

Restorative considerations in children with congenital dental anomalies

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LV Mkhonza¹, LM Sykes²

ABSTRACT

Children with congenital dental manifestations may have to endure challenges due to their abnormal tooth development, altered skeletal growth, compromised masticatory and speech function and poor aesthetics. This may further lead to psychosocial problems. These children may have deficient ridge height and volume in edentulous areas, missing teeth, malformed or malpositioned teeth and often the added complication of xerostomia. These anomalies complicate conventional treatment and have led clinicians to consider alternative restorative treatment options. Recent literature suggests that osseointegrated implant therapy may be a viable option for children

with congenital dental defects. However, studies are scarce, with no standardisation of success or survival criteria, and little long-term follow-up results. This paper explores the various restorative options and highlights the need for a multidisciplinary team approach.

INTRODUCTION

Dentofacial aesthetics may influence how individuals are treated by society. In children a deviation from the accepted dentofacial form can lead to bullying from contemporaries¹. Dental anomalies may have a psychological effect on the self-esteem of children and adolescents, most notably during this early phase of psychosocial development². Congenital defects, trauma and conditions causing oligodontia, aplasia and malocclusion can cause functional, aesthetic and psychosocial challenges for growing patients³. Tooth absence, excluding the third molars, can be divided into hypodontia, oligodontia and anodontia. Hypodontia is the absence of less than six teeth, oligodontia is the absence of six or more teeth (excluding the third molars), and anodontia is the complete absence of teeth^{4,5}.

Conditions affecting odontogenesis can also affect the underlying alveolar bone and, in extreme cases, the maxillofacial skeleton⁴. Oligodontia may be treated with removable complete dentures, partial dentures, overdentures, tooth-supported fixed prostheses or implant-retained prostheses. Conventional treatment to replace

Authors' information

1. Londiwe Mkhonza, BChD, Dip Implantology (UWC), Dip Dent (UP), Dentist, Department of Prosthodontics, University of Pretoria, South Africa
2. Leanne M Sykes, BSc, BDS, MDent, IRENSA, Dip Forensic Path, Dip ESMEA, Head of Department of Prosthodontics, University of Pretoria, South Africa
ORCID: 0000-0002-2002-6238

