Orthodontic management of orofacial problems in young people with impairments: review of the literature and case reports

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Summary. *Objective*. The aim of this paper is to present a series of cases illustrating possible options for the management of malocclusions in a group of young people with impairments.

Design. The study took the form of a case series.

Methods. A review of the literature is presented and a series of illustrative cases are shown to indicate that orthodontic treatment is possible in patients with a range of impairments

Results. Five children with varying intellectual and/or physical impairments, and a malocclusion that resulted in trauma or increased disability, are presented

Conclusion. The risks and benefits of the procedures, and the anticipated oral health outcomes, need to be considered carefully in this group of children.

Introduction

The past 10–15 years have seen significant changes in many countries in the more equitable access to health care for people with impairments. Following legislative changes in the UK, many people with learning and other disabilities are now actively integrated in society, following specifically designed educational programmes, or are in employment.

Physical appearance can significantly influence educational attainment and social interaction, and the mouth appears to be of importance in determining attractiveness, with malocclusion having important social and psychological effects [1,2], as well as imposing functional limitations like impaired ingestion of food stuffs [3]. Attention has been drawn to the consequence of an unstable occlusion, in not

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treating orofacial dyskinesia (abnormal involuntary movements of the facial muscles or muscles of mastication) early in children with Down syndrome, either by myofunctional or orthodontic therapy [4]. In some children with physical impairments, selfinduced trauma can be averted by the judicious use of orthodontic appliances [5]. Mohlin and Kurol made the point that the assessment of treatment need cannot be based on orthodontic indices of need, but depends more on the consequences of the malocclusion for the patient [6]. In another paper, on the extent to which deviations from an ideal occlusion constitute a health risk, the same authors underlined the importance of providing orthodontic treatment to people with disabilities that promotes normal growth and occlusal development [7].

Malocclusion has been found to occur more frequently in children with disabilities than in healthy children [8–10]. The conclusions of a survey of 381 children in six South African schools for children with disabilities using Summers Occlusal Index [11] were that 74% of children required treatment [12].

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Of the 41% of children who were shown, from a questionnaire to parents, to be interested in orthodontic treatment, only 0.5% were planning to seek such care. In a similar survey of 124, 6–18-year-olds with learning disabilities in Ibadan, Nigeria, the authors demonstrated that 58% had a significant need for treatment using the Dental Aesthetic Index [13]. In impairments like Down syndrome, there are specific occlusal features, such as anterior open bite, posterior cross-bite and Class III malocclusion, which are a consequence of the relative underdevelopment of the maxilla, and these may merit orthodontic correction [3,14].

Successful orthodontic treatment requires the active cooperation of the patient, compliance with the wearing of appliances and the ability to maintain an adequate standard of oral hygiene [15]. These requirements may potentially be a problem for children with impairments, especially those with learning disabilities, because the child's understanding of treatment may be limited, their ability to understand and learn new techniques may be impaired, and their manual dexterity is often poor.

In 1967, Jackson was amongst the first to broach the issue of orthodontic treatment for children with disabilities [16]. He suggested that treatment should still be undertaken, although 'ideal' results may not always be possible. Jackson acknowledged the difficulties inherent in the complex issues surrounding elective dental care for children with impairments.

A retrospective review by Chaushu and Becker [17] commenting on the use of behaviour ranking scales in order to ascertain a child's suitability for orthodontic care, concluded that these [18,19] are not suitable for determining the most appropriate orthodontic treatment strategy. It was Chaushu and Becker's view that the specific problems encountered, such as an enhanced gag reflex, uncontrolled movements, inability to submit to prolonged dental treatment procedures, drooling and the possible need for general anaesthesia as an adjunct to care, were more reliable predictors of favourable outcomes. They suggested a scoring system, based on these retrospective observations from the patients' charts, to aid in the assessment of the needs of this group of patients. The most frequently occurring problem was that of the child's inability to remain still for long enough to enable band and bracket placement, and this led Chaushu and Becker [17] to concur with the view of Chadwick and Asher McDade [20] that treatment under general anaesthesia was indicated for some phases of orthodontic treatment plans in children with impairments. Other authors have acknowledged, in a review of the orthodontic treatment of four children with disabilities, that limitations in compliance necessitated a pragmatic approach to treatment planning [2].

