Review Article

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CURRENT TRENDS IN PROSTHODONTICS FOR CHILDREN WITH ECTODERMAL DYSPLASIA: A CRITICAL LITERATURE REVIEW

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ABSTRACT:

Ectodermal Dysplasias are congenital diseases that affect the population in a rate of 1:100,000 live births. The condition is portrayed by the defective development of structures derived from the ectoderm. Oral manifestations of Ectodermal Dysplasia include multiple missing primary teeth, dysmorphosis of primary incisors, taurodontism of second primary molars and reduced number of permanent teeth. The views on a prospective protocol aiming for the prosthetic treatment of children with Ectodermal Dysplasia are very divergent. There are numerous strategies for the management of the many oral manifestations of Ectodermal Dysplasias, they may include removable or fixed partial dentures, dental supported over dentures, implant supported prostheses, and orthodontic care. The purpose of this study is to illustrate, through a critical literature review, the current trends in oral rehabilitation of children with Ectodermal Dysplasia.

Keywords: Ectodermal Dysplasia; Pediatric Dentistry; Rehabilitation; Anodontia; Child.

INTRODUCTION:

Ectodermal Dysplasias are congenital diseases portrayed by the defective development of structures derived from the ectoderm; their manifestations are diverse, with over 160 distinct clinical and genetic expressions.[1] It is estimated that the condition affects population in a rate of 1:100,000 live births.[2]

Oral manifestations of Ectodermal Dysplasias may include a decrease on number of salivary glands[3], multiple missing primary teeth, dysmorphosis of primary incisors, taurodontism of second primary molars and reduced number of permanent teeth. [4] Physiological issues may arise if oligodontia is present; these problems can range from underdevelopment of alveolar ridge to underdevelopment of the maxillofacial skeleton.[5] Due to missing teeth, patients may face speech and mastication problems, unaesthetic appearance and poor self image.[6] Oligodontia or anodontia can also lead to reduced vertical dimension, prominent chin and class III inter-maxillary relationship; therefore rehabilitation treatment should begin as soon as possible. [7]

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The views on a prospective protocol aiming for the prosthetic treatment of children with Ectodermal Dysplasia are very divergent. There are numerous strategies for the management of the many oral manifestations in children with Ectodermal Dysplasia, they may include removable or fixed partial dentures, complete over dentures, implant supported prostheses, and orthodontic care. [4, 8] The enormous diversity of prosthetic rehabilitation treatments to be considered for children with Ectodermal Dysplasias may lead dental surgeons to find it difficult to plan the therapy of these patients. Therefore the aim of this study was to determine which prosthetic resources were chosen for the oral rehabilitation of children (2-15 years old) with Ectodermal Dysplasia, and to determine the outcomes and/or authors considerations of each prosthetic method adopted.

MATERIALS AND METHODS

For the development of this critical literature review, the databases Pubmed, Medline and Google Scholar were used as a research tools. The researched articles ranged from the years 2010-2016, and were written in English. A total of 33 articles were selected for this study. The keywords adopted for research were: “Ectodermal Dysplasia”; “Pediatric Dentistry”; “Rehabilitation”; “Anodontia”; “Child”.

RESULTS AND DISCUSSION

The importance of oral rehabilitation treatments for children with Ectodermal Dysplasia is well documented in the literature researched, with benefits including the improvement of the child’s psychosocial interaction, mastication, speech and esthetics.[9] This critical literature review demonstrates that there are a myriad of valid approaches for the prosthetic rehabilitation treatment of children with ectodermal dysplasia. Different prosthetic resources such as removable partial dentures, fixed partial dentures and overdentures, must be adopted individually or combined, and in appropriate time, in order to achieve satisfactory esthetic and functional results. In order to illustrate the information attained by this review, the interventions performed during the patient’s childhood in each study and the treatment outcomes and/or authors considerations are presented chronologically on Tables 1-6.

