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Dental Management in Children with Ectodermal Dysplasia Using Removable Denture And Acrylic Crown: A Case Report

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Abstract

Oral manifestations of ectodermal dysplasia include the absence of tooth buds and peg shape. The aim of this paper is to report on dental management for children who have ectodermal dysplasia using removable dentures and acrylic crowns. The patient, a boy aged 10.5 years, with the absence of tooth buds 12, 21, 22 and 42 and 43, 41, 31, 32 and 33 were peg-shaped, which disturbed the child's aesthetics. Based on the anamnesis, clinical and radiographic appearances, the child was diagnosed as having hydrotic type ectodermal dysplasia. There are two stages of the treatment for the patient; the first stage of making a removable partial denture on the upper jaw and an acrylic crown on the lower jaw. The procedure for making dentures begins with taking an impression and send to the laboratory. The acrylic crowns was made by using the indirect technique. The patient was asked to undergo regular control every 3-6 months until the child's growth and development is complete, by the age of 18 years. At this age, the second stage of treatment can be carried out by making a final denture using a bridge on the upper jaw or implants and making a porcelain crown on the lower jaw. It was concluded that the choice of dental treatment for the patient depends on the age of the child and the first stage of treatment for pediatric ED patients is quite successful aesthetically.

Keywords: dental management; ectodermal dysplasia; children; removable partial dentures; acrylic crowns

1. Introduction

Ectodermal Dysplasia (ED) is defined as a genetic disorder with two or more congenital defects in the ectodermal structure. Three disorders that often accompany ED, namely alopecia or hypotrichosis (sparse or thin hair and eyebrows), periorbital pigmentation, hyperkeratotic palms due to lack of sweat glands (hypohidrosis), partial or complete absence of primary teeth and/or permanent teeth, and sometimes accompanied by dystrophy nails (ochodysplasia).[1-4] The teeth most often absent in ED cases are the maxillary lateral incisors, maxillary first premolars, mandibular incisors and mandibular first premolars.[5] Other clinical features often found in ED patients are frontal bossing, saddle nose, prominent supra orbital ridge, prominent ears. and oblique, prominent lips, facial concavity, disruption of alveolar ridge development due to anodontic teeth resulting in a short face, dental malformation (peg shape), dry oral mucosa and the child's face looks old.[6-8] The general incidence of ED is 1 in 10,000 births to 1 in 100,000 births.[9] Other researchers state that the prevalence of ED varies between 6-9/10,000 births.[10]

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The types of ectodermal dysplasia are generally divided into: Anhidrotic/hypohidrotic ectodermal dysplasia (HED). This type is also known as Christ- Siemens Toirane Syndrome. HED is reported to be found more frequently, around 80% of cases of ectodermal dysplasia.[11] The etiology of HED is often associated with x-linked recessive, the presence of mutations in genes located at Xq12-q13.[10] HED is more common in men with more severe conditions than in heterozygous women so that defects that occur in women are usually minor (carriers).[12.13] The incidence of HED in men is 1 in 100,000 births, while in women who are carriers it is 17.3 in 100,000 births.[12]

The second type of ED is hydrotic ectodermal dysplasia, also known as Cloustone syndrome. In this type, the sweat glands are not affected but the teeth, hair and nails can be involved. The incidence is 1-7 in 100,000 births, with a 28% mortality rate in boys up to 3 years of age.[14] The etiology of the hydrotic type is autosomal dominant and autosomal recessive.[10]

Dental management for patients with ED requires clinical knowledge regarding growth and development, behavioral management, pedodontics, prosthodontics, orthodontics and oral surgery. [15] A multidisciplinary team approach is needed, since ED cases are often found in early childhood. Oral rehabilitation in ED cases often involves making removable full or partial dentures, followed by a definitive prosthesis in the form of a fixed prosthesis or implant after complete alveolar bone development. Fixed prostheses are started in young adulthood, because rigidly connected fixed partial dentures, crossing the midline, can interfere with the normal growth of the alveolar bone. Crowns that require a lot of tooth preparation are also avoided for children's dental care, because they might cause the pulp to be exposed due to the large pulp chamber.[16] This paper aims to report on dental care management for a 10.5-year-old boy with ectodermal dysplasia using removable dentures and acrylic crowns.

