Case report: severe infraocclusion
ankylosis occurring in siblings

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**ABSTRACT.** Aim This was to report a rare case of strong familiar tendency of ankylosis of maxillary second primary molars. *Case report* Three Caucasian children, male twins of 8.5 years and a sister of 10 years, were diagnosed as having severely infraoccluded maxillary second primary molars with underlying second premolars. In all three cases, the early extraction of the infraoccluded molars and an active treatment with cervical extraoral traction allowed the physiologic eruption of second premolars. Follow-up showed that normal vertical relationship and bone height had been obtained. Conclusion Early diagnosis, as well as appropriate treatment and careful follow-up are very important in the presence of severe infraocclusion, when the marginal ridge of affected primary teeth is at or below gingival level.

**KEYWORDS:** Familiar ankylosis, Extraoral traction, Early diagnosis.

**Introduction**

Ankylosis of teeth is defined as an anatomical fusion of tooth cementum with alveolar bone. It can occur at any time during eruption, before or after a tooth emerges into the oral cavity [Pilo et al., 1989]. Many reports over the years have indicated that these infraoccluded teeth have been in occlusion and are ankylosed to the alveolar bone as a result of some disturbance in their periodontal ligament and therefore remain in a fixed position [Darling and Levers, 1975; Brown, 1981; Kurol, 1984; Kurol and Magnusson, 1984; Rune and Sarvas, 1984], whereas those around them move occlusally with the continuous process of eruption.

Primary molar teeth are often found with their occlusal surface below that of the neighbouring teeth, long after they should have reached occlusion [Koyoumdjisky-Kaye and Steigman, 1982; Kurol, 1984]. The result is that the infraoccluded teeth appear to become progressively more submerged in relation to their neighbours over time [Kurol and Thilander, 1984; Kurol and Koch, 1985]. The progression has been correlated with age in some subjects but not in others [Kurol and Thilander, 1984; Kurol and Koch, 1985]. In some cases infraocclusion remains static or even decreases [Kurol and Thilander, 1984]. Maxillary molar infraocclusion reportedly progresses faster than mandibular molars [Kurol and Thilander, 1984].

Several hypotheses regarding the aetiology have been proposed, including deficient eruptive force, disturbed metabolism of the periodontal ligament, squeezing, trauma, deficient local vertical bone growth, local inflammation, and disturbance in interaction between normal resorption and hard tissue repair [Herman, 1964; Rune, 1971]. Other stated aetiologies for infraocclusion include gaps in the periodontal membrane, localized infection, chemical or thermal irritation, and abnormal pressure from the tongue [Kurol and Thilander, 1984].

A hereditary component has also been suggested, based on the observation of secondary retention in several members of the same family [Via, 1964; Kurol, 1981; Helpin and Duncan, 1986]. Ankylosed primary molars are more common in Caucasians and there is no reported sex predilection [Messer and Cline, 1980]. Primary molars are affected more frequently than permanent molars [Biederman, 1956]. Most reports show that mandibular first primary molars are the most
frequently affected, followed by second mandibular molars, first maxillary molars and second maxillary molars [Kurol, 1981].

 Clinically, it has been observed that several phenomena occur in association with the infraocclusion of primary teeth successor [Biederman, 1956; Brearley and McKibben, 1973; Messer and Cline, 1980; Kula et al., 1984; Pilo et al., 1989; Ertugrul et al., 2002; Kurol, 2002]. These are:

- marked tipping of the teeth adjacent to a severely infraoccluded tooth is frequently seen, with only a minimal degree of space closure occurring;
- the degree of the tipping of the adjacent teeth, which appears to be quite different from that associated with space closure that occurs because of the early loss of primary teeth;
- delayed exfoliation;
- bone defects;
- increased susceptibility to dental caries and periodontal disease of both the neighbouring teeth;
- retained permanent molar.

