Distraction Osteogenesis of the Human Craniofacial Skeleton: Initial Experience with a New Distraction System

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Application of distraction osteogenesis to the human craniofacial skeleton in properly selected cases represents a major advance in the treatment of craniofacial deformities. We report our initial clinical experience with a system of miniature distraction devices that permitted maxillary, orbital, and mandibular distraction in a 4-month-old boy with unilateral craniofacial microsomia and anophthalmia. At 6 months of age, after maxillary repositioning and orbital expansion, a costochondral rib graft was used to construct the missing left mandibular ramus and condyle.

Key Words: Distraction osteogenesis, maxillary distraction, orbital expansion, temporomandibular joint reconstruction

The concept of distraction osteogenesis was championed by Ilizarov, beginning as early as 1954, for the treatment of a variety of congenital and acquired deformities of enchondral bone [1]. In 1973, Snyder and associates reported on gradual distraction of the mandible using an extraoral device in canines [2]. After successfully lengthening the canine mandible [3], McCarthy and his group reported the first clinical application of distraction osteogenesis of the mandible in four patients with a variety of congenital craniofacial anomalies [4]. Because the technique of gradual, osseous distraction of the membranous bones of the human craniofacial skeleton promises major advances in the early reconstruction of severe craniofacial anomalies, numerous reports of clinical mandibular distraction as well as experimental distraction in other areas of the craniofacial skeleton have ensued. Osseous expansion of the cranial vault was reported by Persing and colleagues [5], and distraction of the frontal bone outside of the cranial plane was demonstrated by Barone and coworkers [6]. Successful expansion of the cranial vault using a craniotactic device in rabbits was presented by Remmler and his coworkers [7]. More recently, midfacial advancement was shown to be feasible in adult sheep using lengthening "bolts" mounted on transversely placed pins [8]. Using a modified Hoffman bone-lengthening device, Staffenberg and associates [9] reported successful midface distraction and advancement in the immature canine without osteotomies.

The application of distraction osteogenesis to the human craniofacial skeleton potentially represents a major advance in the treatment of craniofacial deformities. The technique is less invasive than conventional surgery and possibly more stable. In addition, once developed, it is likely that even more complex manipulation of the craniofacial skeleton will be achievable. Herein, we report our initial clinical experience with a system of miniaturized distraction devices that permit three-dimensional movements of the human craniofacial skeleton.

Method

A 4-month-old boy presented with left craniofacial microsomia (Fig 1). At birth the child had upper airway obstruction and was transferred to Scottish Rite Children's Medical Center, where he underwent tracheostomy. Because of extremely poor feeding, a gastrostomy tube was also placed during the same admission. Once the parents were trained in how to care for the tracheostomy and the gastrostomy tube, the patient was discharged home.

At 3½ months of age, the child returned to the operating room, where orbital impressions and a facial moulage were obtained. Preoperatively, a three-dimensional computed tomographic (CT) scan was also procured (Fig 2). In the operating room anteroposterior (AP) and lateral cephalograms were taken with an intraoperative cephalostat (see Fig 2). For the mandibular device, microplates and screws were chosen, whereas for the craniomaxillary device, micro-plus/panfacial-size plates were

