The Current Evidence on Retaining or Prosthodontically Replacing Retained Deciduous Teeth in the Adult Hypodontia Patient: A Systematic Review

Keywords

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ABSTRACT

Background: This systematic review aims to evaluate the survival of retaining or replacing deciduous teeth in hypodontia patients with a variety of prosthetic tooth replacement options, to evaluate prognostic factors associated with retaining deciduous teeth, and report on patient based outcomes with these treatment modalities. Methods: MEDLINE, The Cochrane Central Register of Controlled Trials and Science Direct databases were searched (01/1980 - 08/2017) for studies reporting outcomes associated with retaining or replacing deciduous teeth via prosthetic means in adult hypodontia patients. Results: Twenty-one articles were included. The following survival figures were reported: retaining deciduous tooth/teeth (83%-93%), resin-bonded bridgework (59-96.9%) and implants (86-100%). No survival data was reported for fixed or removable partial dentures. Prognostic factors for deciduous tooth survival, quality of life and patient satisfaction data were also reported. Conclusion: Within the limits of this review, retaining deciduous teeth have reasonable survival: however, studies beyond the third decade of life are lacking. Dental implants appear to be a highly successful long-term tooth replacement option with high patient satisfaction within this patient group, as have resin-bonded bridgework, albeit over the short to medium term. Tooth replacement options in the form of fixed and removable partial dentures were poorly reported upon.

INTRODUCTION

The presence of retained deciduous teeth in the adult patient, associated with absence of the permanent successors, often presents a challenging treatment planning proposition. Frequently, the clinician must decide whether to retain the deciduous tooth, or to commit to its removal and replacement as part of a comprehensive restorative treatment plan.

The retention of deciduous teeth occurs for a variety of reasons, the most common being the developmental absence of the permanent successor (hypodontia).[1] Hypodontia itself is the most frequent dental malformation which varies in severity from a missing single tooth to absence of all teeth.[2] This condition most frequently affects the secondary dentition with a reported prevalence of 2.6%-11.3% in data derived from population studies.[2]

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Hypodontia is more common in females than males,[3] with an approximate 3:2 prevalence ratio;[4] the most frequently missing teeth being the mandibular second premolars (2.9-3.2% of the population), followed by the maxillary lateral incisors (1.6-1.8%), maxillary second premolars (1.4-1.6%) and the mandibular incisors (0.2-0.4%).[2]

Agenesis of the permanent dentition can lead to the retention of a deciduous tooth or teeth. In many cases the prognosis of the retained deciduous tooth is limited. Assessment of the prognosis of the retained tooth or teeth alongside a comprehensive evaluation of the consequences of tooth loss is essential for patient consent and devising predictable and effective treatment plans. In many cases, the management of hypodontia needs to be carried out by specialists as part of a multi-disciplinary dental team due to the potential complexities in planning and delivering dental care.[5,6]

When a deciduous tooth is retained and the permanent successor tooth is missing, there will be a variety of treatment options including;

Retaining the deciduous tooth, with or without restorative modification or extraction of the tooth and either:

- Accepting the presence of an edentulous space.
- Orthodontic closure of the edentulous space.
- Auto-transplantation of another tooth into space.
- Prosthodontic tooth replacement using either; conventional or resin bonded bridgework, a removable prosthesis or an implant retained prosthesis.
- Orthodontic treatment to idealize or redistribute the edentulous space to facilitate prosthodontic tooth replacement.

As there is currently no standard approach in how to treat this patient group, the aim of this review is to evaluate the survival of retaining or replacing deciduous teeth in hypodontia patients in the absence of the permanent successor in the adult patient with a variety of prosthetic tooth replacement options, to evaluate prognostic factors associated with retaining deciduous teeth and report on patient based outcomes with these treatment modalities.

OBJECTIVE

It is the aim of this review to evaluate the survival of either retaining or replacing deciduous teeth in hypodontia patients with a variety of tooth replacement options and to evaluate prognostic factors associated with retaining deciduous teeth and report on patient based outcomes with these treatment modalities.

METHODS

PROTOCOL

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[7,8] for describing and summarizing the results of our review was used.[7,8]

A quality assessment of all selected full-text articles was performed using the Methodological Index for Non-Randomized Studies (MINORS)[9] assessment tool to assess the risk of bias of the included studies. The MINORS scoring list consists of 12 items, eight apply to non-comparative studies and the remaining four items apply to comparative studies. Items are scored as 0 (not reported),1 (reported but inadequate) and 2 (reported and adequate) with this then totalled up to give a score, with the higher scores representing a reduced risk of bias.[9] This was chosen over the Cochrane collaborations' tool for assessing risk of bias for randomised controlled studies since none of the studies were randomised control trials.

ELIGIBILITY CRITERIA

INCLUSION CRITERIA

Studies that met the following criteria were included;

1. Studies limited to humans and restricted to the English language.
2. Studies on patients with hypodontia.
3. Studies reporting on adults or extending into adulthood.
4. Studies reporting on 5 patients or more.
5. Studies reporting on outcomes related to retaining deciduous teeth and/or prosthodontic replacement and addressing the focused question.

EXCLUSION CRITERIA

Studies that met the following criteria were excluded;

1. Studies reporting on non-hypodontia patients.
2. Studies reporting on cleft lip and palate, ectodermal dysplasia or other syndromic conditions associated with hypodontia exclusively.
3. Studies reporting on child patients only.
4. Studies reporting on less than 5 patients.
5. Review articles.
6. Studies that did not address the focused question.

