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Regional Odontodysplasia: A Literature Review

A Project Submitted to
The College of Dentistry, University of Baghdad, Department of oral diagnosis in Partial Fulfillment for the Bachelor of Dental Surgery

By

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Certification Of Supervisor

I certify that this project entitled ***Regional odontodysplasia*** was prepared by the fifth-year student ***Tabarak Saad Hamad*** under my supervision at the College of Dentistry/University of Baghdad in partial fulfilment of the graduation requirements for the Bachelor Degree in Dentistry.

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Introduction

Regional odontodysplasia (RO) is an uncommon, nonhereditary developmental anomaly involving components of both the dental mesoderm and ectoderm. The condition can be differentiated from other odontogenic disturbances since all the histological elements of the dental organ are abnormal in the affected teeth, while other teeth in the same individual are normal. Regional odontodysplasia has been reported under many other names, such as 'ghost teeth', 'odontogenesis imperfecta', 'odontogenic dysplasia', 'nonhereditary segmental amelogenesis imperfecta' and 'unilateral dental malformation'. RO affects a particular segment in either or both dentitions in the maxilla or mandible or both jaws. Radiographic features have consistently demonstrated thin and defective layers of enamel and dentine, resulting in a faint, fuzzy outline, creating a ghost-like appearance.

The RO etiology is uncertain; numerous factors have been suggested and considered as local trauma, irradiation, hypophosphatasia, hypocalcemia, hyperpyrexia.

The altered teeth are commonly yellow or brown and show abnormal morphology. Most patients suffer from recurrent purulent inflammations or tooth mobility. Wide pulp chambers, open apices and blurred demarcation in the dentino-enamel junction are the main radiological features distinctive of RO. The disorder involves both primary and permanent dentitions in the majority of the cases. Histologically, both enamel and dentin appear hypomineralised with poorly organised dentinal tubules and enamel prisms. The pulp chambers very often contain calcifications in the coronary area.

CHAPTER ONE

REVIEW OF LITERATURE

1.1 Etiology and pathogenesis

In 1963, Zegarelli et al. were the first to suggest the term ‘odontodysplasia’. The term ‘regional’ was added because the condition affects a group of several adjacent teeth in a particular segment of the jaw. The age of the patient at presentation is variable, although the condition typically manifests during the mixed dentition **(Zegarelli et al., 1963)(Kahn et al., 1991).**

Regional odontodysplasia is slightly more common in females and there is no tendency for its occurrence in any specific ethnic group. Generally, the disturbance is localized to one arch and the maxilla is involved twice as often as the mandible. The left side of the maxilla is the most frequently affected site followed, in order of decreasing frequency, by the maxillary right, mandibular right and mandibular left regions **(Crawford et al., 1989).**

The number of affected teeth is variable and the affected teeth are usually in a continuous series. In the maxilla or mandible, the central and lateral incisors, and canines are the teeth most commonly affected. In rare cases of RO, a single tooth has been reported to be affected, and the condition has sometimes been seen to ‘skip’ a tooth or a group of teeth **(Koskinen et al., 2019) .**

Not all cases of RO are ‘regional’: some authors have reported cases affecting both the maxilla and the mandible on the same side, and others have reported cases in which both sides of the same jaw were affected. In extremely rare reports, RO affected all four quadrants (**Alotaibi et al., 2019**)**(Upadhyaya et al., 2021).**

If the primary teeth are affected, the permanent successors are usually affected as well. However, affected permanent teeth may succeed normal primary teeth, and very rarely, normal permanent teeth follow affected primary ones **(Crawford et al., 1989) (Ponranjini et al., 2012)**.

1.2 Clinical features

The affected teeth are atypically shaped with surface pits and grooves, hypoplastic, hypocalcified, and show yellowish or brownish discoloration. Some of the affected teeth are whitish in colour at eruption, and later become yellowish or brownish **(Alotaibi et al., 2019)(Silva Cunha et al., 2020)**.

Because their structure is defective, they are usually small in size and more susceptible to dental caries. The alveolar crest in the region of the affected teeth is usually enlarged and covered by fibrous tissue. The eruption of the affected teeth is behind schedule or does not occur at all. When curetted, the unerupted teeth are extremely friable, and the dentine is very soft and could be mistaken for advanced caries **(Das et al., n.d.)(Soxman et al., 2019)(IBSEN et al., 2021)**.