2. Case Report

A 10.5-year-old boy came to a private pediatric dental specialist clinic in Medan- North Sumatra (Indonesia) with his mother with a chief complaint that many of his permanent teeth had not yet erupted and the lower front teeth had grown in a sharp shape, thus disturbing the child's appearance. The patient had a panoramic X-ray taken by another dentist and was referred to a pediatric dentist for further treatment. According to the history with the mother, the patient was the first of three boys. The other two boys had no complaints like the first child. Neither the fathers' families nor the mothers' have such cases. There was no medical history or trauma to the mother during pregnancy or to the patient that contributed to this incident.

Extra oral examination showed that the patient's eyebrows were thin/sparse, frontal bossing, periorbital pigmentation, prominent ears, prominent lips, normal fingernails and no interference with the sweat glands and the palms did not experience hyperkeratosis (Figures 1 and 2). Intra-oral examination revealed hypodontic on 12, 21, 22, and 42. Tooth 11 had shifted to the right, away from the midline (distoversion) due to the absence of tooth 12. Teeth 33, 32, 31, 41 and 43 were peg-shaped so a diastema was visible on the teeth. The teeth that erupted were teeth 16, 15, 14, 13, 11, 24, 25, 26, 36, 75, 34, 33, 32, 31, 41, 43, 44, 85, and 46 (Figure 3).

The panoramic radiograph revealed no sign of tooth buds 12, 21, 22 and 42. Tooth 23 was seen on the panoramic radiograph to grow soon and clinically it was seen that the gums in that region had become inflamed and felt hard as if a tooth was about to erupt. Tooth buds 35 and 45 are visible on the radiograph (Figure 4).

Based on the anamnesis, clinical and radiograph appearance, the patient was diagnosed with hydrotic type ectodermal dysplasia. The patient has two signs of ectodermal dysplasia: problems with the teeth (hypodontia) and hypotrichosis (thin eyebrows). Likewise, other extra-oral clinical features supporting this diagnosis are the presence of periorbital pigmentation, and prominent lips and ears in the patient.

The treatment plan for this patient is in two stages; the first stage is making maxillary removable partial dentures for the three upper front teeth and making dental crowns for the four lower front teeth which are peg shaped. The second stage after the child's growth and development age is 18 years and over, a fixed denture can be made on the upper jaw by making a porcelain bridge or implants. On the lower jaw, crowns can be replaced with porcelain with

better aesthetics than acrylic. However, before the second stage is carried out, it is necessary to have an orthodontic treatment first, to return the position of the distorted tooth 11 to be in the midline and the position of the other teeth which have shifted because the tooth buds next to it was missing. Orthodontic treatment can be carried out in the first stage that is mesialization of tooth 11, but due to aesthetic considerations and the child's location far from the clinic, it was decided to make dentures first, after all the teeth have erupted and the child's age is appropriate, a final denture/crown is made, then orthodontic treatment is carried out before the placement of the denture/crown.

After the parents agree upon the treatment plan, an impression was taken from the upper and lower jaw teeth with alginate material. The impressions were filled with dental stone, and the working models were sent to the laboratory to make removable partial dentures for the upper jaw and acrylic crowns (GC® Tempron, GC Corporation Japan) for the front teeth of the lower jaw.

At the second visit, the removable maxillary denture was tried on the patient. The teeth were made for the removable partial dentures made: 11, 21, and 22. Tooth 11 which has a distortion was considered tooth 12, therefore this tooth looked larger than the second incisor tooth in general and was longer. However, tooth 11 was not grinded or shaped like a second incisor, because this tooth in the second stage will become tooth 11 after being pulled mesially. The removable denture is tried on the patient, asked whether there is pain or not, then the retention and occlusion are checked.

