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ORIGINAL ARTICLE

A Preliminary Study to Analyze the Cranio-facial Growth of an Ectodermal Dysplasia Patient After Prosthetic Rehabilitation

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Abstract An experimental study on craniofacial development and jaw growth pattern of an ectodermal dysplasia patient was performed and was compared with normal individual. An ectodermal dysplasia patient with complete anodontia was prosthetically rehabilitated with complete dentures at age of 6 and 8 years. Two sets of complete dentures were made with age-appropriate denture teeth and a lingualized occlusal scheme. Periodic follow up and adjustment whether needed was done to maintain proper oral function and aesthetics. Serial cephalometric analysis exhibited a marked restriction of forward growth at the ANS point during 6–8 years of age although there was a little change from average in the anteroposterior length of mandibular body and the height of mandibular ramus. So, the maxillary growth was reduced but there was no significant change in the mandibular growth. Cast analysis showed that increase in arch length was greater than in arch width for both maxilla and mandible. There was a little increase in alveolar ridge height in the anterior region but there was a considerable increase in the height of the alveolar ridge in the middle and the posterior region. Our findings concluded that the absence of teeth did not affect the growth of the jaws and probably the denture flange did not arrest the jaw growth, rather it improved the masticatory function by providing good denture stability and retention.

Keywords Age appropriate denture teeth · Cast analysis · Complete anodontia · Complete denture · Cephalometric analysis · Lingualized occlusal scheme

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Introduction

Ectodermal dysplasia syndrome (EDS) has been described as a group of disorders of morphogenesis displaying two or more of the symptoms of trichodysplasia, dental anomalies, onychodysplasia, and dyshidrosis [1]. According to an estimate 150 different subtypes of ectodermal dysplasia can be defined with estimated frequency of one case occurring in every 10,000–100,000 birth [2]. Some EDS types are mild while others can be devastating. Although female carriers outnumber affected males, the female carriers show little or no signs of the condition because it is usually transmitted through a sex-linked recessive gene so the manifestations are predominant in males [3]. In children with ED, the appearance of the teeth is extremely important because it can affect the patients' self esteem which creates challenges for the dentist [4].

Hypohidrotic ED is the most common type of EDS and is characterized by several effects including hypohidrosis, hypodontia or anodontia and hypotrichosis. Complete dentures are fabricated for restorations of oral functions and esthetics, in complete anodontia cases. Some cases of prosthetic management of ectodermal dysplasia have been reported in the literature, although there is little information to notice the effect of dentures on the growth and development of jaws [5–10]. The present study was

performed to discuss the jaw growth and the prosthetic management of such a rare case.

Methods

A 6 year old boy of hereditary ectodermal dysplasia was reported to the Department of Prosthodontics, Faculty of Dental Sciences, CSMMU, Lucknow, Uttar Pradesh, India. The boy exhibited classic features of ectodermal dysplasia—anodontia, hypotrichosis, saddle nose, and prominent forehead (Fig. 1). His parents' main concern was their son's lack of teeth, difficulty to eat properly and, to some extent, the esthetics. Family history was noncontributory as none of the parents or the siblings had similar features. Radiological examination revealed absence of all teeth or tooth buds (Fig. 2). Among the different treatment modalities available, the best treatment option for the child was fabrication of complete dentures. At the age of 6 years the first set of complete denture was fabricated in usual manner. An age appropriate teeth set was fabricated using tooth colored heat-curing acrylic resin (DPI heat cure tooth moulding powder, Dental Products of India, India) [11]. To prepare age appropriate teeth, a patient of similar age was selected, impression of his dentition was made with irreversible hydrocolloid impression material (Zelgan 2002; Dentsply-India, Gurgaon, India) and molten modelling wax (Metrodent, Mumbai) was poured in the teeth impression area. The wax model of the teeth was obtained and invested in plaster of paris. After dewaxing, the mould was filled with tooth colored heat-curing acrylic resin and curing was done. The teeth were retrieved after the curing cycle was completed and were then trimmed, finished and polished.