The findings of this critical review show that removable prostheses are the most conventional oral rehabilitation treatment for patients with ectodermal dysplasia [3-7, 10-29] agreeing with other studies.[30] This tendency may be explained by the fact that conventional treatment is more economical and non-invasive and can be adopted during the child’s growth period initiating as early as age 3.[11] An early prosthetic treatment with removable prostheses can also aid in the maintenance of the vertical dimension and prevent atrophy and further absorption of alveolar ridges.[20]

Although the treatment of Ectodermal Dysplasia children with conventional partial and total removable dentures was the most common in the literature researched with successful outcomes, other types of prosthetic treatments were presented. On their study, Montanari *et al.* [9] presented a study where a new concept of removable dentures was proposed. On their work, a two-way expander screw was installed in mandibular removable prostheses and a three-way expansion screw was installed in maxillary removable prostheses, allowing mandibular growth on the transversal plane without locking it, and maxillary growth on transversal and sagittal planes.

Another prosthetic resource for the treatment of children with Ectodermal Dysplasia vastly presented in the literature was the implant supported overdentures. [5, 10, 12, 31, 32] This course of treatment is indicated for patients with severe hypodontia, where implants can help stabilize partial and complete removable dentures, aiding in the regain of muscular activity, function and adequate growth.[1] There is no consensus in the literature regarding the ideal age for the placement of implants.[2] Filius *et al.*[5] believe implants can be placed in the intraforaminal region of the mandible by age 6. Other authors suggest that implant supported prostheses should be placed in children after the age of 12 [7] or that they should only be placed after completion of craniofacial growth. [6] When location is concerned, implants placed in the intraforaminal region of the mandible, have a better prognosis. [1]

An alternative to the regular implants was described by Mello *et al.* [30] and Sfeir *et al.* [2], where mini-implants, used in orthodontics, were installed in the anterior region of the mandible, for retention of removable prostheses, in patients with Ectodermal Dysplasia. On their study, Sfeir *et al.* [2] also utilized mini-implants with fixed prostheses to replace missing front teeth in the maxilla and mandible. These authors believe that the use of mini-implants can reduce the number of surgeries necessary in conventional implants and avoid complex bone augmentation procedures. [2]

One important tool for the prosthetic rehabilitation of children with Ectodermal Dysplasia is orthodontic care. This review shows that only 21.2 % of the cases presented in our review utilized orthodontics as a tool for the prosthetic treatment of patients with Ectodermal Dysplasia. [3, 9, 14, 23, 28, 31, 33] This may occur due to the numerous absent teeth on many of the patients with the condition. On their study, Kalaskar e Kalaskar [8] stress that orthodontic care plays an important role in the prosthetic treatment of children with Ectodermal dysplasia since it can help improve the facial profile and also align the permanent teeth of those patients. Fraiz *et al.* [14] presented a case where functional orthopedic appliances were combined with removable dentures for the treatment of a young

patient with Ectodermal Dysplasia. This multidisciplinary approach allowed changes in the patient's bone development throughout his growth, including improvement of mandible positioning, growth of the middle and lower thirds of the face, and better inclination of maxillary incisors, preparing the patient for future interventions. [14]

Treatment planning for patients with Ectodermal Dysplasia depends on each case and should consider the long term outcome, reassessments and treatment revisions. [33]

CONCLUSION

This critical literature review reported the current resources chosen for the prosthetic rehabilitation of these patients. These resources consisted in: Restorations with composite resin for conoid shaped teeth; Fixed crowns; Strip crowns with composite restoration; Mandibular and maxillary partial removable dentures; Mandibular conventional complete dentures; Mandibular and Maxillary prostheses with expander screws; Maxillary flexible removable partial dentures; Dental supported mandibular and maxillary overdentures; Screw-retained mandibular bridge; Half screw-retained maxillary bridges; Implant supported overdentures with implants placed on the anterior region of the mandible; Implants at the region of the maxillary first pre-molars; Mini-implant supported mandibular prosthesis with mini implants installed in the intraforaminal region and Mini-implants in the anterior region of the maxilla. Orthodontic treatment was also adopted as a treatment resource, with lingual envelope and mucosa supported functional orthopedic appliances used simultaneously with prosthetic resources to attain more satisfactory long term results.

Each case of children with ectodermal dysplasia is unique, there is no standard formula for a treatment, and therefore, the prosthetic resources adopted in each case should be chosen carefully. They may be used on their own or by complementing each other and should respect the adequate timing of the patient's physical development.