Corresponding author

Name: Prof LM Sykes
Email: Leanne.sykes@up.ac.za

Author's contribution

1. L Mkhonza 50%
2. LM Sykes 50%

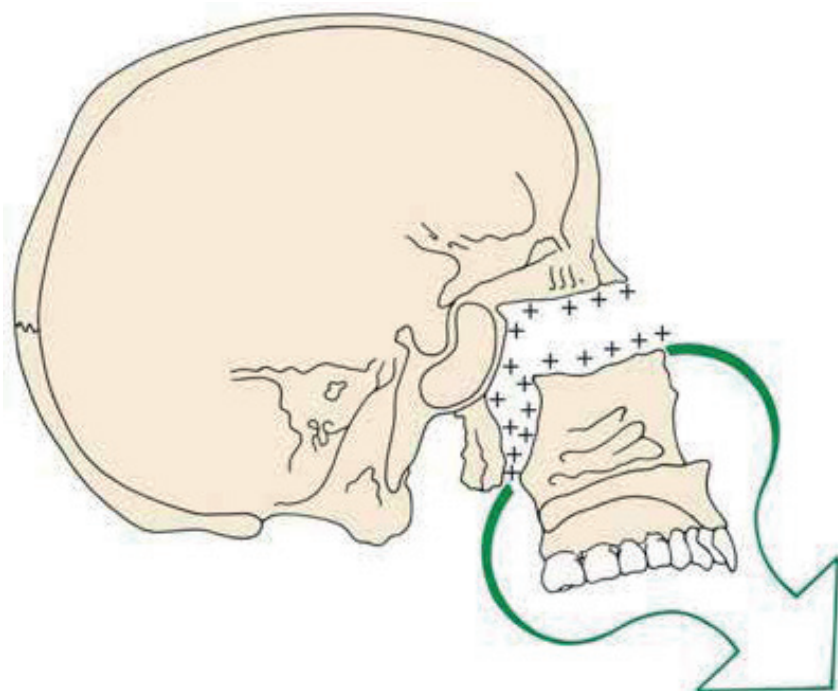


Figure 1. New bone added along the lines of the maxillary sutures⁷

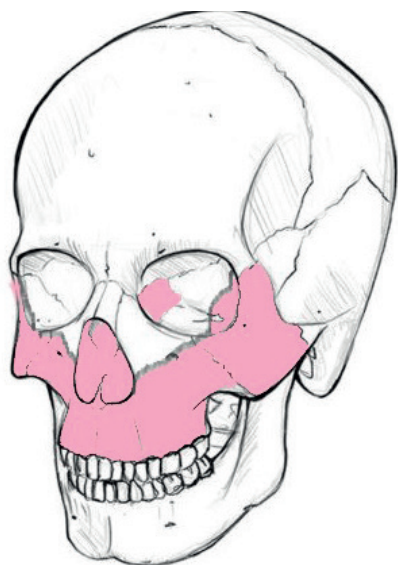


Figure 2. Areas of resorption (yellow)

missing teeth and restore function and aesthetics may pose difficulties in those cases where patients also have severely resorbed, atrophic or knife-edge alveolar ridges in the edentulous areas. It may thus be pertinent to consider implant therapy in children with congenital dental anomalies as opposed to conventional treatment modalities⁶.

Growth of the maxilla and mandible

Growth in the maxilla is due to apposition of bone at the sutures that connect the maxilla to the cranium and cranial base and by surface remodelling. This growth moves the maxilla forwards and downwards relative to the cranium and cranial base, with the forward movement facilitated by the growth of the cranial base and downward movement by growth at the sutures. After the age of 7, sutural growth is solely responsible for the growth of the maxilla (Fig 1)⁷.

As the maxilla grows forward the anterior part simultaneously gets resorbed, as is highlighted by the yellow areas (Fig 2)⁸. Simultaneously, remodelling of the palate moves it downwards and widens it. This is achieved by bone removal from the floor of the nose and bone apposition on the surface of the palate (Fig 3)⁷.

Mandibular growth

Growth of the mandible was originally described in relation to the cranial base, but has more recently been analysed using data from vital staining experiments. In the latter, 2% Alizarin Red S, a dye with a high affinity for calcium, is injected intraperitoneally or intravenously to reveal areas where there is a high calcium concentration, thus revealing where bone is being deposited and resorbed in the mandible. If the cranial base is used as a reference, then the chin appears to move downwards and forwards (Fig 4A). However, contrary to this theory, vital staining data revealed minimal changes in the body and chin area, with only the ramus, condyle and coronoid processes having substantial growth in the mandible (Fig 4B). Thus, staining suggests that the mandible grows longer by apposition of bone in the posterior part of the ramus, with simultaneous

removal of large amounts of bone from the anterior ramus. Essentially the body of the mandible grows longer as the ramus moves away from the chin⁷. These observations are important to bear in mind if fixed restorations or implants are to be considered in children, as mandibular growth patterns will affect their final position⁷.

Definitions of common congenital tooth abnormalities

Anodontia – is the complete absence of tooth development⁸

Hypodontia – is the absence of development of less than six teeth, excluding third molars⁵

Oligodontia – is the absence of development of six or more teeth, excluding third molars⁸

Malformation – refers to any tooth form outside the normal range of size and shape. It includes macrodontia, microdontia, gemination, fusion and concrescence, among others⁸

Malposition – is the incorrect buccolingual or mesiodistal tooth position or angulation of a tooth⁸

Transposition – refers to a normal tooth erupting in an incorrect position⁸

Delayed eruption – is when the tooth has not emerged within a year of its normal range or when the tooth has not emerged even though 75% of the root has formed⁸

Impaction – refers to teeth that stop erupting before they fully emerge into the oral cavity⁸

Ankylosis – is where eruption stops after emergence of a tooth into the oral cavity, but only when this is due to union of the tooth cementum or dentine to the alveolar bone⁸

