Growth After Construction of the Temporomandibular Joint in Children With Hemifacial Microsomia

IAN R. MUNRO, M.D. J.H. PHILLIPS, M.D. G. GRIFFIN, D.D.S.

Free costochondral grafts have been used to construct the absent ascending ramus and condylar head of 22 children with hemifacial microsomia (HFM) who were less than 14 years of age. Evidence of continued growth was demonstrated in all patients. Growth was determined by measurement of cephalograms or by inference; if the patient grew extensively in height but his/her face remained symmetric, the graft was inferred to have grown. In two patients, considerable overgrowth occurred, displacing the chin to the opposite side and causing a class III malocclusion. Variations in age or type of adjunctive operations did not affect growth. There appears to be no single explanation for all findings. Because all of the grafts grew, early joint construction and correction of hemifacial microsomia are advocated.

KEY WORDS: hemifacial microsomia, temporomandibular joint construction, costochondral grafts

Hemifacial microsomia is a commonly seen disorder at craniofacial centers. The facial asymmetry is caused by deficiency in bone and, in some cases, hypoplasia or absence of soft tissue. Severity depends largely on the degree of skeletal deficiency, asymmetry, and displacement (Munro and Lauritzen, 1985). Surgical repair varies according to the presence or absence of a mandibular condyle (Munro and Lauritzen, 1985; Munro, 1987). Patients without a condyle have no mechanical stability of the mandible on the involved side. To reposition and maintain the mandible, a mechanical buttress must be placed between the advanced and rotated mandible and the base of the skull, thereby creating a temporomandibular joint (TMJ). Since 1977 we have constructed the missing TMJ in children and adults by using a costochondral graft, as advocated by Obwegeser (1974). This study reports on some unexpected long-term results.

Method

Between 1977 and 1985, 22 children aged 4 years, 9 months to 13 years, 9 months, with a mean age of 9 years, 7 months, underwent insertion of costochondral grafts to construct TMJs in conjunction with other osteotomies of the face. This was done to correct hemifacial microsomia. The technique for TMJ reconstruction has previously been de-

scribed (Munro, 1980). Other osteotomies were also performed according to the degree of deformity (Lauritzen et al, 1985; Munro, 1987). All grafts were fixed to the mandible by wiring, and the patients were placed in intermaxillary fixation for 6 to 8 weeks.

The patients were followed at the Facial Centre at The Hospital for Sick Children in Toronto. Lateral and posteroanterior (PA) cephalometric and panoramic radiographs and photographs were made before surgery, postoperatively at the time of unwiring, at 6 months, 1 year, and 2 years after surgery, and then at two year intervals. Radiographs taken at the time of unwiring were used to determine the advancement at the incisors, B point, and pogonion achieved by surgical repositioning. The PA cephalometric radiograph showed the horizontal plane of the occlusion (and the nasal floor, if a Le Fort I osteotomy had been used), as well as the position of the dental midlines and the chin midline related to the facial midline. Follow-up ranged from 1 to 9 years (from age 9 to 18). Growth was determined by direct cephalometric measurements or by inference from photographs. When a child grew in height after surgery and the chin point remained in the facial midline, the occlusion remained normal, and the occlusal and labial fissure planes remained horizontal, we presumed that growth had occurred. Four of the 22 patients did not return for follow-up and did not maintain contact. Seven of the remaining 18 children, who lived on other continents, did not return for radiographic examination but supplied photographs.

RESULTS

In nine children (age range at surgery, 6 years, 1 month to 12 years, 7 months with a mean age of 9 years, 7

Dr. Munro was a Staff Surgeon and Dr. Phillips a Craniofacial Fellow in the Division of Plastic Surgery (Department of Surgery), The Hospital for Sick Children, Toronto, ON, Canada; Dr. Griffin was an Orthodontic Fellow in the Department of Dentistry, The Hospital for Sick Children. Dr. Munro is now Director, Humana Craniofacial Institute, Dallas, TX.

