Temporomandibular Joint Ankylosis Caused by Chondroid Hyperplasia From the Callus of Condylar Neck Fracture

*Soung Min Kim, DDS, MSD, PhD,**
Jung Min Park, DDS, MSD,† Ji Hyuck Kim, DDS, MSD, PhD,†
Kwang Jun Kwon, MD,‡ Young Wook Park, DDS, MSD, PhD,†
Jong Ho Lee, DDS, MSD, PhD, Sang Shin Lee, DDS, MSD,§*
Suk Keun Lee, MSD, PhD§

Abstract: A patient who complained of difficulty in opening his mouth after condylar neck fracture 1 year ago presented typical features of temporomandibular joint ankylosis in clinical and radiologic examinations. To demonstrate a possible pathogenesis of temporomandibular joint ankylosis after condylar neck fracture, the fractured condylar portion removed was examined by histologic and immunohistochemical stainings. Interpositional gap arthroplasty was performed by removing the inferomesially displaced fractured condyle, and reconstruction with subcutaneous dermis to the previous vertical height was performed immediately. The fractured condylar portion was almost intact with slight erosion of the condylar cartilage. In the hematoxylin and eosin and Masson trichrome stainings, an extensive chondroid hyperplasia with abundant hyaline cartilage was shown in the removed condylar portion. There were also hyperplastic features of the synovial membrane, which were abnormally distributed throughout the chondroid tissues. In the immunohistochemical stainings of proliferating cell nuclear antigen (PCNA) and bone morphogenetic protein (BMP)-2 and BMP-4, the chondroid tissues were conspicuously hyperplastic and strongly positive for BMP-4 but sparse for BMP-2. From these results, we think that the hyperplastic chondroid tissue was derived from the callus of the primary fractured site of the condylar neck and propose that the chondroid tissue could proliferate continuously because of synovial tissue support from around the temporomandibular joint, resulting in temporomandibular joint ankylosis. This pathogenesis is quite different from those of other diaphyseal fracture of long bones.

Key Words: Chondroid hyperplasia, interpositional gap arthroplasty, immunohistochemical staining, temporomandibular joint (TMJ) ankylosis

From the *Department of Oral and Maxillofacial Surgery, College of Dentistry, Seoul National University, Seoul; and †Departments of Oral and Maxillofacial Surgery, ‡Anesthesiology, and §Oral Pathology, College of Dentistry, Kangnung National University, Gangneung, South Korea.
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Address correspondence and reprint requests to Suk Keun Lee, MSD, PhD, Department of Oral Pathology, College of Dentistry, Kangnung National University, 123 Chibyun-dong, Gangneung, 210-702, South Korea; E-mail: sklee@kangnung.ac.kr
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The temporomandibular joint (TMJ) is a joint that allows mastication and speech. It has a synovial cavity that is formed between the mandibular condyle below and the articular fossa of the temporal bone above.[1,2] The joint is liable to suffer from a number of diseases of the mandible, some of which predispose it to TMJ ankylosis. Ankylosis is defined as a loss of joint movement resulting from the fusion of bones within the joint or the calcification of the ligaments around it.[3–5] Temporomandibular joint ankylosis could be classified according to the type of tissue involved, bony, fibrous, or fibro-osseous, and according to the location, intracapsular or extracapsular.[6,7] Trauma and infection are the main causes of ankylosis.[6] If trauma is the cause of ankylosis, it is hypothesized that the extravasation of blood into the joint, along with the disruption of fibrocartilage integrity, permits the ingrowth of fibrous connective tissue into the joint, which subsequently results in ossification, leading to the fusion of temporomandibular condyle to the articular surface of the temporal bone.[6] However, there were few reports of TMJ ankylosis explaining whether the origin of the calcifying tissue is from the condylar cartilage or from the callus of the fractured site. The aim of this report is to demonstrate a possible pathogenesis of TMJ ankylosis in a patient with a fractured condylar neck.

CLINICAL REPORT

A 38-year-old man who was referred to the Department of Oral and Maxillofacial Surgery at Kangnung National University Dental Hospital complained of difficulty opening his mouth 1 year after a condylar neck fracture. He had typical features of TMJ ankylosis on clinical and radiologic examinations. He had no previous dental history relevant to the symptom but had had a traumatic injury to the head. Computed tomographic scan showed a malpositioned fractured fragment of medial half condyle and broad radiopacity around TMJ demonstrating a possibility of calcifying ankylosis (Fig. 1).

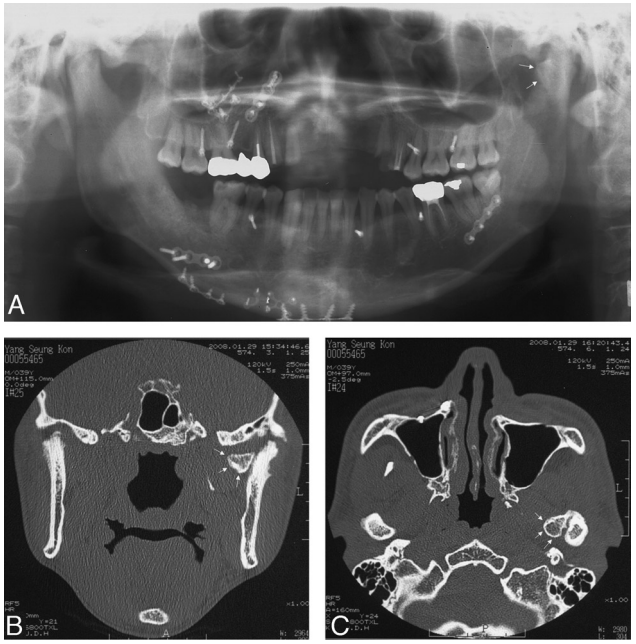

FIGURE 1. Preoperative radiographic views showing a displacement of a fractured condylar head (arrows): panoramic view (A), sagittal view of computed tomographic (CT) scan, (B) coronal view of CT scan (C).

