Restitution of the Temporomandibular Joint in Patients with Craniofacial Microsomia After Multiplanar Mandibular Distraction: Assessment by Magnetic Resonance Imaging

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The purpose of this pilot study was to investigate the response of hypoplastic temporomandibular joints (TMJs) to mandibular distraction osteogenesis. This preliminary study describes changes in 2 male patients with unilateral craniofacial microsomia who were 5 years of age at the time of surgery. Spin echo sequence images of the TMJs without contrast media in axial, coronal, and sagittal views, along with sagittal kinematics studies, were obtained with the use of magnetic resonance imaging (MRI) at: 1 month preoperatively (T1); immediately upon removal of distraction devices (T2), and 14 months postoperatively (T3). At the same time points spiral 3D computed tomography (CT) was used to image the TMJs. Both MRI and CT data obtained were assessed by standard, qualitative interpretation. Predistraction MRI and CT data documented a hemifacial microsomia type IIb deformity with hypoplasia of the condyle and a dysfunctional TMJ on the affected side. After distraction, the MRI and CT data in both patients demonstrated no changes in the nonaffected TMJs. However, on the affected and distracted side the following changes were observed: (1) formation of a rudimentary glenoid fossa and articular eminence; (2) functional displacements of the rudimentary condyle-disk complex; (3) well-defined visualization of the temporalis and lateral pterygoid muscles; (4) increased signal intensity radio-density of the pseudodisk fibrous tissue. Thus, both patients showed improvements in the morphology and function of the TMJ as well as in the associated skeletal and soft tissue components. It was concluded that following mandibular distraction in young children, improved form and function of the TMJ complex is demonstrable using MRI. (Semin Orthod 2011;17:186-196.) © *2011 Published by Elsevier Inc.*

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The temporomandibular joint (TMJ) is a compound, craniomandibular articulation that results from the bilateral articulation of the mandible with the posterior cranial base. The TMJ plays an important role in mastication, swallowing, and speech, and also in respiration, facial symmetry/esthetics, and facial expression.[1] Normal function of the TMJ involves a complex mechanism in which the movable mandible articulates with the immobile, squamous part of the temporal bone at the glenoid fossa. Although movements occurs within each joint independently, both are functionally related and are affected by associated skeletal and soft-tissue components, such as the mandibular condyle, articular disk, and lateral pterygoid muscles, as well as by the dental occlusion.

Craniofacial microsomia (CFM) involves developmental anomalies of the skeletal and soft-tissue derivatives of the first and second branchial arches and, accordingly, its phenotypic expression presents as a variable and progressive craniofacial asymmetry.[2] Thus, TMJ function may be affected by the presence of hypoplastic skeletal and/or soft-tissue joint components on the affected side. The degree of dysfunction, however, may vary as much as the severity of the anomaly itself, with differences in joint morphology and functional displacements of the articular components. However, in unilateral CFM, the contralateral or unaffected joint shows a craniomandibular articulation that is morphologically normal and functionally stable.

The application of distraction osteogenesis techniques to the craniofacial skeleton has expanded the number of treatment alternatives for patients with craniofacial abnormalities. Since its introduction for the treatment of fractures and nonunions in long bones,[3] laboratory studies have provided evidence of its feasibility in the membranous bones of the craniofacial skeleton.[4] Thus, the technique has been applied to treat patients with a wide variety of developmental and acquired craniofacial deformities.[5,6] Indeed, clinical experience over 12 years[7] has demonstrated that mandibular distraction is a safe and effective surgical technique. Aside from other advantages compared with traditional orthognathic surgical procedures, an important additional benefit is that gradual distraction is thought to augment not only the bony skeleton but also the associated soft tissues, such as the masticatory muscles, subcutaneous tissues, and skin.

It is postulated that multidirectional expansion of the skeletal and soft-tissue envelope may be responsible for a reduced skeletal relapse rate.[8] This notion contrasts with the relapse observed in traditional orthognathic procedures, presumably associated with immediate or nongradual surgical elongation of the severely hypoplastic mandible.[9-11] It was noted that although costochondral grafts are suitable for the reconstruction of the mandibular condyle, there is some unpredictability, including infection and "overgrowth," associated with this particular procedure.[12] Therefore, the present study was undertaken to describe changes in TMJ form and function after unilateral mandibular distraction in 2 patients with CFM with hypoplasia of the affected mandible and TMJ.

Methods

Patients

This preliminary study is on the basis of the radiographic studies of 2 male patients who were 5 years of age at the time of distraction. Both patients were diagnosed with unilateral left-sided CFM (type IIb) and had never undergone any type of surgical reconstruction. Both patients were operated on the same day by the same surgeon (J.G.M.).

Orthodontics

After informed consent was obtained, as in the preparation of a patient for orthognathic surgery, predistraction orthodontic therapy was undertaken to remove any dental compensations, coordinate arch widths, correct occlusal plane disharmonies, and correct dental crowding. Fixed orthodontic appliances with passive rectangular arch wires and surgical hooks were placed for the use of intermaxillary elastics.

During the activation phase of distraction, growth guidance of the bony regenerate was undertaken before the consolidation phase. Therefore, a bony regenerate was formed first by activation of the distraction device, and then intermaxillary elastics were used to guide the directionality of skeletal change and adjust the occlusal and facial outcomes of distraction.

