The Pediatrician’s Role in Caring for Patients With Congenital Microtia and Atresia

Burt Brent, MD

As a visible defect, congenital microtia affects the entire family of origin. The patient often receives cruel ridicule by peers, and the parents frequently present with anxiety and feelings of guilt. Experience has shown that such a family presents many questions to the pediatrician: What did I do wrong to make this occur? Can my infant hear? Will my child be able to learn to speak? Will my future children have the same deformity? What can be done about the disfigurement of a missing auricle or a gnarled ear remnant, that marks my child as abnormal and invites teasing and embarrassing questions by outsiders? Being the first physician to deal with such a patient and his or her family, the informed pediatrician can provide a treatment plan that will dispel misconceptions, greatly alleviate anxieties, and wisely council the family.

CASE MATERIAL

The author’s purpose is to provide the pediatrician with practical information that has accumulated from 25 years of practice dedicated to the treatment and management of 1,000 patients with microtia. Of these, 58.2% had microtia on the right side, 32.4% had microtia on the left side, and 9.4% had bilateral involvement. Discrepancy in gender distribution was also seen: 63.1% of the patients were male and 36.9% of the patients were female (Table 1).

EDUCATIONAL OBJECTIVES

1. Develop a treatment plan for congenital microtia and atresia to help dispel misconceptions and alleviate anxieties.
2. Review the etiology of congenital microtia and atresia as well as associated anomalies.
3. Discuss hearing loss in children with congenital microtia, atresia, or both.
4. Review the current surgical repair for congenital microtia and atresia.

ETIOLOGY

Microtia occurs approximately once in every 5,000 births. The ultimate cause is largely undefined. However, McKenzie and Craig\(^1\) and Poswillo\(^2\) theorize that the deformity results from in utero tissue ischemia caused by either an obliterated stapedial artery or a hemorrhage into the local tissues. This speaks in favor of the deformity’s arising from a mishap during fetal development rather than from a hereditary source. In support of this, the author has treated 12 patients with microtia who have an affirmed, identical twin with normal ears. Only one set of identical twin patients have had concordance for outer ear deformities. Of interest is that each of these twins with microtia also had signs and symptoms of pyloric stenosis within 2 days of...
each other and were operated on at 6 weeks of age. However, there is some small familial tendency for this deformity, as described below.

Although one can occasionally find a candidate for a causative event during early development, an analysis of medical histories for the author's 1,000 patients revealed no consistent, unusual contributing factors. It is important to assure the mother that these are random, sporadic occurrences and that her child's deformity was not caused by anything that she did before or during the pregnancy. Reassurance by the pediatrician can do much to ease parents' feelings of guilt.

However, several events during pregnancy have been associated with ear deformities. The occurrence of deafness resulting from rubella during the first trimester of pregnancy is well known, but only one of the mothers in the series of 1,000 patients actually had documented infection by this virus; several others stated they "had been exposed to measles." Also, certain drugs may cause microtia. In this series, three cases of microtia followed the mother's ingestion of the tranquilizer thalidomide during pregnancy. Isotretinoin has also been cited as causing ear deformities when ingested during the first trimester. In the author's series, several mothers had been unaware of this problem and had used isotretinoin to control acne. Other medications that reportedly cause microtia are clomiphene citrate and retinoic acid.

Although inheritance in microtia is felt to be somewhat uncommon, investigation of the author's series of 1,000 patients reveals that recurring major auricular deformities exist within the immediate family in 4.9% of the cases. When the distant relatives are included, this number rises to 10.5%. If a couple has two children with microtia, the risk of recurrence in future offspring is thought to be as high as 15%. Inheritance of this condition is generally felt to be multifactorial, but there have been rare reports of families manifesting autosomal dominant inheritance.

**THE HEARING PROBLEM**

Initially, parents of an infant with microtia are usually most concerned with the hearing problem. They may think the child is completely deaf on the affected side or that hearing can be restored by merely opening a hole in the skin. As these are misconceptions, the pediatrician can do much to alleviate the parents' anxieties by explaining the fundamentals of ear embryology.