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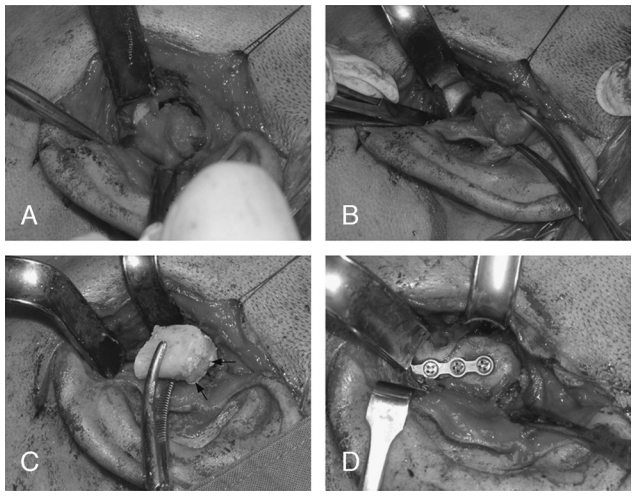

FIGURE 2. Intraoperative photographs. A, Intentional cutting of the condylar portion. B, Removal of fractured condylar head with an almost intact articular surface plus an erosion (arrows). C, Repositioning of intentional cutting fragment. D, The repositioned condyle was fixated by 4-hole titanium plate and screws (Osteomed Co, Addison, TX).

According to the classification of TMJ ankylosis proposed by Kazanjian in 1938, this patient is considered as having true ankylosis, which was defined as any condition that produced fibrous or bony adhesions between the articulated surfaces of the TMJ.[8,9]

Interpositional gap arthroplasty by removing the inferomesially displaced portion of the fractured condylar and reconstruction were performed to the previous vertical height with subcutaneous dermis (Fig. 2).[10] The fractured condylar portion was almost intact with a slightly eroded surface on the condylar cartilage.

DISCUSSION

Temporomandibular joint ankylosis after traumatic fracture of the condylar head is not a frequent complication, if the patient receives the appropriate surgical treatment in time. However, to this date, it has not been clearly explained why the traumatic condyle neck fracture so frequently results in TMJ ankylosis both in young and old patients. The present case showed typical TMJ ankylosis due to an old condyle neck fracture, which was not treated properly for 1 year. Every clinical and radiologic finding was identical to the TMJ ankylosis after the traumatic condyle neck fracture, as reported in the literature.[6,11–13] In particular, we found an extensive proliferation of chondroid tissue admixed with collagenous fibrous tissue in the pseudo-bony lesion of the TMJ ankylosis (Fig. 3).

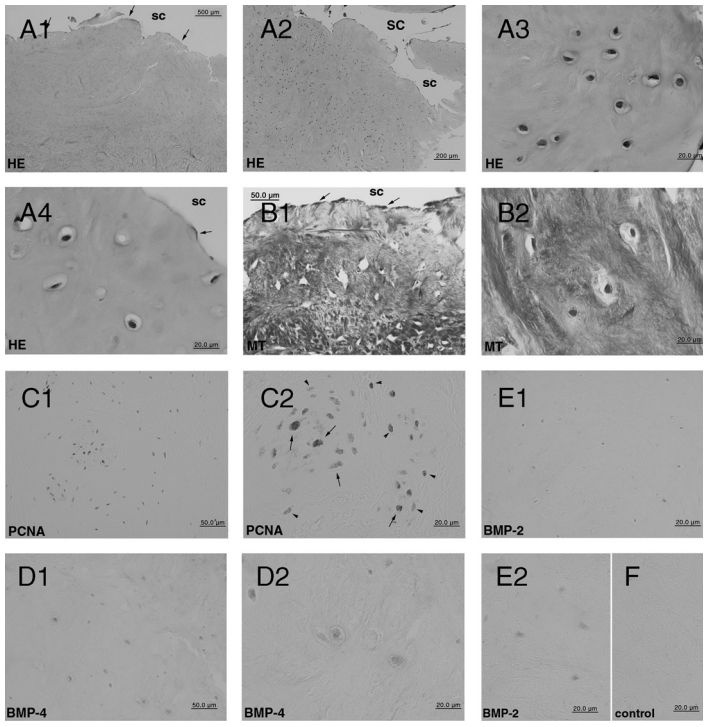

FIGURE 3. Histomorphologic photomicrographs of chondroid tissues. A1–A4, Hematoxylin and eosin staining. A1, Low magnification of chondroid tissue mimicking a round articular surface (arrows) with synovial cavity (SC). A2, The SC had irregular intrusions, and the chondroid tissue cellularity was relatively high in number. A3, The high magnification of chondroid tissue, exhibiting conspicuous chondrocytes and fibrous hyaline stromal tissue. A4, In the margin of the chondroid tissue, there appeared to be clear mesothelial cells (arrows) lining the synovial membrane. B1–B2, Masson trichrome staining. B1, The chondroid tissue stained blue was covered with synovial membrane (arrows). B2, High magnification; the chondrocytes surrounded by fibrous stromal tissue. C1–C2, Immunostaining of PCNA. C1, The chondroid tissue showed frequent positive reaction of PCNA. C2, PCNA was positive not only in the chondrocytes (arrows) but also in the stromal mesenchymal cells (arrow heads). D1–D2, Immunostaining of BMP-4. D1, Low magnification. D2, High magnification; chondrocytes were strongly positive for BMP-4. E1–E2, Immunostaining of BMP-2. E1, Low magnification. E2, High magnification; chondrocytes were strongly positive for BMP-2. F, Immunostaining for the negative control using normal rabbit serum showed no reaction.