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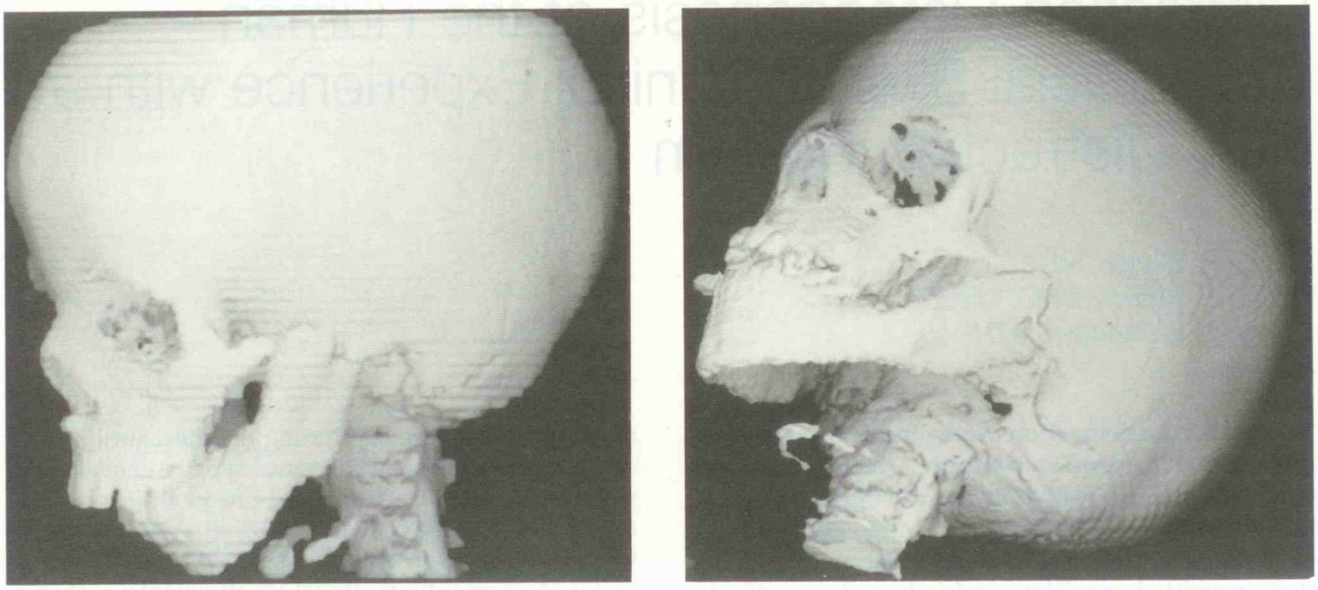

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Fig 4 (A) The left temporomandibular joint of the same patient is also flat and malformed. (B) Postexpansion, the condyle has uprighted with an increase in volume. No displacement of the glenoid fossa was noted.

cartilaginous portion of the condylar head [5]. Excessive compression of the proximal segment is also implicated in temporomandibular joint resorption and relapse after orthognathic surgical procedures [6]. Chronic compressive forces have also been implicated as a contributing factor in the development of temporomandibular joint disorders, including internal derangements [7, 8].

In this clinical study, bone distraction, however, appeared to have a beneficial effect on the temporomandibular joint. Patients with craniofacial deformities manifest abnormalities of the facial skeleton often including the temporomandibular joint. Depending on the degree of disease, the temporomandibular joint may be malformed or absent. The condyle is often malopposed as well as misshaped. Osteodistraction appeared to stimulate the pathological condyle to reorient to a more normally oriented vertical axis as well as to increase in size and volume. In bilaterally expanded cases, such stimulation was expressed on both sides, causing the two condyles to become more closely symmetrical. In unilaterally expanded cases, the affected condyle came more closely to resemble the unaffected side. The unaffected condyle did not appear to be influenced by the contralateral expansion because no gross changes were noted.

Although the condyle is important in growth and development of the mandible, the condyle is only a growth site responding to the soft-tissue forces to which it is subjected [9]. These forces appear to distract the mandible in a downward and forward direction, leading to deposition along the posterior aspect of the mandible with resorption along the anterior ramus region [10]. One can envision that the expansion device may mimic and enhance this soft-tissue developmental effect [1]. Instead of causing deleterious compressive forces on the temporomandibular joint, the bone expansion device appears to stimulate the affected condyle to assume a more upright position and to increase in size and volume.