Before embarking on orthodontic treatment, careful selection of patients is vital and a high standard of oral hygiene should be demonstrable. For patients with a physical or intellectual impairment, greater reliance may have to be placed on carers for the maintenance of satisfactory oral hygiene. Children with disabilities may be more apprehensive than orthodontic patients who are not impaired in the same way [21], and therefore it is important to spend even more time in establishing a good relationship between patient and orthodontist. Once this has been achieved, these children can be extremely compliant.

Because of the patient's initial anxiety, sedation or even general anaesthesia may be required as an adjunct in order to accomplish some or all of the treatment. Although such elective procedures under general anaesthesia raise ethical questions, the parents or carers may exert considerable pressure for some form of treatment that would improve the patient's acceptability amongst their peers and enhance their quality of life [20]. It is important to assess the oral health gain that would arise in the context of the parents' or guardians' expectations and desires, and to offer to provide orthodontic care only when the benefit to the child significantly outweighs the risk [22]. Becker et al. [23] found that fixed appliances were more difficult for children with disabilities to tolerate compared with the use of removable appliances. By contrast, Chadwick and Asher McDade [20] recommended the use of fixed appliances, even for simple tipping mechanics, since, they maintained, the tooth movements can be accomplished more simply and rapidly.

Becker *et al.* [23] reported the results of a study of patients with disabilities and their management of orthodontic appliances. They found that the two main problems encountered were maintaining a high standard of oral hygiene and difficulties in attending for treatment. Overall, the majority of parents were satisfied with the outcome of treatment, and felt that there was a considerable improvement in their child's appearance and function. They considered that the added responsibilities placed on them to ensure a successful outcome, were negligible or tolerable.

In a review of four children with disabilities, differences in treatment need were highlighted in this group compared with nondisabled children [2]. No details were given of the degree of impairment of each subject, the authors only classifying the children as having 'good cooperation' or 'good verbal comprehension' in two of the four cases. Whilst significant improvements, as judged using the peer assessment review (PAR) index, had been possible using a variety of appliances, not all orthodontic tooth movements were maintained. In children with inadequate cooperation, the authors felt it was more important to aim for an improvement in orofacial function, with the aid of orofacial regulation therapy, rather than correction of the malocclusion alone. This paper underlines the difficulty of objectively assessing behaviour, in anticipation of orthodontic treatment, as compared with the more objective indices available to assess malocclusion.

In many parts of the UK, there is limited availability of specialist orthodontic treatment for patients with disabilities. Such care is usually undertaken by orthodontists in a dental or regional hospital setting with a paediatric dentist, if available. The following case reports illustrate the range and severity of both malocclusion and disability that can be managed, and highlight the issues raised by the provision of such care.

Case reports

Case 1

GC was a 13-year-old girl with cerebral palsy as a consequence of surgery as a neonate for aortic stenosis. She had profound learning disability and epilepsy. GC presented for an orthodontic assessment because of severe, self-inflicted trauma. The patient's lip lesion had been monitored by a dentist for a period of months, and he referred her for a biopsy when the lip lesion failed to heal (Fig. 1). The biopsy revealed no significant pathology, only the features of a chronic ulcer that was probably related to a crush injury of the muscles. Placement of upper and lower soft splints, retained by dental cement, failed to prevent the ongoing self-inflicted trauma (Fig. 2). After occlusal analysis, showing a Class II Division 1 malocclusion with a lower lip trap that was exacerbating the lip trauma, a decision was taken to offer orthodontic care in order to procline the lower incisors and so prevent further trauma.



Fig. 1. Case 1: ulcerated area on the lower lip as a consequence of self-inflicted trauma.