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The entire cartilaginous tissue mimicked the round shape of a normal condylar head on low magnification microscopy and showed a hyperplastic proliferation of chondrocytes diffusely scattered in the thick fibrous hyaline stromal tissue. Chondrocytes were frequently positive for PCNA and strongly positive for BMP-4 but were sparse for BMP-2 in the immunohistochemical stainings. However, the PCNA was positive not only for the chondrocytes but also for the stromal mesenchymal cells. Therefore, even though we thought that the growth pattern and distribution of chondrocytes were abnormal compared with the normal condylar cartilage, there seemed to be a conspicuous differentiation in the chondrocytes that seldom undergo osteogenic differentiation, which resulted in the hyperplastic chondroid tissue almost occupying the TMJ area.[14,15]

However, 2 questions were arisen from this TMJ ankylosis; the first question was from which tissue the hyperplastic chondroid tissue was derived, and the second was why the chondrocytes of the hyperplastic chondroid tissue continuously kept on the proliferation without osteogenic transformation.

During the surgical procedure, the downwardly displaced fractured condylar head was removed and fixed again into the normal position of the mandibular condyle. The fractured condyle showed almost intact articular cartilage that was slightly resorbed on its articular surface. Also, the pseudo-bony lesion causing the TMJ ankylosis was tightly attached onto the margin of old fracture of condyle neck. Therefore, it is presumed that the hyperplastic chondroid tissue found in this case of TMJ ankylosis may be derived from the callus formed in the old fractured site of the condylar neck, not from the articular cartilage of the fractured condyle. Fortunately, these findings may directly answer the first question in this report.

For the second question, the hyperplastic chondroid tissue was carefully observed with high magnification microscopy. We found that the hyperplastic chondroid tissue was irregularly associated with synovial tissue forming rudimentary synovial membranes and cavities. The synovial membrane lined by mesothelial cells was directly attached to the chondroid tissue, in which several clusters of chondrocytes were proliferating, admixed with basophilic fibrous stromal tissue. It is known that the differentiating chondrocytes have to be supported by synovial fluid through the synovial membrane, or else chondrocytes cannot mature for the following endochondral ossification.[15–19] The present case demonstrated hyperplastic chondroid tissue in TMJ ankylosis, and this hyperplastic chondroid tissue was closely associated with irregularly formed synovial cavities. Therefore, we suppose that the continuous proliferation of the hyperplastic chondroid tissue in the TMJ ankylosis can be ascribed to the existence of synovial tissue from the nearby TMJ, in contrast to the other diaphyseal fractures in long bones.

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A Triage System for Referrals of Pediatric Skull Deformities

*Hansje Bredero-Boelhouwer, RN, ANP,**
Linda J. Treharne, MSc, FRCS,†
*Irene M. J. Mathijssen, MD, PhD**

Abstract: The clinical pathways for craniosynostosis and nonsynostotic skull deformity are entirely different. At the Dutch Craniofacial Center (DCFC), all patients were assessed in the same multidisciplinary craniofacial clinic, a common practice in countries with developed health care. However, the high volume of referrals of nonsynostotic cases frequently resulted in the capacity of these clinics being exceeded, with some patients being assessed in the general pediatric plastic surgery clinic instead. In these general clinics, not all the multidisciplinary team members are routinely present, so patients with craniosynostosis had to make a second journey for further assessment, causing inconvenience, expense, and

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potential delay in treatment. With triage at the community level unreliable and triage at clinic level inefficient, we decided to trial a triage system to increase efficiency and to ensure patients enter the correct clinical pathway earlier.

The 2 craniofacial secretaries were issued with a flowchart to be completed for each new referral. The flowcharts were designed to triage the patients into true craniosynostosis with an appointment for the multidisciplinary clinic or nonsynostotic deformity with an appointment with the craniofacial nurse practitioner (CNP). During a 3-month period, 107 referrals were made. The triage category listed on the initial flowchart for each patient was compared, with the final diagnosis made in the multidisciplinary and CNP clinics.

None of the patients triaged as nonsynostotic deformity on the flowcharts were found to be true craniosynostosis after clinical assessment by the CNP. Radiographic assessment or assessment by the craniofacial surgeons in the DCFC confirmed this.

The flowchart questionnaire used at the DCFC is a highly sensitive and therefore safe method for detecting craniosynostosis. It has helped to improve efficiency by ensuring patients are seen in an appropriate setting.

Key Words: Triage, clinical pathway, craniosynostosis, nonsynostotic deformity

Craniosynostosis is a relatively rare condition requiring prompt diagnosis and surgical intervention to avoid the possible long-term sequelae of raised intracranial pressure or developmental delay. The child requires assessment in a specialized center familiar with the condition and preferably in a multidisciplinary environment to avoid multiple appointments and inevitable treatment delay. In contrast, nonsynostotic cranial deformity is a relatively common aesthetic problem caused most often by postnatal positioning. Current advice to place neonates prone to reduce the risk of cot death has increased the incidence of nonsynostotic occipital deformity.[1] Treatment is nonsurgical and consists of patient education, patient positioning, and physiotherapy. More severe deformities or insufficient improvement after these treatment modalities may require the use of a molding helmet.[2,3] This clinical pathway is entirely different from that of the synostotic cases.

The 2 conditions can be distinguished by clinical assessment by an experienced practitioner in most cases. The remainder can be diagnosed by plain skull radiographs, but these also require experience to interpret. The rarity of true craniosynostosis means that a general practitioner or pediatrician is unlikely to see the condition more than once or twice in their career and is unlikely therefore to develop the necessary expertise to make a confident diagnosis. The consequence of the large number of nonsynostotic referrals is that the multidisciplinary craniofacial clinics become overburdened with nonsynostotic cases. Not only is this an inefficient use of this expensive resource, but also, more seriously, the true craniosynostoses have their assessment and treatment potential delayed by the far more numerous nonsynostotic cases.