In summary, the literature has shown that partial and total prostheses comprehend an adequate mean of rehabilitation for younger children with Ectodermal Dysplasia, where bone growth is in its early stages. Dental supported overdentures are a great alternative for Ectodermal Dysplasia children with partial anodontia presented by very rudimentary dental formation. Implant supported overdentures, with implants placed in the anterior portion of the mandible, are a good alternative for older children that present difficulties in retention of total and partial prosthesis on the mandibular arch. Implants in the maxillary region and the posterior region of the mandible should only be placed after children with

Ectodermal Dysplasia have completed their bone development

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TABLES:

TABLE 1. SUMMARY OF REHABILITATION TREATMENTS FOR CHILDREN WITH ECTODERMAL DYSPLASIA IN 2010 AND 2011

Authors and publishing year	Interventions performed during childhood	Outcomes and/or author's considerations
Gupta and Tyagi, 2011	The treatment of a 4 year old male consisted on removable partial dentures for the upper arch and complete removable dentures for the lower arch.	Patient's esthetics, chewing and phonetics were improved. He was very cooperative and was satisfied with the prostheses.
Shekar *et al.*, 2010	The treatment of an 8 year old female consisted on removable tissue-supported overdenture in the maxilla and conventional acrylic resin denture in the mandible.	Patient was very pleased with the esthetics; her speech was improved as well as her social interaction. The author believes that these prostheses can stimulate the alveolar ridges and may help with future treatments involving implants.

TABLE 2. SUMMARY OF REHABILITATION TREATMENTS FOR CHILDREN WITH ECTODERMAL DYSPLASIA IN 2012

Authors and publishing year	Interventions performed during childhood	Outcomes and/or author's considerations
Hekmaftar *et al.*, 2012	Case 1: Complete maxillary and mandibular overdentures on a 3 year old male. Case 2: Maxillary and mandibular removable partial prosthesis and restoration of maxillary central incisors (resin) on a 10 year old female.	Case 1: Hygiene instructions were given and follow-up consults were scheduled for every six months for adjustments or replacement of dentures. Case 2: Follow-up consults were scheduled for every six months for evaluation of mandibular development ant teeth eruption. The authors believe that this treatment can be cost effective and improve speech, function, esthetics and psychosocial condition.
Martin and Paulus, 2012	5 mandibular implants between the region of teeth 33 and 44 and autogenous bone graft on the buccal and lingual mandibular symphysis and 7 maxillary implants. Four months after placement of implants a screw-retained mandibular bridge and two half screw-retained implant supported bridges were placed on a 6 year old male.	No implants were lost. The removable jaw complete prosthesis broke itself numerous times what demonstrates the restoration of muscular function in the area. The patient's previous angular cheilitis and perioral wrinkles were resolved, speech is normal, the child is satisfied with his prostheses, has good hygiene as is motivated. Authors believe that in order to endorse this course of treatment more long term studies should be conducted.
Singer *et al.*, 2012	The 11 year old patient received 4 external connection implants between the mental foramen. 3 implants distal to the foramina, 1 on the right and 2 on the left. By age 12 a bilateral provisional prosthesis was installed in mandible. Preprosthetic orthodontic treatment restricted to the maxillary labial segment was completed by 14 years of age when a maxillary removable orthodontic retainer was installed. Two maxillary 3-unit metal ceramic partial fixed dental prostheses and a maxillary tooth supported removable dental prosthesis and 2 mandibular metal ceramic partial fixed dental prostheses supported by implants were placed by age 16.	After 20 years of the last contact with the patient he was reassessed. He had no lost teeth and his prostheses were functioning. On clinical exam all implants placed appeared to be viable. Plaque control was poor, the patient had calculus and gingivitis and periimplantitis was noticed around the four anterior implants. The authors believe that based on this case report, implants can be safely placed during active growth both in the anterior and posterior regions of the mandible. The demand for proper hygiene and regular follow-ups is emphasized.

Bala *et al.*, 2012	Complete removable maxillary and mandibular dentures on a 7 year old male.	The maxillary denture presented good retention, esthetics was improved. Patient and parents received instructions regarding the prostheses. The patient was very satisfied with the results. Future modification or remaking of dentures is expected with growth.
Montanari *et al.*, 2012	The authors described the treatment of 20 patients ages 2-7. Seventeen patients received removable prostheses with expander screws. Two patients received orthodontic treatment with fixed prostheses. One patient received removable prostheses with expander screws and fixed crowns.	In order to allow maxillary and mandible growth, expander screws (three way on maxilla and two-way on mandible) were placed on dentures after 2 or 3 months of denture insertion. Parents received instructions to turn the screws every two weeks. The prosthesis were well accepted by the patients and their families and improved masticatory function, esthetics and speech. The amount of natural teeth was proportional to the acceptance of the removable prostheses and the fixed prostheses showed higher acceptance than the removable prostheses.

