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Infraorbital infection related to odontodysplasia: case report

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Abstract

A case of a 3-year-old, white female with odontodysplasia is presented. Histological considerations and the rapid development of an infraorbital infection in this case encourage the practitioner to pursue a more aggressive treatment plan than might otherwise be considered.

Introduction

Regional odontodysplasia (also known as odontodysplasia, odontogenic dysplasia, odontogenesis imperfecta, or "ghost teeth") is an unusual localized anomaly of the dentition. Thus far, approximately 70 cases have been reported in the literature. A case is presented to highlight the observation and treatment concerns.

Literature Review

Odontodysplasia is a rare defect of tooth enamel and dentin. It can be discovered upon chief complaint, or as an incidental finding upon presentation for other dental concerns. These teeth usually are hypoplastic, often with areas of enamel completely missing on the crown, and can be hypocalcified, yellow, or yellow-brown colored (Walton et al. 1978; Hovinga and Ingenhoes 1979; Kerebel and Kerebel 1981; Dayal and Mani 1981; Kerebel and Kerebel 1982). It appears that both the primary and the permanent dentition are involved, and though the affected area usually is restricted to one quadrant, there are documented cases of both sides of an arch being involved, and one case of both maxillary and mandibular involvement (Lustmann et al. 1975). The literature suggests a 2:1 preference for the maxillary arch over the mandibular, and a similar preference for anterior teeth over the posterior (Lustmann et al. 1975). Thus far, the majority of cases have been reported in females (Lustmann et al. 1975).

Lustmann et al. (1975) reviewed 51 cases and found a significant number were associated to varying degrees with abscesses or swollen gingival areas. Subsequent reports have verified this finding (Walton et al. 1978; Hovinga and Ingenhoes 1979; Anneroth and Ramström 1980; Dayal and Mani 1981). Acute osteitis and facial swellings are not an uncommon finding (Anneroth and Ramström 1980; Dahllöf et al. 1987).

Radiographically, the enamel and dentin appear to have similar radiodensity, and are difficult to distinguish (Walton et al. 1978; Kerebel and Kerebel 1981). The thinness of the dentin and enamel, along with the significantly enlarged pulp chambers, give rise to the characteristic "ghost teeth" appearance of the affected teeth. Sometimes defective or irregular mineralization of teeth can be seen on radiographs, as can incomplete root formation (Anneroth and Ramström 1980; Schmid-Meier 1982). Dilated follicles around the crown also have been reported (Hovinga and Ingenhoes 1979).

The histological findings indicate significantly abnormal enamel and dentin formation. Within the enamel layer are partially formed and irregularly patterned enamel calcifications, remnants of ameloblasts and significant pits (Hovinga and Ingenhoes 1979; Anneroth and Ramström 1980; Kerebel and Kerebel 1982). The DEJ has a unusual scalloped appearance (Sadeghi and Ashrafi 1981; Kerebel and Kerebel 1981). Within the dentin are amorphous areas of hypermineralization, ground substance (collagen-free dentinal matrix), and additional areas of hypomineralization and interglobular dentin (Gardner and Sapp 1977; Anneroth and Ramström 1980; Dayal and Mani 1981; Kerebel and Kerebel 1982). The dentinal tubules have been described as larger and more irregular, and small canals within the dentin have also been observed (Dayal and Mani 1981; Kerebel and Kerebel 1982; Williams and High 1988). Calcified nodules have been noted within the connective tissue surrounding the tooth crown (Hovinga and Ingenhoes 1979; Anneroth and Ramström 1980). The enamel and dentin defects have not been correlated with chronological insults to the tooth-forming tissues.

The pulp tissue has several abnormal characteristics. Wide pulp canals are a common, but not universal, finding (Hovinga and Ingenhoes 1979). A fibrous pulp

with numerous false and genuine denticles has been reported (Hovinga and Ingenhoes 1979; Anneroth and Ramström 1980; Dayal and Mani 1981). Atrophically degenerated odontoblasts with a mild inflammatory reaction pericoronally were noted by Anneroth and Ramström (1980). They and other investigators also have described necrotic pulp with periapical granulation tissue, and infiltration of the pulp with lymphocytes, plasmocytes, and leukocytes (Hovinga and Ingenhoes 1979; Anneroth and Ramström 1980; Sadeghi and Ashrafi 1981).