Under outpatient general anaesthesia, the LLE (75) was extracted, the cement used to retain the splints removed from the upper and lower incisor and canine teeth, and upper and lower alginate impressions were recorded. A lower fixed appliance was placed with 0.018 preprogramed brackets on the lower first premolar teeth (34, 44) and GAC (GAC International, NY, USA) brackets were applied to the lower six anterior teeth, and a 018 Australian alignment arch wire was fitted (Fig. 3). This was all provided under 500 mg Amoxicillin IV with a follow-up oral dose 6 h later. At review, 6 days later, the lip ulcer was healing well, but a small ulcerated area had appeared opposite the LR3 (43). The parent was shown how to apply white wax to the bracket opposite this area and 200 mL 0.15% benzydamine hydrochloride spray was prescribed for application to the ulcerated area. The arch wire was changed at approximately 6-week intervals without recourse to sedation. The lower fixed appliance was debonded 10 months later and a bonded retainer was fitted. This was undertaken at the same time as the removal of a retained primary canine (53) and a carious first permanent molar (26). At review, the ulcerated area opposite the upper right lateral incisor (12) was



Fig. 2. Case 1: same view as in Fig. 1, showing poor healing following an excisional biopsy consequent on the patient removing the sutures. The upper and lower Druformat splints were cemented into place to avoid self-mutilation.



Fig. 4. Case 2: upper sectional fixed appliance in situ.



Fig. 3. Case 1: lower fixed appliance put in place to align the teeth in order to prevent self-inflicted injury.

healing and the latter was smoothed with a Soflex disc.

Case 2

SSV was a girl with Rubinstein–Taybi syndrome. This condition was first documented in 1963 when Rubinstein and Taybi observed the combination of broad thumbs and halluces, characteristic craniofacial dysmorphism, growth retardation, and learning difficulties. Oral and dental features include: talon cusps, a high arched palate, mild micrognathia, and less frequently, bifid uvula, submucous cleft, bifid tongue, macroglossia, short lingual frenum, natal teeth and thin upper lip.

SSV had a high, narrow upper arch, a Class III incisor relationship, and severe anterior crowding in

both the upper and lower arches. The patient had moderate learning difficulties and could not cope with routine dental treatment. She had required general anaesthesia for clinical examination and restorative treatment as an inpatient since the age of 7 years. At the age of 12, a provisional orthodontic plan was made to extract the buccally placed upper canine teeth (13, 23) as well as the LL1 (31) in order to relieve crowding and then align the upper arch. The extractions were carried out under general anaesthesia and the UL6 (26) was removed at the same time because of extensive caries. At a review appointment, the patient had a fixed appliance placed on 5421 124 (Fig. 4). The patient was now very cooperative and keen to maintain good oral hygiene. The fixed appliances were maintained for 6 months until levelling and aligning of the upper arch was obtained.

Case 3

CD was a girl with moderate intellectual impairment, who was very anxious and uncooperative, and had recently been diagnosed with Prader–Willi syndrome. Prader–Willi syndrome is a condition that usually results from deletion on chromosome 15 and is of paternal origin. The presenting features are predominantly obesity associated with hyperphagia, diabetes, crypotorchidism, strabismus, developmental delay and moderate learning difficulties with behavioural problems. CD had had repeated treatment under general anaesthesia as a child for routine dental examination and treatment.

The patient had a Class II Division I malocclusion with an increased overjet of 8 mm, an anterior open bite and incompetent lips.

An orthodontic treatment plan was embarked upon with the extraction of all four carious first permanent molar teeth, and placement of upper and lower fixed appliances for levelling of the arches. This initial treatment was undertaken under general anaesthesia. Thereafter, the patient was seen regularly in the outpatient clinic for the adjustment of the appliance and was happy to receive treatment on a regular basis with no adjunct other than appropriate behaviour management. One year later, when the appliance was to be removed, CD was rather anxious about having treatment under general anaesthesia and was also not happy at the prospect of any form of sedation. A choice was made for oral sedation in the form of Midazolam 0.5 mg kg⁻¹ body weight with routine monitoring throughout the procedure. The patient refused to swallow the oral medication made up as a solution with paracetamol ($250 \text{ mg} 5 \text{ mL}^{-1}$). However, she agreed to the removal of the fixed appliance, but unfortunately, the patient was not able to tolerate impressions for the fabrication of a removable retainer. A further appointment was made for routine restorative treatment, a scale and polish, and dental impressions for retainer fabrication, all under general anaesthesia, the following day.