TABLE 3. SUMMARY OF REHABILITATION TREATMENTS FOR CHILDREN WITH ECTODERMAL DYSPLASIA IN 2013

Authors and publishing year	Interventions performed during childhood	Outcomes and/or author's considerations
Trivedi and Bhatia, 2013	The 4 year old patient received a removable partial denture for the upper arch, a complete denture for the lower arch and strip crowns with composite restoration for central maxillary incisors.	The patient adjusted well to the dentures. The treatment brought satisfactory results such as better facial form and esthetics, improvement of speech and self esteem. The patient and parents received instructions regarding hygiene and after the 3 week period of more constant follow-ups, regular consults were scheduled for every 3 months.
Sadashiva *et al.*, 2013	Removable partial mandibular and maxillary dentures we installed on the 14 year old patient. By age 15 conventional Mandibular and maxillary overdentures without preparing of existing permanent were installed.	During three years after the confection of conventional overdenture with no teeth preparation, the prosthesis had been relined periodically and needed replacement, since the patient returned with complaints of poor retention. The patient resumed prosthetic rehabilitation and had implants placed on maxilla and mandible by 21 years old. The Patient's confidence on the treatment plan and her psychosocial behavior have improved throughout the treatment.
Kalaskar and Kalaskar, 2013	Maxillary and Mandibular flexible dental supported overdentures were installed on a 7 year old female.	Since skeletal maturity is not complete, this was the treatment of choice. Follow up visits were scheduled for 6 months, orientations regarding prosthesis insertion, removing and hygiene were given to the patient.
Ladda *et al.*, 2013	Removable maxillary and mandibular complete dentures were installed on an 8 year old male.	The patient showed improvements on facial profile and expression, speech and mastication. He was able to function well with the prostheses that were well retained. Follow up consults were scheduled for every 6 months.
Aydinbelge *et al.*, 2013	The 7 year old patient received two implants in the anterior mandible followed by one implant-supported mandibular overdenture and a maxillary dental supported overdenture.	The patient and parents received hygiene instructions and the patient was though how to insert and remove the prosthesis. Areas of the dentures causing discomfort were released in a subsequent consult. The patient adapted well to the prosthesis, her mastication, and speech improved. Regular follow-up consults were scheduled and future modification or remaking of dentures is expected with growth.

TABLE 4. SUMMARY OF REHABILITATION FOR CHILDREN WITH ECTODERMAL DYSPLASIA IN 2014

Authors and publishing year	Interventions performed during childhood	Outcomes and/or author's considerations
Fraiz *et al.*, 2014	Reconstruction of teeth 51 and 61 with composite resin, filing of cuspids in maxilla, temporary partial dentures for maxillary and mandibular arches were performed when the patient was 3 years old. By age 9 a replacement of dentures was needed, associated with the use of mucosa supported functional orthopedic appliances and reshaping of primary cuspids to resemble incisors.	The patient's occlusion was stable by 14 years old and he was referred to oral rehabilitation. The authors believe that treatment should be multidisciplinary, and should start at an early age, improving quality of life and preparing the patient for future interventions. The importance of mucosa supported appliances in the rehabilitation is emphasized.
Bahjat *et al.*, 2014	Total removable prostheses on Maxilla and mandible on a 3 year old male.	Only a few adjustments on the flanges of the prostheses were made. The prostheses brought psychological, Esthetic, and functional benefits to the patient.
Mopagar *et al.*, 2014	Complete maxillary and mandibular removable dentures on a 10 year old male.	Minor occlusal adjustments were made over the first month follow up; the patient had no complaints and no signs of mucosal changes or reactions. Improvements on patient's appearance, mastication self esteem and weight were observed.
Huang and Driscoll, 2014	Extraction of compromised maxillary first molars and placement of a Hawley Retainer when the patient was 10 years old. By age 13 two intra-foramen implants were installed on the mandible and one implant was installed at the region of the maxillary right pre-molar. When the patient was 15 years old two intra-foramen implants were installed on the mandible and one implant was installed at the region of the left first maxillary pre-molar. Bone grafting was needed for the maxillary implant.	Five years after the implants were placed, inflammation of the left maxillary implant occurred; since it was nonintegrated it was removed. Several years later the right maxillary premolar implants also failed. The four mandibular implants were successfully osseointegrated.
Sfeir *et al.*, 2014	Case 1: Two mini implants on the anterior region of the mandible and prosthesis with two housings on	Case 1: The implants solved the instability issue observed in the previous prosthesis. Two years

