



A SYSTEMATIC REVIEW: THE PREVELANCE OF PARTIAL EDENTULOURISM IN PATIENTS WITH ECTODERMAL DYSPLASIA

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ABSTRACT

Aim: To compile information regarding how ectodermal dysplasia is managed through various dental treatments.

Materials and Method: A detailed search was performed on articles related to ectodermal dysplasia. Specifically, in this review, it focuses on the treatment plans in children with this disease. A search was performed in the PubMed (Medline) and Web of Science databases to collect relevant information on the restoration of oral function in ED patients using dentures.

Results: 538 in total of related articles and abstracts were obtained from using the electronic search. During the initial screening, 420 articles were excluded. 118 articles were then completely evaluated resulting in only 53 being assessed as it fulfills the inclusion criteria of the systematic review. At the end, only 18 studies were used for assessment.

Conclusion: In a nutshell, the capability of deciding on a treatment for patients that have hypodontia in accordance to the evidence is not yet possible. Although implant therapy for hypodontia patients have a better outcome, when dealing with children, removable partial denture is the most suitable choice.

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INTRODUCTION

Ectodermal dysplasia (ED) is most commonly known to be a group of inherited disorders that are characterized with the presence of developmental abnormalities in either two or more of the four existing ectodermal structures; most commonly the teeth, hair, nails, and sweat glands-including other ectodermal structures^[1, 2]. Out of all these ectodermal structures, the human dentition is the most significant criteria taken into consideration for diagnosing ED patients. Ectodermal dysplasia is specifically caused only through genetic anomalies; which is either passed down from parents or caused by mutation of the genes^[3]. In terms of its classification, it can be either hypohidrotic or hidrotic. Hypohidrotic is mainly characterized by the absence or decrease in sweat glands while hidrotic is characterized by the presence of relatively normal sweat glands^[4]. Hypohidrotic ED is the most common form of ED seen in patients. This form of ED generally affects males more while females will only exhibit minor effects^[5]. Physically, HED patients exhibit common facial characteristics such as frontal bossing, “saddle” nose, maxillary hypoplasia, and hyperkeratotic wrinkles surrounding the eyes (contributing to the similar facial appearance they have)^[6, 7].

HED is the more valuable in the dental community as it predominantly shows more dental anomalies than other symptoms^[8]. During diagnosis, 80% will show hypodontia, while some other symptoms include oligodontia (decrease in the number of teeth) with conical teeth, hypohidrosis and hypotrichosis^[9]. They will also commonly exhibit reduction in salivary secretion, dryness of the oral mucosa and having a hoarse voice^[8]. When dealing with congenital deformities like oligodontia, the absence of dentition starts at an early stage of life; differing them from other oral problems like caries or periodontitis. One of the main advantage to this is that the patient is well adapted to their defects. Treatment wise, prosthodontic treatments are needed even in early childhood. However, many difficulties are faced during treatment mainly because the teeth are not yet ready to be used as an abutment as the pulp cavity is large. Dentures are also not suitable as the jaw is still in the process of development^[10]. Overall, there can be primarily four to five treatments available for these patients. Firstly, the option of preserving the primary deciduous teeth is normally selected. Secondly, there is an option of tooth transplantation; which helps in the healing of functional periodontium allowing for the mastication and development of the alveolar crest. Thirdly, the preparation of prosthetics can be done; primarily unsplinted overdentures. The fourth option would be the placement of dental implants^[11]. The main pupose of this study is to provide a

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systematic review of the literature regarding treatments options for patients that have ectodermal dysplasia.

METHODS

Search strategies

A search was performed in the PubMed of the US National Library of Medicine to obtain relevant information on the restoration of oral function in ED patients using dentures. The following search for relevant literature were used: (tooth OR teeth OR dental) AND (oligodontia OR ectodermal dysplasia OR hypodontia) AND (rehabilitation OR treatment).

Study inclusion and exclusion criteria

During the first stage of study selection, the titles and abstracts were screened and evaluated in accordance with the following inclusion criteria:

1. English language;
2. Human clinical studies including case reports, prospective and retrospective studies, and reviews;
3. Studies aimed at investigation specifically of patients that have hypodontia.

During this procedure, the pre-selected publications were then evaluated according to the following exclusion criteria:

2. More than 5 patients in a publication;
3. Studies in cleft lip and palate patients.

Assessment of quality in studies

A quality assessment of all the selected full-text articles was conducted. Both the exclusion process as well as the quality assessment were conducted independently by the author. Any studies that were believed to be insufficient were excluded during the process.

RESULTS

Study Selection

538 in total of related articles and abstracts were obtained from using the electronic search. During the initial screening, 420 articles were excluded. 118 articles were then completely evaluated resulting in only 53 being assessed as it fulfills the inclusion criteria of the systematic review. At the end, only 18 studies were used for assessment (Fig.1).

Studies on general outcome

Studies on dental implants and implant-supported prosthetics

6 studies in total were related to implant placement treatment for the patient. Almost all of the studies assessed were retrospective in nature.

