences have been reported in literature [14–16]. Shakur *et al.* hypothesised that the overexpression of anti-apoptotic proteins such as Bcl-2 and Bcl-X confirms the neoplastic nature of the tumour. In contrast, Bcl-X expression in odontogenic keratocysts was found to be higher than that of AOTs [17]. Several authors have been suggested that AOT be renamed to “adenomatoid odontogenic cyst”, due to the high prevalence of cystic areas within the tumour, and its similar clinical presentation and simultaneous occurrence with dentigerous cysts [8,18,19]. Thakur *et al.* disagree with this statement, advocating that the cystic presentation stems from a hyperplastic dental follicle with an area of the lining showing an AOT-like proliferation [11]. Other authors have termed AOTs exhibiting prominent cystic features as cystic-AOTs [19]. Another explanation is that the tumour has areas of cystic degeneration, or that they represent hybrid lesions associated with calcifying odontogenic cysts or unicystic ameloblastomas [11,20,21].

AOTs may be associated with primary teeth, supernumerary teeth, unerupted third molars and even odontomas [5,6,20,22–24]. Cases where the tumour was surrounded by a fibro-osseous reaction, or even lesions suspicious for focal cemento-osseous dysplasia, have been reported [25–27]. An AOT with a concomitant odontogenic keratocyst has also been reported in a patient with nevoid basal cell carcinoma syndrome/Gorlin-Goltz syndrome [28].

Multiple AOTs have been reported in association with Schimmelpenning syndrome (SS) or sebaceous nevus syndrome [29]. Postzygotic mutations in the HRAS and KRAS oncogenes have been implicated in the aetiopathogenesis of this neurocutaneous disorder. The majority of these mutations occur sporadically with no sex predilection [30]. This syndrome is characterised by linear epidermal nevi and neurological disturbances including seizures, delayed intellectual development and/or mental retardation [29–31]. The epidermal nevi characteristically run along the lines of Blaschko [29–31]. Less frequently cardiovascular, genitourinary, skeletal or ophthalmic abnormalities may also be present [29,30]. Common intraoral findings include exophytic papillomatous growths of the mucosa that can be continuous with the epidermal lesions [29,31]. In this syndrome, the impacted teeth associated with multiple AOTs often exhibit enamel hypoplasia [29]. Other authors have reported a SS patient with an AOT and misshapen/pigmented teeth, possibly representing another form of enamel hypoplasia [32]. The authors speculate that the involvement of multiple AOTs with deciduous teeth may be due to the KRAS mutation occurring early in embryogenesis [29]. The current case shares many similarities with reports of SS including the linear epidermal nevi, mucosal exophytic papillomatous growths, enamel hypoplasia and multiple AOTs. However, the patient did not exhibit any neurological or other organ system abnormalities. Davies and Rogers reported that the incidence of neurological disturbances was only clinically detectable in 7% of patients in a 196 cohort [33].

The gubernaculum dentis is a normal anatomic structure consisting of a fibrous band, the gubernacular cord, running in a bony canal termed the gubernacular canal. This canal connects the dental follicle of a developing successor tooth to the alveolar crest, forming an eruptive pathway [1]. Authors speculate that remnants of the dental lamina within gubernacular dentis play a role in the aetiopathogenesis of AOT [1,34]. Several reports in literature have highlighted the radiographic presentation of AOTs associated with the gubernaculum dentis [1,35].

Radiographically, AOTs usually present as a unilocular radiolucent lesion with well-defined borders commonly containing irregular radiopacities, referred to as snow-flake calcifications [7,36,37]. These radiopacities represent dystrophic calcifications within the tumour and have been reported in 77% of AOTs [9]. Rare, multilocular variants with ill-defined borders have been reported in the literature [7,9,36]. The radiographic appearance of the current case supports multiple AOTs, rather than the rare multilocular variant, as a unilocular radiolucent lesion surrounds each impacted tooth. Similar findings have been reported in patients presenting

with multiple synchronous AOTs with numerous impacted successor teeth [20,24]. In the current case the position of the primary teeth is most likely due to displacement caused by the individual expansive lesions. It can be speculated that these lesions first occurred in the primary teeth, causing displacement, and only subsequently affected the successor teeth. In most instances AOTs represent small lesions, however, large lesions measuring up to 12cm have been previously reported [9,14,20,38]. The individual lesions in the current case measured up to 2cm in greatest diameter. Expansion is a common feature associated with AOT [7,9,20]. Other uncommon aggressive features, such as root resorption, cortical destruction and soft tissue infiltration have also been reported [7,9,15,16,20]. The current case presented with left maxillary sinus displacement accompanied by mucosal thickening. Whether this represented a nasal blockage or a soft tissue reaction can merely be speculated. Similar findings of nasal blockage with exudate formation have been previously reported in association with AOTs [6].

Larsson *et al.* presented an unusual case of a patient presenting with a 5-year history of 12 AOT-like lesions involving multiple quadrants. In addition, the patient presented with malformed teeth, illustrating a disturbance in enamel formation, as well as taurodontism-like features. Although these lesions showed AOT-like features on histological examination, a final consensus was not reached and the possibility of a new odontogenic entity was speculated [39]. The multiple lesions in the current case were different to the case reported by Larsson *et al.*, as the lesions were limited to one quadrant of the maxilla. A report by Tsaknis *et al* revealed multiple AOTs associated with seven impacted teeth [38].