After completion of activation, the distraction device was maintained in position for approximately 12 weeks (consolidation phase). The device was not removed until there was radiographic evidence of a cortical outline or mineralization of the regenerated portion of the mandible. After satisfactory completion of this stage, the device was removed in the office as an outpatient procedure. Postdistraction orthodontic treatment was undertaken to achieve maximal intercuspation and a stable occlusion. This phase was followed by photographs, cephalogram, panoramic radiograph, 3D computed tomography (CT) scanning, and dynamic magnetic resonance imaging (MRI).

Surgical Technique

A multiplanar, 4-pin extraoral distraction device (Stryker Leibinger Gmbh Co, Freiberg, Germany) was used on both patients. An intraoral incision was performed over the oblique line of the mandible, and its buccal surface was exposed in the subperiosteal plane. A trocar was used to allow percutaneous insertion of the self-screwing half-pins. The osteotomy on the buccal surface, as well as the superior and inferior cortical borders, was completed with a reciprocating saw under saline irrigation. The distraction device was applied and tightened. The osteotomy was completed with an osteotome that was gently tapped and rotated until the bony segments separated. Approximating the segments with the device narrowed the bony gap created by the osteotomy. The intraoral wound was irrigated with saline solution and closed with layer of interrupted 4-0 chromic catgut sutures.

The mandible was maintained in fixation for 5 days (latency phase) before activation was started at a rate of 1 mm per day, with a rhythm of 4 times per day (0.25 mm per screw-turn; activation phase). Upon completion of activation, the distraction device was maintained in position for at least 10 weeks (consolidation phase). All procedures were performed by a single surgeon (J.G.M.).

MRI Scanning

A GE SIGNA Horizon 1.5 T MR Scanner (GE Healthcare, Waukesha, WI) was used to evaluate the TMJ complex both in children before and after multiplanar mandibular distraction. A Burnett TMJ device (Medrad, Inc, Indianola, PA) was used to measure maximum mouth opening and then used intraorally to control mouth openings at each phase of MRI scanning, to minimize mouth movement. A sagittal image of articular motion was scanned every 3 mm, and the data were used to generate cine-mode animations and static prints.

For open and closed mouth imaging, the same imaging parameters were maintained. A sequence of different imaging planes was established, and a comprehensive study of the TMJ was performed. Only T1-weighted images were acquired for high-resolution anatomical depiction of the TMJ. MRIs of the TMJ were performed at the following time intervals: 1 month preoperatively (T1), immediately upon removal of distraction devices (T2), and 14 months postsurgically (T3).

One dentist experienced in oral radiology and 2 medical radiologists independently evaluated each MRI scan by using routine, qualitative description, and reporting protocols. For each time interval, descriptive anatomy, abnormal findings, and function analysis through cine-mode animation were reported for each TMJ.

In addition, standard spiral 3D CT scans of both patients were taken, followed by 3D reconstruction of the data. Superimposition of pre- and postreconstructions was undertaken to identify specific areas of remodeling in the TMJ complex.

Results

3D CT Findings

Preoperative 3D CT reconstructions of the craniofacial skeleton in both patients diagnosed with CFM showed a deficient mandibular ramus and condyle on the affected side (Fig 1A) as well as a nonexistent glenoid fossa and articular eminence (Fig 2A). These deficiencies resulted in skeletal (Fig 3A) and soft tissue (Fig 3B) asymmetry with a mandibular deviation towards the affected side.

The postoperative 3D CT reconstructions showed new bone formation in the area of the ramus (Fig 1B), adequate enough to overcorrect the skeletal (Fig 3C) and soft-tissue asymmetry (Fig 3D). This overcorrection is needed to overcome any anticipated relapse. The postoperative

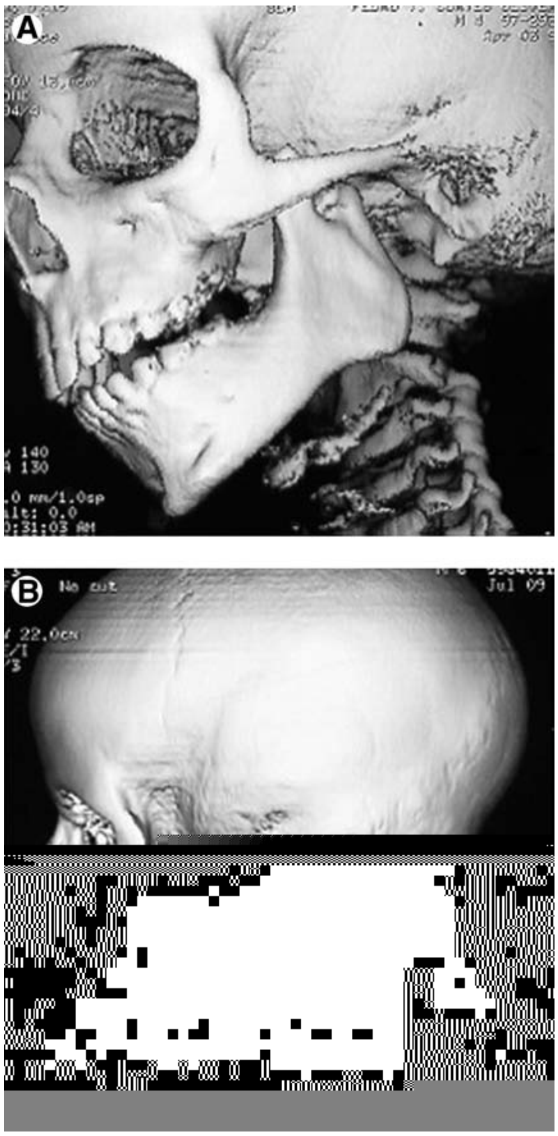

Figure 1. (A) Preoperative 3D CT reconstruction of the craniofacial skeleton in both patients diagnosed with CFM (Type IIA) show a deficient mandibular ramus and condyle on the affected side. (B) The postoperative reconstruction shows new bone formation in the area of the mandibular ramus.