Craniosynostoses in the Netherlands are assessed and treated in a single craniofacial center located at the Erasmus Medical Center–Sophia Children's Hospital in Rotterdam, the Dutch Craniofacial Center (DCFC). The DCFC receives around 400 new referrals per annum, 80 of which are true craniosynostoses. True craniosynostoses are assessed and treated in a multidisciplinary environment, with input from 3 craniofacial plastic surgeons, 2 neurosurgeons, maxillofacial surgery, ophthalmology, otorhinolaryngology, pediatrics, orthodontics, clinical genetics, and all relevant supporting professionals. Nonsynostotic deformities are seen and treated by a craniofacial nurse practitioner (CNP), who runs a parallel clinic alongside the multidisciplinary one, working independently but with backup by the plastic surgeon if required.

Before implementation of our triage system, all new referrals with cranial deformity were seen by the craniofacial surgeons in their general pediatric plastic surgery clinics if the multidisciplinary craniofacial clinic was fully booked. Patients with true craniosynostosis who were seen in these clinics often had to make a second journey to the hospital for further assessment by members of the team not routinely present in the general clinics, causing inconvenience and expense for the patients and families and the potential for delays in surgical treatment. With triage at the community level unreliable and triage at clinic level inefficient, we decided to trial a triage system at a different level—one which combines expertise with efficiency and ensures more patients enter the correct clinical pathway from referral. The aim of a clinical pathway is to improve the quality of care, reduce risks, increase patient satisfaction, and increase the efficiency in the use of resources.[4] For the DCFC, this means adequate planning (extent of urgency), presence of members of the DCFC, decreasing waiting time, and adequate information.

METHOD

All referrals to the DCFC are via telephone to the 2 craniofacial secretaries. We issued the secretaries with a flowchart questionnaire to be completed for each new referral (Fig. 1). Patients with obvious syndromic craniosynostoses, such as Apert syndrome, diagnosed before referral, were excluded from the study. The flowcharts were designed to include all relevant administrative details, in addition to triaging the patients into true craniosynostosis and nonsynostosis. The key questions to differentiate the true craniosynostoses from the nonsynostotic deformities were the following:

(1) "Was the deformity present from birth?" True craniosynostosis is present at birth, whereas nonsynostotic deformities develop in the neonatal period
(2) Presence of preferable position
(3) "Is the deformity improving?" The deformity with true craniosynostosis gets worse with time, whereas the nonsynostotic deformities improve as the child develops head control and the skull no longer has localized pressure for long periods

The patients were then given appointment dates with the CNP clinic if the diagnosis was a suspected nonsynostotic deformity, or for the multidisciplinary clinic if the diagnosis was craniosynostosis. In addition, all parents of patients with suspected craniosynostosis were reached via telephone by the CNP before their appointment. The aims of this telephone consultation were to:

- refine or change the triage category and thus appointment if necessary and at the discretion of the CNP

From the *Dutch Craniofacial Center, Erasmus Medical Center–Sophia, and †Dutch Association for Facial Plastic and Reconstructive Surgery, Rotterdam, the Netherlands.
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Address correspondence and reprint requests to Hansje Bredero, RN, ANP, Department of Plastic Surgery, Erasmus Medical Center–Sophia, Sk-1222, PO Box 2060, 3000 CB Rotterdam, the Netherlands; E-mail: H.Bredero-Boelhouwer@erasmusmc.nl
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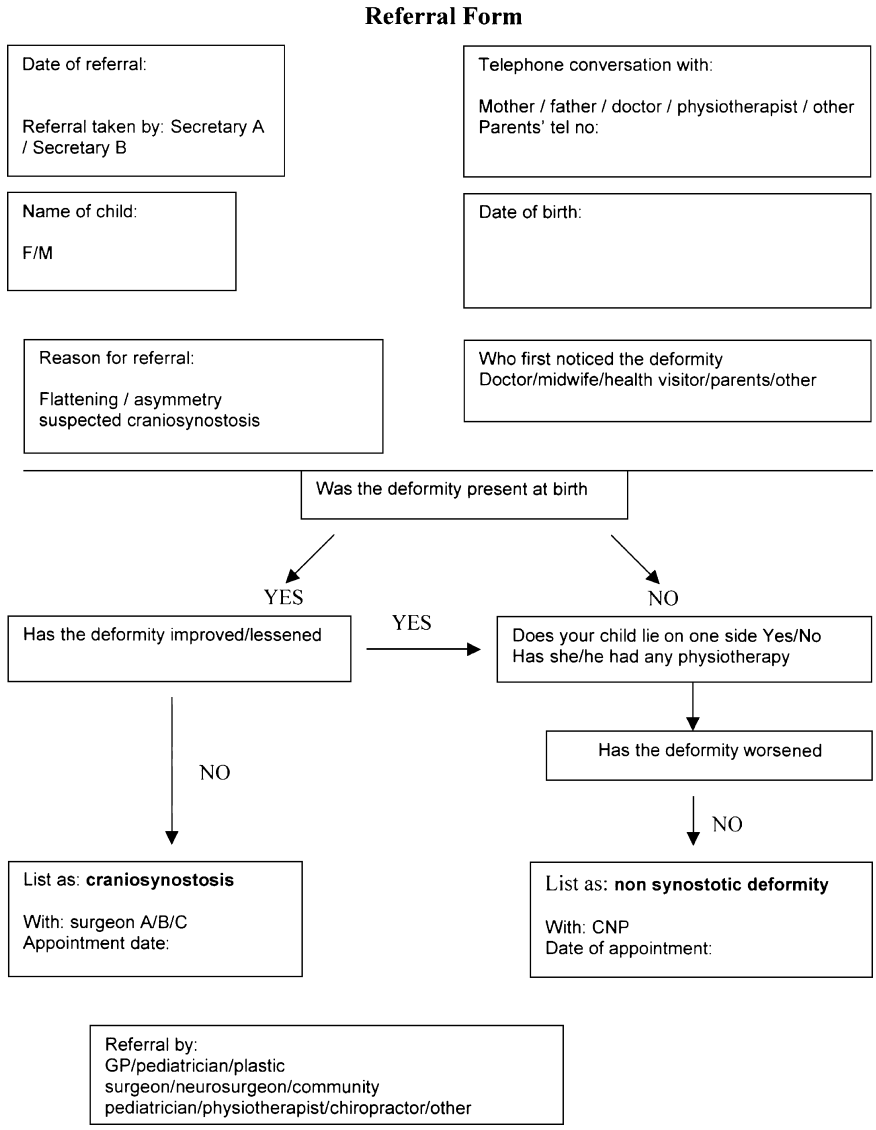

FIGURE 1. Flowchart for triage of new referral via telephone.