INFORMATION SOURCES

Three electronic databases were used to systematically search the available literature: (1) The National Library of Medicine (MEDLINE via Pubmed); (2) Cochrane Central Register of Controlled Trials and (3) Science Direct. The searches were limited to studies involving human subjects and publication dates from January 1980 to August 2017 that satisfied the inclusion criteria.

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SEARCH

The following search terms were used; (tooth OR teeth OR dental) AND (hypodontia OR anodontia OR oligodontia OR tooth aplasia OR congenitally missing teeth OR missing teeth OR deciduous tooth OR developmentally absent OR persistent deciduous) AND (dental implant OR endo-osseous OR crown OR resin bonded bridge OR fixed partial denture OR bridge OR deciduous teeth OR dental prosthesis OR removable dentures) AND (survival OR success OR quality of life OR satisfaction OR self-esteem OR root resorption OR infraocclusion OR submergence) and any MeSH terms associated.

The references of the selected, suitable publications were then hand searched for any additional publications not identified through the electronic search.

STUDY SELECTION

The primary screening (title, abstract) was carried out by reviewer (DL) identifying the studies appearing to meet the inclusion criteria. Studies with insufficient information in the title and abstract to make a clear decision were identified and the full paper was reviewed. Those studies selected for evaluation of the full manuscript were reviewed with reviewers (OA & DL) determining the final inclusion.

DATA COLLECTION PROCESS

Reviewer (DL) then extracted the data using a bespoke data extraction form. Any disagreement was resolved by discussion with reviewer (OA). Studies with missing or incomplete data were excluded and reference lists of the selected studies were checked for cross-references to search for papers that might meet the eligibility criteria for inclusion.

DATA ITEMS

Data was collected for; survival data and outcomes associated with retaining deciduous teeth including the prognostic factors in the form of deciduous tooth type, root resorption, infraocclusion/submergence, caries, periodontal health and the restorative status of the deciduous tooth. Survival data, outcomes and patient based satisfaction for tooth replacement options in the replacement of missing teeth resulting from hypodontia was also collected for the following tooth replacement options; Resin Bonded Bridge (RBB) (adhesive fixed partial denture), Conventional bridgework (fixed partial denture), removable partial dentures and dental implant-supported prosthesis.

RISK OF BIAS IN INDIVIDUAL STUDIES

A quality assessment of all selected full-text articles was performed using the Methodological Index for Non-Randomized Studies (MINORS) assessment tool.[9]

SUMMARY MEASURES

The main outcome measure was related to survival of the treatment modality provided. Other outcomes measures in the form of prognostic factors and patient based satisfaction for each treatment modality where reported on where possible.

SYNTHESIS OF RESULTS

Survival data, patient based satisfaction and other outcomes were taken directly from the study where relevant and appropriate.

ADDITIONAL ANALYSES

No further analysis was carried out.

RESULTS

STUDY SELECTION

Searches of MEDLINE, The Cochrane Central Register of Controlled Trials and Science Direct were carried out and references of selected, suitable publications were hand searched for any additional publications not identified through the electronic search generating 3638 articles. After initial review of the titles and abstracts and removal of duplicates, 126 articles were accepted for further consideration and 3512 articles were rejected. After the full text was attained and reviewed for the 126 articles, 105 articles were rejected leaving 21 articles to be included in the systematic review. (*Figure 1*)

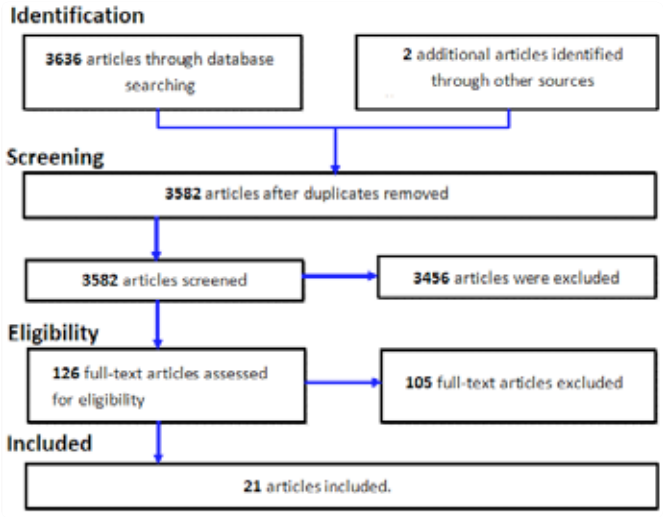

Figure 1: Methodology for data inclusion

STUDY CHARACTERISTICS

The following data was extracted from the studies: study design, the study population, follow up period, patient number, the treatment modality provided, and the number of each treatment modality provided. For retained deciduous teeth the following data was collected: the deciduous tooth type,

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the number of patients, the number of deciduous teeth, survival figure of retained deciduous teeth, and findings relating to; root resorption, infraocclusion, caries, periodontal health, the restorative status of the deciduous tooth/teeth and any other relevant findings. For tooth replacement options the following data was collected: the number of patients provided with the tooth replacement options, the number of each tooth replacement option provided, survival of the tooth replacement option, quality of life and/or patient satisfaction data and the tool used to assess quality of life and/or patient satisfaction outcomes.

RISK OF BIAS WITHIN STUDIES

There were varying scores attained by the studies using the MINORS assessment tool, ranging from 6/16 to 19/24 representing varying degrees of bias within the studies *(Table 1,2 & 3)*.