At review, one month after placement of the removable retainer, CD had not been able to tolerate it and there was slight relapse of the UL2 (Fig. 5). Her oral hygiene was only fair, with generalized marginal gingivitis.

Case 4

JA was a 12-year-old boy with a spastic form of cerebral palsy and moderate learning difficulties.

His medical history included seizures controlled by Epilim. The patient had a history of self-inflicted trauma to the lower lip over a period of 18 months. A number of attempts to stop or reduce the lip trauma had been made using soft splints, all with little success. He had a Class II, Division 1 malocclusion with an increased overjet of 9 mm, and crowding of the upper and lower incisors (Fig. 6).

In discussion with JA and his parents, a treatment plan was made with the aim of reducing the overjet, and relieving the upper and lower crowding. In the lower arch, the first premolars (34, 44) were extracted to allow spontaneous alignment of the incisors. In the upper arch, the palatally placed upper lateral incisor teeth (12, 22) were selected for extraction in order to facilitate tooth movement, using a fixed appliance.

Because of the extent of his intellectual impairment and uncontrolled movements, as well as the need for routine restorations, an upper fixed appliance was fitted under general anaesthesia (Fig. 7). Straight wire brackets (0.018) and bands were placed on the teeth in the upper arch following the extractions. A nickel titanium arch wire (0.16×0.22) was placed, along with power chain, from molar to molar to initiate space closure and overjet reduction. The 0.16×0.22 nickel titanium arch wire was chosen to reduce the chair-side time at subsequent visits.

Over the next 5 months, the power chain was changed twice in the dental chair while JA was conscious. The overjet reduced to 2 mm in this interval (Fig. 8). The fixed appliance was removed under general anaesthesia, and an immediate splint/retainer was put in place and kept *in situ* for 6 months.

Following treatment, there were no further episodes of self-inflicted lip trauma.



Fig. 5. Case 3: at completion of treatment using fixed appliances to align, accepting the slightly increased overjet and anterior open bite. Decalcification is present due to problems with oral hygiene during treatment.



Fig. 6. Case 4: original Class II Division 1 malocclusion and lip trauma.



Fig. 7. Case 4: fixed appliances being placed under general anaesthetic.



Fig. 8. Case 4: reduction of overjet, reducing the possibility of lip trauma.

Case 5

KG was an 11-year-old girl who had been diagnosed with cri-du-chat syndrome. The classic features of this condition are a cat-like cry and hypotonia, the former being attributable to laryngeal abnormalities and the hypotonia. Other notable features are microcephaly, micrognathia, macrostomia and hypertelorism. With age, the rounded facies becomes elongated and scoliosis becomes evident. Profound learning difficulties and self-mutilation are also features of the syndrome. This patient, who spent most of her day in a wheelchair, was initially referred to the orthodontic department for management of her Class II, Division 1 malocclusion. KG's adoptive parents were concerned not only about her appearance, but also about the potential for trauma as a consequence of the episodes of head-banging by the patient. She had mild lower arch crowding and a



Fig. 9. Case 5: increased overjet.

spaced upper arch with a 10-mm overjet (Fig. 9). There was evidence of early caries in both upper first permanent molar teeth.