Acrylic crowns made were for 31, 32, 41 and 43, while the peg shaped tooth 33 was not made because clinically the eruption of tooth 33 was still short (under occlusion). An attempt was made to insert the acrylic crowns on teeth 31, 32, 41 and 43, the retention and cervical parts were checked, then the occlusion was checked along with the maxillary removable denture. After there were no problems, the acrylic crowns for the 4 lower front teeth were cemented using GIC luting, and the patient was prohibited from eating for 1 hour because the cement had not yet hardened. After the teeth were cemented, the mother and patient were very happy because the patient already had artificial teeth and the sharp lower teeth looked normal, so the patient's self-confidence increased.

Patients are instructed to maintain oral hygiene because additional tools and acrylic crowns in the oral cavity will increase the risk of caries if the patient cannot maintain good dental health. Removable partial dentures are cared for by cleaning them twice a day with a brush and soap. At night before going to bed, the denture is placed in a glass filled with water.

Control at the dentist for the removable denture can be done every 1 week, 1 month and every 3-6 months. However, because the patient could not return one week later, it was agreed to have the child checked 1 month later. During the control, they are asked whether there is pain on the device, checked to see if there are red areas due to pressure, the location of the retention wire and most importantly, check the patient's oral hygiene. Oral prophylaxis was done, and dental education about oral hygiene for children and parents is carried out. The next control, patient is asked to check every 3-6 months.

3. Discussion

Considerations in dental treatment for children depend on the child's age, stage of growth and development, abnormal tooth morphology, number of missing teeth, location of missing teeth, and psychological status.[2] Treatment for patients with ectodermal dysplasia might using composite restorations or the use of crowns, making dentures, implants, orthodontic treatment, and prevention in the form of diet, topical fluoride applications, fissure sealants and dental education.[3] Direct and indirect composite crowns can be used to improve the aesthetics of ED teeth that are peg shaped, to increase facial height and articulation. Making dentures in children can start from 2-3 years,[17] but other clinicians recommend the age of 3-4 years.[13] Implants have the advantage of being a fixed prosthesis because they are good at support, retention, and stability. However, for young patients, attention must be paid to the right time of use because it will cause complications in jaw growth.[3] However, Guckes et.al reported the use of implants in young children at 3.5 years and 7-11 years of age. Young children will produce implants that are submerged from occlusion (infraocclusion) due to alveolar bone growth, which requires later correction of the implant.[18,19] Installation of implants in children begins at the earliest age of 15 years for girls and age 17 years

for boys.[6] Orthodontic treatment in hypodontia cases is used to overcome spacing, correct tooth alignment and overbite due to missing teeth.[20]

Prosthodontic rehabilitation is fundamental in children with ED, because it aims to provide a functional and aesthetic solution that allows the child to lead a lifestyle as normal, without damaging self-esteem or psychological development.[21] The treatment plan in this case has two stages, the first stage is making a removable partial denture on the upper jaw and making acrylic crowns on the 4 front teeth of the lower jaw. Next, in the second stage, after the child's growth and development are complete, the upper jaw can be made by a fixed bridge or implant denture and making a fixed crown with porcelain material on the lower teeth. Fixed dentures require conditions such as the apex of the tooth already closed, the pulp chamber is not large and the bone being compact enough to support porcelain dentures heavier than acrylic. Apart from that, the child's gingiva has not yet matured, where the position of the mature gingiva will be more towards the cervical side of the tooth and is achieved after the child's growth is complete, by the age of 18 years and above. If fixed dentures/crowns are used at a young age, the cervical part of the crown will be visible after gingival maturation is complete.[22]

In this case, the choice of acrylic crown is quite good in terms of aesthetics and easy to manipulate. [23] Contraindications to the use of acrylic crowns are in cases of bruxism, deep bite and excessive abrasives, [24, 25] and in this case none of these were found. In this case, the lower anterior teeth are pegshaped, if treatment is not carried out, the teeth can tilt into the empty area, causing the teeth to rotate or shift.[26,27] Making acrylic crowns for patients can improve the aesthetics of the child's teeth, thereby increasing self-confidence. [23]

The main problem in oral rehabilitation in ED cases is that the height of the alveolar bone is disturbed because the tooth buds are mostly missing.[2] Still, in this patient, this does not happen because there are only three hypodontic teeth in the upper jaw (teeth 12, 21 and 22) and one at the lower jaw (tooth 41), so making a removable denture is not too difficult. However, the problem in making dentures in this patient was that tooth 11 had moved distally, so space 12 was occupied by tooth 11 because tooth 12 had hypodontia. This resulted in three teeth being made in the removable denture; 11, 21 and 22. The aesthetics of the denture were not satisfactory because tooth 11 looked bigger and longer, but tooth 11 could not be shaped like a second incisor because in the second stage the tooth would be retract to the mesial and act as tooth 11, while the fixed denture that will be made is tooth 12.