Table 1

Author	Year	Study type	Patients	Age	Treatment (maxilla)	Treatment (mandible)	Follow up [y]
Ladda [12]	2013	Retrospective	1	8 years old	Complete dentures	Complete denture	0.6
Bani [13]	2010	Retrospective	2	-8 years old	Complete denture	Complete denture	Case 1
				-3 years old	Case 2	Case 2	Case 2
Bala [14]	2012	Retrospective	1	7 years old	Complete denture	Complete denture	0.3
Mittal [15]	2015	Retrospective	2	-5 years old	Complete denture	Complete denture	(not recorded)
				-9 years old	Case 1	Case 1	
Hekmatfar [16]	2012	Retrospective	2	-3 years old	Partial denture	Complete denture	(not recorded)
					Case 2	Case 2	
					Complete denture	Implant supported overdenture	
Nallanchakrava [17]	2013	Retrospective	1	12 years old	Case 1	Case 1	Case 1
					Case 2	Case 2	Case 2
Trivedi [18]	2013	Retrospective	1	4 years old	Complete denture	Complete denture	0.6
Jain [19]	2012	Retrospective	1	11 years old	Partial denture	Partial denture	0.6
Pinto [20]	2016	Retrospective	1	2 years old	n.a.	Partial denture	0.6
Marques [21]	2013	Retrospective	1	5 years old	Removable partial denture	Complete denture	1
Moses [22]	2013	Retrospective	1	3 years old	Removable partial denture	Removable partial denture	0.3
					Case 1	Case 1	Case 1
Shigli [23]	2012	Retrospective	3	-10 years old	Removable partial denture	Removable partial denture	(not recorded)
				-9 years old	Case 2	Case 2	Case 2
				-9 years old	Removable partial denture	Complete denture	0.3
Bala [24]	2011	Retrospective	1	8 years old	Case 2	Case 3	Case 3
					Overdenture	Overdenture	1.5
Correia [25]	2013	Retrospective	1	4 years old	Complete denture	Complete denture	(not recorded)
Bhalla [26]	2012	Retrospective	1	6 years old	n.a.	Removable partial denture	(not recorded)
Mello [27]	2015	Retrospective	1	9 years old	Complete denture	Complete denture	(not recorded)
					-Implants	-Implants	0.6
Kramer [28]	2006	Retrospective	1	5 years old	-Partial denture	-Partial denture	
				-6 years old	-Complete denture	-Complete denture	3.0
Bergendal [29]	2008	Survey	5	-6 years old	-Implants	-Implants	
				-6 years old			
				-8 years old	Implants	Implants	0.6
				-12 years old			
				-5 years old			

1. Case reports of patients aged 13 and above;

Eventhough the rate of survival as well as the success information is low, it was still considered for the purpose of

synthesis of data. These studies included varying forms of implants like overdentures over implants as well as implant crowns.

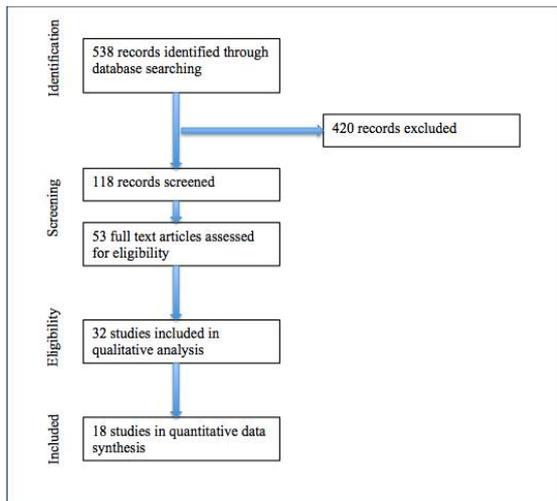


Figure 1

Based on the study by Bergendel, it was recorded that a high rate of implant failures were present in children. Since it was a survey based article, the follow-up time was not included. However, through contact with the author, it was reported that it happened during the healing process (maximum of 6 months) preceding prosthetic restoration^[29]. The study done by Mello focuses on the usage of overdentures following the placement of mini-implants. Though there were reports of implant failure on the patient, the case had a short-term follow up so further clinical evaluations should must be performed for an exact evidence on the matter. Overall, it was found that when dealing with children, the implants placed should be of a smaller size as a result of their smaller sized maxillary and cortical bones^[27].

Studies on treatment with removable partial dentures

In young patients, the process of oral rehabilitation is the most successful with removable partial dentures. The reason behind this is because this treatment option helps in the improvement of the child’s oral function, aesthetics as well decreases their social disadvantage. Based on the report done by Pinto, the young patient as well as his parents were onboard with the treatment since he personally felt different from his other classmates. Throughout the patients treatment, some adjustments were made to the prosthesis. The dentist did not experience any difficulty in the placement of the prosthesis. However, the partial dentures needed replacement within an average of 3-4 years because of it’s short lifespan^[20].