Because the human ear's receptive (inner) portion is derived from embryologic tissue that is different from that of the conductive (external and middle) portion, the inner ear is rarely involved in microtia, so these patients have at least some hearing in the affected ear. Their problem is sound conduction, which is blocked by the malformed middle and external ear complex. Typically, these patients have a hearing threshold of 40 to 60 dB on the affected side. By comparison, normal function allows us to hear sounds between 0 and 20 dB.

The middle and the external ear develop from a common block of tissue, chiefly from the first (mandibular) and second (hyoid) branchial arches. The auricle itself is formed from six "hillocks" of tissue that lie along these arches and can first be seen in the 5-week embryo.

On the other hand, the inner ear first appears at 3 weeks and is derived from tissues of distinctly separate origin. Perhaps this explains why it is usually spared in the developmental mishap that almost invariably involves the middle as well as the outer ear of patients who have microtia. Refinements in radiographic technique (polytomography and computerized tomography scans) have only occasionally demonstrated significant underdevelopment of the inner ear.
Inner ear abnormalities are found in approximately 10% of microtia and atresia cases, but the abnormalities are usually slight (e.g., a dilatation of the lateral semicircular canal). Interestingly, in evaluating approximately 1,500 cases of microtia during a 25-year period, the author has seen only 3 patients who were totally deaf. Remarkably, these were patients with unilateral pinna deformities who had no family history of hearing problems or microtia. Incidentally, one need not wait until a child is old enough to respond to conventional behavioral hearing tests. An infant’s hearing may be objectively assessed shortly after birth by an auditory brainstem response test to determine whether the cochlea reacts normally to sound stimulation and whether the middle ear is normal. This is worthwhile, even in the unilateral cases, because even if the opposite pinna appears relatively normal, its underlying middle ear may be malformed.

Fortunately, 90% of patients who have microtia have one normal ear and are “born adjusted” to the monaural condition. They do well in life even without correction for hearing. The main problems of a patient with microtia arise with sound localization and in noisy environments. This can cause difficulty in school, where following verbal instructions and paying attention are necessary. The teacher’s knowledge of the problem and cooperation in placing the child in a strategic position in the classroom is helpful. Some patients who have microtia may require speech therapy. Also, it is important to protect the normal ear from unusually harmful noise.

Because of their normal inner ears, patients with bilateral microtia usually have serviceable hearing when using bone-conductive hearing aids to overcome the transmission block. If they are referred to an audiologist so that aids can be used as soon as possible, these patients usually develop normal speech. There is no point in waiting for several months. Hearing aids should be applied within weeks of birth. Because these bone-conductive aids are cumbersome and label the child as “different” from his or her peers, it is optimal to surgically correct the hearing deficit to eliminate the aids. However, correcting this conductive problem surgically is difficult because the middle ear beneath the closed skin is not normal.

**OPERATIVE PROCEDURE FOR THE MIDDLE EAR**

Exploration involves cautiously avoiding the facial nerve while drilling a canal through solid bone. One must usually create the tympanum with tissue grafts; the distorted or fused ossicles may be irreparable. Skin graft “take” on the drilled bony canal can be problematic and can result in chronic drainage or mental stenosis. However, in the hands of a competent otologist with a large volume of experience, restoring middle ear function surgically can be rewarding for the patient with microtia—even if only one ear is involved.

Nevertheless, many surgeons presently feel that potential gains from middle ear surgery in microtia are outweighed by potential risks and complications and that this surgery should be reserved for bilateral cases. It is the author’s conviction that, if the otologic surgeon is not comfortable with unilateral cases, he or she certainly should not be operating on bilateral cases either.

As with surgical construction of the outer ear (or any operative procedure for that matter), the outcome of surgery strongly depends on the experience of the surgeon. The otologic surgeon must not only have the expertise and the experience to perform this delicate and sophisticated surgery, but must also be well versed in evaluating the patient’s middle ear development to determine whether the patient is an ideal candidate for surgery. Jahrsdoerfer et al. use a 10-point scoring system in evaluating the computed tomography scan of the middle ear of each patient with microtia. During this evaluation, they look for the presence of a stapes and a patent oval window along with other favorable signs. They will only operate on unilateral cases if the patient scores 6 or higher, but will sometimes operate on a patient with bilateral microtia who has a lesser score. Using these criteria, Jahrsdoerfer et al. discovered that only 50% of patients are suitable candidates for surgery. They found that this grading system not only helps to predict the degree of success one may expect in an individual patient (e.g., a score of 8 translates into an 80% chance of restoring hearing to normal or near-normal levels), but also eliminates impossible surgical candidates. A repair is considered successful if one is able to close the functional dif-
ference between the repaired and the normal ear to within 15 to 25 dB. Thirty decibels is considered borderline improvement and 35 dB is a failure.

