Dimensional Changes in Dental Arches after Complete Dentures Rehabilitation of a Patient with Hypohidrotic Ectodermal Dysplasia: A Case Report with 18-Year Follow-Up

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Background: This case report presents the dimensional changes in dental arches in a patient with hypohidrotic ectodermal dysplasia (HED) after complete denture rehabilitation, with an 18-year follow-up period. Case report: The patient had complete anodontia and was successfully rehabilitated with conventional complete dentures at 3, 4, 5, 7, 9, 12, 16, and 21 years of age. Each successive denture was larger and contained more and larger teeth so as to accommodate for the increase in the size of the developing jaw. A series of diagnostic casts were used to measure the dimensional changes in the arch length and width of the alveolar ridge. Cast analysis revealed that there was an increase in arch length and width in both the maxilla and mandible over time. Cephalometric analysis of craniofacial development was performed at 21 years of age, and suggested protrusion of the maxilla and mandible. Conclusions: The absence of teeth due to HED did not affect the dimensional changes in dental arches after complete denture rehabilitation from childhood to adulthood. The prosthetic treatment improved the patient's social integration and enabled the development of normal dietary habits, speech, and facial esthetics, which in turn led to improved quality of life.

Keywords: ectodermal dysplasia, complete anodontia, complete dentures, cast analysis

INTRODUCTION

Ectodermal dysplasia (ED) is a congenital hereditary disorder, which presents an extensive and complex number of disturbances, and is defined by the abnormal development and/or homeostasis of two or more structures derived from the ectoderm of the developing embryo.[1] One or more of the following tissues are generally impaired: hair (hypotrichosis, partial or total alopecia), nails (dystrophic, hypertrophic, abnormally keratinized), teeth (abnormal or absent), or sweat glands (hypoplastic or aplastic).[2,3]

ED represents a large group comprising more than 170 different clinical conditions.[4] Several ED classification systems have been proposed based on their molecular defects and associated pathways.[2,5,6] Recently, a new classification system for ED has been suggested, based on both clinical and molecular information, as genetic variations influence the phenotypic features.[1]

In general, ED can be classified into two major types according to the number and functionality of the sweat glands: hidrotic (sweat glands are normal) and anhidrotic or hypohidrotic (sweat glands are absent or sparse).[6,7] Dentition and hair are affected similarly in both types, but the hereditary patterns and nail and sweat gland manifestations tend to differ. Although ED is considered a rare condition, the reported prevalence varies widely in the literature.[1] It has been suggested that hypohidrotic ED may be more common than previously described.[8]

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Hypohidrotic ectodermal dysplasia (HED), also called the Christ-Siemens-Touraine Syndrome, is the most common type. It is a non-progressive disease, which presents at birth and is transmitted as an X-linked recessive trait, so the manifestation is predominantly found in men.[6,7,9,10] The typical HED form is characterized by the triad of dysplasia of the sweat glands (dyshidrosis), sparseness of hair (hypotrichosis), and teeth abnormality, with characteristic faces.[7,9,11]

Hypodontia or anodontia is one of the most remarkable characteristics of ED and leads to an atrophy of the alveolar bone;[9,12-14] therefore, it is important to commence prosthodontic rehabilitation as early as possible.[14] The complete absence of the primary and/or permanent dentition is a rare condition in ED,[9,12,15,16] and it adversely impacts on eating, speaking and esthetics, as well as self-esteem in young children.[17] In this context, complete dentures are a simple, inexpensive, and reversible option to provide esthetic, functional, psychological, and social benefits for children with ED.[12-14,17-19] A systematic review concluded that prosthodontic rehabilitation should start in early childhood and needs to be revised in accordance with the patient's growth. Treatment should be carried out by a multidisciplinary team to address the different and variable demands at different age groups.[20]

The literature has demonstrated different options regarding oral rehabilitation in children with ED, such as removable partial or complete dentures, implant-supported prothesis, or in combination.[20] The treatment plan should be based on the child´s age and each patient´s individual needs.[20] As described in the literature, the use of removable complete dentures is the most frequent treatment modality in children with ED.[14,16,18,21-24]

To the best of our knowledge, no previous case report has evaluated the effects of complete dentures on the jaw development of anodontic individuals with HED over an extended follow-up period during prosthodontic rehabilitation. Thus, the present study aimed to report a case of a patient with HED and demonstrate the dimensional changes in dental arches after complete denture rehabilitation, over an 18-year follow-up period, from childhood to adulthood.

