Treatment of severe hypodontia–oligodontia—an interdisciplinary concept


Abstract. The authors’ experience with oral rehabilitation of patients suffering from oligodontia (i.e. six or more congenitally missing permanent teeth, third molars excluded) is reported. The concept is based on an interdisciplinary team approach involving pedodontists, orthodontists, maxillofacial surgeons and prosthodontists. A series of 112 consecutive patients suffering from oligodontia were referred from 1997 to 2001. Ten of the patients (8.9%) suffered from ectodermal dysplasia. The total number of missing teeth was 1126, with an average of 10 per patient. Ninety-two patients had either finished treatment or were on an active treatment schedule. Of these, 97% underwent some kind of orthodontic treatment. Of the 112 patients, 51 had finished treatment at the end of the follow-up period (mean 28 months, range 1–68). Of these, fixed implant-supported prosthetic restoration was used in 90% to replace missing teeth, often combined with alveolar ridge augmentations (73%), sinus floor augmentation (43%), inferior alveolar nerve transposition (18%) and orthognathic surgery (27%). Early diagnosis, and comprehensive treatment planning with good coordination and timing of the individual treatment phases are decisive for a successful treatment outcome. The therapeutic concept is presented with special emphasis on surgical aspects.

In a Swedish study\textsuperscript{4} the prevalence of hypodontia was 7.4\% with 0.19\% missing six or more permanent teeth. Schalk-Van der Weide\textsuperscript{18} defines, in her Dutch study, hypodontia of six or more permanent teeth as oligodontia and estimates the prevalence to be 0.08\%. Oligodontia is therefore a relatively rare condition, probably affecting about 0.1–0.2\% of the population.

Simultaneous with oligodontia are often different positional changes of the existing teeth, their morphology and size\textsuperscript{14,18,20}. Characteristic changes are also growth disturbances of the maxillofacial skeleton and thus the facial appearance\textsuperscript{2,10,17}. Oligodontia may be associated with other ectodermal abnormalities and syndromes\textsuperscript{2,3,8,18,19,20}. The treatment of patients with oligodontia is therefore often complex.

The purpose of this article is to report the authors’ experience with an interdisciplinary, centralized treatment approach to this relatively small group of patients involving pedodontists, orthodontists, prosthodontists and maxillofacial sur-
geons. The present study will focus upon treatment planning and the various surgical treatment options in an interdisciplinary context. The more detailed treatment outcome in the longer term will be reported in a subsequent publication.

Demography

The population of Northern Jutland is approximately 500,000 with an actual birth rate of 6000 per year. Previous experience with treatment of patients with multiple missing teeth had emphasized the need for an interdisciplinary collaborative effort. A centre for management of patients with oligodontia was therefore established in 1997 at the Department of Oral and Maxillofacial Surgery, Aalborg University Hospital. Patients with six or more missing teeth could be referred to the Centre of Hypodontia independent of age.

The interdisciplinary concept: conferences and treatment planning

After a primary examination of the patients, the patients are presented at a conference with participation of pedodontist, orthodontist, prosthodontist, and oral and maxillofacial surgeon. Associated specialties are anaesthesiology, paediatrics, genetics, and nuclear and internal medicine.

The purpose of the conference is:

(1) to establish a treatment plan in agreement with the wishes of the patient (and parents);
(2) to evaluate and adjust an on-going treatment schedule according to a previous treatment plan, and follow up on intermediate treatments in young patients during growth;
(3) to evaluate the final outcome of treatment in order to build up team experience for improvement of treatments in the future;
(4) to inform patients and parents about consequences of missing teeth, treatment options, and to offer genetic counselling.

The clinical and radiographic examinations (Fig. 1), including cephalometric radiographic analysis, orthopantomographic and intraoral radiographs, combined with study casts and diagnostic wax-ups on the casts (Fig. 2), constitute the basic material when treatment planning is performed at the conference.

When young patients during growth are presented, an overall treatment schedule

Fig. 1. Treatment planning. Preoperative profile clinically (A and B) and radiographically (D and E) of a 17.5-year-old patient with 13 missing permanent teeth, deep bite, reduced lower facial height, lip eversion and bimaxillary retrognathia. (B and E) Preoperatively but with a simulated, ‘diagnostic’ bite raising and advancement of the paranasal soft tissue by soft retrievable material placed in the anterior upper vestibular sulcus. (C and F) Three years later, after bimaxillary orthognathic surgery, including a down-sliding Le Fort I osteotomy, bilateral sinus lift, but before implant insertion and final prosthetic treatment.
and planning of the intermediate treatment is outlined. Intermediate treatment is then coordinated with and generally performed in the public health care system. When the orthodontic treatment is finished and the patients have reached an age of 17–18 years, they are rescheduled at a subsequent conference. The patients are then usually ready for planning of the final treatment, i.e. orthognathic surgery, bone augmentations, dental implants and prosthetics.