Treatment approaches. Early treatment of infraoccluded or submerged primary molars has been advocated, especially in the paediatric dentistry literature. Indications for early treatment include ankylosis or fusion of the infraoccluded tooth to the alveolar bone, which will cause occlusal and eruptive disturbances. Over time the progressive infraocclusion may cause tipping of adjacent teeth, bone defects, and hindered or delayed eruption of permanent successors.

Radiographic examination is essential prior to any treatment planning. When considering treatment, the degree of the infraocclusion, the patient’s age, the existence and location of the successor teeth are important. Extraction of the infraoccluded molar has been the treatment of choice to prevent the sequelae due to delayed exfoliation [Kurol and Thilander, 1984; Kurol and Kock, 1985; Ertugrul et al., 2002]. Extraction of the retained molar and the use of extraoral traction is usually the best treatment. The orthodontic treatment was scheduled to provide space for distal uprighting of the first permanent molar that had tipped mesially.

The purpose of this paper is to report a rare case of strong familiar tendency of ankylosis of maxillary second primary molars.

Case reports

A 10-year-old Caucasian female (Case 1) was referred to the Department of Orthodontics, University of Rome “Tor Vergata” by the Department of Paediatric Dentistry for advice and treatment regarding her infraoccluded teeth (55, 65, 84, 85). The patient came with her 8.5-year-old twin brothers (Cases 2 and 3). All three cases showed good oral hygiene, no soft tissue pathology and their general medical history was non-contributory. The parents attended with a panoramic radiograph of 6 month before.

Case 1

Clinical examination. This showed a 10-year-old girl with symmetric face and a normal development for her age. Intraoral examination revealed that all of the permanent teeth were present, some erupted, with the exception of the maxillary left and right second premolars (15, 25), the mandibular right first and second premolars (44, 45), the mandibular right lateral incisor and all the second molars (17, 27, 37, 47). The maxillary permanent molars (16, 26) were tipped with partial closure of the space for 15 and 25 (Fig. 1). The patient presented with a right and left Class II molar and canine relationship and a bilateral open bite.

Radiographic examination. This showed:

- presence of the completely submerged maxillary left and right second primary molars (55, 65) with underlying maxillary left and right second premolars (15, 25). The roots of 55 and 65 had been completely resorbed with only the shell of the crown remaining. There was a tipping of the adjacent first permanent molars (16, 26) and first premolars (14, 24) with closure of the space for 15 and 25;
- presence of the partial ankylosed mandibular right first and second primary molars (84, 85) with their permanent successors (44, 45) in their correct eruption position;
- agenesia of the permanent mandibular right lateral incisor (Fig. 2).

The cephalometric analysis recorded a skeletal Class II malocclusion with a severe deep bite.

Case 2

Clinical examination. This showed a 8.5-year-old boy with a symmetric face and a normal development for his age. Intraoral examination showed an infraocclusion of the maxillary right second primary molar (55). The permanent teeth present were 11, 12, 16, 21, 22, 26, 31, 32, 36, 41, 42, 46. The primary teeth present were 53, 54, 63, 64, 65, 73, 74, 75, 83, 84, 85. The maxillary right permanent molar (16) was tipped with partial closure of the space for 15 (Fig. 3).

The patient was in mixed dentition with a right and
left Class II molar and canine relationship, a maxillary right lateral cross bite and a right lateral open bite.

**Radiographic examination.** A panoramic radiograph revealed:
- presence of the submerged maxillary right second primary molar (55) with its underlying permanent successor (15) displaced high above the roots of the first premolar (14). The roots of 55 had been completely resorbed with only the shell of the crown remaining. There was tipping of the adjacent first permanent molar (16) and first primary molar (54) with partial closure of the space for 15;
- presence of the partial ankylosis mandibular left second primary molars (85) with its permanent successors (45) in the correct eruption position;
- absence of instances of agenesis (Fig. 4).

The cephalometric analysis recorded a skeletal Class II malocclusion with deep bite.