Syndromes associated with congenital dental anomalies

Odontogenesis is a complex process between the enamel epithelium, underlying mesenchyme and signalling factors (FGF, SHH, BMP and Wnt). Many factors, including physical obstruction, disruption of the dental lamina, space limitation, functional abnormalities of the dental epithelium, failure of initiation of the underlying mesenchyme and environmental influences such as trauma, infections, ionising radiation, drugs and hormonal influences may affect odontogenesis^{5,9}. Any interruption in tooth development may affect the morphology, number or differentiation of teeth^{10,11}. Neville et al (2016)⁸ reported on more than 50 congenital syndromes associated with oral and dental manifestations. However, Bohner et al's 2019 systematic review highlighted cleft palate, congenital aplasia, ectodermal dysplasia and non-syndrome agenesis as the most common disorders associated with congenitally missing teeth³.

Oral characteristics of oligodontia

Dentoalveolar characteristics commonly associated with oligodontia include:

1. Occlusal disturbances such as crossbite, deep bite, steep maxillary incisal inclination, abnormal attrition, narrow alveolar ridge and vertical defects^{9,12}.
2. Loss of vertical dimension due to less growth in the maxilla compared to the mandible, leading to a class III skeletal relationship¹³.
3. Eruption disturbances including overeruption of teeth opposing edentulous areas, impacted Ds and Es, infraocclusion of Ds and Es, ectopic eruption, delayed eruption, rotation of teeth, multiple diastemas, severe attrition of retained primary teeth, altered tooth morphology, microdontia, conically shaped teeth and any number of other malformations^{9,12}.

Psychological effects of tooth loss in children

Bzoch stated “the development of a child takes place within the context of interactions with others”, and that “early developmental events can influence later behaviour”. In children with congenital defects, their life experiences may be affected by their disorder as well as the number and types of associated disorders they must bear¹⁴. Congenital oro-facial defects can have an impact on growth and development, speech and hearing, mastication, sight and smell and aesthetics. These in turn can affect the mother-child relationship as well as the child’s interactions with others. Furthermore, these children often need to attend many visits to hospitals and clinics, where they will encounter a number of strangers and undergo various procedures, which can all be frightening and stressful for them. At the same time, they miss out on normal childhood activities and schooling, which can impact their social and academic development and their oral health related quality of life (OHQoL).

OHQoL can be assessed using the Child Perceptions Questionnaires (CPQ). There is a short and a long version of this questionnaire available. Both questionnaires divide children into age categories from 8-10, 11-14, 15-17 and 18+. And both are subdivided into 4 parts that address oral symptoms, functional limitations, emotional wellbeing and social wellbeing. Each question can be answered on a 5-point Likert scale as either never (=0), once/twice (=1), sometime (=2), often (=3) and everyday/almost every day (=4). Final scores can range from 0-148 and 0-64 in the long and short questionnaires respectively. The higher scores correlate with a higher impact of conditions on OHQoL¹⁵. Liang et al (2010) compared the psychosocial impact of

hypodontia with moderate to severe dental malocclusion on children. They reported that patients with moderate to severe malocclusion and those with hypodontia both had a similar psychosocial status¹. Wogelius et al (2011) found an interesting observation that children with fixed orthodontic treatment had a higher CPQ value (22) than those with healthy oral conditions (9), cleft lip and palate defects (9) and those with rare dental anomalies¹⁶.

Conditions associated with hypodontia or oligodontia may negatively affect the OHQoL in young patients who may fear exposing their removable dentures to their peers, being bullied due to their differences or having a fear of dentists¹⁶. Furthermore, as children grow they become more aware of their oral health status, especially girls, who tend to have higher CPQ scores than boys¹⁵. Issues of bullying, embarrassment or shame associated with oligodontia affect the psychological and social wellbeing of children, especially in the important adolescent years.

