in vivo *24*: 895-898 (2010)

Small Peripheral Developing Odontoma of the Maxilla in a 3-Year-Old Patient Depicted on Cone-Beam Tomograms

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Abstract. *A 3-year-old male patient was referred to the Maxillofacial Surgery Clinic due to a painless swelling of the right palatal region. Conventional radiographs revealed no alteration of the dentition and did not delineate a lesion in the region of interest. Cone-beam tomography depicted small radiopaque, extraosseous deposits inside the palatal space. Histological examination revealed a minute mixed epithelial-mesenchymal lesion of odontogenic origin. We made the diagnosis of a peripheral developing odontoma, taking into consideration the components and arrangements of structures of the lesion. Early intervention is advisable to prevent these odontogenic lesions from eventually deforming the jaw and displacing adjacent teeth. Cone-beam tomography was a valuable pre-operative diagnostic tool to assess the lesion as being composed in part of hard tissue.*

Odontogenic tumours account for fewer than 10% of oral pathologies in children (1). Out of a retrospectively designed single center study covering a period of 30 years, more than 80% of odontogenic tumours were subtypes of odontoma (1). Odontomas are considered as hamartomas causing pathologies due to their localization and extension (2-6). Odontomas usually develop inside the jaws and might interfere with the odontogenesis, *e.g.* impairing the eruption of teeth or replacing the tooth germ at the site of development (2-6). Extraosseous odontomas are extremely rare and might cause severe morbidity (7). The current World Health Organization classification on odontogenic tumours distinguishes complex and compound odontomas (3). Differential diagnosis of the entities comprising the group of mixed odontogenic tumours are predominantly the odontoameloblastoma and the ameloblastic fibro-odontoma or fibro-dentinoma. From a clinical point of view, an odontoma might not always show the complete differentiation pattern attributable to the diagnosis of an odontoma, but appears to be more differentiated than ameloblastic fibro-odontoma. These odontomas are referred to as developing odontomas. This subgroup of odontomas is small in number and usually found in children (1). Differential diagnosis of developing odontoma and ameloblastic fibro-odontoma might be difficult to establish (8). In rare cases a developing odontoma can be found outside the bone. These lesions are called peripheral developing odontoma (9). Reports about this variant are distinctly rare (9). These lesions are exceptionally found in adults (10). This report about an odontogenic lesion intends to add some diagnostic information about this entity.

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Key Words: Peripheral odontogenic tumour, cone-beam tomography, odontoma.

Case Report

A 3-year-old male patient was referred to the Maxillofacial Surgery Clinic due to asymptomatic swelling of the right palatal mucosa. Oral examination revealed the deciduous teeth were fully erupted and symmetrically arranged in both jaws (Figure 1). A small and firm protrusion of the right side of the palate was palpable. This region showed no signs of local infection and was painless on palpation. All teeth proved to react sensitively to adequate stimuli.

Radiology. On the cone-beam tomogram (Galileos™ , Sirona, Bensheim, Germany), the dentition proved to be symmetrically developed as shown in the reconstructed pantomographic view (Figure 1). In the region of interest, the deciduous teeth (no. 54/55, nomenclature according to Federation Dentaire International) had completely developed, with mature roots showing a physiological shape. The successors of the deciduous teeth were located inside the maxilla. However, the tomograms revealed a spot-like radiodensity in two layers perpendicular to each other in close proximity to the crown of tooth 55 at the side of the palpable tumour. These minute radiopacities showed a similar density compared to the adjacent teeth and were localized outside the bone (Figure 2).