A lingualized occlusal scheme was preferred over bilateral balanced occlusion scheme to reduce lateral forces and an intercuspal contact area with freedom of movement [12–14]. Thereafter the patient was under regular observation for growth changes, any discrepancy in the occlusal relationship and the fit of the tissue surfaces. The loss of the posterior teeth contact and presence of the anterior teeth contact were the recurring occlusal changes. Self-cure acrylic resin (Trevalon, Dentsply Ltd, Gurgaon, India) was added on the occlusal surfaces of the posterior teeth to produce uniform posterior tooth contact and to eliminate anterior tooth contact.

At the age of 8 years, a new set of complete denture was fabricated containing the teeth suitable to the age of the patient. The steps of fabrication were essentially the same as described earlier for the first denture (Fig. 3).

Fig. 1 Patient's profile view

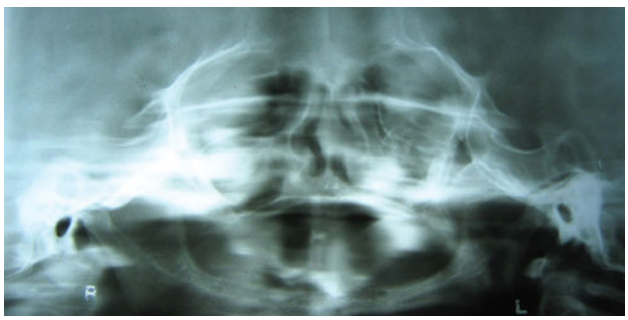

Fig. 2 Orthopantomogram showing absence of all teeth or tooth buds

Analysis of Jaw Growth

Jaw growth analysis was done with the help of cephalometric analysis and analysis of diagnostic casts. Serial lateral cephalograms were taken at 6 and 8 years of age.

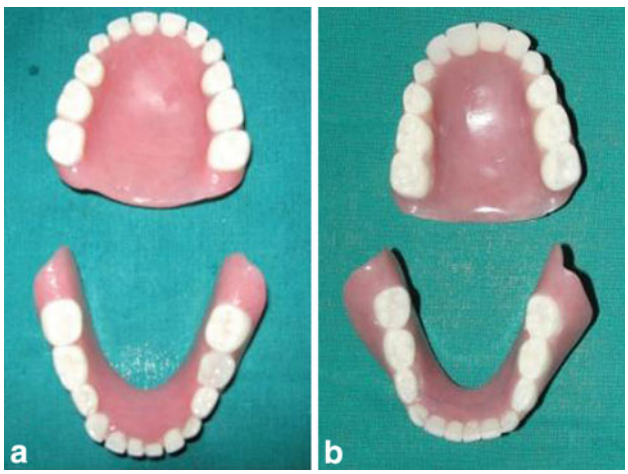

Fig. 3 Two set of complete dentures at age of 6 and 8 years

Table 1 Cephalometric analysis of the patient at 6 and 8 years of age and values compared with the normal average values reported by Lizuka 1958 and Sakamoto 1959

	Patient		Average values
	5 year 3 month	8 year 10 month	7 year 7 month
Angle (°)			
Facial angle	92.2°	92.3°	83.7°
Y-axis	48.6°	50.4°	63.8°
Mandibular plane angle	13°	13.5°	31.5°
Length (mm)			
N-ANS	46.1	44.2	47.1
ANS-ME	52.5	56.1	57.7
GO-X	60.2	64.4	63.6
ANS-PTM	40.3	42.8	46.8
GO-ME	58.7	62.4	64.5
Ratio of lengths (%)			
N-ANS/ANS-ME	87.8 %	84.13 %	81.6 %
ANS-ME/GO-X	89.43 %	87.11 %	90.7 %

These lateral cephalograms were analyzed to study the changes in jaw growth pattern and occlusal relationship. Cephalometric measurements were compared with the average values corresponding with the age as reported by Sakamoto [15] (Table 1). Angular measurements were analyzed to compare the positional relationship of craniofacial landmarks and linear measurements were done to record the change in size and compare it with average values (Fig. 4).