1.3 Radiographical features

The affected teeth show abnormal morphology and hypoplastic crowns. The enamel and dentine are less radio-opaque than unaffected counterparts, and there is little demarcation between enamel and dentine. This faint outline of the affected teeth was the reason for the term ‘ghost teeth’. The pulp chambers and root canals are wide, and the roots are short with wide and open apices. Calcification is occasionally seen within the pulp chambers or root canals. In some cases, the unerupted teeth are surrounded by a pericoronal radiolucency representing an enlarged dental follicle **(Abdel-Kader et al., 2019) (Silva Cunha et al., 2020)**.

1.4 Histological features

All structures of the dental germ are affected. In ground section, the enamel is of variable thickness, producing an irregular surface. The enamel prisms are irregular and the enamel may occasionally lack a prismatic structure, and it is hypoplastic and contains degenerated

globular calcifications **(Ide et al., 2022).** There is also hypocalcification of the enamel because residual enamel matrix is frequently seen in demineralized sections. The dentine is thin, and the tubules are reduced in number and tortuous in shape **(Camacho et al., 2021).** Interglobular dentine and globular masses interrupting the dentinal tubules are frequently seen. Clefts within dentine, some of which establish communication between the pulp and the oral cavity, are common findings. Cellular dentine and amorphous areas within the coronal dentine are usually evident. Closer to the dentino-enamel junction, the dentine is more evenly calcified **(Camacho et al., 2021)(Ide et al., 2022)**.

Although the predentine is of variable thickness, it is usually wider than that seen in normal teeth. Prominent interglobular dentine is also seen in the radicular dentine; however, the radicular dentine and cementum are generally less abnormal compared with the coronal dentine. The pulp chamber is large with occasional long pulp horns, and often contains large irregular, calcified globules or stones. Pulp necrosis is often noted as a result of the communication with the oral cavity through the dentinal clefts and pulp horns **(Silva Cunha et al., 2020)(Ji et al., 2022)**.

Odontogenic epithelial rests, whorled fibrous tissue and foci of calcifications, occasionally coalescing into larger globose areas with an accentuated affinity to haematoxylin, are frequently seen within the dental follicle **(Singh et al., 2021).**

The enlarged gingiva which accompanies the affected teeth in some cases usually shows a parakeratinized surface epithelium with acanthosis and very hyperplastic rete ridges. The lamina propria is composed of fibrous connective tissue and contains chronic inflammatory cell infiltrate. Calcified globules and odontogenic epithelial rosettes similar to those found in the dental follicle are also sometimes observed inside this tissue **(Brown et al., 2022)(Ullah et al., 2022)**.

The calcifications are thought to be the result of: degenerative change of the reduced enamel organ; degenerative change of the connective tissue cells; earlier inflammation; or formation by metaplastic epithelial cells **(Reddy et al., 2022)(Akshatha et al., 2023).**

In spite of its specific clinical, radiographic and histological features, RO is a condition of unknown aetiology. The condition is not hereditary and it appears to be the result of local factors affecting the tooth-forming tissues during development **(IBSEN et al., 2021)(Zhang et al., 2023)** .

1.5 Factors have been suggested as causes

- the activation of a latent viral infection of the tooth germ during development;
- local trauma or ischaemia;
- irradiation;
- metabolic and nutritional disturbances;
- rhesus incompatibility;
- local somatic mutation;
- genetic transmission;
- medications taken during pregnancy;
- failure of migration of the neural crest cells;
- and local vascular defects
- and haemangiomas **(Kahn et al., 1991) (Rosa et al., 2006)(Murthy et al., 2013)**

However, no one factor has been positively identified as the single cause of the condition. In addition, it remains hard to explain why, in the vast majority of cases, particular contiguous teeth are affected with no involvement of others. There are occasional reports of relative normalization and development of the affected teeth in RO after a considerable delay **(Melamed et al., 1994)(Gerlach et al., 1998)**,

suggesting the cessation of the causative factor in these cases. Although the histological features are distinctive, diagnosis is based on clinical and radiographic findings, and coincides with the eruptive periods of the primary and permanent teeth. Many of the cases are discovered incidentally during routine clinical and radiographic examinations **(Pitak-Arnnop et al., 2020)**(**McKinney et al., 2022)**