- introduce the parents to the CNP and the DCFC and give explanation about the DCFC
- ascertain which members of the craniofacial multidisciplinary team were likely to be required at the appointment
- answer any initial questions the parents may have.

During a 3-month period (January 1 to March 31, 2007), 107 referrals were made to the DCFC. The triage categories listed on the initial referral flowcharts were compared with the final diagnosis made in the multidisciplinary and CNP clinics.

RESULTS

Results are shown in tabular form in Figure 2. Of the 107 referrals made, 18 were presented as craniosynostosis and 89 as positional deformities. Based on the flowchart questionnaires, 39 were triaged as true craniosynostoses and 68 as nonsynostotic deformities on the flowchart questionnaires. After telephone consultation by the CNP to the parents of those diagnosed as true craniosynostoses, 7 patients were recategorized as nonsynostotic deformities. Recategorization was based on further investigation of the process of change, questioning the skull deformity and the presence or absence of external influences. A further 8 of the patients were recategorized as nonsynostotic deformities following assessment at the multidisciplinary craniofacial clinic by the craniofacial surgeons. None of the patients triaged as noncraniosynostotic deformity on the flowchart questionnaires were found to be true craniosynostosis after clinical assessment by the CNP or after radiographic assessment or assessment by the craniofacial surgeons in the DCFC.

DISCUSSION

Triage, from the French "trier," meaning "to sort," is most commonly associated with mass casualty situations. Telephone triage has been used in accident and emergency departments and general practitioner surgeries. It seems to improve efficiency by

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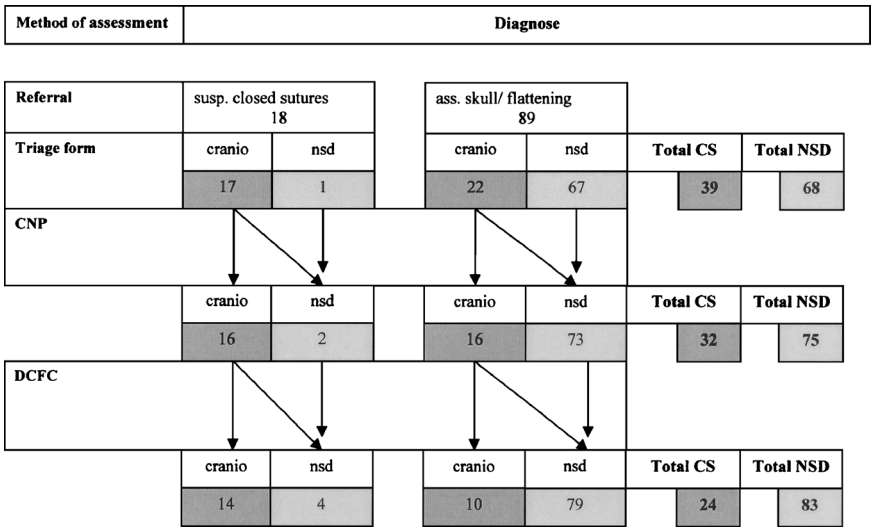

FIGURE 2. Number of patients within each diagnostic group, in the various stages of their intake. CS indicates craniosynostosis; NSD, nonsynostotic deformities; Susp. closed sutures, suspected closed sutures; Ass. skull, asymmetric skull.

reducing unnecessary consultations, but concerns have been raised over the safety of this method of assessment.[5]

The flowchart used by the CFCN is not used as a diagnostic tool but as an assessment tool.

Triage of patients referred to the DCFC with neonatal skull deformities by a flowchart questionnaire had a high false-positive rate of 38.5% for diagnosing true craniosynostosis. The CNP contact reduced false-positive rate to 25%. However, the rate of false-negative diagnoses of true craniosynostosis was 0%, giving this method of assessment a high sensitivity but low specificity. This means that some patients with nonsynostotic deformities are still being seen in the multidisciplinary craniofacial clinic unnecessarily. Although we could modify the flowchart questionnaire to try to decrease the rate of false-positive diagnoses of true craniosynostosis, we believe it is more important to maintain the high sensitivity of the triage system to avoid missing a diagnosis of craniosynostosis.

CONCLUSION

The flowchart questionnaire used at the DCFC is a highly sensitive and therefore safe method for detecting true craniosynostosis. It has helped to improve efficiency in our service by ensuring that all patients with craniosynostoses are seen in the multidisciplinary craniofacial outpatient clinic and reduces the number of patients with nonsynostotic deformities being seen inappropriately in this setting. Telephone consultation with the CNP further decreases the number of false-positive diagnoses of true craniosynostosis while allowing early support and education to parents.