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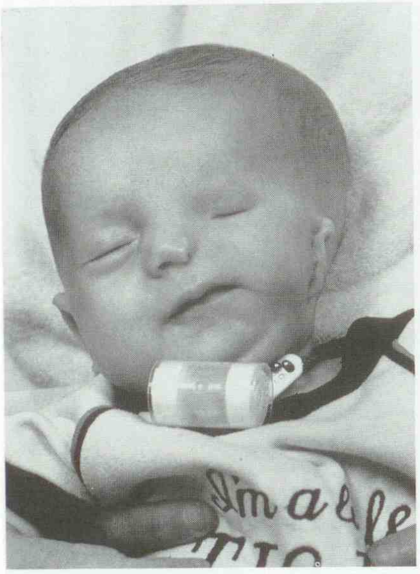
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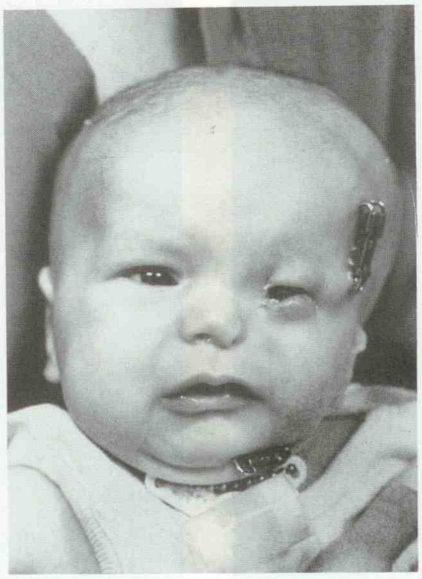
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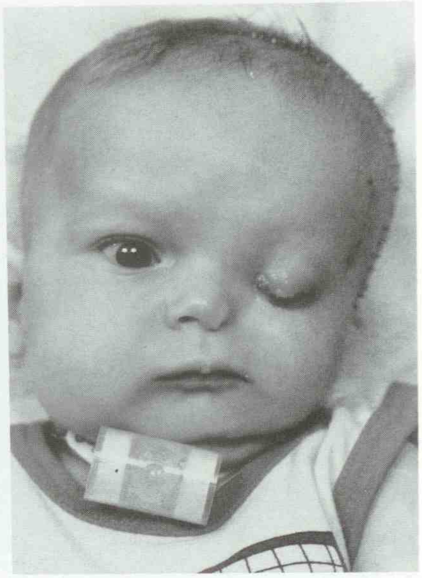
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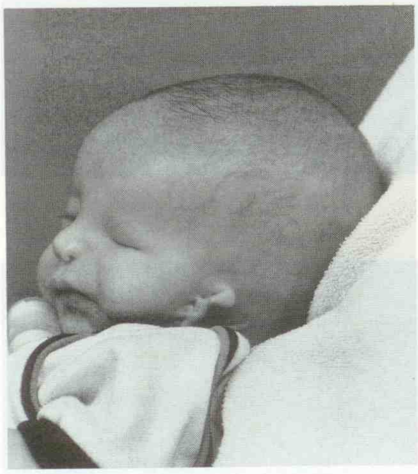
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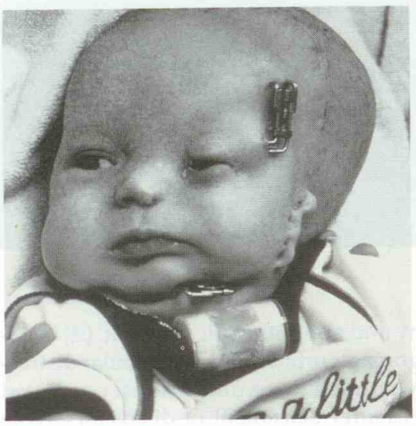
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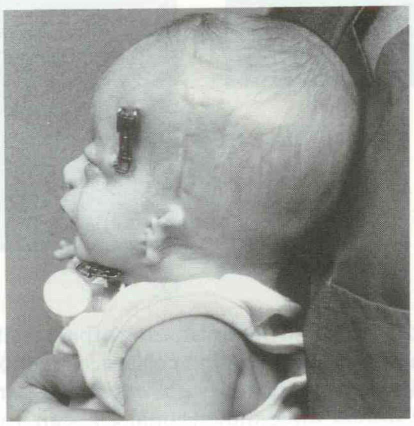
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Fig 1 (A) Preoperative frontal photograph of patient at 4 months of age before undergoing craniofacial distraction osteogenesis. Note left anophthalmia, left microtia, as well as the discrepancy in the oral commissure to lateral canthal distances. (D) Preoperative lateral view. (F) Postoperative fronto-oblique view 3 weeks after placement of craniomaxillary, mandibular, and orbital distractors. Note improvement in vertical maxillary dimensions and occlusal cant. (E) Latero-oblique view 3 weeks after placement of distractors. (B) Frontal view 9 weeks after placement of distractors. Distractors have been fully activated since the third postoperative week. Patient is shown just before removal of devices. (G) Lateral view of affected left side 9 weeks after placement of distractors. (C) Frontal view after distractors were removed and 3 months after costochondral grafting of left ramus and condyle.