Orthodontic treatment takes place either in the public health care system or in private practice. The surgical treatment, including orthognathic, is performed at the Department of Oral and Maxillofacial Surgery, Aalborg University Hospital. The prosthetic treatment is performed by prosthodontists in private practice. The treatment is financed by public means.

Patients
There were 112 patients (54 females, 58 males) with an average age of 20.5 years (range 8–48 years) consecutively referred from July 1997 to March 2001. Children in the public health care system were referred after a diagnosis of oligodontia was established during orthodontic screening procedures, which normally take place at the age of 10–11 years for girls and 11–12 years for boys. Adult persons were referred by private practitioners according to their need for treatment. Many of the adult patients had never had a genuine treatment offer during childhood. The distribution of the 112 patients according to age at the time of referral is shown in Fig. 3. Eighty-one (72%) of the patients were 15 years old or more when referred, and 31 (28%) of the patients were 25 years old or more.

The total number of missing teeth of the 112 patients was 1126 (mean: 10 per patient, range 6–25). The distribution of the 112 patients according to gender and number of missing teeth per patient is presented in Fig. 4. Forty-one patients (37%) were missing 10 or more permanent teeth. The location of the 1126 missing teeth is shown in Fig. 5. The second upper and lower premolars followed by first upper premolars, lateral upper incisors, first lower premolars, central lower incisors and upper canines were the teeth most commonly missing. The second upper and lower premolars accounted for 33% of all missing teeth. Ten patients (8.9%) had a verified diagnosis of hypohidrotic ectodermal dysplasia (HED) (Fig. 6) and one patient incontinentia pigmenti. The 10 patients with HED were on average missing 15 teeth (range 6–25). Changes of the facial soft-tissue profile (Fig. 7), listed in Table 1, and dento-alveolar alterations...
(Fig. 8), listed in Table 2, were prominent features among these patients with oligodontia.

### Table 1. Characteristic growth disturbances and changes of facial appearance in patients with oligodontia

<table>
<thead>
<tr>
<th>Feature</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Maxillary retrognathism and hypoplasia</td>
<td></td>
</tr>
<tr>
<td>Mandibular retrognathism</td>
<td></td>
</tr>
<tr>
<td>Anterior rotation of mandible</td>
<td></td>
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<tr>
<td>Decreased vertical and transversal dimensions of the alveolar process</td>
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<tr>
<td>Large inferior extensions of the maxillary sinuses</td>
<td></td>
</tr>
<tr>
<td>Receding midface</td>
<td></td>
</tr>
<tr>
<td>Low anterior face height</td>
<td></td>
</tr>
<tr>
<td>Increased nasal–labial angle</td>
<td></td>
</tr>
<tr>
<td>Deep labial–mental fold</td>
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</tbody>
</table>

**Intermediate treatment**

**Prosthetic**

For functional, but also aesthetic reasons, temporary replacements of missing teeth, i.e. removable dentures, temporary bridges, as well as morphologic changes of maxillary anterior teeth by composite resin restorations were performed. To prevent overeruption of permanent teeth, composite resin build-ups of the antagonizing deciduous teeth or bonding of the permanent teeth were necessary until the final restorations could be inserted.

**Orthodontic**

Space closure or space reduction combined with extraction of temporary teeth to promote guided eruption of permanent teeth, as well as making and securing space between teeth, according to the
treatment plan, were initiated. Removable appliances were inserted to accomplish bite raising, to correct cross bite, promote sagittal growth of the mandible, and to treat or prevent overeruption and tilting of teeth. Closure of anterior diastemas was sometimes done intercategorically on aesthetic indications. Use of palatal implants for orthodontic anchorage was not yet implemented in the intermediate phase in this study group of patients.

Surgical

Extraction of temporary teeth was performed for guided mesial eruption of permanent upper molars to replace missing second premolars. Impacted temporary molars were also extracted in some cases in order to preserve bone height of the growing alveolar process and to alleviate full eruption and prevent tilting of neighbouring permanent teeth (Fig. 2B).

Treatment

The treatment status of the 112 patients in February 2003 is shown in Table 3. The mean follow-up period of the 51 patients who had finished treatment is range 1–68 months.