differences were detected mainly in the morphology of the squamous part of the temporal bone of the affected joints. Specifically, the change from a nonexistent glenoid fossa and articular eminence (Fig 2A) to a rudimentary, yet incomplete, fossa and articular eminence (Fig 2B) was observed on the 3D reconstructions. Superimpositions of pre- and postreconstructions localized the specific areas of remod-

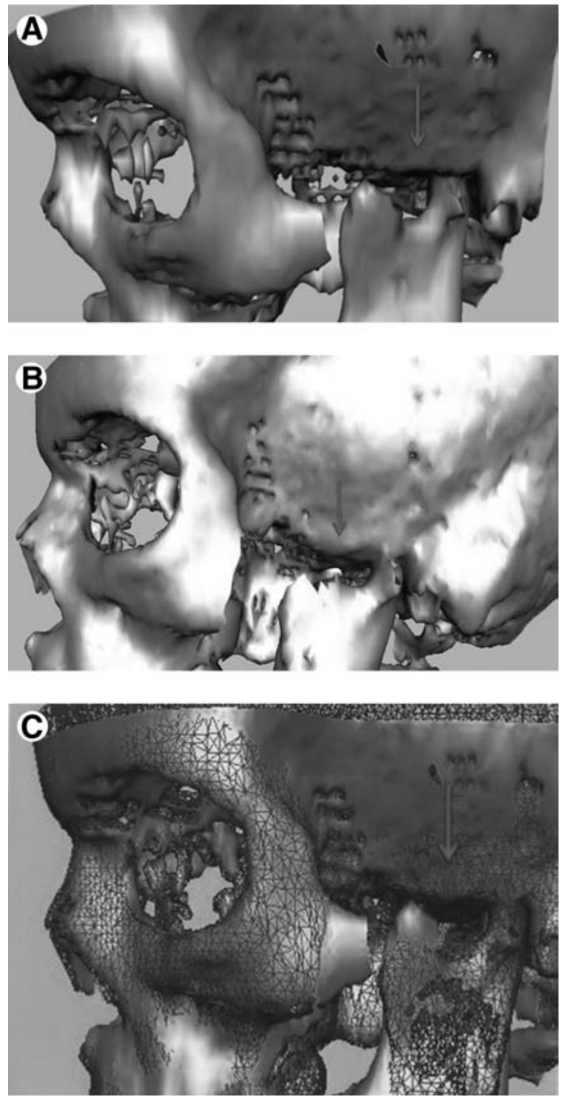

Figure 2. (A) Preoperative 3D CT reconstruction. Note the flat appearance of the squamous part of the temporal bone of the affected joint showing absence of the glenoid fossa and articular eminence. (B) The 3D CT reconstruction after the distraction procedure presents the differences in the morphology of the squamous part of the temporal bone showing a presumptive glenoid fossa and articular eminence. (C) Superimposition of the predistraction (blue, solid model) and postdistraction (red, wire-framed model) using 3D CT reconstructions of the TMJ, localizing the areas of remodeling. Note the formation of a rudimentary, incomplete, glenoid fossa and articular eminence, corroborating the MRI findings reported. (Color version of figure is available online.)