Because nearly 15% of all ears with congenital atresia that De la Cruz et al.\textsuperscript{17} repair have cholesteatoma formation, they feel that all patients with microtia–atresia should have a computerized tomography scan at age 6, even if auditory-enhancing surgery is not desired. To aid in evaluation, they recommend coronal views as well as the usual axial orientation of these scans. De la Cruz et al. also point out that because patients with atresia have eustachian tubes like everyone else, they can develop otitis media, even though they do not have an external canal. Although it is impossible to confirm the diagnosis by otoscopic examination in these cases, if the pediatrician suspects otitis media in the deformed ear, it is prudent to prescribe antibiotics.

One also has to be on the alert for cholesteatoma formation in the child with a normal pinna who has a stenotic canal. Cole and Jahnson have pointed out that a stenosis of 3 to 4 mm can be clinically monitored. However, if the stenosis is 2 mm or smaller, a computerized tomography scan should be done by age 5 and the stenosis should be surgically alleviated with a canaloplasty.\textsuperscript{18}

Any surgical repair of the ear always begins with the reconstructive plastic surgeon. Construction of the auricle must precede the middle ear surgery because once an attempt is made to “open the ear,” the virgin skin is scarred, thus compromising the opportunity for a good external ear construction. When middle ear surgery is contemplated, an accomplished reconstructive plastic surgeon and otologist should plan the procedures as a team. A successful aesthetic and functional outcome requires a carefully integrated approach by two experienced surgeons. In addition, implantable acoustic devices are on the horizon. In the future, these may offer an optimal alternative to middle ear surgery for some.

**ASSOCIATED DEFORMITIES**

Because the auricle develops from tissues of the branchial arches, it is not surprising that a significant percentage of patients who have microtia exhibit deficiencies of facial components whose origins also lie in these embryologic building blocks. When appearing as a flattened side of the face, this condition is known as hemifacial microsomia and embodies an underdevelopment of both the bony jaws and the overlying soft tissues. This obvious facial asymmetry occurred to some degree in approximately 35% of the 1,000 patients in the author’s series (Table 2) and is usually addressed after the external ear is repaired.\textsuperscript{19} From the author’s long-term observations of patients, this asymmetry does not seem to worsen as the child grows. Instead, the asymmetry grows in proportion with existing facial features. This has been borne out by a recent growth analysis investigation.\textsuperscript{20}

Patients with microtia who have underdeveloped jaws should have bony repairs, both to improve facial contours and to correct dental occlusion. These repairs are done after the auricular construction and are accomplished by osteotomies and bone grafting to elongate the jaw, to correct occlusal tilts, and to add bulk.\textsuperscript{21} In the past few years, new methods have emerged to elongate the mandible by controlled serial distraction of surgically divided bony segments with a turn-screw pin device.\textsuperscript{22}\textsuperscript{23} This procedure can advantageously (1) begin at an earlier age, and (2) simultaneously stretch the overlying soft tissues. However, it necessitates the patient’s wearing a cumbersome device for 3 months and may leave significant facial scars. Hopefully, these problems will be resolved as this new method evolves.

**TABLE 2**

<table>
<thead>
<tr>
<th>Microtia: Associated Deformities (Author’s Series of 1,000 Patients)</th>
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<tr>
<td>Branchial arch deformities</td>
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<tr>
<td>Obvious bony and soft tissue deficit—36.5%</td>
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<tr>
<td>Family perceives it as “significant”—49.4%</td>
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<tr>
<td>Overt facial nerve weakness—15.2%</td>
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<tr>
<td>Of these, more than one branch involved—42.6%</td>
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<tr>
<td>Macrostomia—2.5%</td>
</tr>
<tr>
<td>Cleft lip, cleft palate, or both—4.3%</td>
</tr>
<tr>
<td>Urogenital defects—4.0%</td>
</tr>
<tr>
<td>Cardiovascular malformations—2.5%</td>
</tr>
<tr>
<td>Miscellaneous deformities—1.7%</td>
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</table>
The literature cites increased urogenital tract abnormalities in the presence of auricular deformities, particularly when the patient is afflicted with other manifestations of facial underdevelopment. Several patients in the author's series have hypospadias or vaginal agenesis; 4% have proven abnormalities of their collection systems, which include horseshoe kidney, ureteral duplication, unilateral renal agenesis, and pelvic kidney. A routine screening of the urine may detect silent hematuria or proteinuria, but likely will show nothing. Recurring urinary tract infections in patients with microtia prompt renal function studies, and one should start with a renal ultrasound before considering a more invasive technique to detect these deformities. Because of the increased incidence of urogenital abnormalities in patients with microtia, one might consider screening all of them with ultrasound.

