Case report

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Using of modified rapid palate expander with miniscrews in a patient affected by ectodermic dysplasia

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Abstract

Objective. To show the orthodontic treatment in a 8-year-old patient affected by Ectodermal Dysplasia (hypohidrotic type) and presenting multiple agenesiae, contraction of the maxilla and skeletal Class III malocclusion.

Study design. Because of both oligodontia of primary and secondary dentition and no good retention and anchoring, a hybrid modified rapid palatal expander (RPE) was used. It presented dental anchoring with two bands on first upper molars and skeletal anchoring with two miniscrews in the anterior palate. The project included the use of a CBTC for the bone examination and precise silicon dental impression for the insertion of miniscrews.

Results. The procedure was successful and the patient solved the expansion in few days, so RPE has been embedded throughout 6 months in order to develop the bone at the median suture.

Conclusion. This case report can be considered as a valid example for approaching patients affected by Ectodermal Dysplasia with multiple agenesiae and palatal contraction because of the difficult retention.
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Key words: *Ectodermal Dysplasia, Rapid Maxillary expander, miniscrews, primary dentition*

Introduction

Ectodermic Dysplasias (ED) are an assorted group of malformed genetic syndromes characterized by hypoplasia, aplasia as well as dystrophy of ectodermic structures (1-2).

Patients affected by ED have one or more structure abnormalities of ectoderma such as: hair, teeth, nails, sweat glands, cranio-facial structure, fingers (3).

Since 1971 only 8 types of DE existed, while nowadays more than 200 types have been accounted for; the genes involved are 64 and 3 chromosomal loci that cause a genetic transmission both dominant and recessive as well as both dominant and recessive X-linked.

The most intensive kind of DE is the hypohidrotic one with an incidence of 1 to 7 in every 10 000 newborns and is more common and severe in caucasic males (4-5). As regards the oral manifestations, these patients show abnormalities of quantity, shape and structure teeth both in primary and secondary dentition (5). The alterations which concern the number of teeth are mostly hypodontia (genetic absence of teeth), anodontia (total absence of teeth) (6). In secondary dentition the average of absent teeth is 23.7 and the mean number of erupted teeth is 9 (7) (upper central incisors 42%, first upper molars 41%, first lower molars 39%, upper canines 22%) (8-9). Absence of many teeth is often the first signal of the pathology and induces parents to specialistic tests. Hypodontia and anodontia involve not only the alveolar crest but can also cause hypoplasia of both jaws, and conduce to a lost vertical occlusal dimension and to a reduction of facial height of the patient (10-11). These authors identify a third skeletal tendency class because of an insufficient vertical development of alveolar processes (12).

As regards the abnormalities of the shape they concern the presence of conoid teeth in incisal area and globular teeth in posterior zone (13).

The mutation of the structure involves the enamel that appears immature or hypocalcified or hypoplasic. There are many other characteristic signs at mucosae, such as: absence of labial groove, double labial frenulum, keratosis of oral mucosa, gingival fibromatosis, papillomas of gums and lips, aggressive periodontitis (14-15).

The gold treatment of this pathology is early diagnosis and multidisciplinary approach both medical (pediatrician, ophthalmologist, dermatologist, otolaryngologist, psychologist, speech therapist) and dental (pediatric dentist, orthodontist, prosthodontist) one.

The first step of diagnosis is the family anamnesis, in order to research other cases of ED or other abnormalities such as absence of teeth, lateness of eruption, hyperthermia, otitis and commonly pharyngitis. On Literature there are several studies about the use of orthodontic miniscrews which show that miniscrews must have a diameter from 1.2 to 2 mm and length from 6 to 12 mm. The sites of insertion are under the nasal backbone, palate, infrazygomatic crest,

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jaw tuberosity, alveolar processes between roots, symphysis, parasymphysis, alveolar processes and the area back to molars (15-18). The indications for treatment with miniscrews are: molar protraction, indirect anchorage for space closure, intrusion of supererupted tooth, intrusion for anterior openbite, anterior en masse retraction, molar uprighting intrusion for maxillary cant, molar distalization, traction on impacted canine, attachment for protraction face mask (19).

Side effects for using these miniscrews include problems of healing, compromised immune defense, bleeding disorders, an inadequate oral hygiene (20-21).

The incidence of failures on miniscrews is quite low and the most of it is in patients under 20 years of age but it is totally independent on site of insertion as well as sex of the patient (22-23).

There is no side effect for using miniscrews during pediatric age; nevertheless, the presence of all permanent teeth is quite important in order to preserve dental gems.