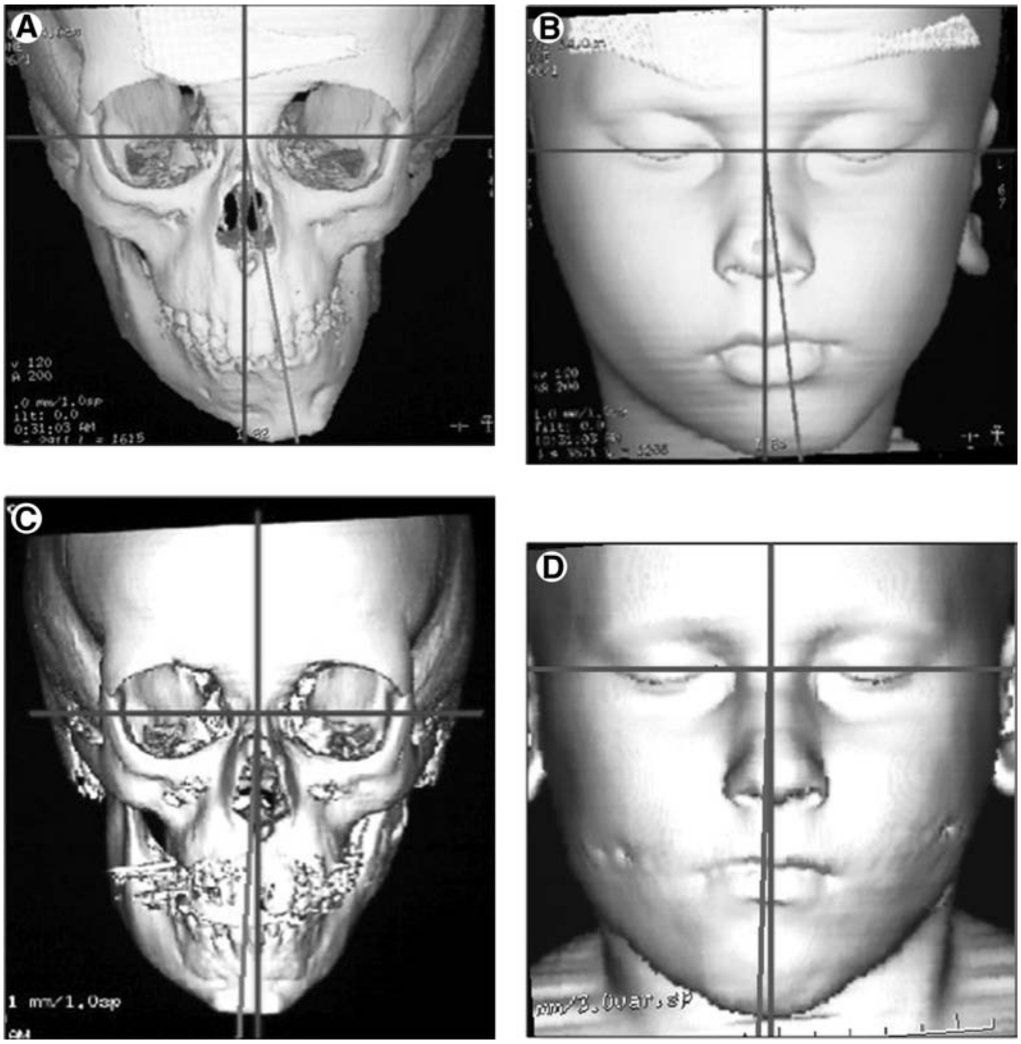

Figure 3. Frontal view of the preoperative 3D CT scan reconstructions showing the skeletal (A) and soft tissue (B) asymmetry. Note the severe mandibular deviation towards the affected side. After distraction, the skeletal (C) and soft tissue asymmetry (D) is corrected by elongating the mandibular ramus. Note the slight overcorrection to the non-affected side needed to overcome anticipated relapse and postdistraction asymmetric growth. (Color version of figure is available online.)

eling of the TMJ complex (Fig 2C) corroborating the MRI findings reported in the next section.

MRI Findings

The MRI technique was found to be time-efficient and did not affect image quality. MRI image of a normal TMJ in a closed mouth position demonstrated a well defined temporalis muscle; a normal contour of the condylar head; an articular eminence and glenoid fossa; and a defined contour of a low signal articular disk (Fig 4). The open mouth position's image displayed well-defined pterygoid muscles; the articular disk at a proper position on the eminence and condyle; and a proper condyle-temporal bone relationship (Fig 5).

A dynamic MRI of a normal TMJ at opening demonstrated rotation around a horizontal axis through the condylar heads, followed by anterior translation of the condyle and meniscus beneath the articular eminence.

The MRI image of the TMJ of the patients diagnosed with CFM in a closed positioned showed an atrophic temporalis muscle; a hyp-

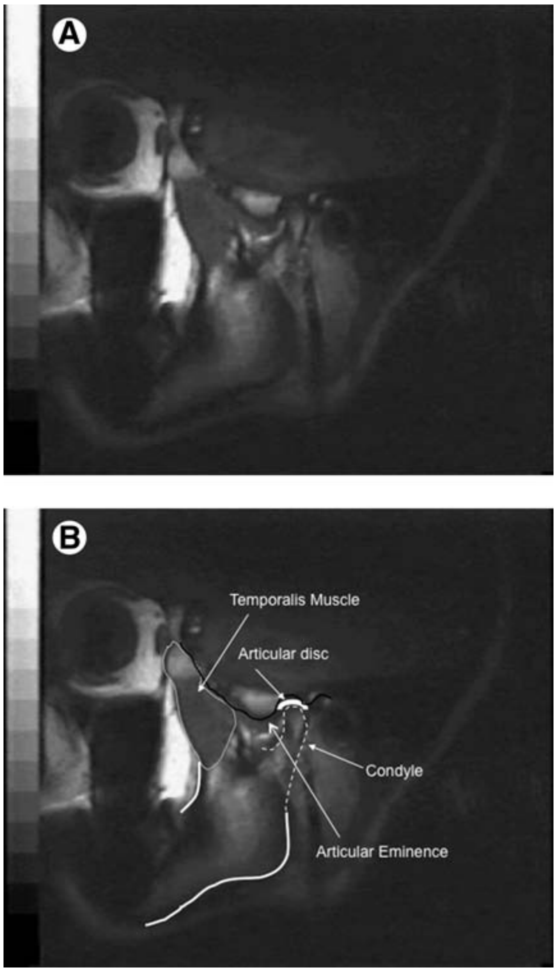

Figure 4. (A) MRI image of a normal TMJ in a closed mouth position presenting a well-defined temporalis muscle, a normal contour of the condyle, articular eminence, glenoid fossa, and a defined articular disk. (B) Outline and labels for A.

oplastic condyle; a low-signal fibrous band (pseudodisk); and a temporal bone without a glenoid fossa or articular eminence (Fig 6). The open position displayed an abnormal displacement of the condyle with no condyle-temporal or condyle-disk relationships, and poorly defined pterygoid muscles (Fig 7). Preoperative cine-mode animations showed that both patients displayed a complex motion of the underdeveloped joints, with an absence of hinge axis and translation displacement movements in the opening and closing motions of articulation.