Reprint requests: Dr. Ian Munro, 7777 Forest Lane, Suite C-700, Dallas, TX 75230.

months), we found measurable growth when the radiograph at unwiring was compared with the latest one available. The mean follow-up period in these children was 4 years, 5 months with a range of 1 through 9 years. The amount of growth ranged from 1 to 8 mm at B point and 1 to 11 mm at the pogonion. Seven patients showed photographic evidence of facial growth.

Five of the 18 patients developed postoperative infection. Two underwent early incision and drainage. They healed satisfactorily and showed subsequent growth. The other three developed temporomandibular ankylosis. Two of them did not return for treatment, but the third underwent release of the ankylosis 1 year after the original operation. At that time, the costochondral graft and TMJ were intact, the joint space was satisfactory, and bony ankylosis was present medial to the joint. The bone was removed, allowing normal movement of the joint and mandible, and one year later radiographic evidence of growth was present. The 16 patients available who had either no postoperative complication or adequate early treatment of complications demonstrated facial growth.

Case Reports

Patient 1 (Fig. 1) was an 8 year old boy with right-sided hemifacial microsomia who underwent a left sagittal mandibular osteotomy to allow advancement of the mandible. To bring the chin into the midline, the right side of the mandible was moved forward 15 mm, down 10 mm at the gonial angle, and laterally 5 mm. A costochondral graft was used to correct the defect in the ascending ramus and to construct the new condyle. A postoperative infection was treated promptly by incision and drainage. The patient's height increased more than 30 cm during the next 7 years and his face remained symmetric.

Patient 2 (Fig. 2) was a 9 year old boy with marked labial fissure tilt, significant displacement of the chin to the left, and absence of the posterior zygomatic arch, mandibular condyle, and ascending ramus. He underwent type IV surgical correction (Munro and Lauritzen, 1985), including a left Le Fort III osteotomy with a right Le Fort I osteotomy. A new zygomatic arch was made from full-thickness rib, and a glenoid fossa was carved out and lined with cartilage. A right sagittal mandibular osteotomy advanced the mandible, and a costochondral graft from the right chest was used to build the left ascending ramus and condyle. The patient was kept in intermaxillary fixation for 7 weeks. The correction of the labial fissure and chin point present at unwiring (Fig. 2B) was maintained over the next 9 years (Figs. 2C, 2D), during which time the patient's height had increased more than 45 cm.

Patient 3 was a 10 year old girl with a right-sided hemifacial microsomia (Fig. 3). She underwent a Le Fort I osteotomy, raising the maxilla 2 mm on the left and lowering it 5 mm on the right, together with advancement and rotation of the right side. The lower position of the maxilla on the right side was maintained with an H-shaped full thickness rib graft (Munro, 1987b). A left sagittal split of the mandible was used for advancement. A glenoid fossa was constructed in the zygomatic arch with cartilage. A left costochondral graft was used for the right ascending ramus and condyle, and an additional 6 mm advancement and rotation genioplasty was used to bring the chin forward and into the facial midline. After 8 weeks of intermaxillary fixation, the labial fissure and occlusion were horizontal, occlusion was normal, and the chin point was in the facial midline (Figs. 3C and 3E). Two years later the patient returned with overgrowth of the graft and deviation of the face and chin to the left with a class III malocclusion (Figs. 3F and 3H).

Patient 4 was a 6 year old girl who underwent a left sagittal split of the mandible with advancement, as well as construction of a right zygomatic arch, glenoid fossa, and ascending ramus and condyle, as described for Patient 2. Immediately after surgery, a panoramic radiograph showed a small ascending ramus (Fig. 4A). Nine weeks later new bone was present in the region of the absent stylomandibular ligament (Fig. 4B). One year after surgery, there was a large amount of growth in both the rib graft and the previously small ascending ramus (Fig. 4C).