STATISTICAL ANALYSIS

Due to the lack of controlled studies and the heterogeneity of the studies a formal meta-analysis would be statistically inappropriate and was not conducted. Descriptive statistics were used to interpret and present the data from these studies.

RESULTS OF THE STUDIES

Descriptive data extraction was carried out for the 21 studies and is summarised in Tables 1, 2 & 3. These 21 studies were published over a range of 32 years (1984 to 2016); 9 studies were retrospective, 8 prospective, and 4 cross-sectional in study design.

STUDIES ON PRESERVATION OF DECIDUOUS TEETH

A total of 7 studies[10-16] (summarised in Table 1) were used to assess the outcomes relating to retaining deciduous teeth and included cross-sectional, prospective observational and retrospective studies. Four of these studies[10-13] were longitudinal observational studies that reported on deciduous tooth survival with survival figures of between 83%[10] and 93%[11] over follow-up periods ranging from 5 to 15 years. However, the majority of these studies,[10,11,13] except the study by Sletten *et al*,.[12] reported on study populations that began in childhood and followed up into adulthood and as such there is unfortunately a paucity of long term follow-up studies available which can inform us of the survival rate of retained primary teeth in a population from the third age decade onwards.[12] Three cross-sectional studies[14-16] were also assessed which identified that deciduous teeth can be retained at a broad range of ages (ranging from childhood up to the fifth decade of life) within the observed populations. Although this tooth survival data is not robust, the reported prognostic factors affecting the deciduous teeth were common in much of the literature to date. These factors were grouped together as follows: the deciduous tooth type, the degree of root resorption, presence of infraocclusion/submergence, presence of active or historical caries, periodontal health and the restorative status of the tooth.

THE DECIDUOUS TOOTH TYPE

The deciduous tooth type and its prognostic effect was reported in one study by Heselden *et al*,.[14] This study reported on the relationship between the deciduous tooth type and tooth survival and concluded that the mandibular deciduous canines had the most predictable prognosis followed by maxillary deciduous canines and that deciduous molar teeth had the poorest and least predictable life span.[14]

ROOT RESORPTION

Root resorption was reported in all 7 studies. In general, most studies interpreted the absence of root resorption as success; however, all studies reported varying degrees of root resorption within their study populations. The age at which root resorption occurred and its rate and pattern of progression varied greatly within the studies. Root resorption within the studies was reported as being unpredictable[10,12,13] and those teeth with more severe root resorption were at higher risk of tooth loss.[10,11,13]

INFRAOCCLUSION

6 of the studies reported on infraocclusion[10-13,15,16] which was a common complication with varying incidence of between 25%[15] to 43.6%[16] within the populations studied. It was also reported that the incidence and severity of infraocclusion increased over time, even into adulthood.[11,13,15] This was highlighted in longitudinal observational studies by Ith-Hansen *et al*,.[13] and Bjerklin *et al*,.[11] and in a cross-sectional study by Garib *et al*,.[15] who reported that the prevalence of infraocclusion was age related and that the highest incidence of infraocclusion occurred in the oldest patient group (patients in the second to third decade of life).[15] These are interesting observations as it contradicts current thinking that infraocclusion ceases once facial growth is complete.[17]

CARIES

3 studies[10-12] reported on caries associated with retained deciduous tooth loss. Overall caries appeared to be low with all aforementioned studies reporting that only an insignificant number of teeth required extraction as a direct consequence.[10-12]

PERIODONTAL HEALTH

Only 1 study reported on the impact of periodontal factors on loss of the retained deciduous tooth. This study by Sletten *et al*.,[12] reported that 3 of the 4 teeth lost during the observational period were a result of periodontal bone loss with this loss being three times as many as those lost due to caries.[12] No other study included reported on periodontal factors and outcomes associated with retained deciduous teeth.

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Table 1. Observational and cross-sectional studies assessing deciduous tooth retention

Author (Date)	Study Design	Quality Assessment using MINORs Assessment Tool	Study Population	Teeth Assessed	Primary Teeth No.	Patient No.	Follow Up	Survival (%)	Infraocclusion	Root Resorption	Other Findings
Haselden (2001)[14]	Cross sectional	8/16	Patients presenting to Joint Hypodontia Clinic, Eastman Dental Hospital, UK	Retained deciduous canine and molars without permanent predecessor	Unknown	249	N/A	N/A	Not Reported	Canine teeth were most likely to show little or no resorption. Upper 1st deciduous molars were most likely to show the greatest and the 2nd deciduous molars did not demonstrate a particular resorptive state.	Lower and upper deciduous canines have a predictable life span. Upper and lower deciduous molars have an unpredictable lifespan. The upper and lower 1st deciduous molars are the least likely to survive with the upper more predictable than the lower.
Ith-Hansen (2000)[13]	Prospective Observational	8/16	Patients attending the orthodontic Department, School of Dentistry, University of Copenhagen, Denmark	Retained deciduous molars without permanent successors.	26	18	15 years	88.5% (3 of 26 teeth lost)	11.5%. (3 teeth showed worsening infra-occlusion)	11.5 % (3 teeth showed progressive root resorption)	The study concluded that retaining primary secondary molars in agenesis of permanent successor can be an acceptable and semi-permanent.
Bjerklin (2008)[11]	Prospective Observational	13/16	Not Specified	Retained deciduous mandibular second molar without a permanent successor	149	99	12.2 years	92.9% (7 of 99 teeth lost)	21% at initial presentation. 50% at age 20-21 years old and 52% at the age of 28–29 years	In almost half of subjects root resorption levels of the primary molars were unchanged from the age of 12-13 years old up to 24–25 years of age. After this age, there was even less change in the resorption level.	During the observation period, only seven of the 99 primary molars were lost due to extensive root resorption, infraocclusion, or caries. Long-term survival may be expected in more than 90 per cent of patients with retained primary molars with agenesis of mandibular second premolars.

table 1 continued overleaf...