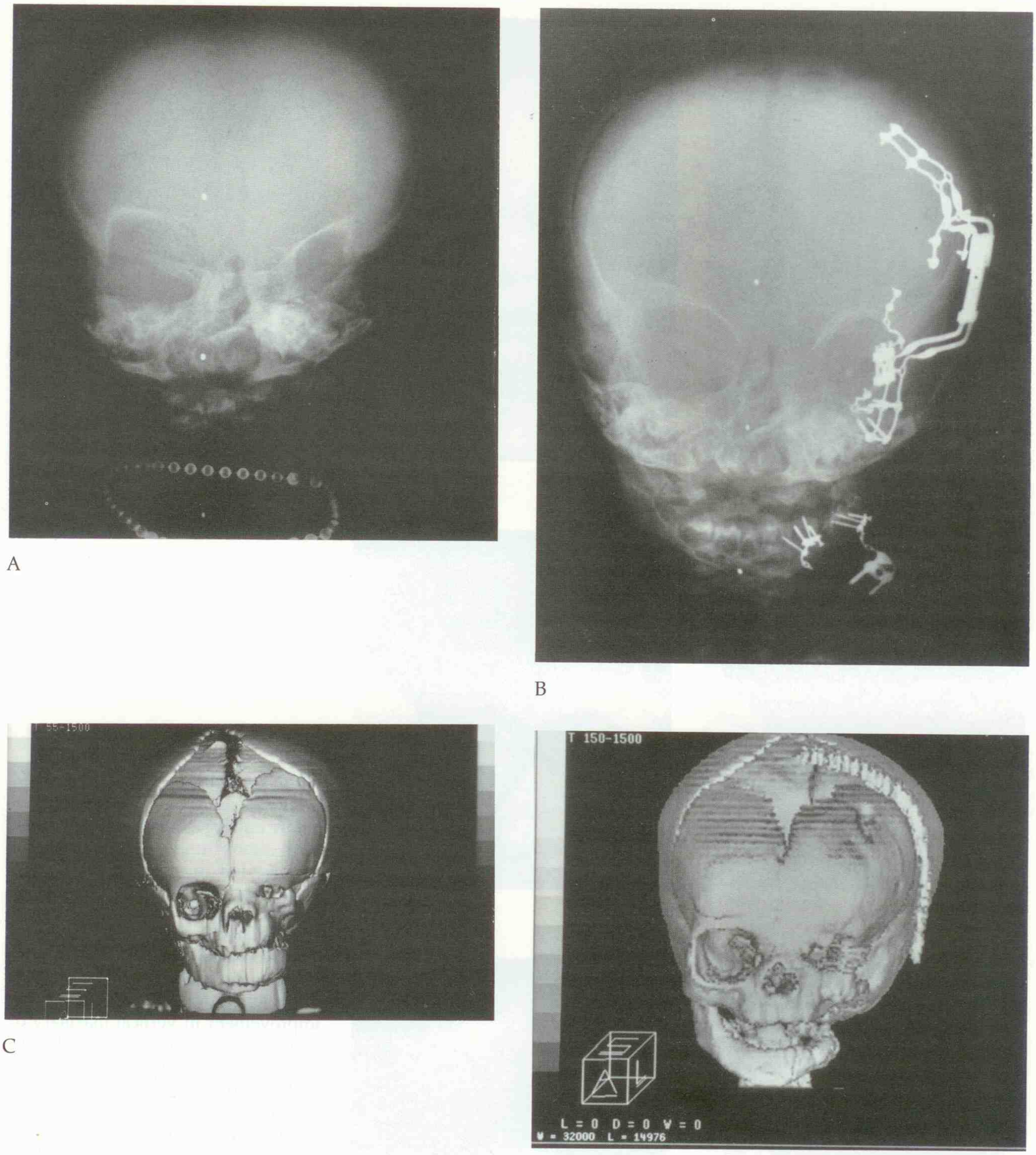

Fig 2 (A) Intraoperative anteroposterior cephalogram demonstrating orbital anatomy and anophthalmic left orbit. (B) Intraoperative cephalogram 9 weeks after first surgery demonstrating position of craniozygomatic, orbital, and mandibular distractors. Note that the mandibular distractor has broken at solder point. (C) Preoperative frontal three-dimensional computed tomographic (CT) scan. (D) Postoperative frontal three-dimensional CT scan 9 weeks after placement of distractors. Note increased bone in the mandibular angle region and improvement of occlusal cant as well as enlargement of orbital cavity. Artifacts are seen along coronal incision line and orbital cavity.