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The Helical Arcade: Anatomic Basis for Survival in Near-Total Ear Avulsion

Detlev Erdmann, MD, PhD, Anthony D. Bruno, MD, Keith E. Follmar, BA, Tracey H. Stokes, MD, Dennis L. Gonyon, MD, Jeffrey R. Marcus, MD

Abstract: Survival of the ear based on a single vascular pedicle has been previously documented. The anatomic basis for this survival remains incompletely described. In the 3 clinical cases of inferior-to-superior near-total ear avulsion presented herein, the authors have observed that the ear can survive based on a narrow pedicle along the helical root. In an anatomic study to further investigate the relevant vascular anatomy, the common carotid artery system of 6 fresh human cadaver specimens was injected with latex. The superficial temporal artery and its branches located at the helical root were examined. This anatomic study showed that the upper auricular branch of the superficial temporal artery continues into the ear as the helical artery supplies an arterial arcade, which we have termed the *helical arcade*. This arcade seems to allow for communication between the anterior and posterior arterial systems of the ear and allows the helical artery to provide sufficient arterial supply to the entire ear.

From the Division of Plastic, Reconstructive, Maxillofacial and Oral Surgery, Department of Surgery, Duke University Medical Center, Durham, North Carolina.
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Address correspondence and reprint requests to Dr. Detlev Erdmann, Division of Plastic, Reconstructive, Maxillofacial, and Oral Surgery, Duke University Medical Center, Box 3181, Durham, NC 27710; E-mail: Detlev.Erdmann@duke.edu
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Key Words: Ear, helical artery, replantation, vascular anatomy

Total and near-total traumatic amputation of the ear can result from a variety of mechanisms, including motor vehicle collisions, falls onto sharp objects, assault with a knife, and occupational injuries. The affected patient population includes all age groups. Total amputation remains a clinical challenge even to the experienced surgeon. Management options include microvascular replantation,[1] replantation without microvascular anastomosis,[2] banking of de-epithelialized auricular cartilage and delayed replantation,[3] local flap coverage of the cartilage,[4] and staged autologous reconstruction with rib cartilage.[5]

Based on the authors' experience, near-total amputation or avulsion of the ear seems to be a very different clinical entity. In 3 recent clinical cases presented herein, it has been observed that nearly the entire ear can survive based on a narrow skin/soft tissue bridge above the tragus, along the helical root. In all cases, cartilaginous reconstruction and layered closure were performed. Venous stasis occurred in 2 cases and required leech therapy, but salvage of the ear was successful in all cases. It is known that the external ear derives blood supply from dual sources: anteriorly via the superficial temporal artery (STA) and posteriorly via the posterior auricular artery (PAA). It was hypothesized that collateralization of the anterior system (specifically branches at the helical root) had provided an adequate single blood supply (axial) to the ear. The anatomic basis for communication between the anterior and posterior vascular systems has not been described, to the authors' knowledge. A series of anatomic dissections was carried out to investigate our hypothesis and identify its anatomic basis.

EMBRYOLOGY AND ANATOMY

The embryologic development of the ear begins at 6 weeks. Six hillocks are seen on the first and second brachial arches. The 3 anterior hillocks arise from the first brachial arch—also known as the mandibular arch—and form the tragus, root of helix, and superior helix. The posterior 3 hillocks arise from the second brachial arch—also known as the hyoid arch—and form the antihelix, antitragus, and lobule. By 8 weeks of development, these 6 hillocks form a primitive ear by the process of fusion or accretion.[6]

The 2 arteries that supply the ear, the STA and the PAA, are each a terminal branch of the external carotid artery. The STA (anterior arterial supply) gives 3 main auricular branches: the upper, middle, and lower branches. The PAA (posterior arterial supply) also provides 3 similarly named main branches, before continuing on to supply the temporal-parietal region. Park et al[7] described 2 arterial networks, the network of the triangular fossa-scapha and the network of the concha. Both eventually communicate on the antihelix. The triangular fossa-scapha network originates from 1 subbranch of the upper auricular branch of the STA and from branches of the PAA that come through the ear lobe and triangular fossa and over the helical margin. The conchal network is provided by 2 to 4 perforators that come from the PAA, piercing the conchal floor. Its piercing points are at the helical root, the cavum conchae, and the cymba conchae. The anterior surface of the ear and part of the superoposterior surface of the pinna are drained by veins emptying into the superficial temporal veins and retromandibular (posterior facial) veins. The posterior auricular veins drain into the external jugular system.[8] The authors have hypothesized that the ears in each of our 3 cases survived because of the interconnections between the 2 arterial systems.

MATERIALS AND METHODS

Three clinical cases of successful replantation of inferior-to-superior near-total ear avulsion were retrospectively reviewed. The anatomy of 6 fresh human cadaver head specimens was studied.

Patient 1

A 23-year-old woman presented with multiple injuries sustained in a rollover motor vehicle accident including a near-total avulsion injury of the left ear. The ear was attached by a 1.5-cm skin bridge superior to the tragus and anterior to the helical root (Figs. 1A, B). The ear appeared venous congested, but otherwise viable. The patient was taken to the operating room approximately 6 hours after the injury, and a 3-layer repair of the ear was performed. Because of the circumferential laceration of the external ear canal and associated potential for stricture, canalplasty was performed at the initial repair using a longitudinal incision and integration of a diamond-shape, full-thickness skin graft. The canal then was maintained with a gauze stent, and the ear was dressed with a bulky, noncompressive dressing.

On follow-up examination, more than 95% of the ear remained viable. Gradual demarcation of the lobule (distalmost aspect) was observed. Once wound healing was complete, the patient underwent an anterior-based postauricular banner flap for reconstruction of the lobule (Fig. 1C). At 1 year, the overall appearance was quite satisfactory with regard to shape; projection was slightly greater on the repaired side.