The following changes in the affected TMJ after mandibular distraction were found in both patients

1. Differences in TMJ morphology between preoperative (Figs 1A, 2A, 6, and 8) and postoperative (Figs 1B, 2B, 9, and 10) images, consistent with the formation of rudimentary elements of a new TMJ on the affected side. These differences were detected mainly in the morphology of the squamous part of the temporal bone of the affected joints. Specifically, the change from a nonexistent glenoid fossa and articular eminence to a rudimentary, yet incomplete, fossa and eminence were depicted on the sagittal views of the joints, in both the static and animated images.
2. Changes in functional displacements of the hypoplastic condyle-disk complex consis-

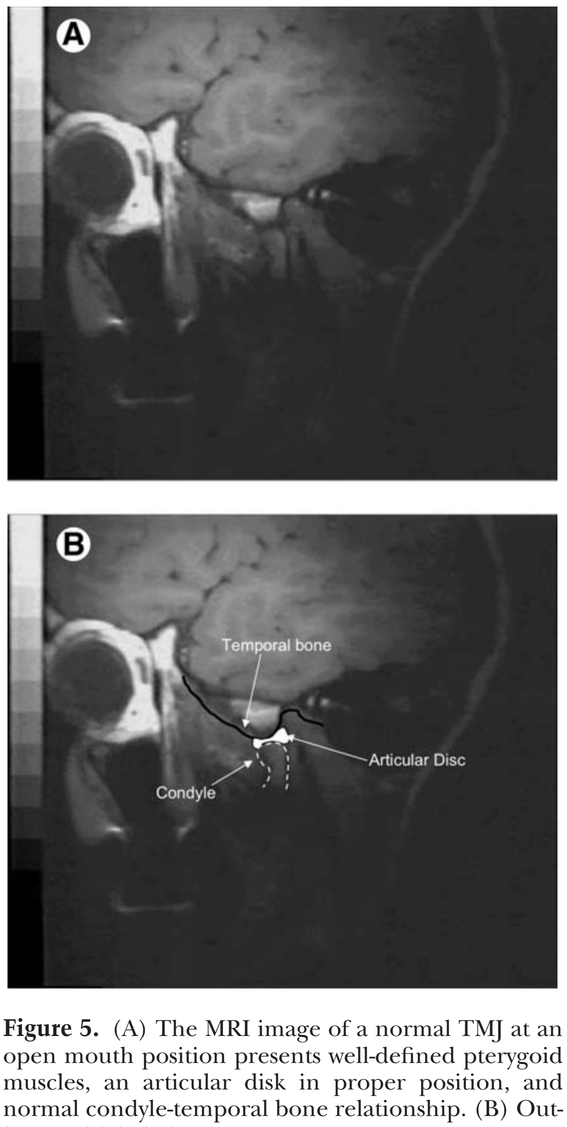

Figure 5. (A) The MRI image of a normal TMJ at an open mouth position presents well-defined pterygoid muscles, an articular disk in proper position, and normal condyle-temporal bone relationship. (B) Outline and labels for A.

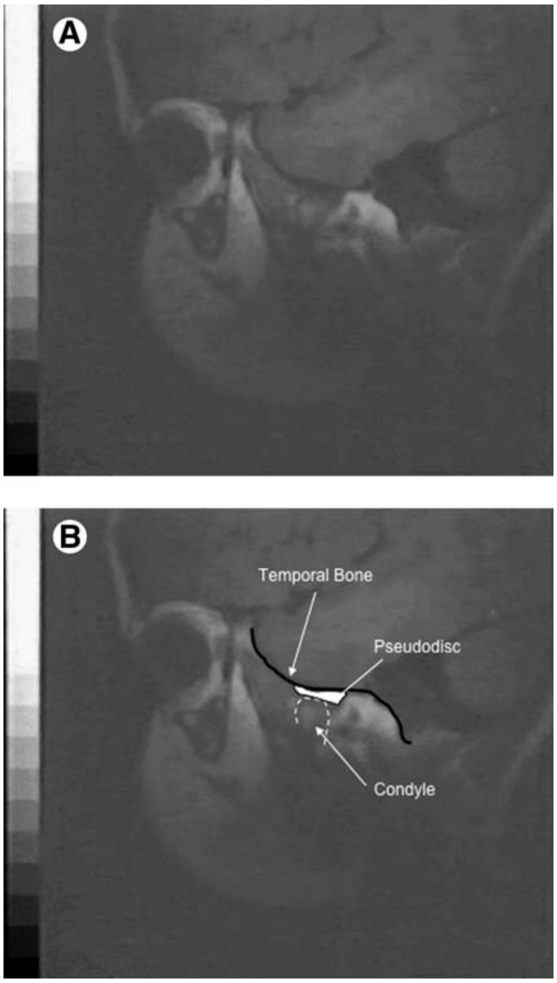

Figure 6. (A) Preoperative MRI of a patient diagnosed with CFM in a closed positioned showing the morphology of the affected TMJ. The squamous part of the temporal bone has a relatively flat surface and the TMJ does not show clear definition of the articular disk. The temporalis muscle is atrophic and the condyle hypoplastic. There are irregular anterior–posterior translatory movements only during opening and closing, with the absence of hinge axis motion during articulation. (B) Outline and labels for A.

tent with normal joint articulation (Figs 10 and 11). Preoperative cine-mode animations showed that both patients displayed a complex motion of the underdeveloped joints, with an absence of hinge axis and translation displacement movements in the opening and closing motions of articulation. The movement was an irregular anteroposterior translation in both cases when compared with the normal motion of a fully developed TMJ, which showed initial hinge motion with subsequent displacement down the articular eminence. Postoperatively, both patients showed improved condylar motion, visible in the cine-mode animations as displacement movements down the rudimentary, articular eminence.