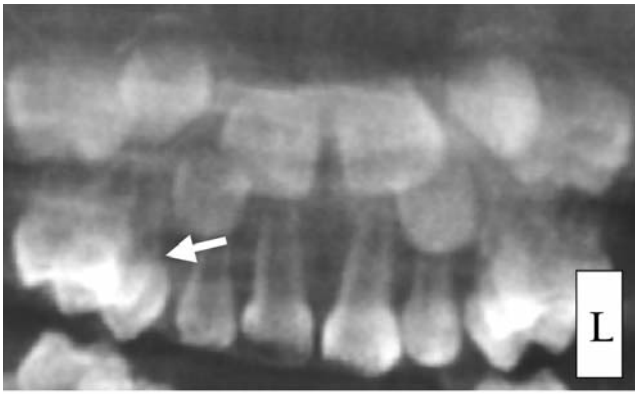

Figure 1. *Detail of the deciduous dentition of the patient as depicted on a pantomogram. The radiopaque parts of the lesion are superimposed by the teeth (arrow) and cannot be discriminated.*

Surgery. Surgery was performed under general anaesthesia. The palatal tumour was excised with covering mucosa (Figure 3 A). A small, tough, space-occupying lesion was identified, attached to the periosteum, and completely excised. The bone was not affected by the lesion. After careful cauterization of the vessels, the healing of the small defect was left to granulation. Healing was uneventful. The follow-up check-ups over 3 years revealed no local recurrence.

Histology. The surgical specimen was fixed in 4% formalin immediately after excision and sent to the laboratory. Tumour tissue was cut into two halves with a knife and completely embedded in paraffin. The resection margins were dye-marked in blue with a tissue marker. Four μm-thick sections were cut and stained with haematoxylin-eosin, periodic acid Schiff, giemsa, elastica van Gieson and Congo red dyes. Microscopical examination showed a circumscribed submucosal ecto-mesenchymal lesion (Figure 3 A) without contact with neighbouring teeth or alveolar bone. The tumour consisted of focal cuboid epithelial portions with discrete areas showing epithelial eosinophilic ghost cells lacking nuclei with focal calcification (Figure 3 B). No Congo red-positive amyloid material was present. More peripherally located, irregular mesenchymal lobules showed spherical, stellate and spindle-shaped cells on a background of bluish extracellular matrix resembling the dental papilla. These were lined with small dark epitheloid cells showing focal pseudoglandular lumina (Figure 3 C) and transition into palisaded cylindrical cells. Within the latter areas, an eosinophilic mineralized dentinoid material displaying parallel tubular structures was found between the epithelial lining and mesenchymal lobules (Figure 3 D). Based on these morphological characteristics, the histopathological diagnosis was peripheral developing odontoma.

