Orthodontic Treatment in Down’s Syndrome Patient with Unilateral Cleft Lip and Alveolus

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Abstract

In this paper, we describe orthodontic treatment in a patient with Down’s syndrome accompanied by reverse occlusion due to skeletal deformity with unilateral cleft lip and alveolus. Orthodontic treatment was not initially indicated due to the potential need for surgical intervention and the presence of mental retardation. The further mental and physical growth of the patient together with the help of their guardians, however, allowed placement of a multi-bracket. Although no skeletal correction occurred as a result, reverse occlusion was corrected by labial inclination of the incisors. The patient and her guardians were satisfied with the correction of the overlap by orthodontic treatment alone.

Key words: Down’s syndrome — Cleft lip and alveolus — Orthodontic treatment — Anterior crossbite — Class III

Introduction

Down’s syndrome is a congenital disorder characterized by specific facial features and physical manifestations, and it is frequently associated with mental retardation and systemic disease.

Patients with Down’s syndrome generally have a characteristic facial bone morphology, including hypoplasia of the cranial base and mid-facial region, brachycephaly, and a flat skull base. The reported dental manifestations include an abnormal number of teeth, exemplified by the congenital absence of permanent teeth; abnormal dental morphology such as microdontia and short roots; and an abnormal eruption/exfoliation cycle such as prolonged retention of deciduous teeth and delayed eruption of permanent teeth. Incomplete lip closure due to muscle hypotonia, a low tongue position, and tongue thrusting have also been reported. While dental caries is not a frequent manifestation, the prevalence of gingivitis and periodontal disease is high. A considerable proportion (approximately 3.3%) of patients with Down’s syndrome have a cleft lip and palate, the reported incidence...
of which is approximately 16.7% among such patients who have been examined by orthodontists.\(^9\)

These characteristic features predispose Down’s syndrome patients to various types of malocclusion, including reverse occlusion, maxillary incisor crowding, posterior crossbite, open bite, a class III molar relationship, and spacing.\(^3,6,9,11,15,16\) The orthodontic treatment of these problems has been reported\(^2,9,16\).

Here, we describe orthodontic treatment in a 9-year-old girl with Down’s syndrome and unilateral cleft lip and alveolus.

**Case Report**

A girl aged 9 years 3 months with Down’s syndrome presented with reverse occlusion and a left-sided cleft lip and alveolus (Fig. 1). She had a brother who was healthy and was undergoing treatment for reverse occlusion due to skeletal malocclusion at another hospital. The patient had undergone cheiloplasty at 11 months of age. Although she was of a short stature (height, 122 cm) and had an atrial septal defect and mental retardation, this did not interfere with any major activity of daily living.

Orthopantomography revealed a wide cleft distal to the left central incisor. The left lateral incisor was unerupted and was assumed to be a microdont. The maxillary second premolar was congenitally absent.

An Angle’s class III molar relationship was observed, with overjet of −8 mm and overbite of 5 mm. Reverse occlusion with significant overlap, bilateral crossbite, and crowding due to a narrow maxillary arch were also observed. The tongue was hypotonic and in a low position.

Skeletal analysis using lateral cephalography revealed an SNA of 81.7°, SNB of 81.6°, facial angle of 92.1°, Y-axis of 53.2°, gonial angle of 111.0°, and FMA of 17.1° (Table 1). Based on a comparison with age-matched individuals with normal occlusion, a brachyfacial pattern and skeletal mandibular protrusion were diagnosed.\(^14\) Dental analysis revealed a U1 to FH of 126.0°, an IMPA of 109.9°, and labial tipping of the maxillary and mandibular incisors. Vertical hypoplasia of the left maxillofacial region was observed when viewed from the front.

The goals of orthodontic treatment in this patient were correction of the anterior and posterior crossbite and incisor alignment after bone grafting. However, the marked mandibular protrusion and labial tipping of the maxillary incisors did not preclude the possibility of future orthodontic surgical intervention. Intraoral imaging and impression-making were initially ruled out as the patient would not have been capable of understanding the treatment, raising the possibility of non-compliance. Therefore, orthodontic treatment was not recommended. The patient’s guardians, however, were extremely keen for orthodontic treatment to be carried out. Therefore, the decision was made to attempt the most feasible treatment.

