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Regional Odontodisplasia Presenting as a Soft Tissue Swelling

Received: May 27.1998

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Abstract: Regional odontodisplasia is a developmental anomaly of dental tissues with characteristic clinical, radiographic and histologic appearances. It most commonly affects the maxillary anterior teeth of both the primary and permanent dentition, and occurs in females twice as often as in males. The pathogenesis is unknown. A case of a

twelve- year old female with regional odontodisplasia is presented. Although the affected teeth displayed the radiographical and clinical features of odontodisplasia, the patient appeared with significant soft tissue swelling on the anterior region of the mandible.

Key Words: Regional Odontodisplasia

Regional odontodisplasia (RO) is a rare developmental anomaly of the dental tissues. The term "odontodisplasia" was introduced by Zegarelli et al(1). About 210 cases have been published in the literature under a variety of names including ghost teeth(2), odontogenesis imperfecta(3) and unilateral dental malformation(4).

Clinically, the teeth appear to be discolored, hypocalcified and hypoplastic((4-6) Radiographically, the teeth have a radiolucent image, with little demarcation between the enamel and dentin (Ghost teeth)(2). They have wide pulp chambers with open apices . The purpose of this article is to report a case of regional odontodisplasia with extensive soft tissue swelling.

A 12-year old girl came to our clinic for evaluation an unusual swelling on the left side of her mandible. Her

complaint was that the area had been increasing in size for about 2 years. Eruption of the permanent lower left first incisor, first and second premolar and second molar teeth were delayed. The child's father stated that the soft tissue associated with the lower first incisor and canine had increased in size. This area was not painful and remained asymptomatic. She had had no previous hospitalizations or systemic illness. The child's mother had taken no medications during pregnancy. At birth no defects or birth marks were noted. No sibling or relatives had had any similar dental condition. The teeth associated the swelling were malformed and discolored. Panoramic radiographs revealed a ghostlike appearance of the clinically affected teeth and the unerupted developing lower first incisors (Figure 1). The pulp chambers were markedly enlarged.

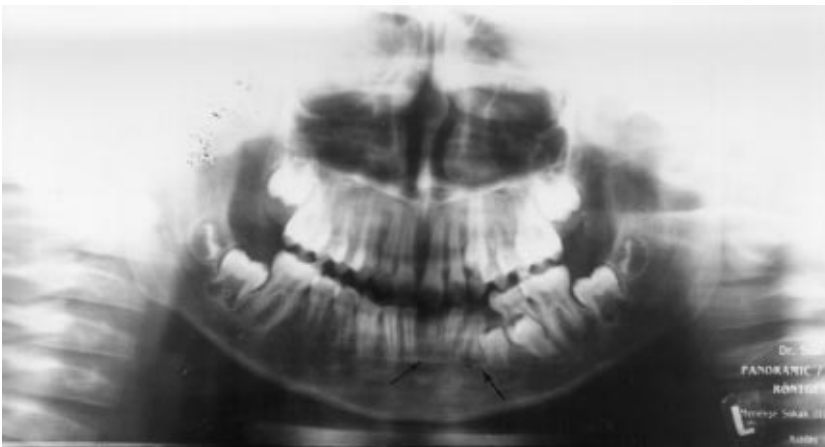


Figure 1. Panoramic Radiograph of Case Showing Several Ghost Teeth in Anterior Region of the Mandible.

In the clinical examination there was a firm nontender swelling in the mandibular anterior region (Figure 2). The lower first and second incisors and premolars were unerupted. We decided to remove the involved teeth and soft tissue. All preoperative laboratory data were within normal limits. Under general anesthesia a full thickness flap from the lower right first incisor to the lower left

first molar was elevated. The lower left first and second incisors and canine were removed. The soft tissue mass was excised but there was no definitive demarcation between pathologic tissue and normal bone. The surgical sites were closed primarily. Healing was uneventful in the postoperative period. On histologic examination, the diagnosis of odontodisplasia was confirmed (Figure 3).

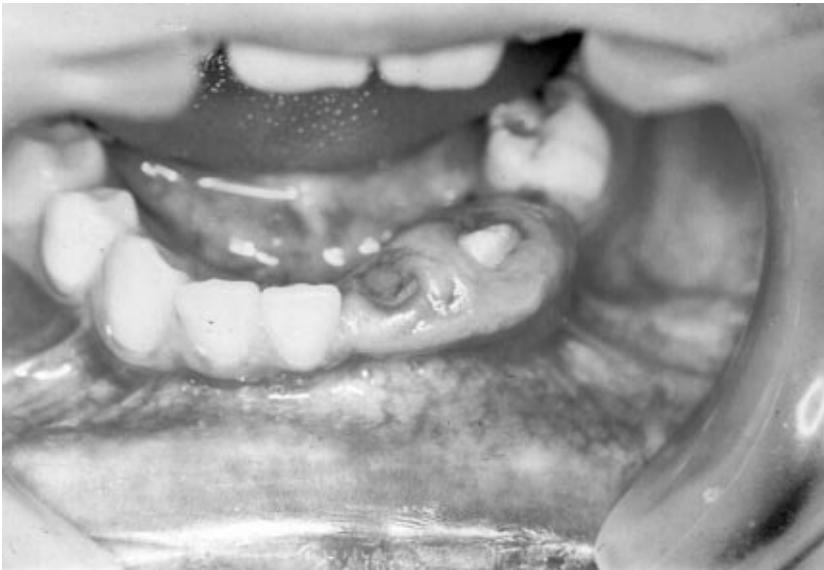


Figure 2. Clinical Presentation of Odontodisplasia as a Soft Tissue Mass.