To curb these negative aspects and emotions, treatment of these patients should aspire to improve aesthetics, promote craniofacial growth, improve the profiles, address masticatory and speech function and, to some extent, lead to better psychosocial development¹⁷. An added complication is that some conditions that affect odontogenesis may also affect the salivary gland development and function, leading to xerostomia, which has several negative repercussions on the teeth and oral environment. Edentulous areas often have narrow, sharp or deficient ridges. When this is compounded by other dental manifestations including missing teeth and xerostomia, restorative treatment becomes that much more difficult. Once again, this will impact negatively on

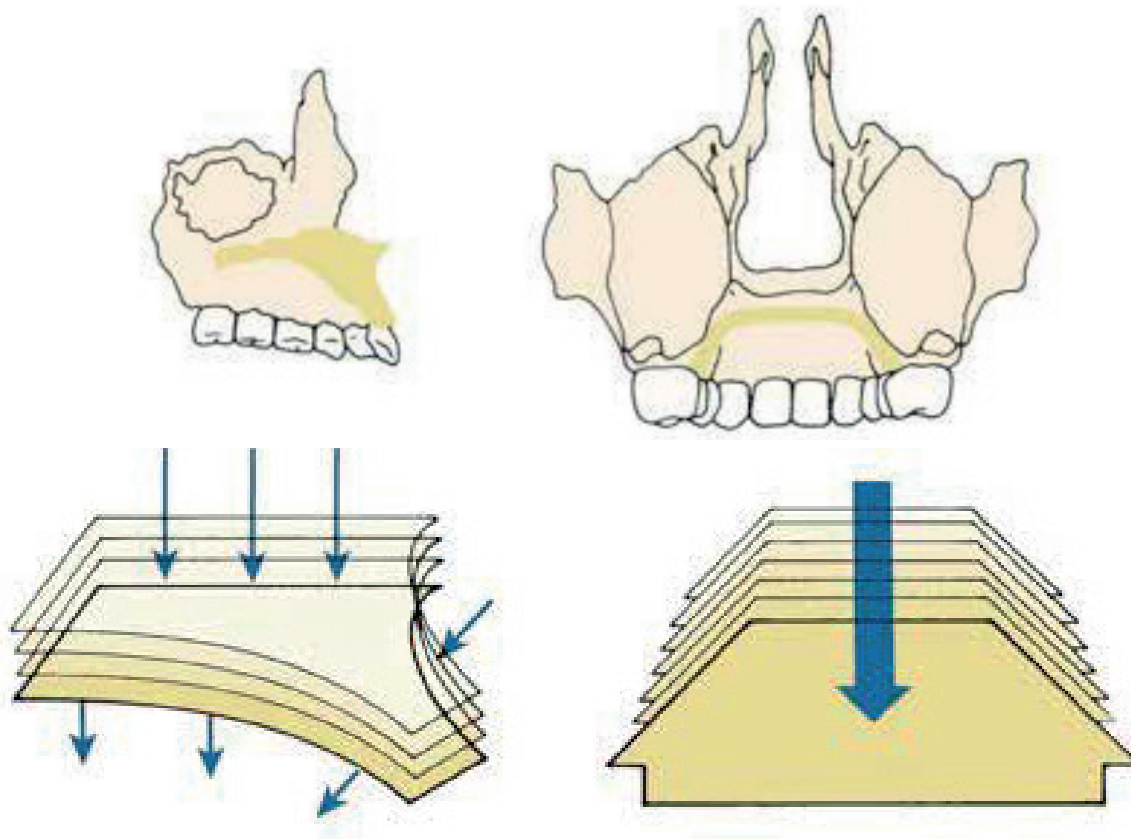


Figure 3. Bone apposition on the surface of the palate highlighted in yellow⁷

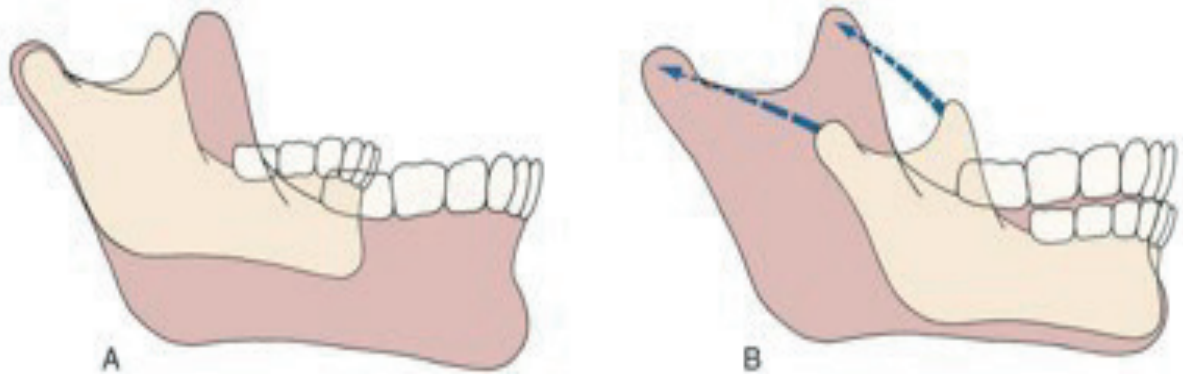


Figure 4. Bone growth in relation to the cranial base (A), and as assessed using Alzarin Red S dye (B)⁷

the child's OHQoL⁶. In recent years, more clinicians have begun to consider using fixed tooth supported restorations or osseointegrated implant-retained prostheses in children with congenital dental defects. These options may improve self-confidence, perceived quality of life, psychosocial health and alleged social acceptability^{6,17}. Careful consideration of the feasibility, timing of implant placement, future growth and potential restorative or implant failures should be made prior to embarking on any fixed or implant-supported prostheses in children with congenitally dental anomalies.

Treatment considerations

Almost all children with congenital tooth anomalies or absence will need some form of prosthodontic intervention, as well as any number of other treatment modalities. It stands to reason then that the overall management of each child should be coordinated by the treating dentist/prosthodontist, as part of a multidisciplinary team. The objective of this team approach is to “bring the child to a point where they do not differ significantly from their peers in terms of health, education or ability to interact socially”¹⁴. Good communication between the different specialities is essential to ensure the patient gets the appropriate treatment at the correct time to achieve the best possible outcomes in terms of speech, language and hearing, facial