There was no evidence of root resorption in the maxillary second deciduous molars, so they were used as substitutes for the missing maxillary second premolars. First, the patient was made to wear a chin cap to shift the direction of mandibular growth backwards. Bone grafting was performed to compensate for hypoplasia in the area of the cleft in the left maxillary region, which facilitated derotation of the central incisor and eruption of the lateral incisor. Subsequently, a palatal expansion plate and face mask were used to correct the narrow maxillary arch. However, the patient exhibited noncompliance with this treatment, and we were unable to correct the overlap. When the patient reached the age of 11 years 2 months, an attempt was made to install a partial wire to apply intrusive force to the extruded mandibular incisors (Fig. 2). Although the brackets were successfully bonded to the incisors, they could not be bonded to the molars because of tongue flaccidity and excess salivation, making installation of the partial wire impossible. Although oral hygiene deteriorated and treatment was delayed during canine and premolar eruption, the incisal overlap gradually transitioned...
Fig. 1  Facial and intraoral photographs, panoramic radiograph and cephalograms at pre-treatment (9 yr 3 mo)
### Table 1 Cephalometric analysis at pre-treatment and post-treatment

<table>
<thead>
<tr>
<th>Measurement</th>
<th>Pre-treatment (9 yr 3 mo)</th>
<th>Post-treatment (16 yr 9 mo)</th>
<th>Normative mean* (9 yr, female)</th>
<th>Normative mean* (15 yr, female)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean</td>
<td>SD</td>
<td>Mean</td>
<td>SD</td>
</tr>
<tr>
<td>SNA</td>
<td>81.7</td>
<td>82.6</td>
<td>80.8</td>
<td>3.1</td>
</tr>
<tr>
<td>SNB</td>
<td>81.6</td>
<td>83.0</td>
<td>77.7</td>
<td>3.4</td>
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<tr>
<td>Facial angle</td>
<td>92.1</td>
<td>94.4</td>
<td>85.5</td>
<td>3.3</td>
</tr>
<tr>
<td>Y-axis</td>
<td>53.2</td>
<td>53.7</td>
<td>62.9</td>
<td>3.5</td>
</tr>
<tr>
<td>Gonial angle</td>
<td>111.0</td>
<td>108.1</td>
<td>124.5</td>
<td>3.0</td>
</tr>
<tr>
<td>FMA</td>
<td>17.1</td>
<td>15.6</td>
<td>27.3</td>
<td>4.0</td>
</tr>
<tr>
<td>IMPA</td>
<td>109.9</td>
<td>99.5</td>
<td>95.8</td>
<td>4.2</td>
</tr>
<tr>
<td>U1 to FH</td>
<td>126.0</td>
<td>137.9</td>
<td>116.7</td>
<td>4.7</td>
</tr>
<tr>
<td>Overjet (mm)</td>
<td>−5.5</td>
<td>1.5</td>
<td>3.1</td>
<td>1.1</td>
</tr>
<tr>
<td>Overbite (mm)</td>
<td>4.5</td>
<td>0.5</td>
<td>3.3</td>
<td>1.9</td>
</tr>
</tbody>
</table>

*: For Japanese normative mean by Masuda."

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Fig. 2 Intraoral photographs at commencement of lower arch leveling (11 yr 2 mo)

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To an edge-to-edge relationship. Therefore, the molars were built up with resin, a wire was placed on the upper incisors, and the overlap was corrected by inducing labial inclination of the teeth (Fig. 3). Although osseointegration was inadequate at the graft site, multibracket treatment was performed, primarily to level the dentition with the microdont lateral incisor (Fig. 4). The patient’s guardians were informed that prolonged use of the left lateral incisor was not feasible. The entire orthodontic treatment lasted for 7 years, and it was considered complete when the patient reached 16 years 9 months (Fig. 5). Compliance with a removable retainer was not expected, so fixed lingual retainers were bonded to the maxillary incisors and mandibular anterior teeth.

Figure 6 shows superimposition of pre- and post-treatment cephalometric tracing. Follow-up assessments revealed forward growth of the mandible, an increase in the SNA and SNB, and a decrease in the FMA (Fig. 6). Commensurate with these changes, the upper incisors showed further labial inclination and the lower incisors lingual inclination. Maxil-
lary and mandibular growth was asymmetrical, with the maxilla inclining upward to the left and the mandible deviating to the left. Tongue thrusting persisted.