Serial diagnostic casts were prepared at 6 and 8 years of age. These diagnostic casts were measured for dimensional changes in arch length and width of the alveolar ridge by segmental method of cast analysis in which each half of the arch was divided into three segments by four reference points: the midline of the arch (a), canine eminence (b), posterior limit of the retromolar pad for mandibular arch or hamular notch for the maxillary arch (d) and mid-point between point (b) and point (d) (Fig. 5).

Results

Cephalograms revealed that the ANS point continued to grow forward during 6–8 years of age but it was posterior to the average values. The Go and Pog points were located posteroinferior and anterosuperior to the average respectively. The facial plane angle was greater than the average while the *y*-axis angle and the mandibular plane angle were smaller than the average.

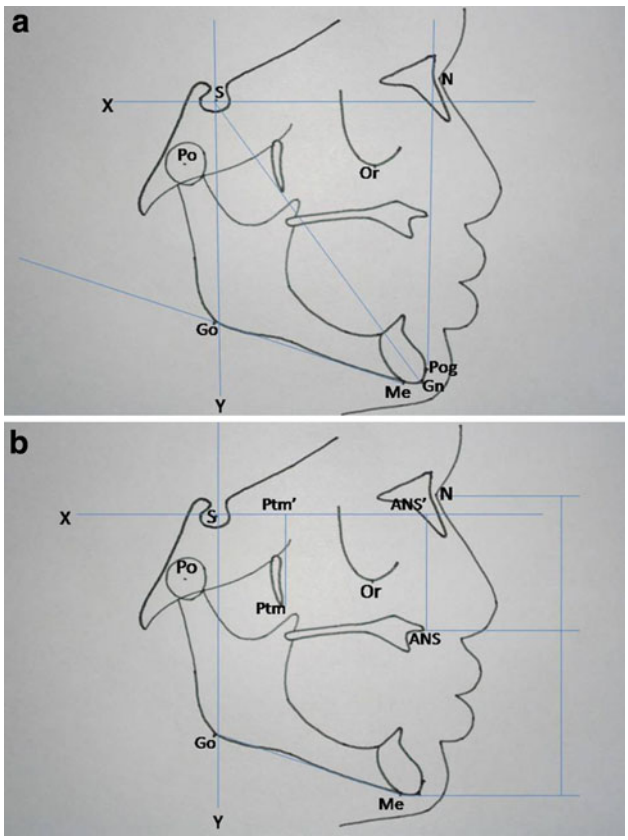

Fig. 4 Craniofacial landmarks used for cephalometric analysis. *Upper*: angular measurements; *lower*: linear measurements

The ANS-Me length was increased with the insertion of new dentures at 6 and 8 years of age although these values are slightly smaller than average values.

The Na-ANS/ANS-Me ratio which represents the ratio between the upper facial height and the lower facial height was greater than the average values, indicating a reduced lower facial height. This deficiency in vertical dimension was addressed during denture fabrication and after the denture insertion the value was almost equal to that of the average values corresponding to the age.

Cast analysis revealed that in maxilla there was an increase of 7 mm in arch length and 1 mm increase of arch width in the anterior segment, 2 mm increase in the middle segment and 3 mm increase in the posterior segment from 6 to 8 years of age. While in mandible, there was an increase of 10 mm in arch length and 1 mm increase of arch width in the anterior segment and 2 mm increase in the middle as well as posterior segments from 6 to 8 years of age. There was a little increase in alveolar ridge height in the anterior region (1 mm) but there was a considerable increase in the height of the alveolar ridge in the middle (4 mm) and the posterior region (4 mm). There was also a decrease in the flat appearance of the alveolar ridge owing