DISCUSSION

The etiology of hemifacial microsomia is generally believed to be intrauterine rupture of the stapedial artery. Poswillo (1974) reproduced the deformity in animals and showed various potential areas of damage. He stated, without proof, that the effect of the damage on the functional matrix would progressively increase the deformity, so that reconstruction should be delayed until growth was complete in adolescence (Poswillo, 1978). In our experience, when untreated, the deformity does not increase with time. Measurements give the absolute difference between the two sides of the face and permit calculation of the difference between them as a percentage of the normal side. As the patient grows the absolute difference will increase, but the percentage does not change, thereby indicating that relative facial symmetry does not change.

A major advantage of correcting this facial anomaly early is psychological, and there is increasing evidence of significant benefit in correcting craniofacial anomalies as early as possible (Lefebvre and Barclay, 1982; Lefebvre and Munro, 1986; Arndt et al, 1987; Lefebvre and Arndt, 1988). when we began correcting the faces of these young children, we warned the parents that the improvement would not be permanent and that asymmetry would recur, with the chin point moving relatively backward and to the side of the original defect as the child grew. This was based on our assumption that the rib graft would not grow longer, although any inherent growth in the mandible would not be affected by the surgery. The actual outcome in these patients has shown that we were unnecessarily pessimistic.

It is reasonable to assume that the bone has grown if a child grows in height after surgery, the chin point remains in the facial midline, the occlusion remains normal, and the occlusion and labial fissure plane remain horizontal. This inference is extremely useful because accurate measurement from serial cephalometric radiographs is difficult. The radiographs can show an increase in the size of the bone graft, in both the sagittal and coronal planes. Many of these patients do not have a normal glenoid fossa preoperatively, and one has to be constructed. The cartilage at the end of the costochondral graft is not visible radiographically and, over time, may become partially ossified. Therefore, it is not



FIGURE 1 An 8 year old boy with right-sided hemifacial microsomia. A, Full-face and B, tilt preoperative photographs. C and D, 1 year after a left sagittal mandibular osteotomy and a right costochondral graft from the body of the mandible to the glenoid fossa.



FIGURE 1—Continued. E, 7 years after surgery. The patient has passed through puberty and is more than 30 cm taller, but his face remains symmetric. F, Preoperative cephalogram. G, Cephalogram taken during intermaxillary fixation immediately after surgery. H, Cephalogram showing contouring of the graft and the position of the dental midlines, 7 years after surgery.



FIGURE 2 Photographs of a 9 year old boy with the Goldenhar variant of hemifacial microsomia and an absent zygomatic arch and ascending ramus. A, Preoperative photograph. B, 1 year after osteotomies and costochondral grafting from the body of the mandible to the new glenoid fossa (see text). C, Photograph 6 years later. The patient has grown 30 cm in height. The visible bandage is where he has just had a cyst removed under local anesthetic. Note that the labial fissure is still horizontal and the chin is still in the facial midline. D, At age 18 years, the chin is still in the midline and the occlusion and labial fissure are horizontal. No orthodontic braces or external devices have been applied since surgery. The patient has grown 45 cm taller since surgery.

A





В



FIGURE 3 A, Photograph of a 10 year old girl immediately before a Le Fort I osteotomy, left sagittal mandibular osteotomy, genioplasty, and costochondral grafting from the side of the genioplasty and body of the mandible to the glenoid fossa of the zygomatic arch. B, Photograph 3 months after surgery. The chin is in the midline and the occlusion is class I and horizontal. C, Photograph 2 years later showing marked overgrowth and a class III occlusion. D, Preoperative cephalogram.