Patient 2

A 3-year-old boy sustained a near-total avulsion of the ear in a fall onto a table. The distal aspect of his ear became caught on a porcelain figurine and was avulsed in an inferior-to-superior direction. He was taken to the operating room within 6 hours of time of the injury. Inspection revealed avulsion of the ear along

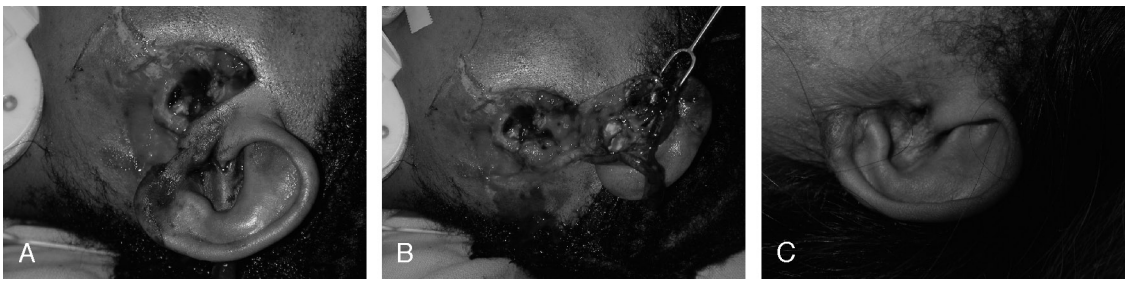

FIGURE 1. A and B, Female patient with a near-total avulsion injury of the left ear; skin bridge superior to the tragus and anterior to the helical root. C, One year follow-up including ear lobe reconstruction.

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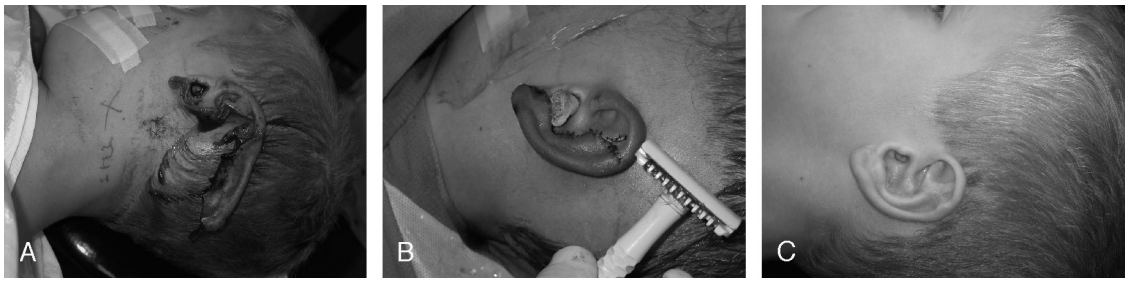

FIGURE 2. A, Near-total ear amputation with attachment at the anterosuperior aspect with a less than 1-cm pedicle of the skin. B, Postoperative venous congestion. C, Satisfactory shape and symmetric projection after 1 year.

the junction of the concha and antihelix. The ear remained attached at the anterosuperior aspect by a less than 1-cm pedicle of the skin (Fig. 2A). The ear appeared congested but was thought to be largely viable, and repair was performed (Fig. 2B). Postoperatively, the ear remained congested, and treatment with intermittent leech therapy was undertaken for 3 days. Congestion improved, and leech therapy was subsequently discontinued. At follow-up, the entire ear remained entirely viable. No tissue loss occurred. At 1 year, the ear maintained a satisfactory shape and symmetric projection (Fig. 2C).

Patient 3

A 52-year-old man sustained a near-complete amputation of his left ear after a work-related fall from a forklift. The ear remained attached at the base of the helical rim superior to the external canal with an approximately 0.5-cm bridge of skin. The patient was immediately taken to the operating room within 6 hours of the injury. As the tissues appeared to be in good condition, a layered reattachment was performed of the posterior skin, cartilage, and anterior skin. Shortly after completion of surgery, the ear appeared to be venous congested. Leeches were applied to the anterior surface of the ear every 4 to 6 hours until venous congestion improved on postoperative day 4. At this point, leech therapy was discontinued. The patient was discharged to home on postoperative day 6 with complete survival and normal appearance of the ear (Figs. 3A, B).

Anatomic Study

The common carotid system of each of 6 fresh human cadaver head specimens was injected with pink latex solution bilaterally. After allowing the latex to polymerize, the branching of the STA at the helical root was investigated by careful anatomic dissection. One specimen had been previously manipulated unilaterally, yielding that ear unusable. Thus, 11 ears were available for study.

RESULTS

In 10 of 11 fresh cadaver dissections (91%), an axial branch of the STA (Fig. 4A) that entered the helical root was identified. In each case, this artery was followed distally to map its course. The artery travels at a 90-degree angle to the STA at the helical root. The branch continued along the margin of the helix. Along its course, the artery formed an arterial arcade (Fig. 4B), which gave off many small branches most posteriorly, but some anteriorly as well. We have termed this arterial arcade the *helical arcade*. Ultimately, the helical artery arcade communicates inferiorly with the branches of the PAA.

DISCUSSION

In our series of 3 patients with inferior-to-superior near-total traumatic ear amputation, the ear survived on a small skin/soft tissue bridge at the helical root. The upper auricular branch of the STA contained in this bridge supplied the entire arterial supply to the ear. The posterior circulation of the ear received blood by anterior-posterior connections via the helical arcade. Venous outflow was significantly compromised, and leech therapy was necessary for 3 to 4 days to allow neovascularization to take place in 2 of the cases.