The typical treatment of AOTs involves surgical enucleation with removal of the associated tooth/teeth [7]. Marsupialisation with successful long-term eruption of the associated tooth/teeth have been reported in the literature [40,41]. Long-term follow-up studies with additional research are needed to evaluate the effectiveness of the different proposed treatment modalities.

Conclusion

The current case has many overlapping features with the case published by Chaves et al [29], and clinical features highly suggestive of Schimmelpenning syndrome. This represents a unique case of multiple AOTs associated with eight impacted and displaced deciduous and permanent teeth. To the authors' knowledge multiple AOTs associated with eight impacted teeth is the highest number reported in English literature. The

multiple lesions were only found in one quadrant of the jaw, with the associated teeth showing enamel hypoplasia. In addition, there was radiographic evidence of the gubernaculum dentis associated with AOTs. These unique features were clearly visible on CBCT imaging and may be used as a reference to strengthen the different theories regarding the development and behaviour of AOTs.

References

1. Ide F, Mishima K, Kikuchi K, Horie N, Yamachika S, Satomura K, et al. Development and Growth of Adenomatoid Odontogenic Tumor Related to Formation and Eruption of Teeth. Head Neck Pathol. 2011;5:123–32.

2. El-Naggar A, Chan J, Rubin Grandis J, Takata T, Slootweg P. WHO classification of head and neck tumours. 4th ed. Cancer IA for R on, editor. Lyon; 2017.

3. Sudhakara M, Vanaki S, Hosur M, Bhullar R. Expression of CK14 and vimentin in adenomatoid odontogenic tumor and dentigerous cyst. J Oral Maxillofac Pathol. 2016;20:369–76.

4. Shruthi D, Shivakumar M, Tegginamani A, Karthik B, Chetan B. Cytokeratin 14 and cytokeratin 18 expressions in reduced enamel epithelium and dentigerous cyst: Possible role in oncofetal transformation and histogenesis- of follicular type of adenomatoid odontogenic tumor. J Oral Maxillofac Pathol. 2015;18:365.

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;36:383–93.

6. Swasdison S, Dhanuthai K, Jainkittivong A, Philipsen HP. Adenomatoid odontogenic tumors: an analysis of 67 cases in a Thai population. Oral Surgery, Oral Med Oral Pathol Oral Radiol Endodontology. 2008;105:210–5.

7. Chrcanovic BR, Gomez RS. Adenomatoid odontogenic tumor: An updated analysis of the cases reported in the literature. J Oral Pathol Med. 2019;48:10–6.

8. Philipsen HP, Khongkhunthiang P, Reichart PA. The adenomatoid odontogenic tumour: an update of selected issues. J Oral Pathol Med. 2016;45:394–8.

9. Becker T, Buchner A, Kaffe I. Critical evaluation of the radiological and clinical features of adenomatoid odontogenic tumour. Dentomaxillofacial Radiol. 2012;41:533–40.

10. Mohamed A, Singh AS, Raubenheimer EJ, Bouckaert MMR. Adenomatoid odontogenic tumour: Review of the literature and an analysis of 33 cases from South Africa. Int J Oral Maxillofac Surg. International

Association of Oral and Maxillofacial Surgery; 2010;39:843–6.

11. Thakur A, Tupkari JV, Joy T, Hanchate A V. Adenomatoid odontogenic tumor: What is the true nature? Med Hypotheses. Elsevier Ltd; 2016;97:90–3.

12. Reichart PA, Philipsen HP, Khongkhunthian P, Sciubba JJ. Immunoprofile of the adenomatoid odontogenic tumor. Oral Dis. 2017;23:731–6.

13. Ide F, Kikuchi K, Kusama K. Adenomatoid odontogenic tumor revisited. Med Hypotheses. Elsevier Ltd; 2017;104:35.

14. 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;10:277–80.

15. Shaikh S, Bansal S, Desai RS, Ahmad I. Aggressive adenomatoid odontogenic tumor of the mandible: A rare case report and review of the literature. J oral Maxillofac Pathol. 2018;2:S11–5.

16. Chuan-Xiang Z, Yan G. Adenomatoid odontogenic tumor: A report of a rare case with recurrence. J Oral Pathol Med. 2007;36:440–3.

17. Shukla P, Prabhu S, Jose M, Sripathi Rao BH. Comparative immunohistochemical study of Bcl-X in ameloblastoma, keratocystic odontogenic tumor and adenomatoid odontogenic tumor. J Oral Maxilofac Pathol. 2017;21:51–7.

18. Manjunatha BS, Mahajan A, Mody BM, Shah V. Adenomatoid Odontogenic Tumor (AOT) Arising from a Dentigerous Cyst: Literature Review and Report of a Case. J Maxillofac Oral Surg. Springer-Verlag; 2015;14:393–7.