In this series of 1,000 patients with microtia, cleft lip, cleft palate, or both occurred in 4.3%, whereas cardiovascular malformations existed in 2.5%. The latter malformations include atrial and ventricular septal defects, dextrocardia, transposition of the great vessels, three-chambered heart, and patent ductus. If any signs or symptoms of cardiopathy are noted in patients who have microtia, one must obtain an echocardiogram and consult with a cardiologist.

Cervical vertebral anomalies are more common in patients with microtia if other defects exist, such as cardiac or renal disorders or cleft lip and cleft palate. Because associated neurologic symptoms are rare, the frequency of these vertebral abnormalities is probably underestimated in patients who have microtia. Goldenhar's syndrome (oculoauriculovertebral dysplasia) is a condition with extreme variability of phenotypic expression, in which the patient with microtia has an ocular dermoid and usually has cervical vertebral abnormalities. When one notes an ocular dermoid or detects limited range of neck motion in any patient who has microtia, the renal function and the cervical spine should be investigated as well. These particular vertebral abnormalities not only pose risky problems for anesthetic positioning during intubation, but, if undetected, also will occasionally cause irreversible neurologic compromise from chronic brainstem compression.

Although a variety of cervical vertebral deformities are seen in patients with microtia, the children chiefly at risk for neurologic compromise are those with occipitalization of the atlas, particularly when there is posterior inclination of the odontoid process. Cephalometric radiograms will screen for this risk, and it is recommended that these patients undergo periodic neurologic examination followed by a computerized tomography scan or magnetic resonance imaging at the earliest suspicion of neurologic disturbance. During any kind of surgery, it is wise to bring this condition to the anesthesiologist's attention so that unusual neck manipulation is avoided during induction and tracheal intubation. The patient's head should not be forcibly turned, but, instead, the head, neck, and body should be "log-rolled" as a unit.

**PSYCHOLOGY AND FUNCTION OF THE EXTERNAL EAR**

Once family members have been educated and reassured somewhat about the child’s hearing, they will turn their attention to the obvious aesthetic deformity. This will then consistently rekindle their anxiety and guilt. Naturally, the family wants this repaired as soon as possible. However, it is not technically feasible before the child reaches school age. Meanwhile, the family continuously seeks guidance and support from the pediatrician. Understanding the psychological impact of having any congenitally deformed facial part, especially a deformed ear, and the possibility of its surgical correction provides the pediatrician with the necessary information to counsel and direct the family.

At approximately age 3, the child will discover that he or she is "different" from other children. Classically, the parents find the child in front of the mirror comparing the normal ear with the deformed remnant. The child begins to refer to the ear with microtia as the "little ear" or "closed ear." The parents are advised to be honest with their child, to explain that he or she was born with a "different" ear or ears and that before school begins (age 6) the ear or ears can be repaired and made to look more normal. Optimally, the parents should be counseled to treat the child as normal, without making a fuss about the deformity. The only children who seem
disturbed by microtia before the age of 6 or 7 are those whose parents have projected their anxieties onto them, thus making them feel "flawed."

The first big psychological trauma occurs in approximately the first grade in school, when children begin to socialize with a large group of peers for the first time. At this time, one’s self-awareness begins to develop through referencing and comparing body image. Also at this time, teasing begins and the patient with microtia learns that he or she is "different" (Table 3).

The second trauma takes place during adolescence, when the developmental venue of "fitting in" to a social group begins. With "fitting in," how one looks becomes important. Because they are different, teenagers with microtia are self-conscious and become particularly motivated to have the ear deformity repaired. However, they are also critical and may have unrealistic expectations of what can be produced through surgery.