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...table 1 continued

Table 1. Observational and cross-sectional studies assessing deciduous tooth retention

Sletten (2003)[12]	Retrospective	9/16	Patients presenting at University of Iowa College of Dentistry, USA	Retained deciduous mandibular second molar without a permanent successor.	20	28	12.4 years	86% (4 of 28 teeth lost)	No changes in infraocclusion were observed.	Root resorption progression was negligible. Mean root length shortening was 0.16 (0.62) mm over the observational period with .01 (0.05) mm mean annual root length shortening.	4 mandibular deciduous molars were lost because of caries or periodontal breakdown. These teeth were lost at a mean age of 51.0 years over a mean observational period of 14.3 years; that approximates the lifespan of some prosthetic appliances and is a viable treatment options.
Hvaring (2014)[16]	Cross sectional	11/16	Patients attending Interdisciplinary Clinic at University of Oslo, Faculty of Dentistry, Norway.	Retained deciduous mandibular molars with lack of a permanent predecessor	188	111	N/A	N/A	43.6% of teeth with significant and 18.8% with severe infraocclusion	The mesial and distal root exhibited resorption in 81.1% and 66.7% of cases.	Most primary molars had no restorations (78.4%). A significant correlation was found between root resorption and infraocclusion as was age. With infraocclusion reported as being a critical factor for the prognosis of retained primary molars.
Rune (1984)[10]	Prospective Observational	9/16	Patients attending the centre for Craniofacial Anomalies, General Hospital, Malmo, Sweden.	Retained deciduous second molar without a permanent successor.	123	77	5 years and 3 months (males) and 5 years and 4 months (females)	82.9% (21 of 123 teeth lost)	Submergence affected mandibular molars more frequently than maxillary molars	50 of 104 Mandibular Molar underwent continued root resorption	In the maxilla 26% of the molars were lost due to total root resorption and none due to submergence while in the mandible 5% of the molars were removed because they were submerged and none were lost due to root resorption.
Garib (2014)[15]	Cross sectional	11/16	Orthodontic patients from a Brazilian Dental School and from eight private dental offices.	Retained deciduous second molar without a permanent successor	251	158	N/A	N/A	25% of the teeth were infraoccluded. (15% in the 1st decade of life, 27% in the 2nd, and 45% in the 3rd)	Root resorption progressed from the 1st to the 2nd decade of life and remains stable until the 3rd decade.	The amount of root resorption was positively correlated with infraocclusion.

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Table 2. Survival of Tooth replacement options in Hypodontia patients

Author (Year)	Study Design	Quality Assessment using MINORs Assessment Tool	Study Population	RBB Patient No.	RBB No.	Follow Up	Survival (%)	Additional notes
Garnett (2006)[19]	Retrospective	10/16	RBBs provided for post-orthodontic hypodontia patients with missing maxillary lateral incisors.	45	73	Upto 100 months	59.0%	30 RBBs debonded on at least one occasion (41.1%) with six of as a result of trauma (20%). The mean survival time of the prosthesis was 59.3 months.
Allen (2016)[18]	Prospective	10/16	RBBs provided for post-orthodontic non-syndromic hypodontia patients favorable for tooth replacement with resin bonded bridgework.	44	65	24 months	96.9%	RBB Survival = 96.9% with 2 RBB failures (due to debonding on more than 1 occasion) RBB Success = 92.3% with 3 RBBs (due to debonding on one occasion)
Author (Year)	**Study Design**	**Quality Assessment using MINORs Assessment Tool**	**Study Population**	**Removable prosthesis Patient No.**	**Removable Prosthesis No.**	**Follow Up**	**Survival (%)**	**Additional notes**
Hobkirk (1989)[22]	Retrospective	6/16	Patients with severe hypodontia	Total:138	Total: 138 Maxilla: 48 Mandible:90	6 years	Not reported	Partial dentures had a short lifespan; particularly partial dentures in the maxilla, which needed replacement 3.5–4 years on average after delivery. The most common reported reasons for replacement included; dissatisfaction of the patients with regards to the aesthetics of the prosthesis, fracture, wear or oral changes
Author (Year)	**Study Design**	**Quality Assessment using MINORs Assessment Tool**	**Study Population**	**Implant Patient No.**	**Implant No.**	**Follow Up**	**Survival (%)**	**Additional notes**
Créton (2010)[23]	Retrospective	19/24	Patients referred to academic centre of special dental care and classified with “oligodontia” or “severe hypodontia”	44	214	Mean observation period of 2.9 years. Range of 0.1 to 18.3 years)	89.8%	
Finnema (2005)[24]	Retrospective	11/16	Oligodontia patients treated with dental implants.	13	87	Mean follow-up 3 +/- 2 years. (Range 1 to 8 years)	86% Maxilla and 96% Mandible	Implant survival 86% maxilla and 96% mandible. 11 of 13 patients underwent autogenous bone grafts.