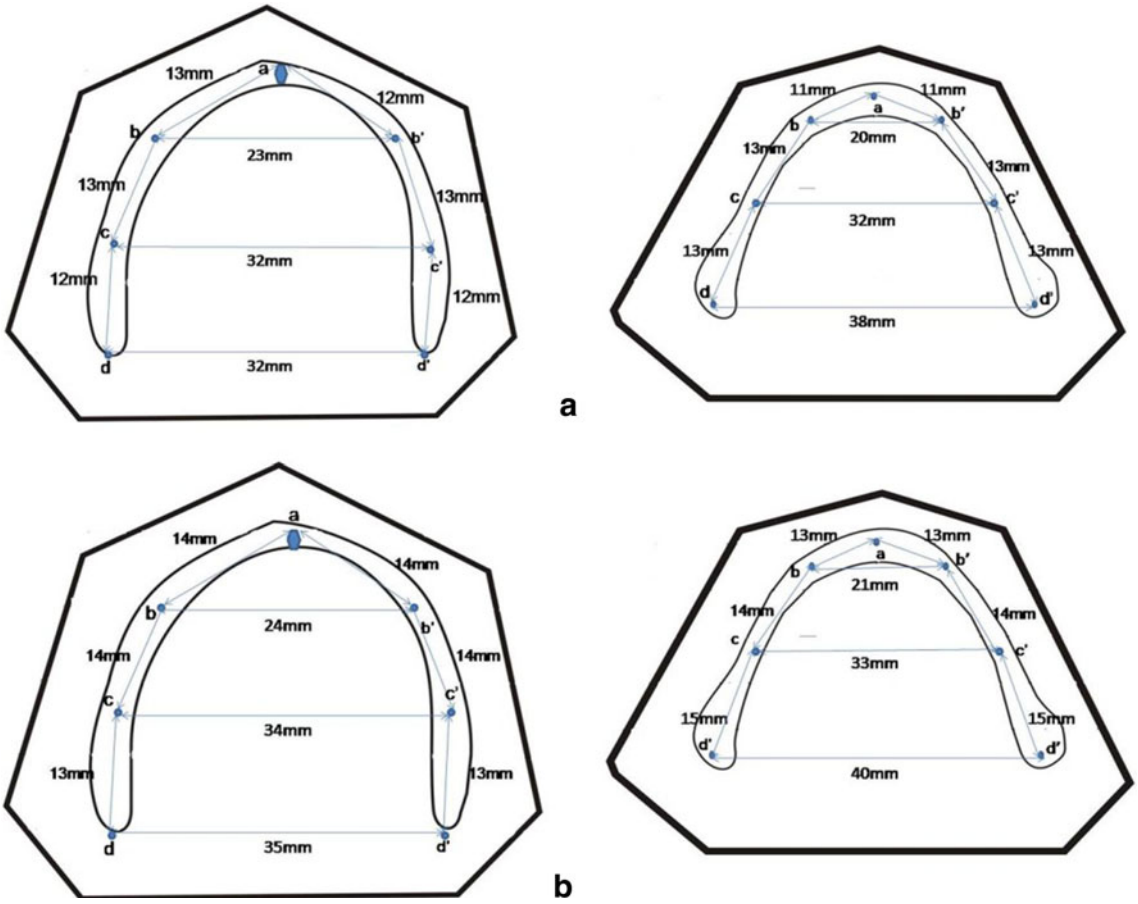

Fig. 5 Diagnostic cast analysis at age of 6 years **(a)** and 8 years **(b)**

to slight increase of the concavity of the palatal surface along with the increase in height of the alveolar ridge.

Discussion

Various treatment modalities are possible for ectodermal dysplasia patients depending upon the age, alveolar bone volume and presence of teeth. Though some preliminary studies have been performed by some researchers suggesting that endosseous implants can be placed in ectodermal dysplasia patients successfully [16], however, if placed at an early age the vertical dentoalveolar growth results in submergence of the implant and evidence are also available indicating that there is high risk of implant failure in ectodermal dysplasia patients if placed earlier than 18 years of age [17]. So, in the case discussed here, a tissue supported removable complete denture was planned because of the young age and ongoing growth and development in the patient. The influence of the complete denture on the growth and development of facial as well as alveolar structures was evaluated with the help of cephalometric and cast analysis.