The patient was seen jointly by an orthodontist and a paediatric dentist, and a treatment plan was formulated to include management of the carious teeth as well as the proposed orthodontic treatment. The latter involved reduction of the overjet using an upper fixed appliance in conjunction with upper first premolar extractions. The lower arch crowding was accepted. The dental extractions, the temporary restoration of both first permanent upper molar teeth in anticipation of placement of preformed crowns on completion of the orthodontic treatment and placement of the upper fixed appliance were undertaken under general anaesthesia. Since the upper incisors were very proclined, a 0.016 nickel titanium arch wire was put in place. Subsequent changes of archwire and power chain were undertaken successfully at the chair side with oral sedation (Midazolam, 0.5 mg kg^{-1} body weight). The active treatment period was 14 months. The appliances were removed under general anaesthesia, at the same time as both first upper permanent molar teeth had stainless steel crowns put in place. An upper bonded retainer was



Fig. 10. Case 5: bonded retainer in situ.

fitted on the day of appliance removal (Fig. 10). The overjet was reduced to 3 mm.

Discussion

The patients described in this report demonstrate the range of clinical care that can be provided for young people with disabilities. The majority required some help in overcoming anxieties or moderate impairment in order to accomplish the treatment aims [21], and the synergistic alliance of the paediatric dentist and orthodontist in accomplishing health gains is not to be underestimated. Whilst an objective measure of which patients will be able to cooperate for such involved care is lacking, an interdisciplinary appraisal of all the factors which favour treatment must be considered in the context of the potential risk of having to resort to extreme behaviour management techniques, such as general anaesthesia to accomplish the treatment objectives. General anaesthesia is the ultimate in physical intervention and must not be utilized before consideration has been given to the alternative management strategies to accomplish the safe delivery of dental care [24].

It is important that the provision of orthodontic care is appropriate to the child's needs and demands, and not driven by unrealistic parent/carer aspirations [22]. At the same time, efforts should be directed towards correcting significant malocclusions where psychological harm in an already compromised child may be an additional and unnecessary burden [25–27].

Orthodontic treatment for many patients with disabilities is entirely possible, and denying this treatment could be viewed as neglectful, especially in the circumstances of a child who is vulnerable to selfmutilation [5]. Reducing an incisor overjet may also reduce the risk of anterior tooth trauma, as well as preventing self-inflicted soft-tissue trauma in those who are predisposed to it. In children who have impaired understanding and/or a physical impairment that compromises their manual dexterity, considerable reliance will need to be placed on parents and carers to ensure that not only is oral hygiene scrupulously maintained throughout the active phase of treatment, but also during retention, especially if bonded retainers are fitted. In one of the cases described above (case 3), oral hygiene temporarily lapsed at a time when the child's confidence had been lost because of her inability to accept the final stages of the orthodontic plan without any pharmacological help.

It is essential that treatment goals are not set at the ideal, but aim for a reasonable, sustainable improvement in function/aesthetics and self-confidence [2,26]. Each stage should be reassessed and the following treatment modified accordingly, if required. If part of the treatment can only be achieved by resorting to general anaesthesia as an adjunct, parents need to be appraised of the potential risk:benefit involved, in order to be able to make any informed decision, in consultation with the child, where appropriate.

In contemplating orthodontic care for compromised children, it is evident that the pre-assessment appraisal period is crucial. Parents and carers need to be involved in ongoing discussions about the indications for treatment and the role that they will have to play in achieving the treatment objectives in each individual case. A useful predictor of the behaviour management techniques that may be required is the child's acceptance of, and ability to cope with, the exposure of diagnostic radiographs. Another factor is the child's previous dental care and how this was provided. In all these cases, previous dental care had been undertaken under general anaesthesia, although the clinicians involved made assessments independently of this information during the planning stages. Behaviour in children is subject to change not only with maturity, but also as a result of familiarity with, and confidence in, the operator and the approach adopted. For this group of children, parents needed to demonstrate their ability to maintain the child's mouth in a clean state before orthodontic treatment was contemplated.

The tooth movements required in the examples given above would not have been accomplished without the use of fixed appliances, and whilst a compromise in treatment objectives may be necessary in these situations, there is little point in embarking on care that is costly in terms of both everybody's time and money for little or no health gain. Effectiveness as well as efficiency are also factors in the care of children with impairments. All the children required other, restorative and preventive dental care.