3. Increased signal intensity of the pseudodisk was noted postoperatively in both patients. The TMJs did not show clear definition of the articular disk in the preoperative images. The relative signal density between the hypoplastic condyle and the temporal bone (articular space) was presumed to be fibrous tissue, rather than the fibrocartilaginous density of a

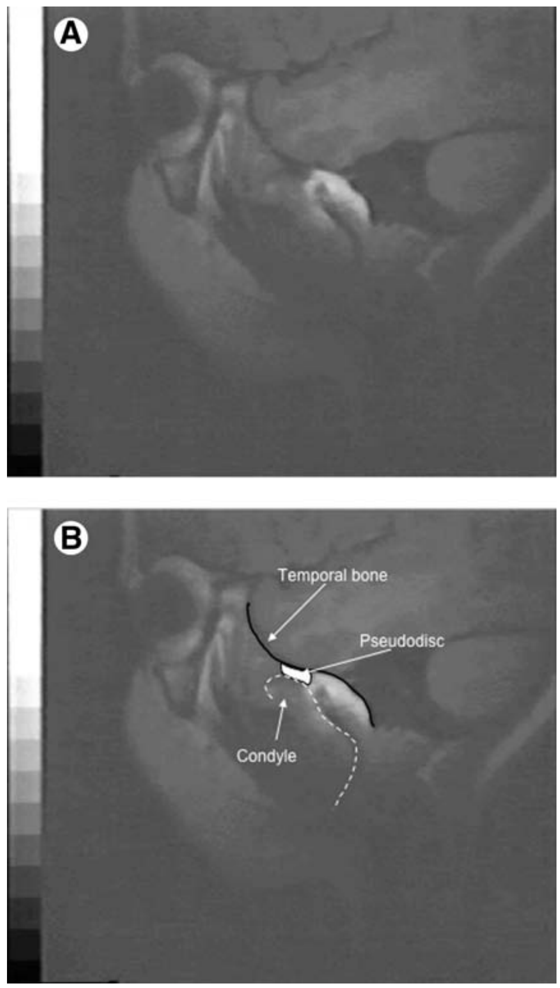

Figure 7. (A) The MRI of the same CFM patient seen in Figure 6 in an open position shows atypical displacement of the condyle. The relationship between the condyle, temporal bone and disk is abnormal. The pterygoid muscles are poorly defined. (B) Outline and labels for A.

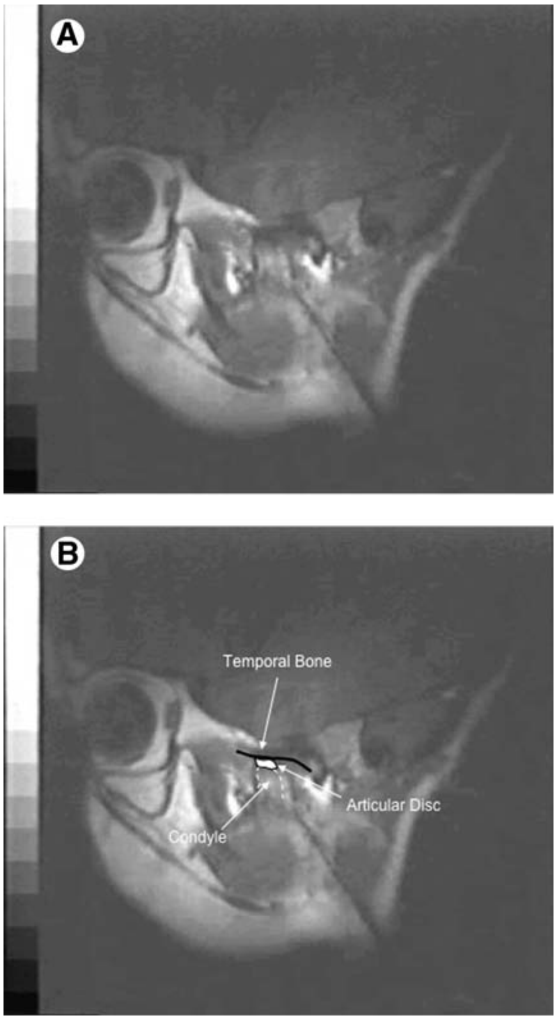

Figure 8. (A) The preoperative MRI in an open position of the TMJ of a second patient diagnosed with CFM showing the abnormal anatomy and displacement of the TMJ components. (B) Outline and labels for A.

normal disk. In the postoperative images, the affected joints showed increased signal intensity with a “pseudodisk,” presumably still consisting of fibrous tissue, but with increased intensity when compared with the preoperative images. These MRI findings are consistent with improved joint structure and function.

Discussion

Imaging of the TMJ is an important part in the evaluation of the patient with CFM before, during, and after treatment, so the distraction procedure can be rigorously tailored to match the specific anomaly, and monitored to ascertain if the expected outcome had been achieved.[13] Since its first use,[14] MRI of the TMJ has become a popular adjunct in the diagnosis of TMJ dysfunction and degenerative joint diseases.[1] The benefits of MRI are that it is a nonionizing, noninvasive technique that can produce high-resolution 3D images of the TMJ. For example, during (Herbst) orthodontic treatment in adolescents, condylar and glenoid fossa remodeling was demonstrated with MRI.[15] Thus, MRI is an excellent technique for joint visualization, and these advantages may make this method an op-