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...table 2 continued

Table 2. Survival of Tooth replacement options in Hypodontia patients

Becelli (2007)[25]	Retrospective	10/16	Oligodontia patients.	8	58	Mean follow-up 8.5 years.	96.6%	Implant survival 97.1% maxilla and 96.2% mandible. 5 patients underwent bone grafting or sinus lift procedures.
Hosseini (2013)[27]	Prospective	11/16	Patients with tooth agenesis.	59	98	3 years	100%	Comparing all-ceramic and metal-ceramic single implant crowns.
Nissan (2011)[28]	Retrospective	7/16	Patients with hypodontia that required a cancellous bone block-allografts to accommodate an implant fixtures with a bony deficiency of ≥3 mm horizontally and ≤3 mm vertically from a CBCT.	12	21	Mean follow-up time was 30 ± 16 months	95.2%	All patients underwent grafting with cancellous allogeneic bone-blocks were used.
Worsaae (2007)[26]	Cross sectional	9/16	Patients suffering from oligodontia.	46	283	28 months	97.8%	43% of patients underwent sinus floor elevation and 73% underwent bone grafting procedures.
Branzén (2016)[29]	Retrospective	13/16	Patients with congenitally missing upper lateral incisors	36	54	Mean follow up 6.8 years	100%	1 patient underwent bone augmentation due to a buccal fenestration defect at the mid part of the implant.

Table 3. Patient satisfaction with tooth replacement options in Hypodontia Patients

Author (Year)	Study Design	Quality Assessment using MINORs Assessment Tool	Study Population	Treatment Modality	Follow Up	Patients No.	QoL assessment/ Satisfaction Questionnaire used	Findings	Conclusion
Allen (2016)[31]	Prospective	12/16	Non-syndromic mild hypodontia with missing teeth in the anterior maxilla. Single implants with single crowns.	Implants	24 months	12	OHIP-20 scores at 12- and 24-month. Pink (PES) and white (WES) aesthetic scores	There was a significant improvement in oral health-related quality of life following treatment (P = 0.026), and the effect size of this change was 1.17 indicating a large clinically meaningful change. Mean PES and WES scores were high, indicated a satisfactory aesthetic outcome at 24 months.	Favorable clinical outcomes were observed. Implant retained crowns had a large and clinically meaningful impact on quality of life of patients with hypodontia

table 3 continued overleaf...

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...table 3 continued

Table 3. Patient satisfaction with tooth replacement options in Hypodontia Patients

Dueled (2009)[21]	Retrospective	19/24	Adults with tooth agenesis restored with FDPs or implants	Implants vs FDPs vs Control Group	46 months	129	OHIP questionnaire. Aesthetic index score.	The median scores for all five aesthetic variables were acceptable in 92% of the implant reconstructions and for 83% of FDPs. The six OHIP questions concerning patient-based aesthetic problems was reported in 41% of implant-supported reconstructions and 47% of tooth-supported FDPs. 98% of the group treated with implant-supported reconstructions and 84% of the patients in the tooth supported FDP group were very satisfied or satisfied with the treatment outcome.	The total OHIP scores were inferior in rehabilitated patients with tooth agenesis in comparison to the control group without tooth agenesis. Improved outcomes were obtained with implant-supported reconstructions than with tooth-supported reconstructions both from patient and professional based assessments.
Finnema (2005)[24]	Retrospective	11/16	Oligodontia patients treated with dental implants	Implants	Follow-up 3 +/- 2 years, range 1 to 8 years	13	Own Satisfaction Questionnaire. Mandibular Function impairment Questionnaire	Patients were at least satisfied with implant based reconstruction. Mean overall satisfaction was 8 out of 10 and 9 of 13 patients had improved self-confidence.	Patients were generally satisfied with the overall treatment experience and reported significant functional improvement
Goshima (2009)[30]	Prospective	12/16	Patients with agenesis of permanent teeth treated at the Department of Oral Rehabilitation, School of Dentistry, University of Copenhagen.	Implants - single implant cement retained crowns	1 month after crown placement	18	Masticatory function and OHIP-49	Patients reported high satisfaction (moderate to very satisfied) with treatment and also experienced a significant overall reduction in the severity of their oral health related problems.	The OHRQoL was improved 1 month after crown cementation and subjects with tooth agenesis may benefit from this type of treatment, both on subjective and objective levels.
Hosseini (2013)[27]	Prospective	11/16	Patients with tooth agenesis.	Implants	3 years	59	Danish version of OHIP-49	QoL improvement was shown from pre-treatment to baseline (after fit of the implant crown). However, at 3 years after the implant crown was fitted QoL improvement had reduced slightly. This was reported to be as a result of patients reporting sensitivity and pain with their natural teeth.	All scores decreased during the course of the study, but the summary scores of the 49 OHIP questions indicating the over- all OHQoL increased slightly from the baseline to the 3- year observation.