Clinical Report

A 13-month-old male child was referred to the Pediatric Dentistry Dental Clinic at a Public University, Brazil. His mother reported tooth eruption delays and feeding difficulties. Prior to the treatment of the patient, his parents signed an informed consent form.

During anamnesis, the mother informed that right after birth the child presented a medical condition of severe hyperthermia and dry skin, leading to a neonatal diagnosis of lamellar ichthyosis. Based on the clinical findings of hypohidrosis, hypotrichosis, and saddle nose (Figure 1A), the parents were instructed to consider genetic mapping, which confirmed the diagnosis of X-linked recessive hypohidrotic ectodermal dysplasia (HED) or Christ-Siemens-Touraine Syndrome at the age of sixteen months. At that time, the mother reported that a previous son had passed away after birth due to hyperthermia without any defined cause. After HED was diagnosed, the child managed by a multidisciplinary team, and management included dermatological and endocrinological counseling. The child was very introspective, and he was also monitored by a dental professional in order to establish collaborative behaviors for future dental treatment.

Approximately 2 years later, the child returned to an oral evaluation. He presented with a total absence of tooth eruption clinically. Radiological examination revealed the absence of all teeth or tooth buds (Figure 1B). Intraoral examination revealed complete anodontia of the primary dentition, thin alveolar ridges, and reduced vertical bone height (Figure 2A), resulting in a lower vertical dimension of the face and prominent lips (Figures 2B). Among the treatment modalities available, the best treatment plan option consisted of removable complete dentures to provide better quality of life by improving appearance, mastication, and speech.

At the age of 3 years, the child received his first set of complete dentures (Figure 3A), which provided a corrected vertical dimension of the face and enabled proper lip sealing (Figure 3B). The impression was made with a silicone-based impression material (Zhermack; Badia Polesine, Italy). An occlusal plane using modeling wax was obtained to define the correct midline, smile line, vertical dimension of occlusion, centric relationship position, lips support to correct labial contour of the face, and free-way space. An age-personalized tooth set was fabricated and mounted following the occlusion references obtained earlier. Phonetic and esthetic properties were also evaluated, and stability was confirmed. After prosthesis acrylization, balanced bilateral occlusal adjustments were performed. Internal relief was obtained and a resilient tissue reconditioning material (Coe-Soft; GC América, USA) was used for better adaptation to the thin and resilient alveolar edges. During the first week of use, parents and the child were instructed about oral care, correct denture maintenance procedures, and adequate soft diet for better accommodation. Moreover, advice was given

Figure 1. (A) Patient`s facial aspect. (B) Panoramic x-ray showing absence of all teeth and tooth buds.

Figure 2. (A) Intraoral examination showing complete anodontia of the primary dentition. (B) The patient's profile showing a reduced vertical dimension and everted lips.

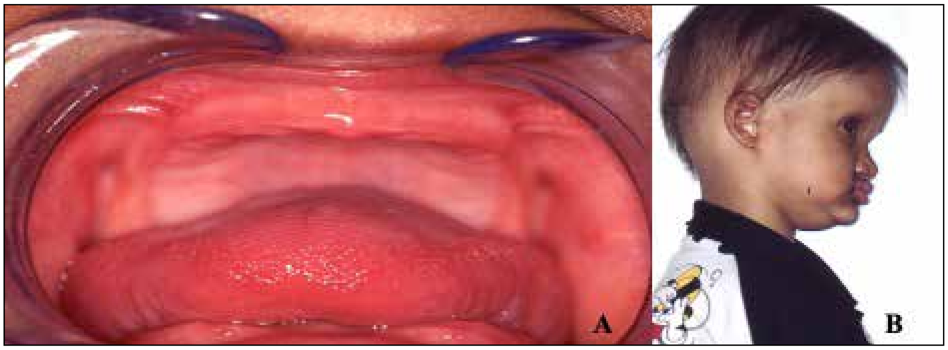


Figure 3. (A) The first set of age-appropriate complete dentures installed. (B) Patient`s profile shows harmonious proportion of the face and good lip seal.

for the child to remove the dentures at night to promote healing of the oral soft tissues.

During the first month of oral rehabilitation, the child was monitored every week and seemed to have adapted well. Later, the child was followed-up every six months, or earlier, if any pain or discomfort was noted.

As part of the multidisciplinary management approach, the young patient was also referred to speech therapy and dental care, to assist with phonation development and appropriate feeding movements when using complete dentures. Dermatology and endocrinology management were also indicated, as the absence of sweat glands and calcium deficiency (osteopenia) were part of the patient's condition.