19. Gadewar DR, Srikant N. Adenomatoid odontogenic tumour: Tumour or a cyst, a histopathological support for the controversy. Int J Pediatr Otorhinolaryngol. Elsevier Ireland Ltd; 2010;74:333–7.

20. Mehkri S, Rajkumar GC, Nagesh KS, Manjunath GS. Bilateral adenomatoid odontogenic tumour of the maxilla in a 2-year-old female - The report of a rare case and review of the literature. Dentomaxillofacial Radiol. 2012;41:342–8.

21. Yamazaki M, Maruyama S, Abé T, Babkair H, Fujita H, Takagi R, et al. Hybrid ameloblastoma and adenomatoid odontogenic tumor: Report of a case and review of hybrid variations in the literature. Oral Surg Oral Med Oral Pathol Oral Radiol. Elsevier Inc.; 2014;118:e12–8.

22. Bonardi JP, da Costa FH, Matheus RA, Ito FA, Pereira-Stabile CL. Rare presentation of adenomatoid odontogenic tumor in a pediatric patient: a case report. Oral Maxillofac Surg. Oral and Maxillofacial Surgery; 2016;20:215–7.

23. Mohanty R, Singh V, Dey AK, Behera S. A rare nonsyndromic case of adenomatoid odontogenic tumor associated with multiple impacted supernumerary teeth. Natl J Maxillofac Surg. 2019;10:114–7.
24. Bartake AR, Punnya VA, Sudeendra P, Rekha K. Two adenomatoid odontogenic tumours of the maxilla: A case report. Br J Oral Maxillofac Surg. 2009;47:638–40.
25. Rezvani G, Donoghue M, Reichart PA, Pazuhi N. Case Presentation of Concomitant and Contiguous Adenomatoid Odontogenic Tumor and Focal Cemento-Ossifying Dysplasia. Open Dent J. 2015;9:340–5.
26. Li B, Xie X, Jia S. Adenomatoid Odontogenic Tumor With Fibro-osseous Reaction in the Surrounding Tissue. J Craniofac Surg. 2013;24:100–1.
27. Naidu A, Slater LJ, Hamao-Sakamoto A, Waters P, Kessler HP, Wright JM. Adenomatoid odontogenic tumor with peripheral cemento-osseous reactive proliferation: report of 2 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol. Elsevier Inc.; 2016;122:e86–92.
28. Shepard M, Coleman H. Simultaneous adenomatoid odontogenic and keratocystic odontogenic tumours in a patient with Gorlin-Goltz syndrome. Aust Dent J. 2014;59:121–4.
29. Chaves RRM, Júnior AACP, Gomes CC, de Castro WH, Gomez RS. Multiple adenomatoid odontogenic tumors in a patient with Schimmelpenning syndrome. Oral Surg Oral Med Oral Pathol Oral Radiol. 2020;129:e12–7.
30. Gorlin RJ, Cohen MM, Hennekam RCM. Syndromes of the Head and Neck. 4th Editio. Oxford: Oxford University Press; 2001.
31. Warnke PH, Schimmelpenning GW, Happle R, Springer ING, Hauschild A, Wiltfang J, et al. Intraoral lesions associated with sebaceous nevus syndrome. J Cutan Pathol. 2006;33:175–80.
32. Ernst LM, Quinn PD, Alawi F. Clinical Report: Novel Oral Findings in Schimmelpenning Syndrome. Am J Med Genet. 2007;143:881–3.
33. Davies D, Rogers M. Review of neurological manifestations in 196 patients with sebaceous naevi. Australas J Dermatol. 2002;43:20–3.
34. 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.
35. Friedrich RE, Zustin J, Scheuer HA. Adenomatoid odontogenic tumour of the mandible. Anticancer Res. 2010;30:1787–92.
36. Adisa AO, Lawal AO, Effiom OA, Soyele OO, Omitola OG, Olawuyi A, et al. A retrospective review of 61 cases of adenomatoid odontogenic tumour seen in five tertiary health facilities in Nigeria. Pan Afr Med J.

2016;24:2–7.

37. Al-Shimari F, Chandra S, Oda D. Adenomatoid odontogenic tumor: Case series of 14 with wide range of clinical presentation. J Clin Exp Dent. 2017;9:e1315–21.

38. Tsaknis PJ, Carpenter WM, Shade NL. Odontogenic adenomatoid tumour: report of case and review of the literature. J Oral Surg (Chic). 1977;35:146.

39. Larsson Å, Swartz K, Heikinheimo K. A case of multiple AOT-like jawbone lesions in a young patient - A new odontogenic entity? J Oral Pathol Med. 2003;32:55–62.

40. Erdur EA, Ileri Z, Ugurluoglu C, Cakir M, Dolanmaz D. Eruption of an impacted canine in an adenomatid odontogenic tumor treated with combined orthodontic and surgical therapy. Am J Orthod Dentofac Orthop. American Association of Orthodontists; 2016;149:923–7.

41. Motamedi MH, Shafeie HA, Azizi T. Salvage of an impacted canine associated with an adenomatoid odontogenic tumour: a case report. Br Dent J. 2005;199:89–90.