	implants, on a male age 11. Case 2: Two mini implants on the anterior region of the mandible, four mini maxillary implants to replace the four incisors on a 12 year old male. Case 3: Two mini implants on the maxilla on the region of teeth 11 and 22, two mini implants at the region of the mandibular central incisors, metal ceramic crowns teeth 31 and 41 on a 12 year old male.	later they remained well integrated. Case 2: Four years after the placement of the first implants and two year after second placement of implants, good function and esthetics were achieved. Case 3: No further considerations were made regarding that one specific case alone. The authors believe that mini-implants have an undeniable role on the enhancement of function, aesthetics and psychosocial development of children with Ectodermal Dysplasia.
Filius *et al.*, 2014	The treatment was common for all 3 cases of children age 6-8, and consisted of: two dental implants in the intraforaminal area of the mandible, temporary prostheses for 3 months, followed by conventional partial maxillary denture and implant-retained mandibular overdenture.	The esthetics was improved in all patients, no implants were lost and there was minor need for prosthetic after care. No changes in interimplant distance were reported, peri-implant bone loss was minimal. Patients were satisfied with the implant treatment. Proper oral hygiene was a slightly challenging.
Callea *et al.*, 2014	Orthodontic treatment with lingual nocturnal envelope, maxillary removable partial prosthesis and mandibular removable total prosthesis on a 6 year old male.	The return of function and facial harmony were observed. The authors emphasize that this is not a final prosthetic solution.

TABLE 5. SUMMARY OF PUBLISHED REHABILITATION TREATMENTS FOR CHILDREN WITH ECTODERMAL DYSPLASIA IN 2015

Authors and publishing year	Interventions performed during childhood	Outcomes and/or author's considerations
Mittal *et al.*, 2015	Case 1: Removable maxillary partial denture and mandibular complete denture on a 5 year old male. Case 2: Removable maxillary complete denture and mandibular implant supported overdenture with two implants placed on the anterior region of the jaw of a 9 year old male.	The authors found that the prosthetic rehabilitation may be difficult but also rewarding. The esthetics and function of overdentures supported by implants can be satisfactory for young children; therefore it should be a treatment option. Periodic recall is mentioned as necessary as well as the progression of the treatment as the child grows.
Rao and Gounder, 2015	The 11 year old patient received a maxillary flexible removal partial denture and a mandibular conventional complete denture.	The patient attended the schedule recalls with minor problems only and presented improvement on speech, social interaction, masticatory function and esthetics. The authors believe that the conventional prosthetic treatments bring many benefits to the child and should start in an early age, and that definitive prosthetic treatments such as implants should be conducted after completion of growth.
Bergendal *et al.*, 2015	The 3 year old patient received a partial maxillary removable dental prosthesis. By age 6 two Implants were installed in the anterior region of the mandible- A. Mandibular Overdenture was installed by age 7.	The implants placed at age 6 brought good function and appearance; they were not lost, although there was exposure of one of the implant treads in the buccal surface due to the small width of the mandible of the young patient. The authors recommend postponing implants for a few years or using the now available smaller diameter implants. This long term case report supports the use of implants as a viable treatment for children with X-linked Hypohidrotic Ectodermal Dysplasia.
Mello *et al.*, 2015	The procedures executed on a 9 year old patient consisted on: Restoration of Maxillary incisors with composite resin, partial removable conventional prosthesis on maxilla and mini implant supported mandibular prosthesis with implants placed on the	During a 6 year follow up there was no bone resorption and the prosthesis was well retained by the mini implants. The patient adapted well to the prosthesis, mastication, speech, self esteem and socialization were improved.