After 12 months of using the complete dentures, they were replaced due to growth of the jaw bones. The first set of dentures was used as trays. Thereafter, the patient was regularly observed for growth changes and any discrepancies in the occlusal relationship. During the 18-year follow-up period, from childhood to adulthood, eight sets of complete dentures were designed and inserted at the ages of 3, 4, 5, 7, 9, 12, 16, and 21 years (Figure 4). Age-appropriate teeth anatomy was always respected. Every new denture was larger and contained more and larger teeth, to accompany the increased in jaw size. The steps of fabrication for each denture were essentially the same as described earlier for the first set of dentures.

Serial diagnostic casts were prepared at 3, 4, 5, 7, 9, 12, 16, and 21 years of age. A precision silicone-based impression material (Die Silicone; Voco, Germany) was used to copy and transfer the anatomical characteristics of each set of complete dentures. These diagnostic casts were used to measure the dimensional changes in arch length and width of the alveolar ridge. Measurements were taken using the segmental method of cast analysis, in which each half of the arch was divided into three segments by four reference points: (a) the midline of the arch, (b) canine eminence, (c) posterior limit of the retromolar pad for mandibular arch or hamular notch for the maxillary arch, and (d) the mid-point between points b and d.[25]

Table 1 shows the changes in the arch length and width of the maxillary and mandibular alveolar ridges of all the diagnostic casts. In general, cast analysis revealed that there was an increase in arch length and width in both the maxilla and mandible. Figure 5A shows the diagnostic cast analysis of the maxillary arch at ages 3, 7, 12, and 21 years. Cast analysis showed a significant increase in the maxilla arch length of 9 mm from 3 to 7 years, 18 mm from 3 to 12 years, and 37 mm from 3 to 21 years. In relation to arch width, in the anterior, middle and posterior segments there was an increase of 1.5 mm, 1.5 mm and 6 mm, respectively, from 3 to 7 years; of 4 mm, 6 mm and 10 mm, respectively, from 3 to 12 years; and of 6 mm, 12 mm and 12 mm, respectively, from 3 to 21 years.

Figure 5B shows the diagnostic cast analysis of the mandibular arch at ages 3, 7, 12, and 21 years. Regarding arch length, there was an increase of 14 mm from 3 to 7 years, of 24 mm from 3 to 12 years, and of 44 mm from 3 to 21 years. For arch width, in the anterior, middle and posterior segments there was an increase of 3 mm, 3 mm and 6 mm, respectively, from 3 to 7 years; of 3 mm, 6 mm and 6 mm, respectively, from 3 to 12 years; and of 5 mm, 11 mm and 8 mm, respectively, from 3 to 21 years.

Figure 4. Eight sets of complete dentures according to the ages evaluated (in years).