Orthodontic

The type of orthodontic treatment of the 92 patients who had finished treatment or were in some kind of active treatment is shown in Table 4. In patients with very few permanent teeth, temporary or strategic implants were inserted to establish sufficient orthodontic anchorage. Before implant placement, and while implants ossointegrated, retention of the accomplished orthodontic treatment was secured by removable splints, temporary bridges or bonding of teeth.

### Table 2. Dento-alveolar characteristics associated with oligodontia

<table>
<thead>
<tr>
<th>Occlusal disturbances</th>
<th>No. of patients (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Deep bite</td>
<td>10 (20%)</td>
</tr>
<tr>
<td>Cross bite</td>
<td>10 (20%)</td>
</tr>
<tr>
<td>Steep inclination of maxillary incisors</td>
<td>10 (20%)</td>
</tr>
<tr>
<td>Abnormal attrition</td>
<td>10 (20%)</td>
</tr>
</tbody>
</table>

### Table 3. Treatment status of 112 patients with oligodontia

<table>
<thead>
<tr>
<th>Status</th>
<th>No. of patients (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Finished treatment</td>
<td>51 (45)</td>
</tr>
<tr>
<td>Ongoing treatment</td>
<td>41 (37)</td>
</tr>
<tr>
<td>Growth observation</td>
<td>8 (7)</td>
</tr>
<tr>
<td>Other*</td>
<td>12 (11)</td>
</tr>
<tr>
<td>Total</td>
<td>112 (100)</td>
</tr>
</tbody>
</table>

*Primary teeth still well-functioning, patients have moved from the county, or have chosen not to be treated.

### Table 4. Orthodontic treatment of 92 patients who have finished or are in active treatment

<table>
<thead>
<tr>
<th>Type of orthodontic treatment</th>
<th>No. of patients (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Conventional</td>
<td>57 (62)</td>
</tr>
<tr>
<td>Orthognathic</td>
<td>32 (35)</td>
</tr>
<tr>
<td>None</td>
<td>3 (3)</td>
</tr>
<tr>
<td>Total</td>
<td>92 (100)</td>
</tr>
</tbody>
</table>

### Table 5. Surgical procedures used for 51 patients who had finished treatment, and rate of associated complications

<table>
<thead>
<tr>
<th>Type of treatment</th>
<th>No. of patients (%)</th>
<th>No. of procedures</th>
<th>No. of complications (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Orthognathic surgery</td>
<td>14 (27%)</td>
<td>19</td>
<td>2 (22%)</td>
</tr>
<tr>
<td>Le Fort I osteotomy</td>
<td>9 (18%)</td>
<td>9</td>
<td>2 (22%)</td>
</tr>
<tr>
<td>Sagittal split osteotomy</td>
<td>10 (20%)</td>
<td>10</td>
<td>2 (20%)</td>
</tr>
<tr>
<td>Inferior alveolar nerve transposition</td>
<td>9 (18%)</td>
<td>14</td>
<td>4 (29%)</td>
</tr>
<tr>
<td>Sinus lift</td>
<td>22 (43%)</td>
<td>34</td>
<td>0</td>
</tr>
<tr>
<td>Bone grafting</td>
<td>37 (73%)</td>
<td>96</td>
<td>Minor bone loss</td>
</tr>
<tr>
<td>Implant placement</td>
<td>46 (90%)</td>
<td>283</td>
<td>6 (2%)</td>
</tr>
<tr>
<td>Other*</td>
<td>10 (20%)</td>
<td>12</td>
<td>None</td>
</tr>
</tbody>
</table>

* Alveolar osseodistraction, vestibular plasty, genioplasty, nasal lift.
So far, 283 implants have been inserted in 46 patients. Two implant systems were used, Brånemark (Nobel Biocare, Göteborg, Sweden) and Astra (Astra Tech, Mölndal, Sweden). Six implants, three in two patients each (2%) were lost, all before abutments were connected. In both patients the implants were placed in anterior maxillary alveolar ridge augmentations.

In some cases where implants were inserted either simultaneous with or shortly after extraction of temporary molars, an accelerated resorption of vestibular marginal bone was seen from implant insertion until abutment connection, resulting in visible titanium buccally at the marginal level on the final prosthetic restorations.

The number of inserted implants in proportion to the total number of missing teeth in the 51 patients who had finished treatment was 283:536 (53%).