**Case 3**

**Clinical examination.** This showed a 8.5-year-old Caucasian boy with a symmetric face and a normal development for his age. Intraoral examination showed an infraocclusion of the maxillary left second primary molar (65). The permanent teeth present were 11, 12, 16, 21, 22, 26, 31, 32, 36, 41, 42, 46. The primary teeth present were 53, 55, 63, 64, 73, 74, 75, 83, 84, 85. The patient was in mixed dentition with a right and left Class II molar and canine relationship

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**Fig. 1** - Clinical view of the retained maxillary molars being covered by the surrounding tissues, Case 1.

**Fig. 2** - Panoramic radiograph showing: submerged maxillary second primary molars with underlying second premolars; partial ankylosis of mandibular right first and second primary molars with their permanent successors; agenesis of the permanent mandibular right lateral incisor.

**Fig. 3** - Clinical view of the infraocclusion of the maxillary right second primary molar.

**Fig. 4** - Panoramic radiograph showing: submerged maxillary right second primary molar with underlying permanent successor displaced high above the roots of the first premolar; partial ankylosis mandibular left second primary molars.
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and a left lateral open bite (Fig. 5).

Radiographic examination. A panoramic radiograph revealed:
- presence of the submerged maxillary left second primary molar (65) with its underlying permanent successor (25) in the correct eruption position. The roots of 65 had been completely resorbed with only the shell of crown remaining. There was light tipping of first primary molar (64) with partial closure of the space for 25;
- presence of the partial ankylosis of mandibular left and right second primary molars (75, 85);
- agenesia of mandibular left second premolar (35) (Fig. 6).

The cephalometric analysis recorded a skeletal Class II malocclusion with deep bite.

FIG. 5 - Clinical view of the infraocclusion of the maxillary left second primary molar.

FIG. 6 - Panoramic radiograph showing: submerged maxillary left second primary molar with its underlying permanent successor in the correct eruption position; partial ankylosis of mandibular left and right second primary molars; agenesia of mandibular left second premolar.

FIG. 7 - Clinical view of the case 1 year after orthodontic therapy. The second maxillary premolars have erupted normally.

Treatment. In all three cases, a diagnosis of infraoccluded primary molars was established and the treatment consisted of extraction of the submerged teeth. This was followed by orthodontic therapy, in all three cases, with cervical extraoral traction for about 8 months (Fig. 7).

Follow-up. In Case 1 the patient was reviewed monthly for 1 year, until the complete eruption of second premolars. In Cases 2 and 3 the patients were seen monthly for 2 years, until the physiologic eruption of second premolars. For all three cases at the end of the follow-up period a normal vertical relationship and a bone height have been obtained.

Discussion

In the management of infraoccluded primary molars, early diagnosis as well as proper treatment and careful follow-up are very important. The indication for treatment depends on the age of the patient, the degree and the extent of infraocclusion, the amount of root resorption, the severity of tilting of neighbouring teeth and the presence and location of the permanent successor.

On the basis of some previous reports [Kurol, 1981; Via, 1964; Helpin and Ducan, 1986], we concluded that there was a hereditary component of ankylosis based on the observation of infraocclusion of second maxillary primary molars in several members of the same family. Ankylosed primary molars are common in 9% of young people and more so (18%) in siblings, indicating genetic factors at work [Kurol, 1981].

It should be emphasized that the extraction of
retained molars is important in the presence of severe infraocclusion, when the marginal ridge is at or below gingival level. In all three cases presented here the early extraction of the infraoccluded molars and an active treatment with cervical extraoral traction for 8 months allowed the physiologic eruption of second premolars obtaining a normal vertical relationship and normal bone height.

Conclusion
Early diagnosis as well as proper treatment with cervical extraoral traction and careful follow-up are very important in cases of severe infraocclusion of the primary molars when their marginal ridge is at or below gingival level.

References