In addition to holding up our eyeglasses and funneling sounds toward our eardrums to improve hearing, external ears make us look more symmetrical and create a self-image as a whole person. This, then, is the driving reason for surgical construction of the outer ear. It is a psychoemotional, aesthetic endeavor to restore self-esteem through restoring a symmetrical, normal self-image. Far from being "cosmetic surgery," repairing a congenital deformity permits a person to have a normal self-image and a normal life and to be a normal, productive member of society. If the ear goes uncorrected or a poor result is achieved by an inexperienced surgeon, the patient tends to feel flawed, "less than" others, and tends to be plagued by low self-esteem that may last a lifetime.

Because of these implications, it is important for the pediatrician to direct the family to an experienced surgeon, even if distant travel or a battle with the insurance company is necessary to obtain subspecialized expertise.

CONSTRUCTION OF THE EXTERNAL EAR

Total construction of the auricle is one of the greatest challenges that confronts the reconstructive plastic surgeon. Composed of a delicately convoluted cartilage frame covered by a fine skin envelope, the ear is a difficult structure to draw or sculpt, let alone surgically reproduce. The foundation of the surgical repair consists of creating and placing an ear framework underneath the periauricular skin and then refining the auricle with several soft tissue procedures. Generally, artificial, alloplastic substances are not well tolerated as frameworks, may cause skin ulceration and infections, and are frequently extruded and rejected. Homograft cartilage (maternal, paternal, or "banked") resorbs, distorts, or disappears within 18 months. For all of these reasons, the preferred material for auricular construction is autogenous rib cartilage. Although its use necessitates a sizeable operation and subspecialized expertise, unlike a reconstruction using artificial alloplastic materials, a successful construction with autogenous tissue grows, heals, and is far less susceptible to trauma. This helps reduce patient concern about injury during normal activities.

The age at which an auricular construction should begin is governed by psychological and physical considerations. Because the concept of body image usually begins forming at approximately 4 or 5 years of age, it would be ideal to begin construction before the child enters school and is psychologically traumatized by cruel peer ridicule. However, surgery should be postponed until age 6, when rib growth provides substantial cartilage to permit a quality framework fabrication.

In the author's experience with more than 1,000 patients with microtia between the ages of 1 month and 62 years, patients and their families consistently state that their psychological disturbances rarely begin before age 7 and usually become overt from ages 7 to 10. Thus, it is prefer-
able to delay the initial cartilage graft until the child is 6 years old when (1) there is usually sufficient rib cartilage for the repair, (2) the child is aware of the problem and usually wants it fixed, and (3) the child can comprehend and cooperate during the surgery. At age 6, the normal ear has grown to within 6 or 7 mm of its full vertical height. This permits one to construct an ear that will have reasonably constant symmetry with the opposite normal ear. In the author’s series, most of the surgically constructed ears have grown symmetrically with the opposite, normal ear. Some have slightly overgrown the normal ear. None have shrunk, softened, or lost detail. Therefore, one should try to match the opposite, normal ear during the preoperative planning session. Certainly there is no reason to construct the ear larger, as some investigators have thought in the past. If the patient is small for his or her age, if the opposite normal ear is large, or both, it is prudent to postpone the surgery.35

In conclusion, it is best to initiate the repair before the child has been psychologically traumatized, but the surgeon must not be pressured to begin until rib growth provides substantial cartilage for framework fabrication.

CORRELATING AURICULAR REPAIR WITH THE CORRECTION OF OTHER FACIAL DEFORMITIES

When repairing the ear with microtia, one needs to correlate timing with any other facial surgery that may be necessary (eg, surgical correction of bony deficiency, soft tissue deficiency, or both). In the author’s experience, the patients and their families have been more concerned about the ear defect initially, so the auricular surgery is usually under way before other corrections begin. By carefully planning the auricular location with reference to the opposite normal side, one should be able to meet this psychological urgency without compromising the other facial repairs. It has been argued that ear placement would be easier if the facial symmetry was corrected first.36 But the author finds this unnecessary when appropriate topographic facial landmarks are used. If mandibular repairs, soft tissue repairs, or both are begun before the ear repair, every effort must be made to preserve the virgin auricular site and to keep it scar free. If the bony work is done first, it is imperative that scars be peripheral to the proposed auricular site and that they not lie precariously over the future region of the ear.