Studies on treatment with complete dentures

Complete dentures in young children are predominantly used to help them obtain a healthy diet; especially when dealing children below the age of 3 years old. Based on case report by Bani, for the initial insertion, the patient experienced some difficulty in keeping the denture in place. However, with a few months, the patient was said to have an improvement in his speech and was pleased with the denture. This overall has helped in the maintenance of a healthy diet for the child; which was the main concern before treatment^[13].

Studies on quality of life, self-esteem, and patient satisfaction

Most of the patients satisfaction level increased only after a few months after the initial placement of the prosthetic. A large number of the patients were in a range of satisfied to very satisfied with their treatment.

DISCUSSION

The most recent information about oral treatments including outcomes were assessed in this review.

Table 2

Publication	Year	Design	Outcome
Mittal [15]	2015	Retrospective	n.a. Case 2
Shigli [23]	2012	Retrospective	-the facial profile as well as the expression improved with the usage of the prosthesis. Their mastication and dietary patterns also improved significantly.
Mello [27]	2015	Retrospective	The patient adapted very well with the dental prosthesis. She showed great satisfaction with an increase in self-esteem and an improvement in her social skills.
Kramer [28]	2006	Retrospective	The patient was easily accustomed to the prosthesis and had an excellent oral hygiene. The parents reported that the patient’s diet has improved and changed for the better.
Bergendal [29]	2008	Cross-sectional survey	4/5 of the patients had lost at least one implant throughout the years.

Table 3

Publication	Year	Design	Outcome
Nallachakrava [17]	2013	Retrospective	There was functional occlusion with an improvement in esthetics. The nutrition as well as their self-confidence has showed an improvement.
Hekmatfar [16]	2012	Retrospective	The retention and stability of the denture was hard to obtain initially. Overall, the patients are more self-confident, has improved of facial esthetics, speech as well as the mastication.
Trivedi [18]	2013	Retrospective	The parents were very pleased with the outcome. There was an improvement in the patients speech and esthetics that lead to a better psychological well-being.
Jain [19]	2012	Retrospective	The patient reported to be comfortable because of it’s light weight quality. There was also also an improvement in both speech and mastication.
Pinto [20]	2016	Retrospective	The child was able to experience a wider range of foods. The patient’s esthetics and self-esteem also improved significantly.
Marques [21]	2013	Retrospective	There were no signs of pressure points and no interferences between both dynamic and static occlusion. It showed an excellent retention with an improvement in speech as well as mastication.
Moses [22]	2013	Retrospective	After intitial prosthesis insertion, the patient was depressed. After a week of usage, it became very comfortable for him. Overall, his speech and masticatory function has improved significantly.

Table 4

Publication	Year	Design	Outcome
Ladda [12]	2013	Retrospective	The patient's facial profile as well as expression has improved greatly. Case 1
Bani [13]	2010	Retrospective	There was good retention and a significant improvement in both speech and mastication. The child also enjoyed wearing the prosthesis. Case 2
Bala [14]	2012	Retrospective	After initial insertion of the denture, the patient was reported to have a difficult time in accepting it and keeping them in the mouth. After a while, it was reported that his speech has improved.
Mittal [15]	2015	Retrospective	The retention of the denture was good as the patient slowly adjusted to the prosthesis. Both his aesthetics and self-esteem has improved significantly.
Trivedi [18]	2013	Retropective	n.a.
Jain [19]	2012	Retrospective	The parents were very pleased with the outcome. There was an improvement in the patients speech and esthetics that lead to a better psychological well-being.
Marques [21]	2013	Retrospective	The patient reported to be comfortable because of it's light weight quality. There was also an improvement in both speech and mastication.
Bala [24]	2011	Retrospective	There were no signs of pressure points and no interferences between both dynamic and static occlusion. It showed an excellent retention with an improvement in speech as well as mastication.
Bhalla [26]	2012	Retrospective	Both the patient's masticatory function and aesthetics has significnatly improved.
			There was good denture stability as well as retention.

These studies that were assessed had a generally low quality as most of them are about removable dentures (both complete and partial) and only a few reported about implants. Even though the quality is low, it cannot be denied that dental implants showed a higher rate of failure with patients that have severe hypodontia when compared to healthy patients. Since this review is more focused on children that are affected with severe hypodontia, generally, removable dentures are more targeted towards them. One reason being that implants require more bone availability in the jaws and another reason being that it is a much more lower cost treatment option^[30].

Generally, it is not recommended for growing children to have implants except for the interforaminal area of the mandible^[31, 32]. The main reason behind this is because an ongoing eruption of teeth will result in the implant submerging; similar to that of secondary retention of teeth^[32]. Children on the other hand, are more easily adapted to removable partial dentures; as it can be known as their 'interim phase'^[30].

CONCLUSION

In a nutshell, the capability of deciding on a treatment for patients that have hypodontia in accordance to the evidence is not yet possible^[30]. Although implant therapy for hypodontia patients have a better outcome, when dealing with children, removable partial denture is the most suitable choice.

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