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...table 3 continued

Table 3. Patient satisfaction with tooth replacement options in Hypodontia Patients

Branzén (2016)[29]	Retrospective	13/16	Patients with congenitally missing upper lateral incisors	Implants	Mean Follow up 6.8 years	36	A modified Swedish version of the questionnaire used by Chang. (Chang M. The peri-implant tissues from an esthetic perspective. Thesis. Gothenburg, Sweden: Sahlgrenska Academy, University of Gothenburg, 2009)	The patient rating was high for the overall satisfaction, with 21 being completely satisfied and 14 fairly satisfied. However, 12 patients wished for the replacement of their single implant crown,	One-third of the patients wished for the replacement of their ISCs. Soft tissue adaptation seems to be an important factor for overall satisfaction
Anweigi (2013)[20]	Prospective	20/24	Patients with a confirmed diagnosis of hypodontia participated who had undergone orthodontic treatment to idealise spacing for tooth replacement with RBB.	Resin bonded Bridge	After Resin bonded bridge fit	82	OHIP-49	There were no differences between the groups prior to treatment. For the test (RBB) group, there was a significant improvement in median OHIP summary scores ($p<0.001$) after treatment	Hypodontia has a significant impact on oral health related quality of life. Provision of resin bonded bridges has a positive impact on oral health related quality of life of patients with hypodontia

RESTORATIVE STATUS OF THE TOOTH

2 studies reported on the restorative status of the retained deciduous teeth. A cross-sectional study by Hvaring *et al.*,[16] reported that 20.8% of retained deciduous teeth were restored (9.0% approximal and 11.8% occlusal restorations)[16] and a longitudinal study by Sletten *et al.*,[12] who reported on the restorative status of the retained deciduous teeth over time, noted that 16 retained deciduous teeth (of 27 retained deciduous teeth followed) were initially restored either with a full coverage gold crown, composite onlay or an amalgam restoration and that during the observational period, 5 retained deciduous teeth that were previously restored had replacement restorations and 1 unrestored retained deciduous tooth was restored.[12] This would suggest that these teeth are commonly restored; however, no report was made on the restorative status of the tooth and the prognosis of the retained deciduous tooth.

PROSTHODONTIC TOOTH REPLACEMENT

STUDIES ON RESIN BONDED BRIDGE (RBB) (ADHESIVE FIXED PARTIAL DENTURE)

3 studies were included in total with 2 studies reporting on RBB survival[18,19] and 1 study reporting on patient satisfaction with the provision of RBB work in hypodontia patients.[20]

A prospective study by Allen *et al*,.[18] reported a high survival rate of 96.9% with a follow up of over 24 months in RBBs replacing a variety of missing permanent teeth. However, Garnett *et al.*,[19] reported a survival rate of 59% with a mean survival rate of 59.3 months in a retrospective study which assessed RBBs replacing maxillary lateral incisors with a follow-up of up to 100 months *(Table 2)*.[19] Both of these studies reported on patients who had undergone pre-restorative orthodontic treatment to idealise the edentate spaces prior to tooth replacement.[18,19] When assessing patient satisfaction with RBBs in hypodontia patients *(Table 3)*, 1 study was included. This study by Anweigi *et al.*,[20] reported RBB restoration of the edentulous spaces had a positive and significant difference on oral health related quality of life (OHQoL) post-treatment in comparison to the control group (hypodontia patients undergoing orthodontic treatment).[20]

STUDIES ON CONVENTIONAL BRIDGEWORK (FIXED PARTIAL DENTURE)

No studies reporting on fixed conventional bridgework survival in hypodontia patients were identified during the search of the literature. Patient satisfaction with FPDs was reported in 1 study by Dueled *et al.*;[21] *(Table 3)* this study compared professional and patient-based evaluation of implant- and tooth-supported reconstructions in patients with tooth agenesis. However, no description was given of the type of fixed partial denture that was evaluated (whether that be adhesive or conventional).

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This comparative study *(Table 3)* reported that 41% of patients treated with implant-supported reconstructions and 47% of patients treated with tooth-supported FDPs had aesthetic problems within the preceding months occasionally, fairly often or very often and that satisfaction (rated as either very satisfied or satisfied) with a tooth supported FPD was lower (84%) in comparison to an implant-supported reconstruction (98%).[21]

STUDIES ON REMOVABLE PARTIAL DENTURES

1 study by Hobkirk *et al.*,[22] was included. This study assessed the failure rates and modes of failure of 138 removable partial dentures in patients with severe hypodontia. This study reported that removable definitive partial dentures had a short lifespan; particularly partial dentures in the maxilla, which needed replacement 3.5–4 years on average after delivery. The most commonly reported reasons for replacement included; dissatisfaction of the patients with regards to the aesthetics of the prosthesis, fracture, wear or oral changes.[22]

No studies reporting on patient satisfaction with removable prostheses were identified during the search of the literature.

STUDIES ON DENTAL IMPLANTS AND IMPLANT-SUPPORTED PROSTHESIS

7 studies were included[23-29] in total with all 7 reporting high implant survival in hypodontia patients of between 86%-100% with varying follow up[23-29] *(Table 2)*. 2 studies compared implant survival in the maxilla and the mandible. Finnema *et al.*,[24] reported higher implant survival in the mandible (86% maxilla & 96% mandible) however, Becelli *et al,*.[25] reported higher implant survival in the maxilla (97.1% maxilla & 96.2% mandible) within their patient cohorts. 5 studies reported on outcomes related to use of bone augmentation at the implant site;[23-26,28] this appeared to increase the risk of implant failure with Créton *et al.*,[23] and Worsaae *et al,*.[26] reporting this. However, studies by Finnema *et al.*,[24] Becelli *et al,*.[25] and Nissan *et al.*,[28] reported similar implant survival rates in grafted vs ungrafted sites in hypodontia patients.[24,25,28]