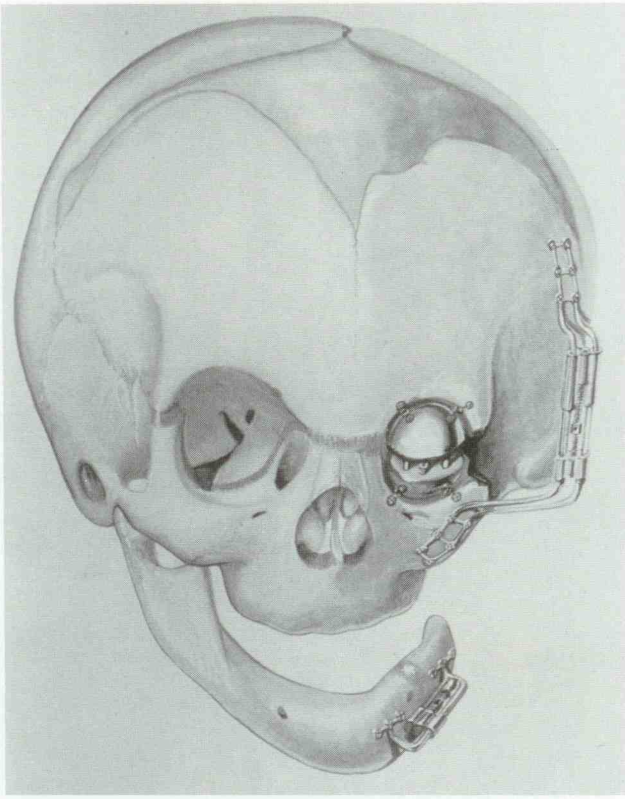

Fig 3 Artist's depiction of craniomaxillary, orbital, and mandibular distractors activated with gradual correction of facial asymmetry.

used. The devices were then fabricated with orthodontic expansion screws soldered to the metallic plates. Two weeks later, the child underwent a modified Le Fort type III osteotomy with 270-degree internal orbital osteotomies and a mandibular osteotomy. A craniomaxillary device was placed to correct the severe vertical maxillary deficiency and occlusal cant. An orbital prosthesis was placed to expand the anophthalmic orbit and distend the residual hypoplastic upper and lower eyelids. The mandibular distraction device was used to distract the mandibular body, which itself was hypoplastic (Fig 3). The ramus and condyle were absent. Total operative time was less than 3 hours with an estimated blood loss of 50 ml. The patient was brought to the intensive care unit and transferred to the floor the next morning. The patient's diet was progressed rapidly, and on the third postoperative day, before discharge, the expansion screws were activated 1 mm. The parents were instructed to activate the expansion-distraction screw 1 mm every other day.

Three weeks after discharge from the hospital, the family returned to the office. The mandibular and craniomaxillary distraction screw had each been opened 11 mm. The orbital screw had been expanded 5 mm. No complications had occurred, and the family did not indicate problems with the distraction devices. All plate exit sites and pin sites were inspected and found to be clean without evidence of infection of the tracts. Over the ensuing 2 weeks, the family expanded the craniomaxillary device to a total of 17 mm, its maximal expansion dimension. The mandibular device was expanded to 13 mm and the orbital expander to a total of 8 mm.

On the return visit, no infections of the pin tracks were noted. Crazy Glue was applied to each of the expansion screws to lock them in position. Preparations were made to return the patient to the operating room 6 weeks later for removal of the devices and replacement of the orbital expander. Substantial correction of the patient's occlusal cant was noted. The eyelids themselves had expanded considerably as had the anophthalmic orbit. Clinically, the body of the mandible appeared to be increased in its AP dimension, and the maxillary and mandibular midlines appeared to be more coincident with the true facial midline. At reoperation, the devices were removed and the osteotomy sites inspected. The mandible appeared to have new bone formation, and there was no evidence of nonunion. Some bony ingrowth over the plates had occurred in the mandible but not in the cranium or maxilla. The osteotomy site in the region of the lateral orbital wall appeared to be filled with new bone. The craniomaxillary expansion device did not demonstrate buckling from its initial position. The screws were secure. The orbital device was well seated. A new orbital distractor was placed. To maintain the improvement in craniofacial form, a costochondral graft was used to reconstruct the left mandibular ramus and condyle. Total operative time was 4 hours, with an estimated blood loss of 75 ml. AP and lateral cephalograms were taken in the operating room. A three-dimensional CT scan was procured in the immediate postoperative period. The patient tolerated the procedure without difficulty. The patient, who is now 14 months old, has been followed for approximately 4 months since costochondral rib graft reconstruction. Some recurrence of occlusal cant has been noted. Incisal opening is excellent. Attempt at a decannulation of the patient's tracheostomy is anticipated after 18 months of age.

Preoperative Evaluation

Patients undergoing craniofacial distraction osteogenesis have complex three-dimensional deformities. A careful preoperative physical examination is essential to develop a proper treatment plan. The sites of osteotomies and the position of the distraction devices must be determined. Multiple distraction devices can be placed. It is generally best to pick a vector of distraction for each of the