appearance, dento-occlusal development and oral function. Each phase of treatment and treatment modality impacts on the others, and thus requires careful co-ordination. For example, in cleft palate patients if timing of surgery is not correctly planned, too early closure of the palatal defect could impede dento-occlusal development and orthognathic growth¹⁴. Similarly, dental prostheses may address aesthetics and masticatory concerns but could affect speech and articulation at a crucial time in the child's social development. Thus, all aspects of the treatment regime need to be pre-planned and integrated rather than individually administered. Figure 5 is a diagrammatic representation proposed by the authors of how the various role players may interact. The vertical pillars represent the family; the prosthodontist/restorative dentist and technician; and various crucial allied disciplines including social workers, psychologists, geneticists, speech and hearing therapists, surgeons and nurse aids. The horizontal arms illustrate key dental disciplines that may be needed individually or in combination with each other.

1. Assessment and planning phase

This stage requires cephalometric, panoramic and hand wrist radiographs as well as articulated study casts as diagnostic aids³. During this phase of treatment, the clinician should

assess the age and developmental stage of the child, the anatomy of the hard and soft tissue, the number and location of missing teeth, presence of permanent successors, arch shape and the condition the child is suffering from^{3,18}. The quality and quantity of bone available in edentulous areas should also be evaluated as some areas may need bone augmentation to help support a prosthesis or to facilitate implant placement and integration¹⁹.

Any previous or currently worn prosthesis should also be examined to visualise arch relations and facial dimensions, and to identify any shortcomings with the hopes of improving these in the future. The assessment phase also includes planning to blueprint a phased approach that will address both functional and aesthetic needs. This will entail use of above-mentioned radiographs and diagnostic wax ups on articulated study casts. These help clinicians explore the various possible treatment options and facilitate communication between patient and clinician, and between members of the treatment team⁹.