The chief complaint is often delayed eruption or a non-inflammatory gingival swelling in the vicinity of the affected teeth. Some patients present with pain and abscesses in the affected area even in the absence of gross caries. These symptoms have been attributed to bacterial access to the pulp through clefts in the defective enamel and dentine **(VIJAYAN et al., 2021) (Elmezwghi et al., 2023).**

The patient and/ or parent may also present complaining of unpleasant appearance, problems in speech or mastication, draining fistulas, or facial asymmetry related to the loss of vertical dimension. In some reports, affected patients also had vascular naevi **(Nijakowski et al., 2022),** hypoplasia of the affected side of the face and facial asymmetry , hydrocephalus, dolichocephaly and clinodactyly, epidermal nevus syndrome, hypophosphatasia , and other abnormalities **(Slootweg et al., 1985)(Bagherpoor et al., 2010).**

1.6 Differential diagnosis

- Amelogenesis imperfecta
- Dental dysplasia
- As well as other disease states affecting the tooth formation apparatus.

Although conditions such as dentinal dysplasia, amelogenesis and dentinogenesis imperfecta show some similarities to RO, these conditions affect the entire dentition without segmental involvement. When the

affected teeth erupt, RO can be misdiagnosed as dental caries, and therefore, the teeth will be extracted without being submitted for histological examination. Many cases of RO are also misdiagnosed as malformed teeth or odontomes **(Soxman et al., 2019).** Most dentists elect to extract the teeth involved immediately and later rehabilitate the patient with a temporary removable partial acrylic prosthesis because, even if they erupt, the teeth are defective and of undesirable appearance.

The longer the affected teeth are retained, the higher the chances of pathology developing. This will necessitate extraction of the teeth, and their removal may be more difficult, especially if they are unerupted **(Abdel-Kader et al., 2019).**

The temporary prosthesis can be maintained till the age of 17 or 18 years, a time when the gingival margin is stable and restoration with a fixed prosthesis can be considered. Others clinicians have argued that removal of teeth at a young age may lead to undesirable psychological effects, and a substantial reduction in alveolar ridge height. The subsequent defect would also pose immense problems for future restorations should the patient want a fixed prosthesis or implants **(Elmezwghi et al., 2023).** In addition, loss of the vertical dimension on the affected side might lead to defective jaw development and subsequent facial asymmetry. Placement of osseo-integrated implants in growing children with hypodontia is well documented in the literature. Since the general quality of bone is not affected in RO, there may be a role for implants in such cases. Nevertheless, care must be taken when carrying out such procedures since there have been reports of a lower density of bone around affected teeth **(Tankittiwat et al., 2021).** Care must also be taken to place mandibular implants forward of the mental foramen, and a certain amount of implant impaction is expected in the maxilla because of relocation of

the maxillary sinus and nasal floor with growth **(Tankittiwat et al., 2021).**

1.7 Treatment

Restorative procedures to protect the affected erupted teeth have been suggested. Some clinicians have even suggested moving those teeth which have the most-developed roots orthodontically with subsequent restorations or prosthesis fixed on the pillar elements remaining **(Ziegler et al., 2012).** Extreme caution must be exercised if movement of affected teeth is to be attempted. Since the roots of these teeth are short and a lower density of bone has been observed around affected teeth, forces used for orthodontic tooth movement must be kept very low to avoid both root resorption and undermining bone resorption. Furthermore, the atypical shape of the crowns of these teeth and their hypoplastic nature make it very difficult to bond orthodontic brackets to these teeth. Once the permanent dentition has been established, joint orthodontic/restorative treatment aimed at uprighting any unaffected teeth that are severely tilted into extraction spaces may be commenced **(Hamdan et al., 2004).**

Treatment for other malocclusions may be carried out at the same time. Treatment of RO is controversial and no consensus has yet been reached. The dentist should take into consideration factors such as the age of the patient, the medical history, previous dental experience, the number of affected teeth, the presence or absence of any pathology, as well as the attitude and wishes of the child and parent. The aims of treatment should include aiding mastication and speech, improving aesthetics, reducing the psychological impact, allowing normal jaw growth and development, and if possible, protection of any erupted affected teeth. We present a case of RO that affected the primary and permanent dentitions in the anterior mandibular region, crossing the midline.