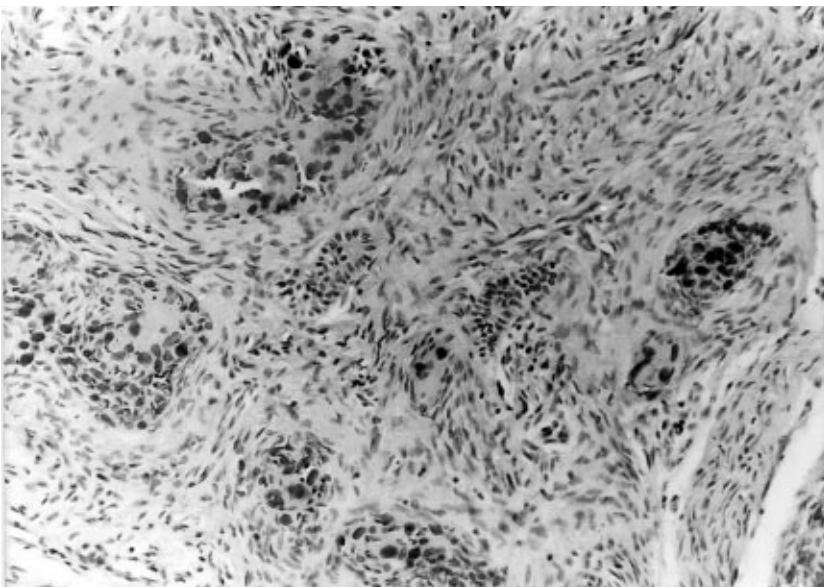


Figure 3. Photomicrograph Displaying Calcifications and Odontogenic Epithelial Rests in Cellular Fibrous Connective Tissue (Haematoxylin and Eosin x 200).

Regional odontodysplasia, a developmental defect in hard tissue morphogenesis, has been reported under various names(3, 6). Although several causative factors such as local trauma(1, 3, 6) infection(3), Rh incompatibility(7), irradiation(1), neural damage(2), local vascular defects(2) have been suggested, the pathogenesis remains uncertain. Odontodysplasia seems to be slightly more prevalent in females, has no racial predilection and most commonly involves the maxillary arch (2:1 over mandible)(3). Although no average age at diagnosis has been stated, the condition typically manifests during the time of primary tooth eruption and mixed dentition. Most often the central and lateral incisors and canine are involved, but any teeth usually consecutive, may be affected(7). Often tooth eruption is delayed or fails(5).

Previous reports of RO have consistently described its unique characteristic radiographic features(4-6). Because of defective thin layers of enamel and dentin, subsequently reduced radiodensity results in a faint, fuzzy outline, creating a ghostlike appearance. The pulp chambers and canals are correspondingly enlarged, whereas the roots appear case most of the clinical and radiographic features were evident. Clinical and radiographical evidence of our patient were consistent with previous reports. However, reports on RO have consistently described its unique characteristic radiographic features and there are no findings about differential diagnosis of regional odontodysplasia in the literature.

Treatment of RO has been somewhat controversial. Most clinicians elect to extract the involved teeth as soon as possible and insert a prosthetic replacement(8-9).

Other clinicians have emphasized restorative procedures, if possible, to protect the erupted affected teeth(1). Timing is the critical factor for treatment planning. In young children unrestorable abscessed teeth should be extracted, but ideally as many affected teeth as possible should be preserved to retain normal jaw development and to lessen the psychological effects. In older children abscessed permanent teeth should be extracted, with others retained until a permanent prosthesis can be constructed(8).

Because of the young age of our patient, the short-term treatment philosophy was to maintain the present dentition so that normal jaw development could proceed for as long as possible. First, we extracted the affected lower left and second incisors and deciduous first molar. Then, deciduous prosthetic rehabilitation of the region was planned.

The present case demonstrates the normal proliferative capabilities of the soft tissues affected by RO. The importance of recognizing and documenting these changes may be interpreted as neoplastic, a possibility mentioned by previous authors(7).

We anticipate long-term treatment to include extracting the deciduous molars and fabricating an acrylic partial denture. If the lower first and second premolars fail to erupt, further prosthetic construction will be needed to avoid drifting and supraeruption of the opposing dentition. Thus the treatment goals include aiding mastication and eruption of uncovered teeth, increasing function, improving esthetics, decreasing psychological effects, and protecting erupted affected teeth.

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