2. Orthodontic phase

Malocclusion, rotated teeth, tilted teeth, microdontia and multiple diastemas are a few common traits seen in patients with oligodontia. These patients may require initial active orthodontic treatment¹², and often need prolonged use of retainers⁴. Orthodontics can help create or maintain space in the arches and correct adverse tooth relationships. Orthognathic surgery may also be needed to correct the jaw relationship⁶. It is imperative to remember that oral rehabilitation in growing patients must not only focus on immediate restoration of facial dimensions but must also facilitate craniofacial growth to obtain the ultimate ideal jaw relationship – if possible – as this will result in more optimal treatment in the adult patient. Jaw relationships may be improved using maxillary dentures with expansion screws to try to promote sutural growth¹³.

3. Surgical phase

The goal of this phase is to preserve bone and provide treatment that will benefit the patient immediately and in the future. The most crucial aspect is to try to preserve bone by retaining primary teeth for as long as possible²⁰. This may also be achieved by autotransplantation of teeth when possible. In children this procedure can be successful if the tooth apices are still open as this allows for healing with a functional periodontium. This ligamentous attachment enables the tooth to participate in growth of the alveolar ridge and allows for orthodontic tooth movement²¹. While auto transplants are only successful in children and adolescents,

deciduous teeth may, at times, be retained well into adulthood²¹. Although implants have not been widely used in children with congenitally missing teeth, Terheyden and Wüsthoff (2015) noted that implant supported prosthesis had a better outcome and higher survival rate than tooth autotransplants. Thus, the possible use of osseointegrated implant therapy warrants further debate²¹.

4. Prosthetic phase

This phase is important because of the dynamic oral environment, where there are many factors to consider. The first is the arch length. Bu et al (2008) found that in children with oligodontia the maxilla was on average 4.40mm shorter than its dentonormative counterpart, while the mandible was approximately 2.80mm shorter. The next factors are those of prosthesis retention, support, bone loading and follow-up treatment or maintenance. Kearns et al (1999) noted that loading of the basal bone at an early age leads to gradual resorption of the basal bone. Thus, any teeth that could viably be used to retain and support a prosthesis should be considered. Even small, malformed or malaligned teeth may be prepared for telescopic crowns and used to help support an overdenture. In the mandible, these are particularly useful to inhibit tilting of the mandibular denture and make cleaning easier than a fully fixed prosthesis¹⁷. Follow up should be every 3-6 months until growth is complete as new dentures may be needed to accommodate the growing jaw²². These appointments should monitor crown discolouration, gingival impingement, occlusal interferences and prosthesis stability¹⁶. In addition, radiographs should be taken every 2-3 years, with adjustment of restorations being made when needed, until growth is complete^{3,4}. (Note: The same principles for overdenture abutments will apply with regard to growth, monitoring, adjustment and maintenance when osseointegrated implant retained restorations are to be considered.)

In children with ectodermal dysplasia salivary gland hypoplasia is common leading to xerostomia and difficult adaptation and retention of removable dentures^{6,21}. Conventional treatment poses challenges due to previously mentioned absence of teeth, conical shape of existing teeth, knife-edged alveolar ridges and compounding xerostomia. This all results in poor denture retention, instability, pain on the ridges and mucosa, speech and masticatory difficulty and poor aesthetics²³. In these children one may thus begin to explore the possibility of placing endosseous implants. However, Terheyden and Wüsthoff (2015) expressed caution when placing implants in children with ectodermal dysplasia, as they often have delayed development in comparison to their peers and this could lead to an unfavourable final position of the implants.

4.1. Considerations for the use of osseointegrated implants in children with congenital dental anomalies

Implant treatment in children has historically only been considered if all other options have been exhausted and have failed^{24,25}. However, with the emergence of many new implant systems, mini-implants and possibility of custom-made implants, this is an area of dentistry that warrants more research and deliberation. Implant treatment planning needs to consider factors such as gender and age of the patient, skeletal maturation, available supporting and retentive structures, proposed implant location and the current and future needs of the patient²⁴. In the maxilla, growth may cause implants to develop diastema between them, to become infra-occluded, to end up in the sinus or to lose stability due to loss of buccal bone²⁶. It has been advised that 10 years is the minimum age that implants be placed in the anterior maxilla and only after the age of 15 in girls and 17 in boys on the posterior maxilla to help avoid later growth-related implant complications^{3,24}. In the