In a study available since 2010, it is described the use of hybrid rapid palatal expander with dental anchoring on first upper molars and skeletal anchoring with two miniscrew on the anterior palate (24). There are other similar studies with the use of RPE (Rapid palatal Expander) with miniscrews (25-29), but there is no evidence of the use of RPE with miniscrews in a patient affected by DE on Literature. So the aim of this paper was to show the orthodontic treatment in a 8 year-old patient affected by Ectodermal Dysplasia (hypohidrotic type) with contraction of the maxilla and skeletal Class III malocclusion.

Case Report

An 8-year old patient affected by Hyprohidrotic Ectodermic Dysplasia as well as his mother and brother presented to Pediatric Dentistry Unit, Department of Dental and maxillofacial Sciences, Sapienza University of Rome.

The pathologic anamnesis showed the presence of DE, OSAS (30) and oral breathing.

The extraoral exam revealed brachifacial type, small chin, acute nose-labial angle, protruded lips, projecting and square-shaped forehead, protruded eyebrows, creases and pigmentations in periocular area, small nose, big ears, blonde thin fine hair (Fig.1a-1b) .

The intraoral exam underlined the multiple agenesis of primary and secondary teeth verified with orthopantomography(31) (Fig 1c).

The maxilla presented: central primary incisors, primary canines, the second primary molars and the first permanent molars, while in the orthopantomography were the gem of the permanent incisor.

In the mandible only second primary molars and first permanent molars were present.

The objective exam and plaster models showed as hypotrophy of both jaws and the maxilla decreased both on transversal and on sagittal planes. Exams also showed thin and small edentulous crests.

Cephalometric analysis underlined first skeletal class with a light tendency of third skeletal class, hypo-divergence, counterclockwise growing, reduced anterior facial height. (SNA=72,4°; SNAB=73.2°; ANB=-0.8°; N-S-Ar= 130.1°;

Fig. 1a: intraoral frontal view; 1b: intraoral occlusal view; 1c: orthopantomography

S-Ar-Go= 130.3°; Ar-Go-Me=127.6°; ∑= 388°; S-Go= 52.3; N-me=81.2; FMA=1.7°).

After signing an informed consent, the first step of the therapy was solving the transversal contraction of the maxilla also for the breathing problems. The treatment protocol was conformed to the ethical guidelines of the 1975 Declaration of Helsinki.

Because of both oligodontia of the primary and secondary dentition and no good retention and anchoring, we decided to devise a hybrid modified rapid palatal expander: dental anchoring with two bands on first upper molars and skeletal anchoring with two miniscrews in the anterior palate (32).

A study published in 2011 suggested the insertion of miniscrews in the anterior palate because of the good density of the bone and the optimal anchorage for every movement, especially for the expansion of the palate(33). Because of the presence of dental gem and the hypotrophic bone it was necessary to realize a CBCT to verify the quantity of available bone and space for the correct insertion of the miniscrew (34) (Fig.2).

In order to have a very precise indication for the placement of miniscrews it was created a surgical template (vacuum-formed) with two overstuffed holes with guttapercha that the patient put on to execute a CBCT. The CBCT showed a reduction of the maxilla with the possibility to insert miniscrews having 6mm length and 1.8 mm diameter. The vacuum-formed template was also used during surgery as a guide for miniscrew placement (Imtec ORTHO Implant, IMTEC Corp, Ardmore, OK). Also, there was an O-Cap integrated on the head of the miniscrew where it was possible to settle the RPE. The whole orthodontic appliance was made in titanium.

Fig. 2. CBCT

For the surgery, under local anesthesia the surgeon placed miniscrews used a 1.1-mm-diameter pilot drill rotating no greater than 1000 rpm in order to prepare a cautiously shallow perforation of the cortical plate under sterile irrigation.

The implants were placed using the straight driver and tightened to approximately 40 N/cm.

It was important to reach a good parallelism between miniscrews for an optimal distribution of the forces of RPE (Fig. 3a). After the insertion of miniscrews, the surgeon put the O-Cap and took a high-precision silicon dental impression. The laboratory produced a titanium modified RPE with other two miniscrews similar to the ones on the dental impression.

RPE was fixed with vetroionomer cement on bands on first upper molars and the protocol consisted of two activations a day for 12 days (Fig.3b).

During the following check, the patient was exposed to occlusal X-ray in order to verify the diastases of the median palatal suture. The protocol was successful and the patient solved the expansion in a few days, but RPE remained fixed for 6 months for the development of the bone at the median suture (fig. 3c). After that, there was the resolution of the transverse contraction but esthetics had to be preserved (35). In order to guarantee a functional occlusion and esthetic smile, the laboratory produced two prostheses with artificial teeth in the anterior area with a dental and mucosal anchorage: 4 upper incisors and 4 lower incisors (Fig.3d).