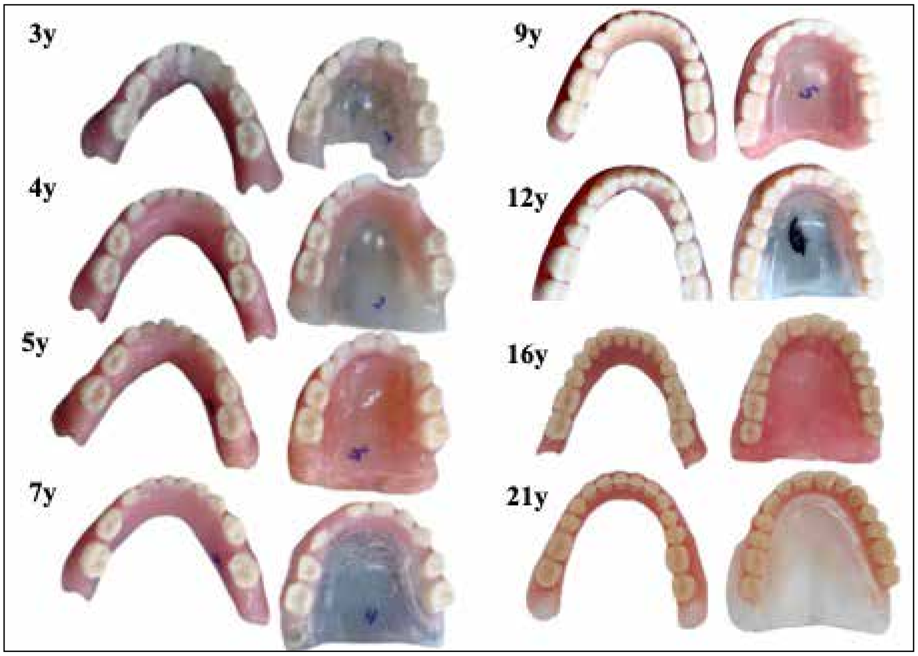
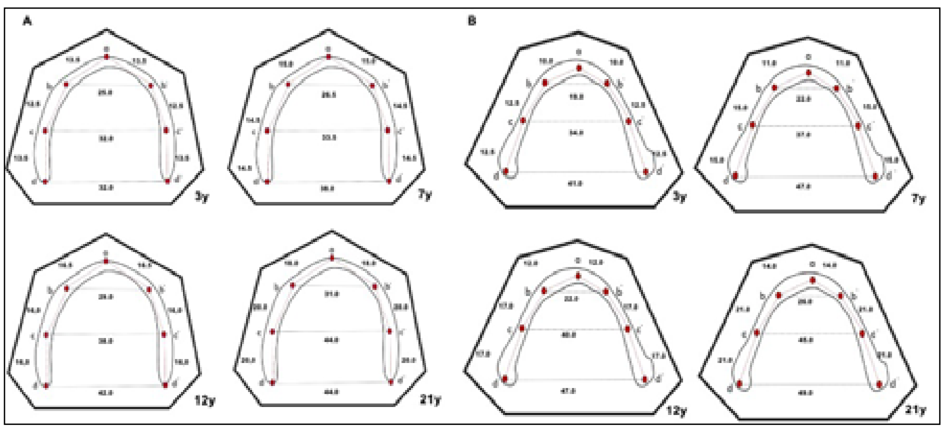


Figure 5. (A) Maxilla diagnostic cast analysis at age of 3, 7, 12 and 21 years. (B) Mandible diagnostic cast analysis at age of 3, 7, 12 and 21 years.



The cephalometric analysis was performed only at 21 years of age. The lengths of the maxilla (Co-A) and mandible (Co-Gn) were calculated and compared with the normal average values for age and sex derived from the McNamara Standards (Table 2). Craniofacial landmarks in Figure 6A show the average values of maxilla (87.54 mm) and mandible (122.80 mm) within the normal standard variation. The patient's anterior facial height (Ena-Me) (57.89 mm) also showed a value within the normal standard variation. The anterior-posterior orientation of the maxilla and mandible relative to the cranial base indicated a protrusion of 4.04 mm and 21.92 mm, respectively, as observed from the patient's lateral profile (Figure 6B).

Over the years, the possibility of rehabilitating the patient with an implant-supported prosthesis was discussed with the parents. However, the patient has always been averse to dental surgery procedures till date. The patient was comfortable using removable dentures and had a good quality of life. The most recent recall was in September 2020, and the patient, at 25 years of age, is still wearing the set of dentures issued at 21 years of age, with no complaints regarding prosthesis stability (Figure 7A, 7B).

Table 1. Cast analysis of the patient at different ages (in years) showing measurements of the dimensional changes in arch length and width (in mm).

Arch	Reference Points	Cast Analysis							
		3y	4y	5y	7y	9y	12y	16y	21y
Maxillary	midline of the arch/ canine eminence (a-b)	13.5	13.5	15.0	15.0	15.0	16.5	17.0	18.0
	midline of the arch/ canine eminence (a-b')	13.5	13.5	15.0	15.0	15.0	16.5	17.0	18.0
	canine eminence/ mid-point (b-c)	12.5	13.5	14.5	14.5	16.0	16.0	17.0	20.0
	canine eminence/ mid-point (b'-c')	12.5	13.5	14.5	14.5	16.0	16.0	17.0	20.0
	mid-point/ posterior limit of the hamular notch (c-d)	13.5	13.5	14.5	14.5	16.0	16.0	17.0	20.0
	mid-point/ posterior limit of the hamular notch (c'-d')	13.5	13.5	14.5	14.5	16.0	16.0	17.0	20.0
	anterior segment (b-b´)	25.0	25.0	26.5	26.5	26.5	29.0	31.0	31.0
	middle segment (c-c´)	32.0	33.5	33.5	33.5	38.0	38.0	42.0	44.0
	posterior segment (d-d´)	32.0	35.5	35.5	38.0	42.0	42.0	43.0	44.0

Table 1. Cast analysis of the patient at different ages (in years) showing measurements of the dimensional changes in arch length and width (in mm) (continued).