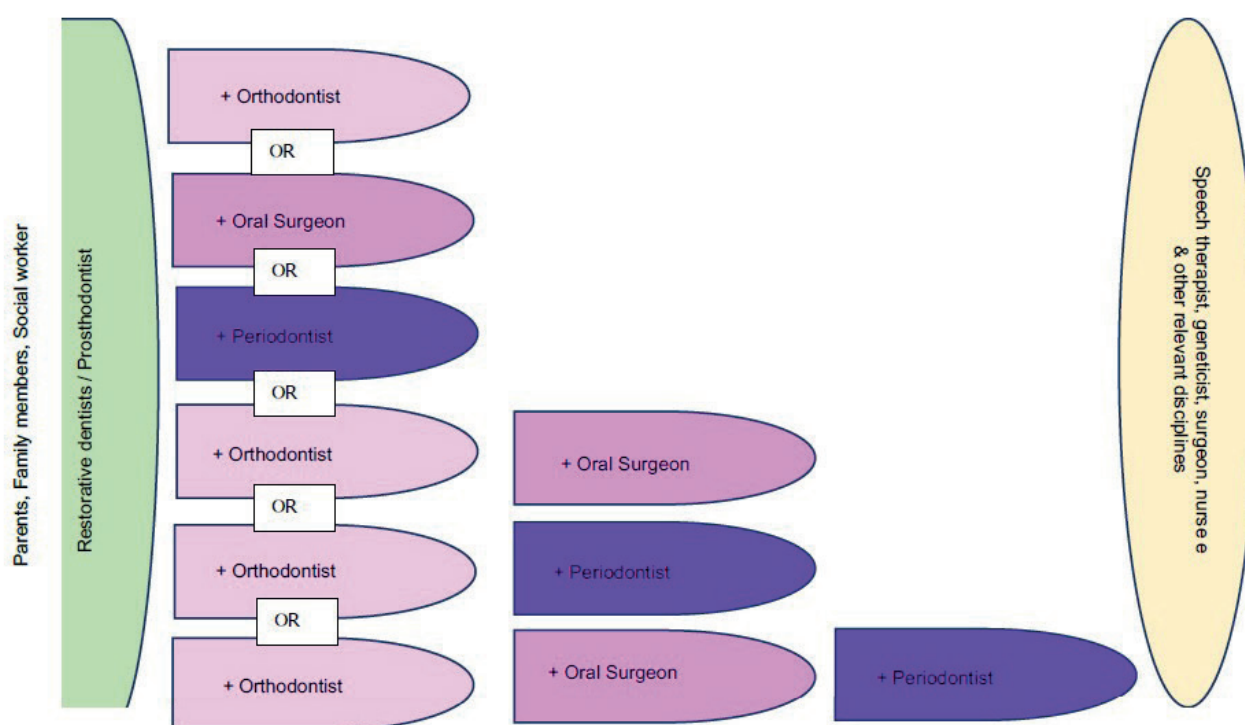


Figure 5. Diagrammatic representation of multidisciplinary team interactions (Proposed by LM Sykes and LV Mkhonza)



Figure 6. The Onplant implant.

maxilla placement of implants more coronally may help prevent infra-occlusion of implant crowns at a later stage, but could complicate the occlusal and aesthetic needs of the immediate restorations. In patients where the maxilla is unsuitable for implant placement, zygomatic implant placement may even be considered¹⁹.

In the mandible, the interforaminal region is the best site for implant placement, as there is little growth in this area after 6 years of age in the dentate mandible and after 3 years in a mandible with an edentulous interforaminal region^{3,17,19,24}. Posterior mandibular implant placement should be avoided until completion of skeletal maturation. This is because of the unpredictable nature of the anteroposterior dimensions of the mandible²⁴. It must also be noted that a high number of implants in children are lost in the healing phase²¹.

4.2. Contraindications to implant placement

Contraindications include use of implants in children with systemic conditions such as uncontrolled diabetes, vascular conditions, active chemo/radiation therapy and psychiatric disorders; those with metabolic disorders related to wound or bone healing; arches with insufficient bone height or width for implant placement; children with uncontrolled parafunctional habits (for example bruxism, lip or nail biting); uncooperative patients and those who fail to follow and maintain proper oral hygiene practices²².