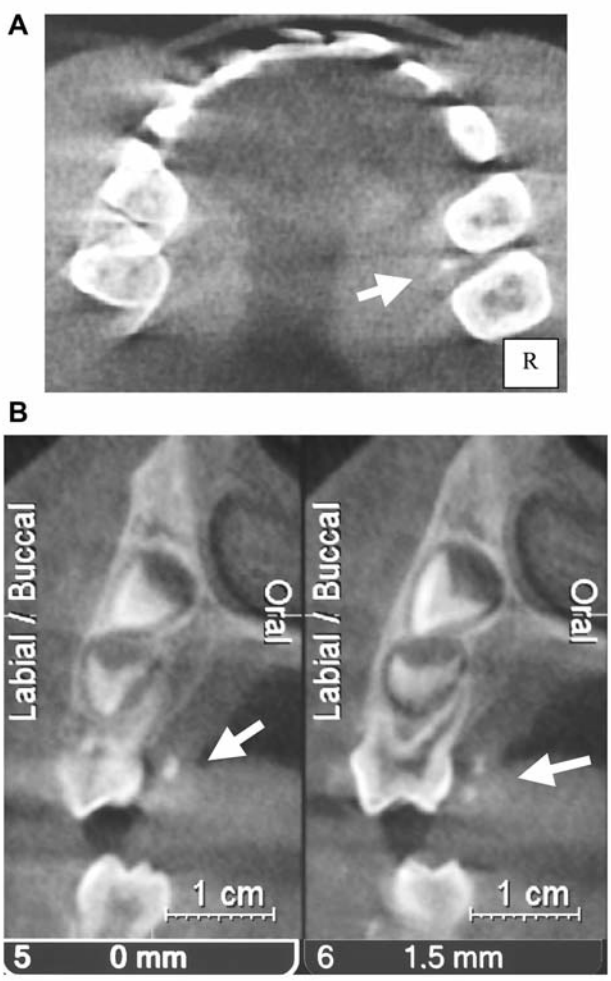

Figure 2. *A: The axial tomogram depicts a radiopaque lesion (arrow) between the approximal surfaces of both right upper molars on the palatal side. B: On the photographs of sagittal sections, these radiopacities (arrows) have distinct borders, are located at the level of the molar crown and outside the bone. The distance between slice 5 and 6 is 1.5 mm.*

Discussion

Odontoma is a rare benign lesion, preferentially found in children and young adults (1, 5). Diagnosis of odontomas in patients older than 20 years is extremely rare (4, 10). Some authors strictly recognize odontoma not as a neoplasm but a hamartoma (5). This judgement is accordance with the clinical experience that these lesions usually do not recur after local excision. Exceptions are extremely rare but point to the neoplastic nature of at least some of these lesions (11).

The diagnosing of this case was challenging and comprised both radiological and histological techniques. Interestingly, although the radiopaque part of the lesions was