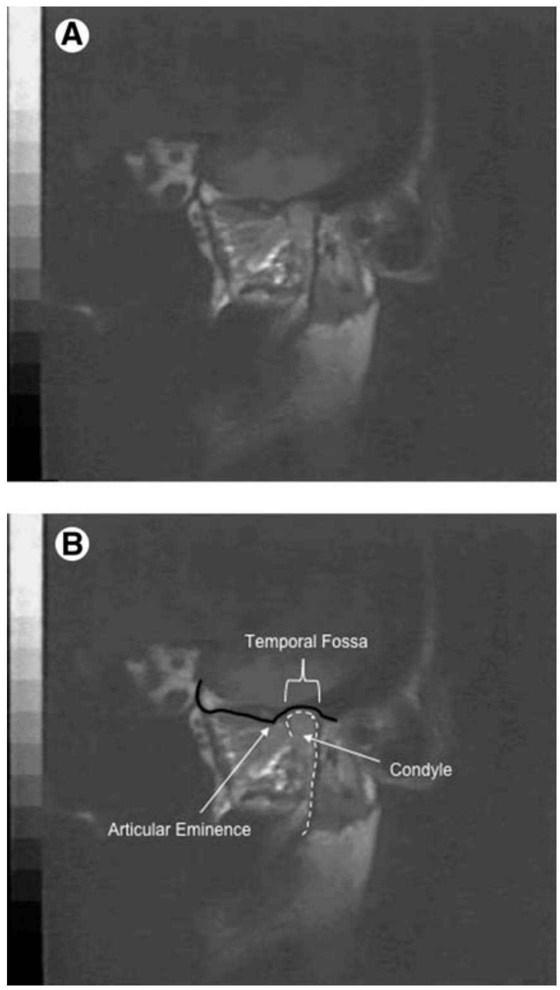

Figure 9. (A) Postoperative MRI of the TMJ in a closed position showing formation of rudimentary elements of a new TMJ. Note a more defined temporal fossa, condyle and articular eminence with an improved anatomical relationship between them. (B) Outline and labels for A.

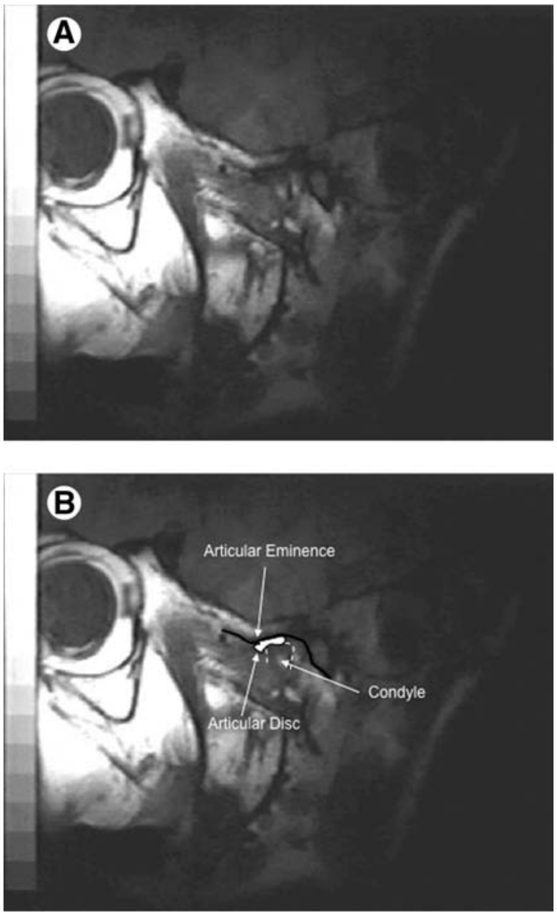

Figure 10. (A) Postoperative MRI image of the second patient showing improved anatomy and relationship of the temporal bone, articular disk and condyle, resembling a normal joint. (B) Outline and labels for A.

timal choice in the clinical evaluation of the TMJ. MRI also provides simultaneous multiplanar assessment of both skeletal and soft-tissue components of the TMJ. Indeed, functional assessment is possible using simulated computer animation (cine MRI),[16-18] and findings using MRI concur with those of arthrography 67%-95% of the time (15%-53% for false-positives).[19] Although the reliability of MRI has been quantified, its major drawback, is mostly the overdiagnosis of disk displacement. Slice thickness is another important scanning parameter. Although MRI permits slice thicknesses as fine as 0.7 mm in 3D mode, the chemical-shift artifact increases with very fine slicing when compared with the spin-echo technique. Thinner slices also increase the scan time because of signal-to-noise ratio maintenance. Nevertheless, use of standard 3-mm slicing produces diagnostic-quality images of the TMJ.[13]

The purpose of this pilot study was to use static and dynamic MRI sequences before and after treatment with mandibular distraction to investigate the response of the TMJ in patients diagnosed with unilateral CFM. In support of the functional and spatial matrix hypothe-