Arch	Reference Points	Cast Analysis							
		3y	4y	5y	7y	9y	12y	16y	21y
Mandibular	midline of the arch/ canine eminence (a-b)	10.0	11.0	11.0	11.0	11.0	12.0	14.0	14.0
	midline of the arch/ canine eminence (a-b’)	10.0	11.0	11.0	11.0	11.0	12.0	14.0	14.0
	canine eminence/ mid-point (b-c)	12.5	14.0	14.0	15.0	17.0	17.0	18.0	21.0
	canine eminence/ mid-point (b’-c’)	12.5	14.0	14.0	15.0	17.0	17.0	18.0	21.0
	mid-point/ posterior limit of the retromolar pad (c-d)	12.5	14.0	14.0	15.0	17.0	17.0	18.0	21.0
	mid-point/ posterior limit of the retromolar pad (c’-d’)	12.5	14.0	14.0	15.0	17.0	17.0	18.0	21.0
	anterior segment (b-b´)	19.0	22.0	22.0	22.0	22.0	22.0	26.0	26.0
	middle segment (c-c´)	34.0	35.0	36.0	37.0	40.0	40.0	42.0	45.0
	posterior segment (d-d´)	41.0	43.0	43.0	47.0	47.0	47.0	48.0	49.0

Table 2. Cephalometric measurements of the patient at 21 years of age compared with the normal average values for age and sex, derived from the McNamara Analysis.

Structural Relations	Means	Normal Standard	Deviation	Interpretation
Maxilla X Cranial Base				
A-Na Perp	5.14mm	1.1 ±2.7	4.04	Maxillary protrusion
Mandible X Cranial Base				
Pog-Na Perp	21.62mm	-0.3 ±3.8	21.92	Mandibular protrusion
Maxilla X Mandible				
Co-Gn	122.80mm	109-112 ±6.8	17.08	————-
Co-A	87.54mm	99.8 ±6.0	-12.26	————-
Ena-Me	57.89mm	61-63 ±5.0	0.76	————-

Figure 6. (A) Craniofacial landmarks (McNamara Standards) used for cephalometric analysis at age of 21 years. (B) Patient`s facial profile using the last set of complete dentures at age of 21 years.

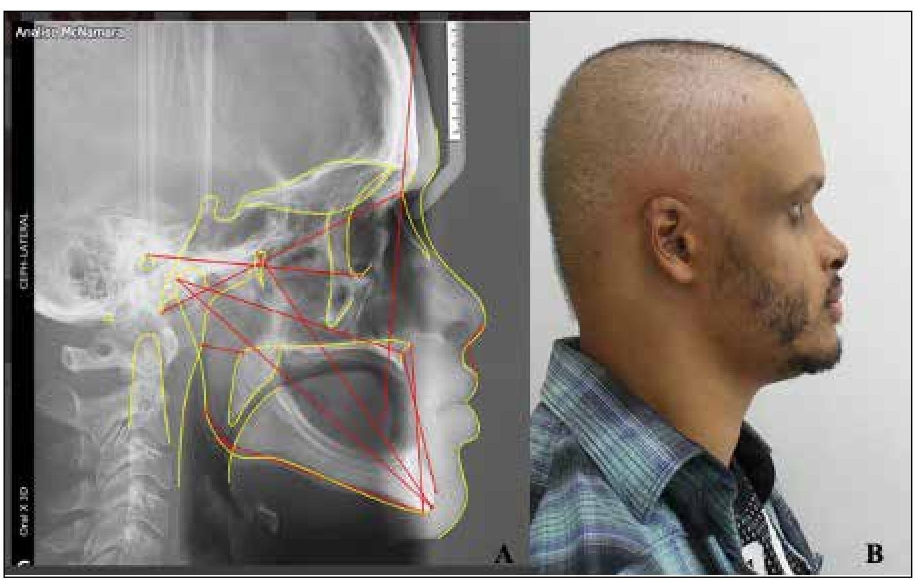


Figure 7. (A) The set of dentures issued at 21 years of age.
(B) Patient's facial aspect showing comfortable using the set of removable dentures.

DISCUSSION

Regarding the treatment modalities for ED patients, removable complete dentures are the most successful in complete anodontia cases among children.[14,16,18,21-24] However, to date, the literature has not demonstrated the chronic effects of complete denture rehabilitation on jaw growth and development through a longer follow-up period.