As there is no medical treatment for this condition, the affected individuals with dental defects could be subjected to early dental evaluation and intervention beginning at an early age. This would not only help the child's psychological growth but also his physical growth and development. Denture fabrications at an early age lead to significant improvements in appearance, speech and masticatory functions. Such positive changes increase the self confidence of the child and aid in establishing lifelong dietary patterns.

During denture fabrication, vertical dimension was established clinically by physiologic rest position and was verified by observing facial support and with the help of cephalometric analysis of the patient and the normal individuals of same age. Thus the correct vertical dimension can not only help preventing development of class 3 malocclusion but also improving skeletal relationship during growing period of the child. Considering the individual variation in growth and development, direct comparison between the measured values and the average values can't be justified. So, the ratio between upper and lower facial height was used for evaluation of occlusal vertical dimension.

With the construction of new dentures, we were able to bring the N-ANS/ANS-ME value (89 %) quite close to the average value (82 %). This helped to ascertain that the vertical dimension was adequately established. Though ANS-ME/GO-X value (80 %) was smaller than the average value (90 %), it doesn't mean a lack of vertical dimension, but it might be influenced by greater value of GO-X in this case. This was also supported by Sarnart et al. [5] who in their longitudinal study on craniofacial growth in patients with anodontia through serial cephalometric measurements obtained from individual cases, described the growth of facial structures and jaws as lying within the lower range of normal and they concluded that the absence of teeth did not affect the growth of the jaws.

In this case, the cephalogram showed a marked restriction of forward growth at the ANS point during 6–8 years of age which was also reported by Sarnart et al. [5] in their study. There was a little change from average in the anteroposterior length of mandibular body and the height of mandibular ramus. So, the maxillary growth was reduced but there was no significant change in the mandibular growth. In a study performed by Tocchini et al. [10], it was found that the forward growth of maxilla was reduced in case of anodontia but it was difficult to determine whether this decrease in growth was due to absence of teeth or due to the effect of prosthetic replacement of the teeth. Hamano et al. [18] in their study on craniofacial growth in anodontia cases with anhidrotic ectodermal dysplasia concluded that the lack of forward growth of maxilla was due to the interplay between the absence of teeth and the atrophic rhinitis.

In our case, the increase in arch length was greater than in arch width. This was also supported by the studies performed by Shirakawa et al. and Tocchini et al. [10] independently. Shirakawa et al. measured the alveolar ridge arch on the study casts during the period of 3 years 2 months to 5 years 11 months of age and reported that there was an increase of 5 mm in arch length and 6 mm arch width in the maxilla and 12 mm arch length and 6 mm arch width in the mandible. There are some variations in the results of increase in arch length and width between our case and that of Shirakawa et al. which may be due to different ages of the patients.

In a case report of anodontia, reported by Shaw [7], underextension of dentures and posterior open bite were recurring problems which he attributed to the growth of the jaws. In this case also the lack posterior contact was managed by adding autopolymerizing acrylic resin to the occlusal surface of the posterior denture teeth. We support that, use of acrylic teeth and autopolymerizing acrylic resin are suitable choice for coping the growth changes in such poor patients. Periodic recall examination and the necessary adjustments are very important to address the growth changes occurring in a child.

Conclusion

Within its limitation, this preliminary study concluded that the absence of teeth does not affect the growth of the jaws and probably the denture flange did not arrest the jaw growth, rather it improved the masticatory function by providing good denture stability and retention.

The presented study is limited for no. of subjects and the time period chosen for the observation for any confirmatory conclusions. In future, this has to be followed up at a later date, with more subjects and for a minimum time period of 5–10 years for more confirmatory and substantial results.

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