	anterior region of the jaw.	
Bharadwaj *et al.*, 2015	Partial mandibular removable denture on a 6 year old female.	The authors emphasize that follow up is required until the child has reached adult dentition, when more permanent approaches like implants can be adopted.
Bector *et al.*, 2015	The 3 year old patient received a removable partial denture on the maxilla and a complete removable denture on the mandible.	There was improvement of facial expression and profile. Follow up consults were scheduled and discomfort areas were relieved. The patient adapted well to the prostheses.
Vashisht *et al.*, 2015	The 12 year old patient received a conventional upper partial denture and a lower complete denture.	The child adapted well to the prostheses that had good retention. His social skills and self esteem have improved.
Renahan *et al.*, 2015	Case 1: The 7 year old child received a maxillary removable partial denture and a mandibular removable total denture with removable space maintainers. Case 2: The 5 year old child received a maxillary removable partial denture with removable space maintainer and a mandibular fixed partial denture with a removable space maintainer that was trimmed and attached to an orthodontic wire that was soldered on to bands on the lower molars. The 13 year old child received a mandibular conventional complete removable denture.	On both cases function and esthetics were successfully restored. The prosthetic management brought improvement of the lower vertical height of the face, reduced angular folds of lips, restored masticatory function, improved quality of life and brought better self confidence to the patients. Implant supported prostheses are planned after completion of craniofacial growth.
Vilanova *et al.*, 2015		The authors observed improvements on the patient`s physical and emotional states, speech and esthetics. Recalls for relining or remaking of dentures are planned.

TABLE 6. SUMMARY OF PUBLISHED REHABILITATION TREATMENTS FOR CHILDREN WITH ECTODERMAL DYSPLASIA IN 2016

Authors and publishing year	Interventions performed during childhood	Outcomes and/or author's considerations
Tavargeri *et al.*, 2016	The 13 year old child received provisional complete upper and lower dentures with magnets on posterior teeth.	The magnets were inserted on both upper and lower prostheses in order to enhance retention and stability. The patient was required to report for regular follow ups for 3 months for prostheses adjustment and trimming. The patient accepted the prostheses well; mastication was improved as well as social interaction and weight. Magnets brought better acceptability of the prostheses due to improved retention.
Kale *et al.*, 2016	The 7 year old child received a maxillary partial denture with a window for eruption of upper central incisors, a mandibular removable partial denture and metal crowns on primary second molars.	The patient received instructions regarding the removal of the prostheses during the night and recalls were scheduled every 3 months. In the future the patient should undergo reshaping of incisal edges, relining or replacement of dentures, and eventually placement of endosseous implants in order to achieve better prostheses support.
Retnakumari *et al.*, 2016	The 5 year old patient received total removable prostheses.	The patient was prescribed a dry mouth gel in order to enhance salivary secretion. His social interaction has improved. Regular follow ups are required in order to monitor ridge growth, prostheses retention and irritation due to use. In the future the prostheses should be replaced due to growth and further ahead implants may be placed depending on the growth of the ridge.
Dutta *et al.*, 2016	The 12 year old child received orthodontic correction with approximation followed by retraction and intrusion of maxillary central incisors. Then, the patient underwent a maxillary labial frenectomy and a mandibular vestibuloplasty using a Diode laser. Then prosthetic and restorative rehabilitation started with an increase of the occlusal surfaces of primary second molars by 1 mm with composite resin, followed by cementing of polycarbonate crowns on maxillary central incisors and finally the	The patient's facial expression and profile were greatly improved. Follow ups were scheduled for 1 week, 4 weeks, and the every 6 months. After 4 weeks the patient showed good function with both prostheses. Patient's speech and mastication were improved and good retention of the prostheses was observed.

	placement of acrylic partial prostheses on maxilla and mandible. Case 1: The 8 year old patient received maxillary and mandibular complete removable dentures over natural teeth. Case 2: The 6 years old patient received temporary partial orthodontic resin dentures on maxilla and mandible.	
Maatouk *et al.*, 2016		Case 1: The patient and his parents received instructions on how to place and remove the dentures, feeding, speech, hygiene and maintenance. They were also instructed to attend follow up consults every 6 months. Case 2: The patient could not attend a second appointment therefore the temporary prostheses were made on the same day. The father received instructions regarding hygiene and denture insertion. The child was referred to a dentist in their hometown.