As stated before, the auricular repair must likewise be done before a canal is created during an otologic procedure. Unscarred skin and the surgeon’s ability to sculpt a realistic ear framework from rib cartilage are imperative for a successful outcome37 (Figs. 1 and 2).

SUMMARY OF LONG-TERM RESULTS OF REPAIR OF MICROTIA

To evaluate the benefits of auricular repair, a questionnaire survey was sent to the 1,000 patients who had microtia surgically repaired. Although many of the author’s patients are scattered throughout the world, a 50.8% response rate was obtained. This volume (508 respondents) provided data to present information regarding
the benefits of auricular construction. Follow-up ranged from 1 to 18 years, with an average of 7.7 years.

**Durability of the Constructed Auricle**

The respondents indicated no instances of softening or shrinkage of the cartilaginous frameworks due to resorption or trauma. Despite more than 70 reports of major trauma to surgically constructed ears, all healed without incidence. These traumatic episodes included severe blows from soccer kicks, baseballs, and hockey pucks; abrasive injuries from football tackles and wrestling matches; insect bites and bee stings; and even a human bite and a dog bite.38

**Psychological and Emotional Benefit**

The data showed that the impact of microtic deformity on the patient and the family sequentially becomes greater as the child enters school, approaches adolescence, and reaches adulthood (Table 3). Before 10 years of age, 64.8% of families and patients rated the deformity’s impact as “moderate” to “severe,” whereas this jumped to 77.6% in patients aged 10 to 14 years and 86.7% in patients older than 15 years. Respondent families of children 6 to 10 years old who rated the deformity’s impact as only “mild” (35.2%) also often stated that the full impact had not yet been realized and that they sought surgery “not to solve a current problem but to prevent a future, anticipated crisis” as their child grew older.

In families where the deformity’s impact was rated “moderate” to “severe,” the psychological and emotional relief attained after surgical repair was also rated high (Table 4). The respondents indicated that relief was attained in 100% of patients in all age groups when the deformity’s impact was rated “severe,” and the emotional improvement was graded “significant” in 86.6% of these families and “somewhat” in the remaining 13.4%. When the impact of microtia was rated
### TABLE 4

**Emotional Relief by Repair of Microtia**

**(Author’s Series of 1,000 Patients)**

<table>
<thead>
<tr>
<th>Age (yr)</th>
<th>Severe</th>
<th>Moderate</th>
<th>Mild</th>
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<tbody>
<tr>
<td></td>
<td>Significant Relief</td>
<td>Some Relief</td>
<td>Unchanged</td>
</tr>
<tr>
<td>5–10</td>
<td>91.8%</td>
<td>8.2%</td>
<td>—</td>
</tr>
<tr>
<td>11–14</td>
<td>75.0%</td>
<td>25.0%</td>
<td>—</td>
</tr>
<tr>
<td>15–20</td>
<td>66.7%</td>
<td>33.3%</td>
<td>—</td>
</tr>
<tr>
<td>21–62</td>
<td>87.5%</td>
<td>12.5%</td>
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</tr>
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</table>

as being only “moderate,” 78.4% of families still reported the emotional relief as being “significant,” except in ages 15 to 20 years where 76.9% deemed the procedure “somewhat” beneficial. Perhaps these data reflect the critical nature of adolescents, or suggest that these patients had become resolved to their deformity or psychologically “numbed” to peer criticism.

Data describing the emotional benefits of the surgical repair were further reflected in the patients’ activities. In the youngest age group, operated on before 10 years of age, 42.1% of parents found their children to be “more outgoing.” The 10- to 14-year age range was the most impressive of all, in that 62.5% of the respondents indicated that they were more willing to socialize and to participate in sports after their surgical repair and 20% had improved their performance in school. In ages 15 to 20 years, 37.5% of the respondents indicated that they were more outgoing, 26.1% that they were more social and sports oriented, and 20% that they had improved scholastically. After age 20, 64% of the respondents ranked themselves as more outgoing, and 50% felt that they were “more social.”

### REFERENCES

22. McCarthy JG. The role of distraction osteogenesis in the reconstruction of the mandible in unilateral craniofacial