С





FIGURE 3-Continued. E, Cephalogram taken 2 months after surgery at unwiring. F, Cephalogram taken 2 years after surgery showing deviation of the jaw to the opposite side of the original deformity and a class III occlusion. G, Panoramic radiograph 2 months postoperatively, showing the costochondral graft and a small ascending ramus. H, Panoramic radiograph 2 years postoperatively, showing increased length and size of the graft and modeling with the ascending ramus.

possible to determine a fixed point radiographically at the proximal end of the graft. The rib is beveled distally (Munro, 1980 and 1987), and the inner cortex is removed so that the cancellous bone and outer cortex lie flush with the anterior part of the body of the mandible and, after a few weeks, fuse with the mandible cortex and become radiographically invisible. The anterior reference point (usually B point) may be altered by a genioplasty, which also induces bone growth at this point, or by the rib graft that has been brought forward to overlap part of the genioplasty (Munro, 1987a).

Metal bone markers and the wires used to fix the bone have been used as reference points for growth. These techniques assume that the metal is pushed along by bone growth. Bone growth in the face, however, occurs by a combination of apposition and resorption. On reoperation, metal wires are often buried partially or completely in the bone, which indicates that bone changes can occur around a piece of metal without necessarily moving it. Thus, change in position of a piece of metal cannot accurately measure bone growth when the bone may grow without moving the metal. Cephalograms are therefore not sufficiently reliable



FIGURE 4 A, Panoramic radiograph of a 6-year-old girl with right hemifacial microsomia who has just undergone a left sagittal mandibular osteotomy and construction of a right zygomatic arch with a full-thickness rib. A glenoid fossa has been carved out and lined with cartilage, additional split rib has been overlaid onto the zygoma, and a costochondral graft passes from the anterior body of the mandible to the new glenoid fossa. An acrylic bite wafer has been used to produce an open bite on the involved side. *B*, Panoramic radiograph taken immediately after unwiring 9 weeks postoperatively. Note the lengthening of the previously small ramus and signs of bone formation toward the styloid process. *C*, Panoramic radiograph taken 1 year after surgery. The open bite has remained even without treatment. The costochondral graft has enlarged and changed shape, and the patient has grown most of a new ascending ramus. She has full mouth opening.

to differentiate between the growth of the costochondral graft and that of the original mandible. In addition, measurements on lateral cephalometric radiographs are made in a straight line between two points, but the graft grows longitudinally, transversely, and over a curve that cannot be measured from a lateral or anteroposterior cephalograph (Fig. 3E). In any case, whether the growth was from the cartilaginous or bony part of the costochondral graft or from the basal mandible is largely academic. The fact that the face maintained the correction is the only factor of importance to the patient or parent.

Ware and Taylor (1965 and 1966) showed the continual growth of both transplanted rib cartilage and metatarsal epiphyses when transplanted in growing rhesus monkeys. Schatten et al (1958) found that costochondral grafts placed heterotopically in the axilla of rats grew in only 70 percent of those left in situ. Autologous grafts used to construct TMJs have included metatarsal bone, the clavicle and sternoclavicular joint (Daniels et al, 1987), as well as costochondral grafts. The morbidity and unesthetic scarring produced on the foot or anterior neck by other grafts make them unacceptable, especially because no growth or technical advantage has been shown. Pedicle flaps of sternomastoidclavicle or pectoralis-rib and free flaps such as composite iliac flaps have a much greater morbidity than simple costochondral grafts. Although pedicle or free flaps are indicated in irradiated patients, there is probably little or no indication for their use to reconstruct the condyle or to repair the bony deformity in patients with hemifacial microsomia.

The amount of soft tissue deficiency is often overestimated because the underlying skeleton is grossly malpositioned. Once the skeleton is correctly aligned, absent bone constructed, and hypoplastic bone augmented, most patients will not need soft tissue augmentation by a free flap. Free autogenous costochondral grafts are now accepted widely for the reconstruction of the TMJ in a variety of conditions for which irradiation is not a problem (Obwegeser, 1974; McIntosh and Henny, 1977; Ware and Brown, 1981; James and Irvine, 1983; Lindqvist et al, 1986). Ware and Brown (1981) showed definite growth of costochondral grafts in 10 children, but the etiology was either for the correction of temporomandibular ankylosis or for removal of the condyle and ramus for a benign tumor.