The follow-up, after two years from the expansion with modified RPE, showed the keeping of the expansion and the eruption of another conoid in the incisal zone (Fig.4a-4b).

Discussion and Conclusions

Ellis and McNamara (36) found that 65–67% of all Class III malocclusions were characterized by maxillary deficiency. In subjects whose mandible is not markedly affected, orthopaedic forces can be used in order to stimulate and guide maxillary growth. Different extraoral appliances have been used to correct maxillary deficiency, such as facemasks and reverse pull headgears but they are linked to patient compliance due to their size and appearance (37). Recently, miniscrews (mini-implants) have resulted most used because they are easier to both insert and remove (38-39). There is no evidence-based evaluation of the allergic/inflammatory reaction either to orthodontic titanium miniscrews in adolescents and young adults (40). In order to minimize the failure of miniscrew implants, proper oral hygiene instruction and effective supervision should be given for < 12-year-old patients(41). Although this popularity, Rodríguez de Guzmán-Barrera J et al (42). concluded that skeletal anchorage is an effective treatment for treating skeletal Class III malocclusion, but when compared with other traditional treatments such as disjunction and face mask, there is no clear evidence that skeletal anchorage improves the results. In Literature regarding patients affected by genetic diseases there is a study about Down' s

Fig.3a: the inserted miniscrews in frontal wiew; 3b: RPE banded on teeth; 3c: intraoral frontal view after expansion
3d: esthetic prostheses

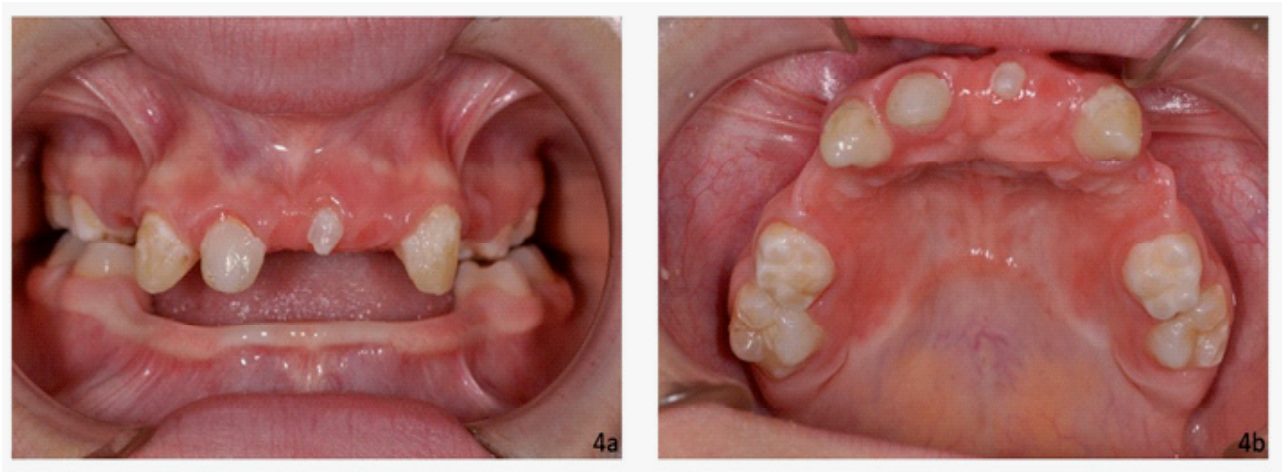

Fig 4a: 2-year follow-up after with the eruption of another conoid; 4b: occlusal view of 2-year follow-up

syndrome patients(43) treated with miniscrews but there is no evidence of using miniscrews in patients affected by ED. In these patients the lack of tooth eruption and consequent decreased functional stimulation contribute to alveolar bone dysplasia, osteopenia, height reduction, and a knife edge alveolar ridge, which can complicate their oral rehabilitation. The oral alterations of ED may affect patient mastication, nutrition, and maxillary development. Patients might also have difficulties with phonetics and esthetics, commonly resulting in emotional problems such as low self-esteem and limited social interactions (44). Despite international efforts toward a standard dental treatment for patients with ED, no specific clinical treatment protocol has yet been defined for the dental management of the heterogeneous anomalies in ED (45-46). An early oral rehabilitation of young patients is fundamental to improve their social interaction and restore their speech and masticatory function. So miniscrews could be a possible key to solve some skeletal problems in an optimal way. In order to have a good answer to the therapy, it is important to select the patient for: systemic health, age, compliance, oligodontia, hypotrophy of the jaws, location of dental gems. Miniscrews can give the good distribution of forces without dental effects. This case report can be considered as a valid example for approaching patient affected by DE with multiple agenesiae and palatal contraction because of the difficult retention.

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