The present study reports a patient with anodontia followed up for a period of 18 years during prosthetic management, including the critical period of normal eruption of the permanent dentition. To our knowledge, there is no other similar study reported with a follow-up from childhood to adulthood. A few articles published in the literature reported a follow-up period varying from 4 to 12 years (from childhood to adolescence) during prosthodontic rehabilitation.[9,25,26]

In the present case report, the use of complete dentures from childhood to adulthood did not inhibit the jaw growth and development, as confirmed in the cast analysis and also previously demonstrated in the literature.[9,25,26] The increase in the size of the dentures as well as the increase in the size and number of teeth can be confirmed in the eight sets of full upper and lower dentures made between 3 to 21 years of age, and also as shown by Sarnat et al.[26]

It is known that bone growth is based in two fundamental physiologic processes: modeling (apposition and resorption) and remodeling. The following basic phenomena are involved in the growth mechanisms: conversion of cartilage (synchondroses, nasal septal cartilage, condylar cartilage), sutural deposition, and periosteal remodeling.[27] According to the theory of Moss[28] the determination of bone and cartilaginous growth is a response to the intrinsic growth of associated structures, called functional matrix, noting that the genetic code for skeletal growth is outside bone growth. Each component of this functional matrix performs a necessary function such as breathing, chewing, speaking, while skeletal tissues support and protect these associated functional matrices. Skeletal growth occurs in response to soft tissue growth, such as muscle function in muscle insertions. In the present case, the correct use of complete dentures from an early age, allowed patient´s functions of chewing, speaking, breathing, and possible influenced the bone growth and development of the system, regardless of the presence of teeth in the alveolus. Periodic follow-ups and multidisciplinary approach were important for the decision to change the complete dentures from time to time, suggesting that this type of oral rehabilitation did not prevent the arches to grow.

Looking for our results, when comparing the first set of complete dentures at 3 years of age with the last set, which was replaced at 21 years of age, there was a 37 mm increase in maxillary length and 44 mm in mandibular length. In our patient, the increase in arch length was greater than the arch width as described in previous studies.[9,25,29,30] It should be mentioned that the reason for a lack of forward growth of the maxilla is unclear in the literature but may possibly be due to the absence of teeth,[30,31] the effect of prosthetic replacement,[30] and/or atrophic rhinitis.[31]

According to Thilander[32] significant changes of the dental arches occur from the primary until the adult period, with individual variations in patients with normal occlusion between the ages of 5 and 31 years. The presence of the teeth influences the growth in width, length and depth of the arches. A continuous increase of palatal height up to adulthood seems to be a result of a slow continuous eruption of the teeth. However, this cannot be extrapolated to the cases of patients with complete anodontia because of the non-development of alveolus. Moreover, in the absence of teeth, width, length, and depth measurements follow reference points of anatomical structures different from the cases of normal dentition.

Previous studies used cephalometric analysis as an aid to clinically locate the physiologic rest position and by face support to determine the occlusal vertical dimension during complete denture construction.[9,19,25] A limitation of our study is that cephalometric analysis was not performed during all rehabilitation appointments. As such, the comparison of these measurements from childhood to adulthood was not possible. This case report is different from previous studies with shorter follow-up, serial cephalometric analysis over a longer follow-up period would expose the young patient to the hazardous effects of radiation. Moreover, the radiographic set-up was not available in the dental clinic.

According to Alhaji et al,[33] the establishment of vertical dimension by facial esthetic appearance can be utilized as a guide in young or middle-aged patients. Clinical judgment, dentist and patient preference, and individualized patient treatment still play an important role in the assessment of the vertical dimension. The complete dentures resulted in normal facial proportions between the upper and lower facial heights, as verified by serial photographs of the patient, taken at periodic intervals. The dentures also improved masticatory functions, speech, and the overall well-being of the patient, which was described by Bhalla et al.[25] Prosthodontic treatment can result in psychological and social benefits for young patients.[13] Oral rehabilitation should be initiated as early as possible and the treatment provided needs special attention and care to ensure normal jaw growth and prevent the occurrence of adverse factors. Regular recalls are necessary for timely adjustments and for replacement of the dentures when necessary, so that a harmonious facial appearance can be maintained.[9]

CONCLUSION

Our findings concluded that the absence of teeth due to HED did not affect the dimensional changes in dental arches after complete denture rehabilitation over an 18-year follow-up period, from childhood to adulthood. The prosthetic treatment also improved the patient's social integration and enabled the development of normal dietary habits, speech, and facial esthetics, which in turn led to an improved quality of life.

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Conflict of interest:

The authors declare no conflicts of interest.

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