Prosthetic

Fixed implant-prosthetic restorations were used in 90% of the patients who had finished treatment (Table 6). Screw-retained prosthetic superstructures were chosen when possible. Cantilever units were often used in non-weight-bearing areas with critical mesial-distal space (Fig. 9E and F).

Discussion

Epidemiology

Based on previous epidemiological studies, the prevalence of patients with six or more congenitally missing permanent teeth can be estimated to be about 0.1–0.2%. In an area the size of Northern Jutland, Denmark, at least six children with oligodontia should therefore be born on average per year. The high average age of 20.5 years of the referred patients, with 28% more than 25 years of age, is probably a temporary phenomenon, and due to an accumulation in the county of adults with oligodontia with an uncovered requirement for treatment.

In the present study, the upper and lower second premolars, followed by the upper first premolars, the upper lateral incisors, the lower central incisors and the upper canines, were the most frequent regions of hypodontia. In other studies of oligodontia and in several larger epidemiological studies of hypodontia, the upper canines are not reported missing as frequently. As in other studies, the regions of the upper central incisors, and the upper and lower first molars were only rarely involved.

Syndromes with hypodontia/oligodontia

Hypodontia is seen in connection with more than 120 different syndromes, but most frequently with X-linked ectodermal dysplasia, and more rarely in autosomal recessive and dominant forms of HED. In the present study, 10 patients...
(9%) had HED. In a Swedish study of 88 patients with eight or more missing teeth, seven patients (8%) had ED and a further six patients had other syndrome diagnoses. In the study by Schalk-Van der Weide et al. involving 167 selected patients with oligodontia, 48 patients had a verified syndrome diagnosis (25% with ED). From this study it is concluded that with a high number of missing teeth, and especially within the upper central incisors and the upper and lower first molars are absent, a possible association with ED should be considered, even in cases with only a discrete expression of the syndrome, as it may be seen in heterozygous females with HED. In the present study, these regions were affected in four of the ten patients with HED, and further in seven other patients, all with severe hypodontia, but without known HED. Since HED is clinically and genetically heterogeneous, an on-going study is being performed to estimate the proportion of possible undiagnosed cases of HED.

Organization of management of oligodontia

The low prevalence of patients with oligodontia necessitates a certain degree of centralization of the more complicated cases, if sufficient accumulation of knowledge and experience is to be obtained. With the present number of referred patients, 60–70 patients will continuously be under treatment planning, treatment, coordination of treatment and review appointments. Optimized conditions for the treatment of these patients include a fully integrated interdisciplinary team approach of orthodontists, oral and maxillofacial surgeons, and prosthodontists working under the same roof. So far, the orthodontic and prosthetic treatments are still mainly performed in private practice associated with the centre.

Based on collaborative efforts and gained experiences, the following considerations may enhance a more successful treatment outcome in the management of patients with oligodontia.

Orthodontic treatment

Without intermediate treatment patients with oligodontia may end up with steep and over-erupted upper and lower incisors, deep bite, bimaxillary retrognathism, and a hypoplastic and narrow maxilla. Intermediate treatment is therefore of utmost importance to secure and stimulate sagittal, vertical and transversal growth of the jaws, including the alveolar processes. Reduced vertical growth of the alveolar process should, if possible, be prevented by extraction of impacted primary molars in good time, eventually combined with mesial guided eruption from posterior to the permanent molars, especially in the upper jaw. Cross bite should be eliminated, and sagittal growth of a retrognathic mandible should be promoted. A hypoplastic maxilla with missing lateral incisors should not be closed but opened, and the occlusal plane should be levelled by intrusion and uprighting of steep inclined and over-erupted incisors.

In this way, patients with oligodontia may avoid a deep impact on facial appearance and thus later major surgical interventions. In case orthognathic surgery is not indicated, the final orthodontic treatment needs mainly to focus upon harmonizing the dental arches and securing sufficient space for dental implants.

The size of the edentulous space between existing teeth should be precise and in accordance with the planned implant treatment. Bilateral and intermaxillary variations in tooth size (microdontia) may aggravate the space problems. Even small deviations from the plan may result in a compromised situation with insufficient space for implants at the cervical level as well as between roots of adjacent teeth. Uprighting of the adjacent teeth is often critical due to the subsequent implant placement and to segmentalization of the maxilla during orthognathic surgery.

Extrusion of permanent molars, when combined with bite raising, should be postponed until fixed implant–prosthetic constructions have secured the height. The potential of implant anchorage for orthodontic treatment, especially in this group of patients, is high. By this technique it will be possible to close posterior spaces orthodontically without retraction of anterior incisors and without compromising the soft-tissue profile. In this way it will be possible not only to reduce the number of implants, but also the number of sinus floor augmentations.