1.8 Case report

An 8·5-year-old girl was referred by her general dental practitioner to the Department of Paediatric Dentistry, Jordan University Hospital, Amman, Jordan, because of missing lower permanent incisors. The patient had neither other local abnormalities nor any relevant medical history. According to her parents, there had been a delay in the eruption of the lower primary incisors and canines, which had been grossly abnormal, with altered morphology, and yellowish/brownish in colour. The parents had not sought dental care for the child at that time and the lower anterior primary teeth had been lost very early after their eruption. There had been no similar cases among other members of the family.

1.8.1 On examination,

The patient had no facial asymmetry or other extra-oral abnormality. In the maxillary arch, there were partially erupted permanent central incisors, primary lateral incisors, canines, and first and second molars in addition to the first permanent molars. In the mandibular arch, only the first and second primary molars and the first permanent molars were present; these teeth were clinically normal. The lower incisors and canines were absent, the associated alveolar mucosa was enlarged and covered by fibrous tissue (Fig. 1). The oral hygiene was poor, but there were no active carious lesions.

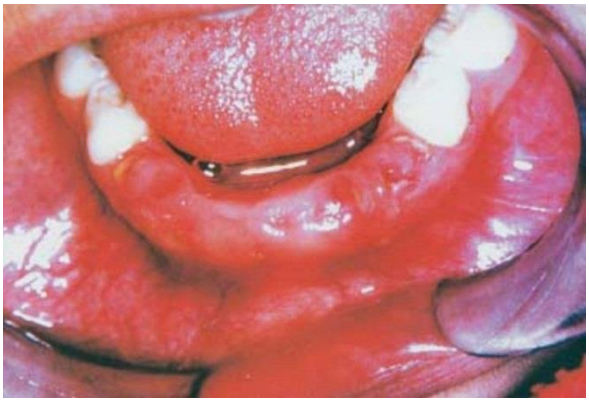

Fig. 1-1. Intra-oral view of the lower arch showing unerupted permanent incisors and canines, and gingival enlargement.

1.8.2 On Radiograph

Panoramic, occlusal and periapical radiographs were taken. The maxillary dentition was normal, as were the mandibular primary molars, premolars and permanent molars In the anterior mandibular region, permanent incisors and canines were unerupted and showed retarded development for the age of the child. The teeth had thin radio-opaque contours with no distinction between enamel and dentine, and wide pulp chambers giving a 'ghost-like' appearance. The crowns of the affected teeth were surrounded by large radiolucent areas, probably representing enlarged dental follicles. No, or only an insignificant amount of root formation, was visible radiographically (Fig. 2a,b).