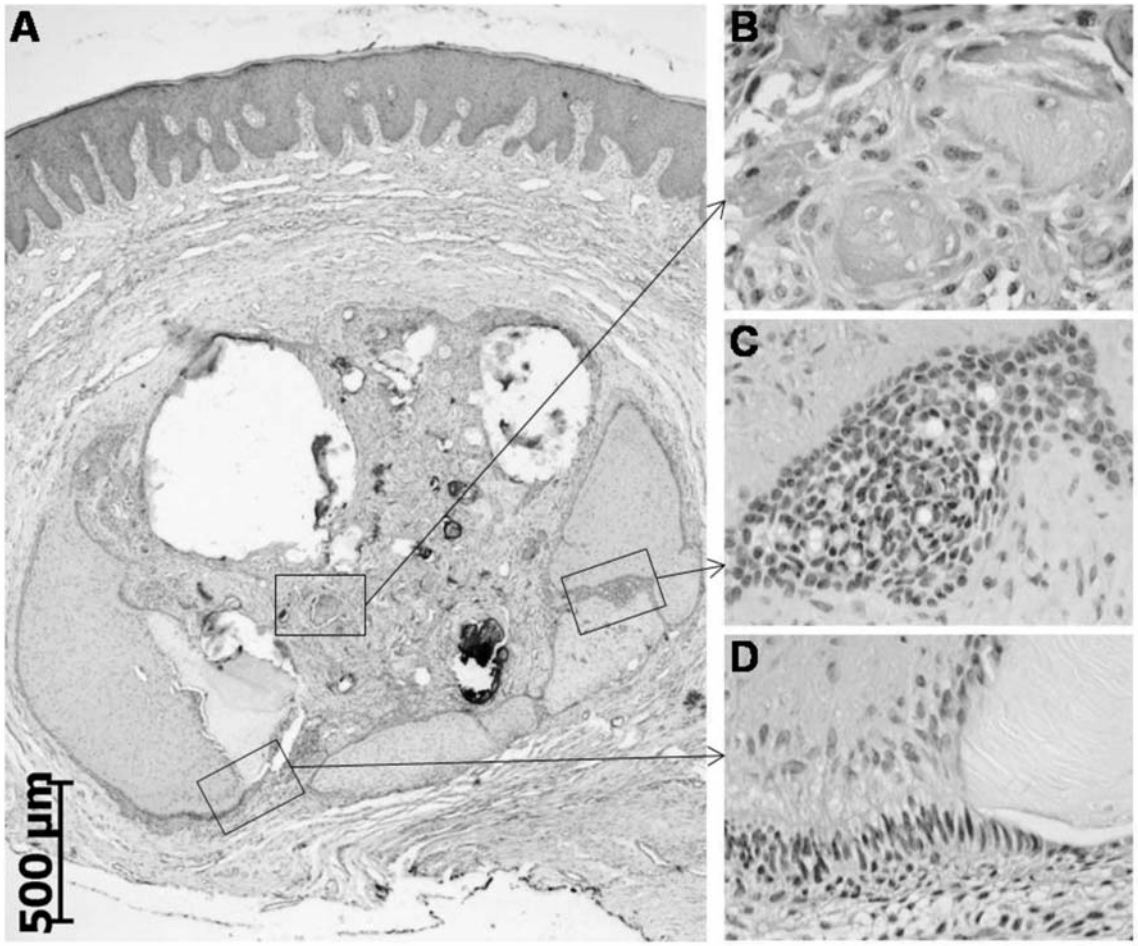

Figure 3. *Histopathological findings. A: At low magnification, a localized admixture of mesenchymal, epithelial and calcified structures was found in the gingival fibrous tissue. The lesion was excised in toto with intact blue dye-marked resection margins. Neither contact with the superficial epithelium nor bone and dental structure were apparent (stain: haematoxylin-eosin, original magnification: ×25). B: Intralesional epithelioid structures lacking stainable nuclei (ghost cells) were found in the centre of the tumour. C: More peripherally, irregular nodular mesenchymal structures were covered with smaller epithelial cells which were focally arranged in small solid to cribriform or pseudoglandular nests. D: At the periphery of the tumour, the formation of dentinoid material with tubular structure was focally associated with proliferation of tall epithelial cells with palisading nuclei and odontogenic mesenchyme, consistent with developing odontoma. Cystic changes or ameloblastoma-like differentiation were not observed (B-D: stain: haematoxylin-eosin, original magnification: ×200).*

not detectable on the conventional panoramic view, the assumption of a lesion forming hard tissue outside the bone could be drawn from adequate tomograms. This report illustrates the value of cone-beam tomography of the jaws in the differential diagnosis of tumours and tumour-like lesions of odontogenic origin in small children. The finding of a radiopaque lesion, closely related to teeth and with an adsorption quality of X-rays similar to odontogenic hard tissue, points to a benign lesion in the vast majority of cases. However, malignant tumours, *e.g.* the very rarely diagnosed ameloblastic sarcoma, might occasionally produce hard tissue of odontogenic origin (4). Furthermore, several other benign odontogenic tumours might also show radiopaque areas (3). Peripheral developing odontoma is characterized by a mixed radiopaque-radiolucent appearance, with irregular calcifications. These calcifications are more dense than the radiographic appearance of adjacent bone and resemble the density of teeth (5, 11). Usually a distinct delineation of the lesion to the bone and adjacent teeth is seen on radiographs. This feature was not visible on the radiographs of this case, due to the extraosseous localization of the lesion.

Histologically, the incomplete formation of a dental papilla and dental hard structures supported the differential diagnosis of an ameloblastic fibro-odontoma (11). However, the ameloblastic fibro-odontoma develops preferentially in the region of permanent teeth and is located inside the bone.

Indeed, it was suggested that ameloblastic fibro-odontoma be considered as an exclusively central (intraosseous) lesion (4). In line with this estimation is the frequent incidental finding of ameloblastic fibro-odontoma in the differential diagnosis of non-emerging (permanent) teeth (3, 11). Our patient presented with a lesion located superior to the direction of the emerging anlage of the successors. This topography is often found in ameloblastic fibro-odontoma (11). However, this lesion had developed completely extra-osseously and was not likely to interfere with the change of the dentition. Recent reports on the rare findings of peripheral odontogenic tumours did not include ameloblastic fibro-odontoma (10, 12, 13). However, some reports on developing odontoma confirm that this entity can be located strictly outside the bone (9, 14-17). Interestingly, the palatal mucosa is noted as the region where the lesion emerged in all these reports on peripheral developing odontoma (9, 14-17), excepting the report of Ide *et al*. (12). Complete local excision of the lesion is the therapy of choice. Recurrence of peripheral developing odontoma has not yet been reported.