Surgical treatment

As outlined in Table 3, this covers a wide spectrum, and the procedures involving bone augmentation, and especially orthognathic surgery, are more demanding in this group of patients. In case several surgical procedures had to be performed in the same patient, general anaesthesia was often used, although fundamentally some of them could have been done under sedation and local anaesthesia. Approximately one third of the treatments included orthognathic surgery, due to the hypoplastic growth of the jaws. Information on the treatment options, but also the possible complications are therefore very important at an early stage for treatment planning.

With lack of sufficient anchorage and an unstable occlusion, there is a potential risk of relapse after advancement osteotomies, and this may have caused the relapse of two of the maxillary osteotomies. Both patients had very few permanent teeth, and both might have benefited from insertion of implants in a preceding procedure to achieve a more stable peri- and postoperative occlusion. Surgically assisted rapid maxillary expansion should also be considered in a preceding sequence, if severe transverse deficiency and maxillary retrognathia coexists. Relapse after inferior repositioning of the maxilla may be reduced by using a down-sliding osteotomy which also reduces the need for bone grafting.

If closing of space in the upper premolar regions is not done orthodontically it may be performed by advancement of the posterior maxillary segments during Le Fort I osteotomy as an alternative to sinus floor augmentation and subsequent implant treatment.

Inferior alveolar nerve transposition, combined with implant insertion, is facilitated when managed simultaneously with a sagittal split osteotomy. In general, nerve transposition is more easily performed in patients with oligodontia due to the often extensive edentulous regions, but neurosensory disturbances have to be expected. In the present series they were mild and did not cause significant problems to the patients. When sinus floor augmentations are performed in a preceding sequence, before the maxillary osteotomy is done, the sinus membrane is more easily elevated intact, probably due to fewer adhesions. Alternatively, augmentation can be done simultaneously with the maxillary osteotomy, but revascularization of the augmented region may be critical. Displacement of bone chips may occur if uncovered by a sinus membrane. In such cases fibrin glue is a beneficial adjunct.

Implants were widely used to replace missing teeth in the present group of patients. Orthodontic, orthognathic, and prosthetic treatment reduced the number of missing teeth replaced by implants to 53%. At the end of the study period only 2% of the implants had failed to integrate, and none were lost after abutment connection.
Unless special circumstances indicate the use of implants in adolescents\(^1,2,11,19\), they should not be inserted until skeletal growth has finished. Slight continuous eruption of teeth and vertical growth of the alveolar process may proceed, even in adults, after skeletal growth is completed, resulting in infraposition of the implant/superstructure\(^2,21\).

Increased vertical resorption of the alveolar process may occur after extraction of deciduous teeth. This should be taken into account when implants are inserted shortly after or simultaneously with extraction of primary teeth to avoid exposure of titanium at the cervical level.

### Prosthetic treatment

No more implants than necessary should be inserted, or it may be difficult to achieve a satisfactory aesthetic prosthetic result and natural configuration of the peri-implant mucosa. The use of cantilever bridges supported by one or more implants in non-weight-bearing areas, like the lateral upper and lower incisor regions, may be a solution for the frequent space problems in these areas. Screw-retained solutions are to be preferred. Most treatments are performed in young patients and the prosthodontic works may later in life need to be repaired.

The vast majority of the prosthetic superstructures in this material were implant borne, but one should not exclude the possibility of using conventional fixed bridges when indicated. Sometimes, this may even spare the patient from a long and sometimes complicated treatment with orthodontics, bone augmentations and implants. It should be noted that conventional bridges have to await pulp retraction in young patients, which may delay the final treatment significantly, and conventional bridges are also very dependent on number and distribution of existing permanent teeth.

Patients with more severe oligodontia, with a high number of missing permanent teeth and compromised facial profile, may endure not only functional but also speech problems and psychological distress. It is therefore especially important in such cases to supply clear material information about future treatment goals and the associated time perspective. To promote a higher self esteem and better social acceptance it may also be necessary to insert implants at a young age, and in this way establish support for temporary fixed or partially fixed prosthetics until the final treatment can be performed later on.

From specialization and centralization of treatment follows longer travelling distances for the patients, as well as for their relatives. Most of the patients have to wait several years, often at a critical age, before the final treatment can be performed. A good will to cooperate, confidence and patience are important qualities among the patients, as well as their parents, during this type of long-lasting treatment with multiple patient appointments.

### References


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