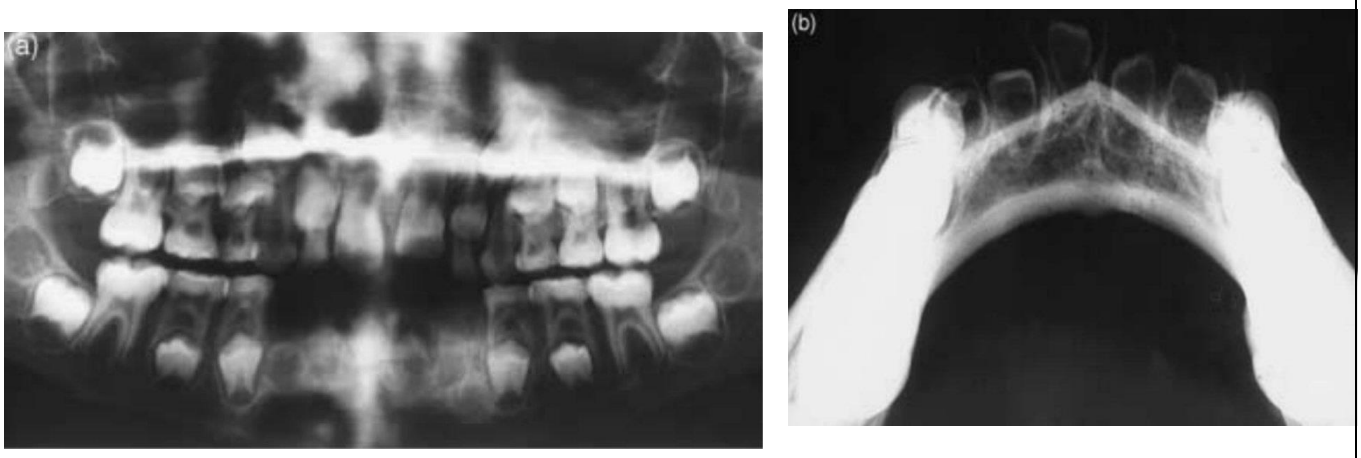

Fig 1-2 (a) Panoramic tomogram showing normal maxillary dentition and the ghost-like appearance of the mandibular incisors and canines. (b) Occlusal view

On the basis of the clinical and the radiographic findings, a provisional diagnosis of RO was made. Under local anaesthesia and intravenous sedation, all the unerupted mandibular incisors were enucleated, but the unerupted mandibular canines were retained. The enucleated teeth were of altered morphology, yellowish in colour, soft or rubbery in consistency, and had very short or unformed roots, and wide open apices. Following decalcification, the extracted teeth were prepared for microscopic examination in the usual manner and stained with haematoxylin and eosin. Light microscopic examination showed dental hard tissue and associated fragments of dental follicles. The dental follicles were hyperplastic, and were composed of dense fibrous connective tissue containing odontogenic epithelial rests and whorled fibrous tissue. These were scattered foci of calcification within both the odontogenic epithelial rests and the whorled fibrous tissue (Fig. 3).

The quantity of enamel matrix remaining after decalcification was inadequate for evaluation. The dentine was generally thin (Fig. 4) and contained a reduced number of dentinal tubules with irregular courses.

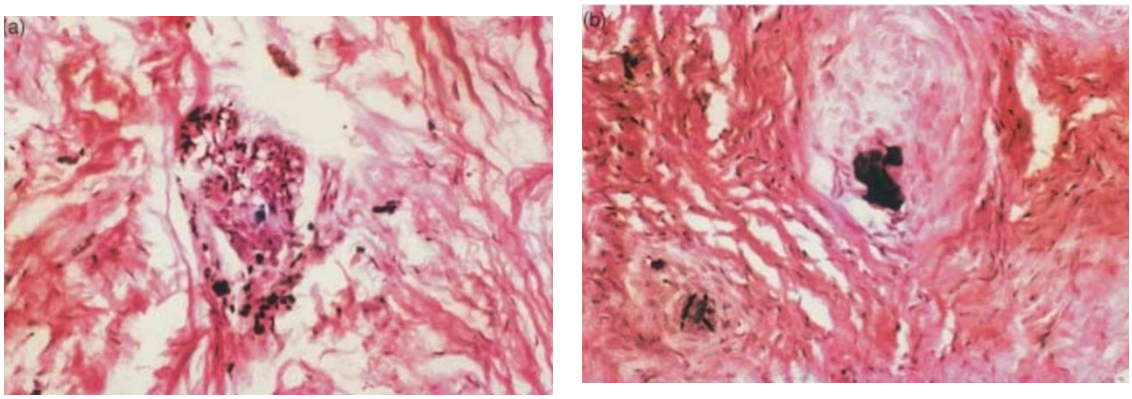

Fig 1-3 (a & b) Calcifications within whorled fibrous tissue in the dental follicle (H & E, × 200).

Large amounts of interglobular dentine and globular masses interrupting the dentinal tubules were evident. Closer to the dentino-enamel junction, the dentine was more evenly calcified. Generally, the predentine zone was wider than that seen in normal teeth (Fig. 4).

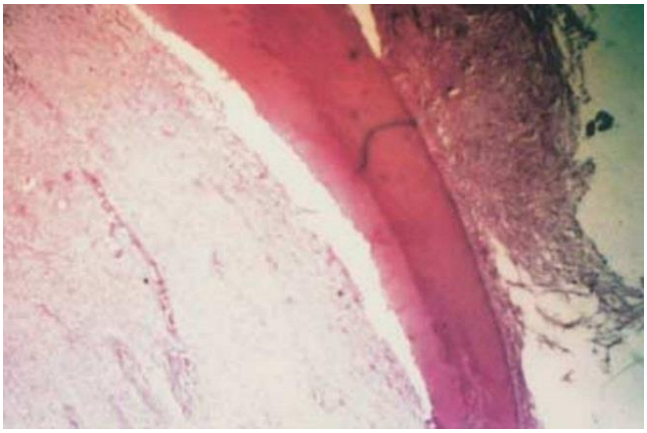

Fig 1- **4** Demineralized section showing the thin coronal dentine and a wide predentine zone (H&E, × 100).