Conclusion

The peripheral developing odontoma is an exceedingly rare benign odontogenic lesion that can be treated by local excision. Thorough radiographic and morphological investigation is mandatory to establish the differential diagnosis, in particular to other mixed odontogenic lesions (tumours or hamartomas), in order to prevent extensive surgery.

References

1 Jones AV and Franklin CD: An analysis of oral and maxillofacial pathology found in children over a 30-year period. Int J Paediatr Dent *16*: 19-30, 2006.

2 Kramer IRH and Pindborg JJ: Histological Typing of Odontogenic Tumours, 2nd edition. Springer, Berlin, pp. 16-21, 62-70, and 82, 1992.

3 Barnes L, Eveson JW, Reichart P and Sidransky D: Pathology and Genetics of Head and Neck Tumours. Chapter 6: Odontogenic Tumours. World Health Organization. IARC Press, Lyon, 2005.

4 Reichart PA and Philipsen HP: Odontogenic Tumors and Allied Lesions. Quintessence Publ., London, pp. 105-115, and 135, 2004.

5 Marx RE and Stern D: Oral and Maxillofacial Pathology. Quintessence Publ., Chicago, pp. 675-683, 2003.

6 Reichart PA and Jundt G: Benigne epitheliale odontogene Tumoren. Der Pathologe *29*: 175-188, 2008.

7 De Faria PR, Cardoso S, Rocha A, Gomes DC, de Castro SC and Loyola AM: Intracranial compound odontome. J Craniomaxillofac Surg *37*: 376-379, 2009.

8 Gardner DG: The mixed odontogenic tumors. Oral Surg Oral Med Oral Pathol *58*: 166-168, 1984.

9 Silva ARS, Carlos-Bregni R, Vargas PA, de Almeida OP and Lopes MA: Peripheral developing odontoma in newborn. Report of two cases and literature review. Med Oral Patol Oral Cir Bucal *14*: e612-e615, 2009.

10 Ide F, Shimoyama T and Horie N: Gingival peripheral odontoma in an adult: case report: J Periodontol *71*: 830-832, 2000.

11 Friedrich RE, Siegert J, Donath K and Jäkel KT: Recurrent ameloblastic fibro-odontoma in a 10-year-old boy. J Oral Maxillofac Surg *59*: 1362-1366, 2001.

12 Ide F, Mishima K and Kusama K: Rare peripheral odontogenic tumors: report of 5 cases and comprehensive review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod *106*: e22-e28, 2008.

13 Abughazaleh K, Andrus KM, Katsnelson A and White DK: Peripheral ameloblastic fibroma of the maxilla: report of a case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod *105*: e46-e48, 2008.

14 Gans BJ: Ectopic tooth: report of a case. J Oral Surg Anesth Hosp Dent Serv *20*: 435-437, 1962.

15 Diekmann SL, Cohen DM and Gutz DP: Ectopic soft-tissue mesiodens. Oral Surg Oral Med Oral Pathol *53*: 391-394, 1982.

16 Lin CC, Chen RJ and Lai S: Ectopic soft-tissue mesiodens. Report of a case. J Oral Med *41*: 124-125, 1986.

17 Kintarak S, Kumplanont P, Kietthubthew S and Chungpanich S: A nodular mass of the anterior palatal gingiva. Oral Surg Oral Med Oral Pathol Oral Radiol Endod *102*: 3-6, 2006.

Received September 1, 2010
Revised October 20, 2010
Accepted October 21, 2010