The etiology of odontodysplasia is relatively unknown. Possible causes have been described and discussed in the literature, but no consensus has developed concerning the ones that are most probable. The suggested etiologies of odontodysplasia include localized vascular disorders, neural crest defects, toxic maternal drug effects, genetic mutation, trauma, localized infection, inherited genetic defect, somatic mutation, neural damage, localized viral infection that alters amelo- and dentinogenesis, and maternal hypoxia caused by fainting (Walton et al. 1978; Hovinga and Ingenhoes 1979; Anneroth and Ramström 1980; Ferguson and Geary 1980; Dayal and Mani 1981; Mock et al. 1986; Dahllöf et al. 1987; Williams and High 1988). The effect on both primary and permanent dentitions makes the discussion of an isolated event more difficult.

Case Report

A 3-year-old, white female presented to the dental emergency room for facial pain and swelling. The patient's medical history was unremarkable and the physical examination, except for oral findings, was within normal limits. Upon clinical examination, a nonfluctuant swelling that extended from the left infraorbital region to the left lateral border of the nose and upper left lip was noted. The parent said that the swelling had begun 2-3 hr before the child was brought to the clinic. The child appeared tired and restless, and was febrile at 100°F (oral). Intraorally, there was diffuse swelling of the left mucobuccal fold adjacent to severely dysplastic and mobile maxillary left primary central and lateral incisors, and canine. No draining fistulas were noted, nor were there any carious lesions on the affected or adjacent teeth. There was no pharyngeal swelling, nor any palpable nodes or masses. A maxillary occlusal radiograph showed three severely dysplastic teeth with a periapical radiolucency associated with the lateral incisor.

The child's condition worsened while she was in the emergency room area. Within 2 hr, her temperature was 103.6°F, and she had become more distraught. Her facial swelling clearly had enlarged during this 2-hr time span (Fig 1). After consultation with doctors in oral surgery and pediatrics, the patient was admitted and received intravenous antibiotics and hydration. The patient remained febrile into the next morning, at which time the swelling began to localize. Her temperature was steady at 101°F.

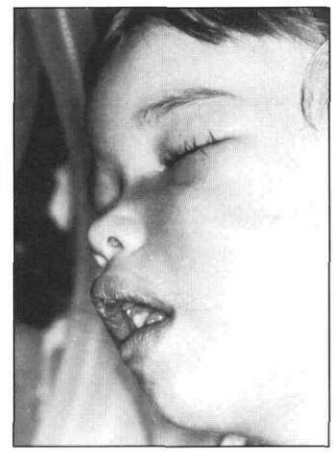

Fig 1. Patient had developed significant swelling of the avleolus, infraorbital and orbital areas while receiving care in the emergency room.

Because of the child's discomfort, lack of cooperation, and the need for adequate visualization and removal of the offending teeth, she was prepared for general anesthesia. Informed consent was received from the parent for the anticipated procedures, and for general anesthesia. Under general anesthesia, adequate maxillary occlusal radiographs of the involved area (Figs 2a and 2b, see next page) were exposed. The maxillary primary left central and lateral incisors and canine were extracted. An incision and drainage of the left maxillary mucobuccal vestibule also was performed. The patient was stable postoperatively, and her temperature continued to decrease. Intravenous hydration and antibiotics were continued for one additional day. The child was discharged one day postoperatively with oral antibiotics. The patient returned to the dental clinic for follow-up evaluation five days postoperatively. No swellings were present, and the extraction sites were healing well. The patient was seen one month postoperatively for evaluation, and then for regular dental recall visits.

Discussion

Although odontodysplasia can present as a rather asymptomatic condition in either the primary and/or permanent dentition, this case history shows why the condition must be monitored carefully and continuously as long as the affected teeth remain in the oral cavity. Some of the frequently observed negative consequences of odontodysplasia suggest the need for more aggressive treatment.

The child presented had been examined when she was 21 months old by a pediatric dentist, and the diagnosis of odontodysplasia was determined at that time by clinical and radiographic examination (Fig 3, see next page). The family was aware of the nature and possible consequences of the problem, including the prognosis of the primary teeth, and the probable involvement of the permanent teeth. The child was taken to the pediatric dentist every four months for follow-up exami-

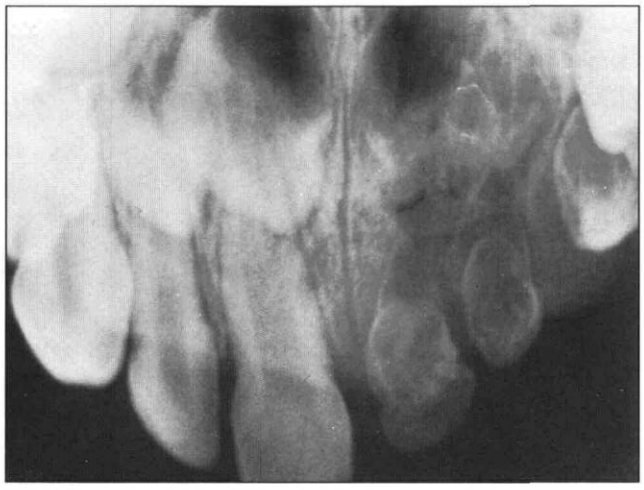

Fig 2a. Occlusal radiograph at the time of general anesthesia (patient age - 3 1/2 years) showing the affected left primary and permanent anterior teeth.