The disadvantage of removable dentures is that they easily retain food on the abutment teeth and are less aesthetic. Therefore the solution for using this device is only for a short period.[17] However, this situation can be overcome with regular check-ups, where during the check-up the most important thing is oral hygiene and dental health education. Existing teeth are always checked, and if there are cavities, restorations are in order. Prevention of cavities can be done at regular check-ups every 6 months by administering topical fluoride applications. [3,28] Regular control is very important for children who use dentures, to avoid any disruption to the growth and development of the jaw.[15] During control, you need to pay attention to the retention wire, reassure that it does not interfere with the growth and development of the teeth. Do not use an occlusal rest on back teeth that have not yet completed their growth and development. In this case, the retention wire used on teeth 16 and 26 with two finger clamps and tooth 11 with C clamps, the apex of tooth 11 is completely closed so there is no problem using retention clamps even though the aesthetics are not good.

Removable dentures in children are replaced several times. The greatest growth of the jaw and dental arch occurs when the teeth start to erupt. Vergo stated that rebasing or relining needs to be done every 2-4 years for dentures and need to be replaced every 4-6 years due to jaw growth.[29] Replacement of dentures in cases is likely to be carried out at the age of 14-15 years, when the permanent canines, second premolars and second molars have erupted.

4. Conclusion

Dental treatment for pediatric patients with ED requires consideration, including the patient's age. In this patient, a removable partial denture was made on the upper jaw and acrylic crowns were placed on the lower front teeth considering that the child was in a state of growth and development. After the growth and development period is

complete, final treatment can be carried out by making fixed teeth with porcelain or implants. Routine periodic control is very necessary for children who use dentures to prevent abnormalities in the growth and development of the jaw. Dental care carried out on children increases children's self-confidence.

Figure 1. (a) Extraoral profile (b) normal fingernails

Figure 2. Intraoral profile

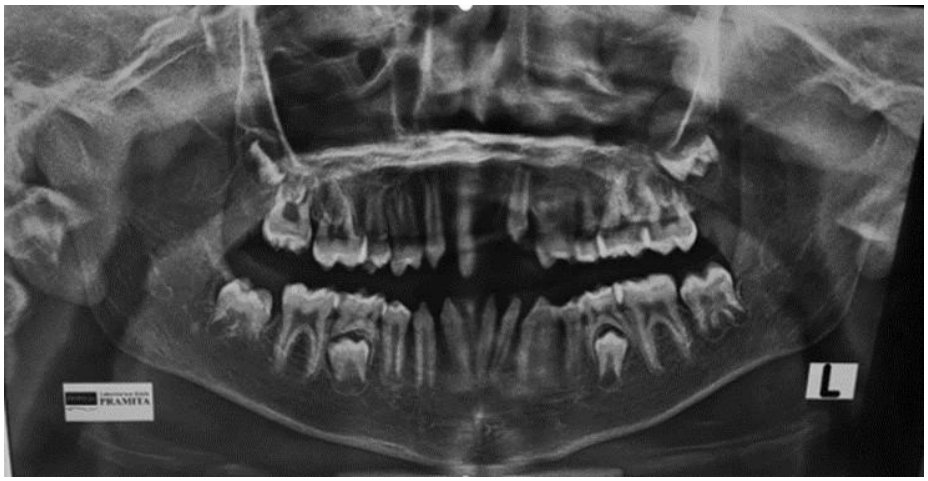

Figure 3. Panoramic radiograph

Figure 4. Rehabilitation with removable denture in maxilla and acrylic crown in mandibula

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