Four published reports show evidence of growth in a free costochondral graft in a child with hemifacial microsomia. Figueroa and colleagues (1984) found that a graft in an 8-year-old boy grew 10 mm in 6 years and that the mandible moved forward substantially. McIntosh and Henny (1977) reported growth of a costochondral graft placed in a 5year-old child after 4 years. Lindqvist et al (1986) reported on the use of costochondral grafts in 60 patients for a variety of problems at various ages. They stated that postoperative growth of the graft was present in a 9 year old child but gave no details. Ortiz-Monasterio and Fuente Del Campo (1985) reported on 12 children who underwent a Le Fort I osteotomy and construction of the TMJ with a costochondral graft between 4 and 5.5 years of age. Throughout follow-up, up to 6 years, the face stayed in the midline. They anticipated that further surgery may be necessary after the pubertal growth spurt. Nine of our patients have reached puberty, but none has shown relapse and two have experienced overgrowth.

It is difficult to explain fully what is happening in these children. Initially we speculated that the increased prepubertal production of growth hormone was significant. However, many of these children showed evidence of growth before this could be expected. The functional matrix theory of Moss (Moss and Rankow, 1968; Moss and Salentijn, 1969) could explain the continuing growth in most patients but not the overgrowth in two children. By anecdote at various professional meetings we have heard of other cases in which overgrowth occurred.

The costochondral graft is always taken from the chest opposite to the side of the reconstruction so that the best convexity to the graft is obtained to produce maximum facial width. We choose the rib that will produce the least conspicuous scar, usually the fifth, sixth, or seventh. It would be interesting to know the potential longitudinal growth of a costochondral graft in situ in the chest and whether the growth potential is transferred to the face or whether the amount of growth in the graft is determined by the functional matrix of the face. Unfortunately, we have been unable to find out how much longitudinal growth occurs in ribs and their adjacent cartilage in the chest wall.

The type of surgery performed in our patients varied according to the age of the patient, the degree of abnormality, and the geographic origin of the patient. Although, like Ortiz-Monasterio and Fuente Del Campo (1985), we have performed Le Fort I or even hemi-Le Fort III plus Le Fort I osteotomies in very young children, this was not routine. However, when the mandible is leveled, we have advanced the normal side as well with a sagittal split osteotomy, even in young children. If the child could be followed orthodontically, we sometimes omitted the Le Fort I osteotomy, created an open bite on the side of the deformity, and allowed progressive eruption of the teeth with a bite wafer. Apart from this short-term use of a bite wafer for a few weeks after removal of intermaxillary fixation in some cases, no other orthodontic devices were used to maintain the corrected position or to "stimulate" growth of the jaw or graft. There was no correlation between specific technique, patient age, the amount of surgery done, the number and variety of osteotomies, and subsequent growth.

Construction of a TMJ in a child carries considerable surgical and postoperative risk and should be undertaken only by surgical teams who perform such surgery regularly and only in a facility fully equipped for and experienced in managing major pediatric problems. However, it now seems that early surgery for patients needing construction of the TMJ is indicated for growth as well as for psychosocial reasons. A few more years will be needed to determine whether further surgery after adolescence will always be obviated. Costochondral grafts definitely grow. They are associated with low morbidity when used in a child. The cartilage cap prevents the occurrence or recurrence of temporomandibular ankylosis in hemifacial microsomia or correcting established temporomandibular ankylosis (Munro et al, 1986). Unless it can be proven experimentally in animals that alternatives such as cranial grafts or composite free-flap iliac grafts produce better growth or appearance, we feel that their use is not indicated.

Drs. Arlene Dagys and R. Bruce Ross and members of the orthodontic team at The Hospital for Sick Children for help with these cases. Dr. Ross also helped us prepare this article. The authors would like also to thank Carla Salvador for editorial assistance.

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Acknowledgments. The authors would like to acknowledge the support of