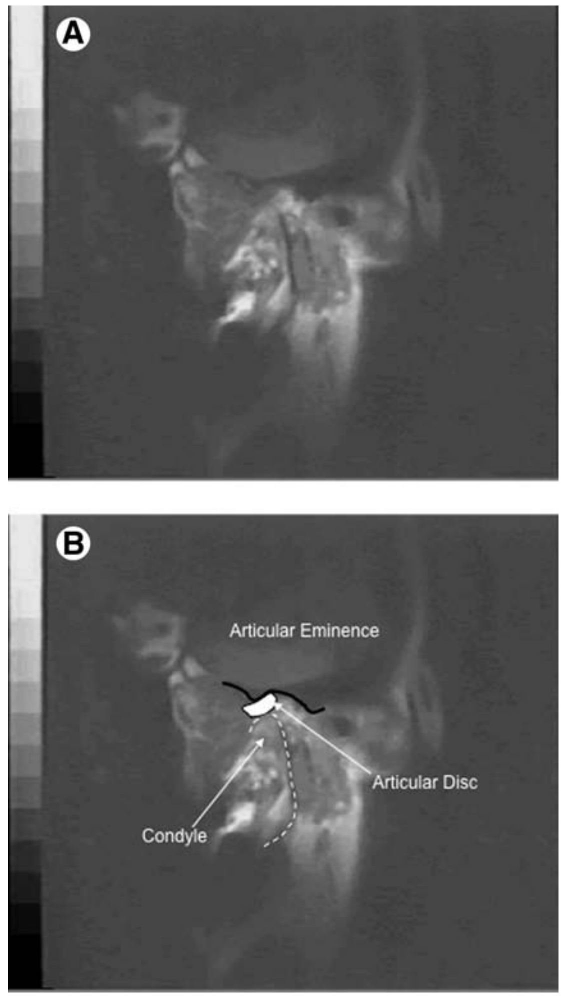

Figure 11. (A) Postdistraction MRI in an open position showing morphology of the affected TMJ. Note the change in the morphology of the squamous part of the temporal bone, which now has a putative glenoid fossa and articular eminence, consistent with the formation of the rudimentary elements of a new TMJ. There is also increased signal intensity in the region of the pseudodisk. There is improved condylar motion, with hinge movement followed by displacement down the rudimentary articular eminence consistent with a normal joint articulation. (B) Outline and labels for A.

ses,[20,21] it was suggested that a dysfunctional TMJ might adapt to normality after appropriate therapeutic alteration. Concurrently, it was suggested[22] that, in a small proportion of refractory TMJ dysfunction cases, condylotomy of the affected condylar neck is a safe and effective procedure. This concept was, presumably, based upon the findings that, in macaques, the condylar cartilage regeneration that occurred after condylectomy possessed inherent growth potential, and that the glenoid fossa was found to be capable of adaptive changes.[23] Similarly, corresponding adaptive TMJ changes in young sheep were reported,[24] which were thought to be similar to the reactions expected in young children.

As expected, the unaffected TMJs of both patients in this current preliminary study, showed normal growth and development during the 1-year period of the investigation. However, the anatomical and functional improvements noted in the affected TMJs after mandibular distraction cannot be attributed to normal growth alone; the observed changes being associated with the distraction process. Nevertheless, the cranio-mandibular articulation is considered to function as a single (bilateral) joint. Thus, it is possible that changes in the joint associated with the distraction procedure could also affect the nondistracted side. In this present study, however, unilateral mandibular distraction showed no deleterious effects on the nondistracted TMJ morphology or function before or after the distraction procedure, using MRI images and cinemode animations. These findings support the notion that unilateral distraction of the mandible has no major effect on the function or morphology of the contra-lateral TMJ, presumably because of adaptive responses during the correction of the dysfunctional (TMJ) matrix.

In contrast, significant improvements in the TMJ on the distracted side were observed in both cases. The postoperative improvements in TMJ morphology consisted of formation of rudimentary elements of a new TMJ on the affected side (Figs 1B, 2B, 9, and 10). Specifically, functional displacements of the new condyle-disk complex were consistent with normal joint anatomy, indicative of the formation of a rudimentary glenoid fossa and articular eminence with quasi-normal articulation (Figs 11 and 10). Indeed, masticatory muscle definition was improved postoperatively and increased radiodensity of the pseudodisk after mandibular distraction was demonstrable. These findings are consistent with the correction of the dysfunctional matrix, and with observations made in animal studies, which document the biological, adaptive capability of the TMJ complex.[24]

On the contrary, it is thought that in traditional orthognathic surgery, skeletal elements predominantly change with little, if any, amelioration of the surrounding, dynamic soft tissues. Therefore, the hypoplastic mandible undergoes size-, shape- and positional change[9–11] but corresponding soft-tissue change may not be adequately corrected,[25] so that relapse is perhaps more likely. By contrast, it appears that the gradual application of strain used during distraction osteogenesis provides a mechanism that permits correlated soft-tissue and skeletal size-, shape- and positional change, which may mimic a "growth spurt." For example, during puberty growth of long bones occurs first, followed by the neuromuscular response. Presumably, distraction osteogenesis activates growth genes because of physical stretch and tension within the regenerate, and deposition of osteoid within the callus, which then mineralizes. Thus, downstream growth factors are biosynthesized and secreted, which provide the developmental mechanisms for a multitissue response that includes signal transduction, mesenchymal proliferation and extracellular matrix deposition, leading to bone formation,[4] as well as muscle hypertrophy and epithelial hyperplasia. A recent study documented significant increase in the volume of the medial pterygoid muscle following mandibular distraction.[26] Therefore, it is possible that this integrated tissue response may be able to provide increased stability (decreased relapse). Thus, distraction histogenesis attains physiological equilibrium for a complex adaptive (TMJ) system.

In summary, MRI proved to be a useful, noninvasive means of assessing the pre- and postoperative morphology and function of the TMJ, during and after mandibular distraction. The results of the present study demonstrate that the form and function of the TMJ complex can be improved significantly by mandibular distraction in young patients, findings not previously demonstrated after other types of mandibular surgical augmentation techniques. Thus, MRI of the TMJ should be considered in the protocols of patients undergoing mandibular distraction, even though the craniofacial distraction phenomenon remains incompletely understood at this time. The author's current studies are now investigating longer-term postdistraction responses in a larger sample of older children.

Acknowledgments

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