6 studies reported on patient satisfaction or quality of life (QoL) associated with implant based tooth replacement in hypodontia patients. 5 studies reported improved satisfaction and QoL with the provision of dental implants to replace missing teeth.[21,24,27,29-31]

Allen *et al,*.[31] Finnema *et al,*.[24] and Goshima *et al,*.[30] all reported significant improvement in OHQoL in hypodontia patients provided with implant based prostheses; this was also true in a study by Hosseini *et al.*, 27 however, after 3 years this improvement reduced slightly which was reported to be as a result of patients reporting sensitivity and pain with their natural teeth. Brazén *et al,*.[29] reported high overall satisfaction of single implant crowns after an average follow up of 6.8 years, despite this however, almost one-third wished for the replacement of their single implant crown.[29]

A comparative study by Dueled *et al,*.[21] comparing implant- vs tooth-retained fixed partial dentures in hypodontia patients with a control group without hypodontia and the need for prosthetic tooth replacement reported higher satisfaction and aesthetic ratings with the provision of implant-retained in comparison with tooth-retained fixed partial dentures. However, the total OHIP score was inferior in rehabilitated patients with tooth agenesis to that of the control group without tooth agenesis.[21] *(Table 3)*

DISCUSSION

SUMMARY OF EVIDENCE

Current evidence regarding preservation or extraction and prosthodontic tooth replacement of patients with retained deciduous teeth with permanent tooth agenesis was reviewed.

The studies included generally reported on young adult patients or reported on patients from childhood into adulthood. The evidence level for the majority of the literature was low with observational and cross-sectional studies being the mainstay of the literature available. In some instances, there was a clear lack of literature available which was particularly true when assessing prosthodontic tooth replacement options in the form of removable prosthesis and fixed conventional bridgework in this patient group. This is understandable as these tooth replacement options are infrequently carried out in young patients due to their removable and destructive nature respectively.[32]

Survival of retained deciduous teeth with associated tooth aplasia has been shown to be high with tooth survival figures of between 83%[10] and 93%[11] over follow-up periods ranging from 5 to 15 years *(Table 1)* with such survival rates rivalling that of implants or other fixed restorations.[1,33-35] However, very few studies reported on patients beyond the third decade and those that did only reported on a small number of teeth. As a result, a long-term conclusion cannot be extrapolated. A number of prognostic factors were reviewed within the literature which included; the deciduous tooth type, the degree of root resorption, presence of infraocclusion/submergence, presence of active or historical caries, periodontal health and the restorative status of the tooth.

With regard to the deciduous tooth type, it is difficult to truly assess the effect of the deciduous tooth series on prognosis since certain deciduous teeth are more commonly retained than others, predominantly due to aplasia of the permanent successor. As a result, most of the studies included predominantly reported on the deciduous mandibular second molar due to the frequent absence of the permanent second premolar.[1] Root resorption was also assessed which revealed that the presence and the rate of root resorption in retained deciduous teeth was unpredictable but the absence of a permanent successor appeared to reduce the rate of root resorption progression.[10,12,13] Age also appeared to affect progression,

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with those patients beyond their mid-twenties showing much slower progression of root resorption in 2 studies.[11,15] This could be a good indicator that if a tooth gets to this age then it has a reasonable long-term outcome; however, there are very few studies that go beyond this age group. The current evidence indicates that careful monitoring of the root morphology of deciduous teeth is required. Those teeth with extensive root resorption or greater rates of progression clearly have a poorer prognosis and must be considered for extraction and replacement if warranted. Whilst teeth with mild root resorption and a slow rate of progression clearly have a better prognosis, it is also important to recognise that root resorption can be unpredictable.[10,12,13] Infraocclusion presents when a deciduous tooth fails to follow the facial growth and fails to move occlusally with the adjacent teeth, thereby leaving it apical to the occlusal plane and appearing submerged[20] and is a common presentation in hypodontia.[11,13,15,16] The tendency for infraocclusion has been shown to increase over time in most reports.[11,13,15] An interesting observation to note is that the severity of infraocclusion appears to increase with age even into adulthood which goes against current thinking that infraocclusion ceases once facial growth is complete;[17] however, this observation is based upon a single cross-sectional study and as such should be interpreted with caution. The presence and progression of submergence has been shown to be a critical factor for retained deciduous tooth prognosis.[11,13,16] Importantly, positive correlations between root resorption and infraocclusion have been noted in some studies[15] whilst others have failed to identify this association.[10] Infraocclusion itself can lead to a non-functional tooth which would be a poor outcome, particularly in this group of patients. Therefore, the degree and rate of submergence/infraocclusion should be closely monitored, and it should be recognised that even if a retained deciduous tooth is restored to become functional it may become non-functional over time as infraocclusion progresses.[1] Other prognostic factors were assessed and would suggest that caries and periodontal disease are low within this patient cohort and that caries, periodontal health and the restorative status of the tooth showed no link to clinical tooth survival and long-term tooth outcome.

Overall, the literature would tend to support retaining deciduous teeth in adults when they are non-mobile, functioning with good root morphology, minimally affected by caries and are aesthetically acceptable.[1,36,37] The advantages of retaining a healthy deciduous tooth include the psychological benefits of a person keeping their own tooth. It also incurs minimal maintenance,[38,39] reduces treatment costs and the risks associated with tooth removal and replacement. Where deciduous teeth are retained, careful monitoring of the tooth is required due to the risk of progressive root resorption, infraocclusion and other pathological processes that occur within the rest of the dentition. Overall, the patient must understand that there is a lack of long term follow-up studies showing the survival rate of retained primary teeth in a population beyond the third age decade.