4.3. Advantages of implant therapy

The main advantage is the perceived improvement in OHQoL due to implant's potential to provide more successful treatment in patients who have had difficulties with conventional treatment^{1,19}.

4.4. Disadvantages of implants in children

In children with congenital dental anomalies the main disadvantage is that implant survival is lower in augmented areas. Vertically augmented bone is particularly susceptible to resorption, development of peri implant pockets and soft tissue recession within the first year after implant placement¹⁹. In addition, the survival rate of implants in children is lower than that reported for adults and geriatrics. Furthermore, growth in children can lead to changes in implant position as already mentioned⁹.

4.5. Possible implant types for use in children

Based on the available space, ridge volume and the age of commencement of treatment, different implant systems can be used. Mini implants have been used in orthodontics to help anchor and support active appliances. Their diameters range from 1.8mm to 2.7mm and were designed to be used in areas where there was limited bone²². Mini implants have a smaller surface area and roughness, and this decreases the likelihood of osseointegration and long-term survival. Their main advantage is that they may be loaded immediately if primary stability is achieved or after 6 months if not initially stable¹⁶. These implants should be large enough to provide retention and stability, but small enough to allow bone growth. Literature suggests that mini-implant supported prostheses can be functionally stable for up to 8 years¹⁶, but that they should be replaced with standard implants once bone growth is complete²⁷. Further advantages are that due to their small diameter they are easier to insert than standard implants, are cheaper, do not need submerged healing, and can often be immediately loaded¹³. Some authors also believe that they could preserve alveolar bone volume and may even stimulate remodelling of bone.

Standard implants maybe used for in children if their bone volume is sufficient or if augmentation is possible. Kearns et al (1999) reported high success rates in a series of patients where a total of 36 Titanium plasma-sprayed, press-fit cylindrical implants (3I Implant Innovations) and 5 titanium screw-type implants (Nobelpharma) were placed in the maxillae and mandibles of 6 children²³. Worsaae et al (2007) placed 283 Brånemark (Nobel Biocare) and Astra (Astra Tech) implants in 46 children, and also showed high success rates (98%)¹². Filius et al (2014) successfully placed 8 Strauman SLA implants size 10-14mm in the mandibles of children with congenital defects. All these studies seem to suggest a high survival rate for standard implants in children; however, success and survival criteria were not clearly defined and long-term follow-up results not reported on²⁸.

Another rather unconventional type of implant to consider is the Onplant from Nobel Biocare (Fig 6)¹⁷. It has a disc-like design that can help overcome bone deficiency in the maxilla. Heuberer et al (2011) used it successfully for anchorage of an upper overdenture¹⁷. In 2019, Kamatham et al stated that

there was insufficient evidence on the use of implants in children to establish any definitive guidelines²⁹. To date there is still a paucity of literature and long-term follow-up studies reporting on the use of implants in children, most especially those with congenital dental anomalies.

Conclusion

The type and severity of congenital dental defects can influence both the psychosocial development and oral rehabilitation in children. If conventional treatment has been unsuccessful, it may be prudent to consider implant therapy. However, the timing, placement, monitoring and adjustments, and anticipated future needs must all be carefully considered and planned for by a multidisciplinary team. At present there is little literature regarding recommended implant protocols. Neither are there many long-term follow-up studies of survival and success rates, or the psychological impact and benefits that may be derived from their use. There is also very little data on failed implant treatment in children and how these could be addressed. The aim of all dental and restorative treatment in children with congenital dental manifestations should be to restore function and aesthetics and to “bring the child to a point where they do not differ significantly from their peers in terms of health, education or ability to interact socially” – Bozch 1997¹⁴

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CPD questionnaire on page 372

The Continuing Professional Development (CPD) section provides for twenty general questions and five ethics questions. The section provides members with a valuable source of CPD points whilst also achieving the objective of CPD, to assure continuing education. The importance of continuing professional development should not be underestimated, it is a career-long obligation for practicing professionals.