The roots were short and thin-walled with noticeable interglobular dentine and wide open apices (Fig. 5). The pulp chambers were large and contained focal areas of calcifications or stones. The odontoblasts were flattened and lying along the margin of predentine. The pulp tissue was intact and no necrosis was seen. Based on the histological features, the provisional diagnosis of RO was confirmed. Two weeks postoperatively, a temporary acrylic mandibular partial denture was made to preserve the

alveolar ridge during the period of skeletal growth (Fig. 6). Oral hygiene instructions, and dietary analysis and advice were given, and regular fluoride application was planned in order to prevent caries and periodontal disease. The patient was placed on periodic recall to review the unerupted mandibular permanent canines and to monitor the development of the mandibular arch.

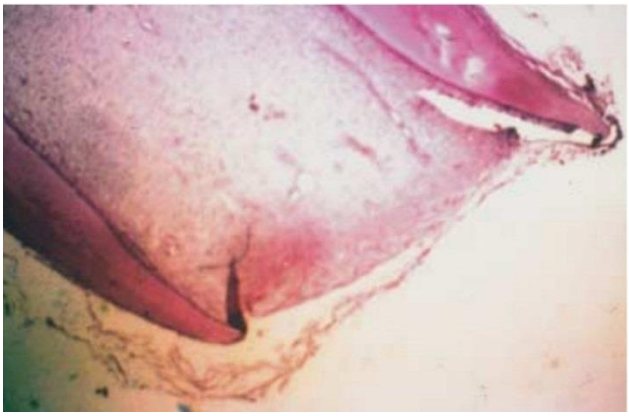

Fig 1-5. Demineralized section showing the short and thin-walled root of an affected tooth with a wide open apex (H & E, × 100).

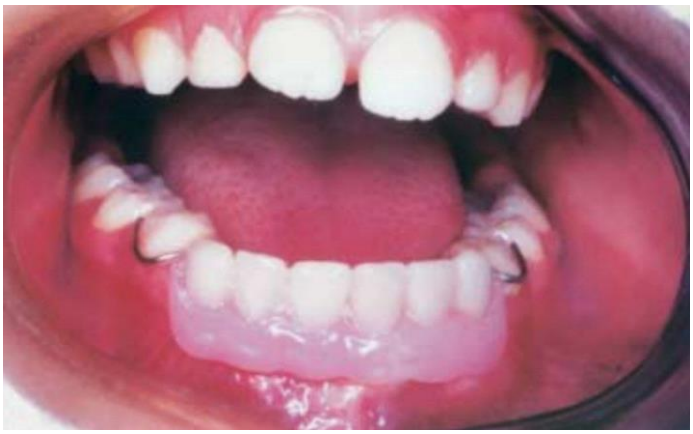

Fig 1- 6 Clinical view taken after fitting the removable prosthesis replacing the lower incisors and canines.

1.9 Treatment planning should be designed for each individual case of RO, taking into account factors such as

- The age of the patient,
- The medical history,
- The extent of involvement,
- The eruption of the teeth,
- Aesthetics,
- The development of pathology,
- And the wishes of the patient and parents.

CHAPTER TWO

Conclusion

Conclusion

Regional odontodysplasia is a rare developmental anomaly affecting deciduous and permanent teeth, which for several decades has been described only by authors from a few countries. Nevertheless, the radiological picture of ghost teeth accompanying clinical discolouration of tooth crowns due to enamel hypoplasia can be considered as a pathognomonic symptom. Although there is no consensus on the treatment method of choice, most cases ended with the surgical removal of the affected teeth. Similarly, the potential etiopathogenesis of this disorder has not been determined so far. It must be emphasised that patients with regional odontodysplasia require long-term specialised care due to the rapid onset and progression of the disease, as well as the gradual need to restore aesthetic and functional properties of the dentition.

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