In conclusion, the UK Disability Discrimination Act [27] requires that dentists do not discriminate on the basis of disability alone in providing safe and effective care to patients. The above cases have demonstrated that orthodontic care should be part of this, provided that:

1 the treatment is indicated on clinical grounds where to refuse treatment would further compromise the patient's, oral, dental or general health, including their psychological well-being;

2 parents and carers are aware of, and fully accept, the not-insignificant risks and side-effects associated with orthodontic treatment, especially where part of the treatment plan has to be delivered under general anaesthesia; and

3 parents and carers are committed to maintaining demonstrably good oral hygiene and regular dental visits

To guide the less-experienced clinician, some form of objective assessment tool to decide who should receive orthodontic care, and how it might best be delivered, for people with impairments is long overdue. In clinical governance terms, there must be a demonstrable oral health gain that is sustainable following such orthodontic intervention. Unless the child's oral or general health is at risk from nontreatment, general anaesthesia should not be considered. It is the clinician's duty of care to provide treatment that is in the patient's best interests.

What this paper adds

• This paper reviews published literature and describes the orthodontic management in five young disabled people.

• The paper shows that improvements in orthodontic status are possible for people with impairments and dentists should not discriminate on the basis of disability alone.

• Treatment must be effective and efficient and should only be contemplated when there are clear clinical indications to do so and the parents or carers and the patient can achieve good levels of oral hygiene.

Why this paper is important for paediatric dentists

• Provision of specialist orthodontic treatment requires backup from paediatric dentists. All of the cases described needed other restorative and preventive dental care. **Résumé.** *Objectif.* L'objectif de cet article est de présenter une série de cas illustrant les options possibles lors de la prise en charge des malocclusions dans un groupe de jeunes personnes avec déficience. *Protocole.* Série de cas.

Méthodes. Une revue de littérature est présentée et une série de cas montre que le traitement orthodontique est possible chez des patients présentant des déficiences.

Résultats. Cinq enfants sont présentés. Ils ont une déficience intellectuelle et/ou physique et une malocclusion entraînant un trauma ou une déficience accrue. *Conclusion*. Les risques et bénéfices des procédures et les résultats attendus en matière de santé buccale doivent être soigneusement pris en considération dans ce groupe d'enfants.

Zusammenfassung. *Ziel.* Zweck dieser Veröffentlichung ist es, eine Reihe von Fallberichten vorzustellen, in welchen Optionen der Behandlung von Malokklusion in einer Gruppe von jungen Behinderten illustriert werden.

Design. Fallserie.

Methode. Eine Literaturübersicht wird dargestellt und eine Reihe von Beispielen, welche zeigen, dass eine kieferorthopädische Behandlung bei Patienten mit unterschiedlichen Behinderungen möglich ist.

Ergebnisse. Fünf Kinder mit verschiedenen intellektuellen und/oder körperlichen Behinderungen sowie Malokklusion, die zu Trauma oder Symptomverschlimmerung führten, werden vorgestellt.

Schlussfolgerung. Die Risiken und Vorteile der Behandlungsmaßnahmen sowie der erwartete Gewinn für die Mundgesundheit müssen in dieser Gruppe von Kindern sorgfältig geprüft werden.

Resumen. *Objetivo*. El objetivo de este artículo es presentar una serie de casos que ilustran opciones posibles en el tratamiento de maloclusiones en un grupo de jóvenes con minusvalías.

Diseño. Serie de casos.

Métodos. Se presenta una revisión de la literatura y se muestra una serie ilustrativa de casos para indicar que el tratamiento ortodóncico es posible en pacientes con un abanico de minusvalías.

Resultados. Se presentan cinco niños de varias minusvalías intelectuales y/o físicas y una maloclusión que resultó en un traumatismo o aumento de la discapacidad.

Conclusión. Los riesgos y beneficios de los procedimientos, los resultados de la salud bucal anticipada, necesitan ser considerados cuidadosamente en este grupo de niños.

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