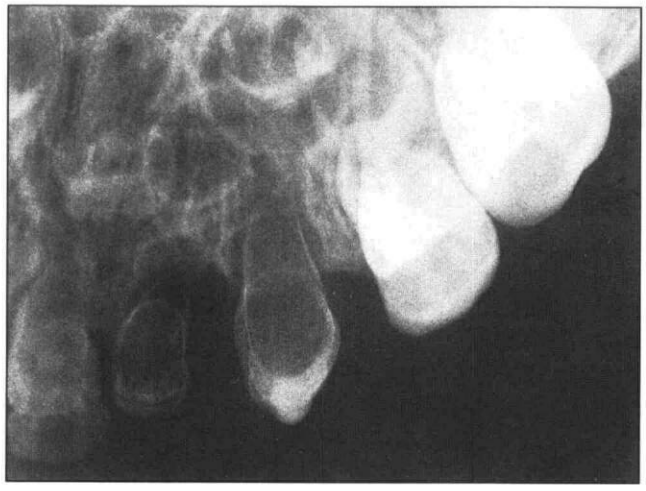

Fig 2b. Occlusal radiograph giving a clear picture of the affected permanent successors. (Primary teeth F through J are shown.)

nations. There had been no problems until the acute episode requiring hospital admission occurred.

In the review of Lustmann et al. (1975), almost half of the cases which presented symptomatically for odontodyplasia did so because of gingival swelling. Though the reason for these frequent infections is not clear, the nearness of the pulp to the oral fluids and the reported large dentinal tubules probably encourage bacterial contamination of the pulpal tissue. Inflammatory cells frequently have been reported in the pulp of odontodysplastic teeth, even those not associated with gingival swelling. Leaving affected teeth untreated in the oral cavity appears to render them significantly susceptible to pulpal infection. Extraction of the affected teeth or immediate restoration with full crowns would seem warranted for preventive reasons. However, since these teeth are often only partially erupted, as in the case presented, restoration may not be a viable choice, and allowing the teeth to remain may make the patient susceptible to infection. The speed with which infection proceeded in the case presented heightens this concern. In addition, pulpal therapy under restorations probably is contraindicated, given the large size and irregular shape of the pulp (and roots). Also, preventing pulp exposure during preparation for full crown coverage restoration may be difficult.

The radiograph (Fig 2b) shows that the permanent successors also are likely to be affected. Given what happened with the primary teeth, aggressive treatment of the permanent teeth when they erupt probably is warranted.

Although the case presented seems to represent an extreme outcome among reports of odontodysplasia, there have been enough other reports of gingival and facial swelling to suggest the need for aggressive preventive treatment of this anomaly (Lustmann et al. 1975; Dahllöf et al. 1987). The histological literature suggests that this developmental defect is predisposed to pulpal infection, and other associated sequelae. Therefore, "preventive" full-crown restoration, or extraction of affected teeth, seems to be the appropriate treatment options for patients with odontodysplasia. Decisions to wait and observe the condition before committing to any treatment (as decided by parents and/or the dentist) require continuous and frequent monitoring. It should be pointed out that frequent observation did not prevent the detrimental outcome in this case.

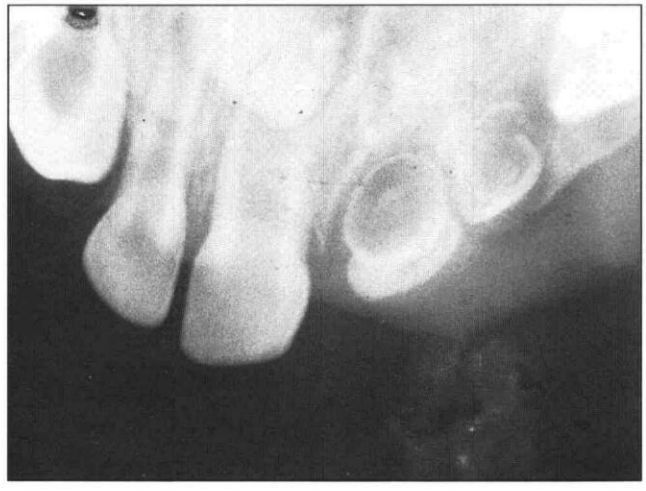

Fig 3. Occlusal radiograph taken at 21 months showed malformations of the partially erupted primary left central and lateral incisors.

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