Discussion

1. Characteristics of occlusion and maxillofacial morphology

We previously examined occlusion in patients with unilateral cleft lip and alveolus and found an increased prevalence of anterior crossbite (72.7%), while posterior crossbite was relatively infrequent (18.2%). We also reported that the incidence of reverse occlusion and edge-to-edge bite was 72.3%, that of posterior crossbite 55.6%, and that of maxillary incisor crowding 50.9% in individuals with Down’s syndrome. Furthermore, hypoplasia of the cranial base and mid-facial region and a brachyfacial pattern were

Fig. 3  Intraoral photographs at commencement of upper arch leveling (13 yr 3 mo)

Fig. 4  Intraoral photographs at 15 yr 4 mo
Fig. 5  Facial and intraoral photographs, panoramic radiograph and cephalograms at post-treatment (16 yr 9 mo)
observed, and the patients often exhibited mild reverse occlusion with slight overlap and labial tipping of the maxillary incisors\textsuperscript{15}.

In the present patient, malocclusion had resulted from inherited reverse occlusion in combination with the type of occlusion characteristic of Down’s syndrome and unilateral cleft lip and alveolus.

2. Course of treatment and outcome

A palatal expansion plate and chin cap were used to pull the maxilla forward and rotate the mandible downward. However, the patient was unable to wear these devices long enough for skeletal correction to take place, and the overlap was eventually corrected by the tipping movement of the teeth. Excess inclination of the anterior teeth is best avoided in such cases, as Down’s syndrome patients are prone to periodontitis.

Short roots have been reported in Down’s syndrome patients\textsuperscript{7}, particularly in the maxillary and mandibular central incisors and canines and mandibular lateral incisors. Short roots were not observed in the present patient, however, and there was no root resorption. The left lateral incisor was maintained in position by a fixed retainer due to inadequate osseointegration at the graft site. The patient’s guardians, however, were informed of the worsening stability of the tooth, and the patient’s course was followed.

In addition, the molars showed inadequate alignment.

3. Indications and prognosis in orthodontic treatment in Down’s syndrome patients

In a clinical survey of Down’s syndrome patients, Horie \textit{et al.}\textsuperscript{9} reported that orthodontic treatment was chosen in accordance with the patient’s status. Therefore, the goals of orthodontic treatment in such patients differ from those in typical patients. For example, posterior crossbite may be corrected or crowding decreased while overlooking reverse occlusion, if present.

When planning orthodontic treatment in such patients, congenitally absent teeth, teeth with short roots, and delayed eruption must all be taken into account. Moreover, numerous problems may be encountered, including non-compliance due to a lack of understanding and mental retardation, the need for cooperation from a guardian or parent, difficulty in placing bands or brackets because of abnormal dental morphology and macroGLOSSIA, the presence of heart disease, difficulty in maintaining oral hygiene, and the prevalence of periodontal disease. Nevertheless, patients with Down’s syndrome often have a cheerful disposition and are more amenable to orthodontic treatment than might be expected. Treatment goals should be initially set at the lowest level possible, and these goals can then be upgraded according to feasibility. In some patients, the best option may be to limit orthodontic treatment to correcting crowding of the incisors to facilitate oral hygiene. The present patient initially detested her appliance. Her treatment proceeded gradually as she grew older, and she eventually got accustomed to treatment and was also able to comply with a multi-bracket appliance. Although she was unable to brush her teeth at first, she was eventually able to cope with this task up to a point after repeated instruction, even though a final brushing by her mother was still necessary.

Some problems were encountered in fully implementing orthodontic treatment in this patient, including non-compliance with the
proper use of an extraoral appliance and the unfeasibility of bonding brackets to the molars. Moreover, the prognosis of maxillary expansion is not promising in such patients as the maxilla is reportedly susceptible to narrowing and reverting to its previous shape. Therefore, the achievement of occlusal stability in the current patient was presumed to be difficult.

The results of the present case suggest that orthodontic treatment in Down’s syndrome cases should only be undertaken after a complete evaluation of the patient’s condition. The wishes of the patient and his/her guardians should be acknowledged and treatment provided in accordance with the patient’s overall status.

References


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