Tooth replacement options in adult patients with hypodontia were also assessed and included; resin bonded bridgework, conventional fixed bridgework (fixed partial dentures), removable prosthesis and implant based tooth replacement. However, there was a clear lack of literature reporting outcomes associated with conventional fixed bridgework (fixed partial dentures) and removable prosthesis in this patient cohort as previously discussed.

Dental implants in hypodontia patients demonstrated high survival rates *(Table 2)* with these figures well aligned with currently published data on implant prognosis in conventional dental implant patients.[34,35] High patient satisfaction and improvement in QoL was also shown with this treatment modality *(Table 3)*.[21,24,27,29-31] When specifically looking at the implant location and implant survival within this review, no strong evidence was found; however, two systematic reviews have concluded that implant failure is higher in the maxilla than in the mandible in this patient cohort.[39,40] When assessing the best time to extract and replace the deciduous tooth in the adult patient prior to implant placement, there is no high-level evidence available to help guide this decision.[41] However, once the decision has been made to extract the deciduous tooth and replace with an implant, then placement of the implant is best carried out soon after the time of extraction or exfoliation to achieve maximal preservation of alveolar bone height and width.[42] It has been shown that the alveolar ridge narrows by 25% in the four years following extraction of retained lower primary second molars.[43] The availability of bone to accommodate a correctly angled implant fixture is a key concern when considering implant-based reconstruction of the hypodontia patient. It is common for patients to have narrowing of the bone or a lack of bone apical to the retained deciduous tooth creating a concavity of the alveolar process beyond the root apices and generating an 'hour glass' ridge morphology in cross-section.[18,32] The lack of a permanent tooth successor leads to this underdevelopment of the alveolus[41] which may also be exacerbated by root resorption of the retained primary tooth. There may also be pneumatisation of the maxillary sinus making implant placement into the posterior maxilla challenging. Addressing deficiencies in bone volume is a common pre-prosthetic necessity in this patient group and typically involves bone augmentation and sinus lifting procedures.[42] Within this study a variety of procedures were reported upon which included maxillary sinus floor elevation, guided tissue regeneration and bone grafting, with or without the use of a membrane barrier [23-26,28] with these adjunct procedures appearing to increase the risk of implant failure.[23, 26]

The current evidence available would suggest that resin bonded bridgework has a reasonable short to medium term survival rate of between 59% and 96.9% *(Table 2)*[18,19] with positive OHQoL being reported in this patient group *(Table 3)*.[20] Survival of such prostheses appeared to be lower in this study in comparison to current published data on resin-bonded bridge prognosis within the general population.[44] However the potential limitations of its long-term survival are offset due to its minimally invasive nature and ease of clinical application for replacing missing teeth in young patients with hypodontia in comparison to other tooth replacement options.[19]

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Unfortunately, there is very little literature reporting on the use and outcomes of conventional bridgework (fixed partial dentures), specifically in hypodontia patients. This is probably due to the fact that the majority of adult patients are treated early in adulthood, where abutment teeth are often unrestored with large pulp chambers and where preparation of the abutment teeth would be destructive of healthy tooth tissue. This risks exposure of the pulp during preparation and/or pulpal necrosis with its subsequent sequlae.[42] Use of conventional bridgework is generally limited to the older patient and where resin-bonded bridgework is contra-indicated by the presence of large restorations.[42] Although the reported survival of conventional bridgework in the general population is better than that of resin-bonded bridges,[45] it is still short in relation to the expected life span of a young adult patient.[42] Failure of conventional bridgework commonly occurs as a result of recurrent caries and loss of tooth vitality necessitating endodontic management.[42]

This lack of literature was also found when assessing outcomes associated with removable prosthesis in hypodontia patients, with Hobkirk *et al*,.[22] reporting poor outcomes in oligodontia patients treated with removable prosthesis and the need for frequent replacement. In general, removable partial dentures are usually applied to treat severe hypodontia (oligodontia) and often used as 'interim prosthesis before definitive treatment such as implant therapy can be carried out, particularly in children.[18] This has been shown by Lexner *et al*.,[46] who reported on children with hypodontia associated with ectodermal dysplasia and described successful use of removable prostheses, from the perspective of the patient, parents and dentist, with young patients adapting well to their prostheses and the prostheses being found to be well retained and stable.[46] Most patients, however, prefer fixed restorations as opposed to removable for definitive tooth replacement[42] and this is a common reason why few hypodontia patients are definitively restored with a removable prosthesis.

CONCLUSIONS

The key decision of whether to retain or extract and directly replace retained deciduous teeth in the adult patient is related to the prognosis of the retained primary tooth and its ability to provide the patient with appropriate function and aesthetics. Retained deciduous teeth can last well into adulthood; however, studies going beyond the third decade of life are lacking and these studies report a degree of unpredictability in relation to the long-term prognosis of such teeth. Where such teeth are compromised or unable to provide the desired function or aesthetics, extraction is warranted with subsequent replacement. Dental implants have been shown to be a highly successful long-term tooth replacement option with high patient satisfaction within this patient group, as have resin bonded bridgework albeit over the short to medium term whilst offering a minimally invasive tooth replacement solution. Other tooth replacement options such as conventional fixed bridgework and removable prosthesis are poorly reported within the literature which appears to be as a result of such tooth replacement options being unsought and undesired by patients.

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