The findings from our fresh cadaver dissection corroborate our clinical findings and provide anatomic insight into survival of the ear based on a single arterial pedicle. In 10 of 11 ear dissections, the upper branch of the STA was identified and followed to a richly

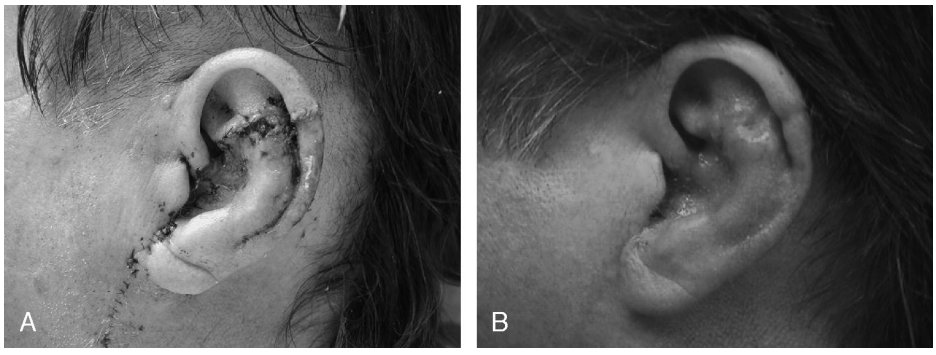

FIGURE 3. A, Day 6 after near-complete amputation of the left ear due to a work-related fall from a forklift. B, Follow-up after 6 months.

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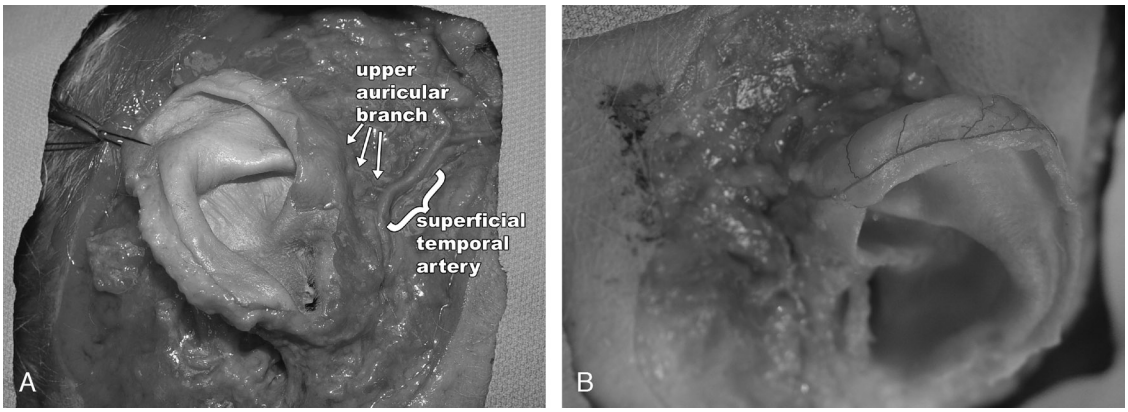

FIGURE 4. A, Anatomic study with latex injection of small branches off the STA. B, Helical arcade.

anastomotic arterial arcade. The arterial branches of the STA to the ear have been described by Park and Roh[6] as the upper, middle, and lower branches based on dissection of 20 cadaver ears. Parkhouse and Evans[9] also demonstrated this anatomy in a series of 40 cadaver dissections.

Park suggested that the ear can survive via a single vessel, either a posterior pedicle or superior pedicle.[6,7] Our findings confirm ear survival via a single axial vessel in the situation of traumatic partial ear amputation. Park and Chung[10] discussed the relevance of the vascular system in designing flaps for auricular reconstruction, and Parkhouse and Evans[9] described this anatomy for an upper anterior helix composite free flap. Recently, the blood supply to the auricle was analyzed in terms of feeding vessels to auricular arteriovenous malformations, yielding again findings similar in terms of anatomic blood supply to the ear and the existence of a watershed area between the STA and the PAA.[11]

Our findings concur with this previously described anatomy and also with Park's hypothesis[6,7] that the ear may survive on either of its 2 dominant vascular systems. Specifically, the upper auricular branch of the STA, continuing as the helical artery and dividing into the helical arcade, which communicates with the posterior vasculature, is demonstrated to provide an adequate sole source of blood supply in 3 cases of inferior-to-superior avulsion with only a minimal skin bridge. It is this helical arcade, which has not been previously described or named, that is the basis for ear survival in the near-total ear amputation. In our anatomic study, the quality and continuity of the helical arcade between the anterior and posterior systems are clear and consistent.

Primary closure of the nearly avulsed ear is possible with good results. For cases in which the avulsed ear is neither viable nor suitable for replantation, other options for ear salvage have been described. These include banking of the de-epithelialized ear,[3] local/regional flap coverage of the auricular cartilage,[4] and finally, discard of the amputated part followed by staged autologous reconstruction.[5] The decision regarding the management of a near-total ear avulsion should be made at the time of presentation based on the viability and condition of the ear, surgeon experience and expertise, hospital resources and support, and associated injury or morbidity.

CONCLUSION

Clinical observations suggest that the external ear can survive attached at a small bridge anterior to the helical root. Fresh human cadaver dissections confirm a consistent upper auricular branch of the STA, which terminates as the helical artery. A vascular arcade was found along the helical rim proceeding through preinjury watershed areas of communication with branches of the PAA; it is this arcade which we have named the helical arcade. The supply of the upper auricular-helical artery seems to be sufficient for the arterial vascular supply of the entire ear. Thus, in the case of near-total ear avulsion that is still attached at the helical root, the ear can be successfully reattached with venous outflow supported by leech therapy until neovascularization takes place.

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Survival After Lip Cancer Diagnosis

Katayoun Sargeran, DDS, PhD,†*
*Heikki Murtomaa, DDS, MSc, PhD,**
Seyed Mohammad Reza Safavi, DDS, MSc,‡
Miira M. Vehkalahti, DDS, PhD, Olli Teronen, DDS, PhD§*

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Abstract: The purpose of this study was to analyze the 5-year survival rates of 82 patients with lip cancer attending 5 university hospitals during 1999–2003 in Tehran, Iran. We used information from patient records, telephone calls, and death register files of the Iran Ministry of Health to ascertain the patients' vital status. Associations between survival and the variables of sex, age, stage of the tumor at the time of diagnosis, treatment modality, and tumor histopathologic type were analyzed with Kaplan-Meier, log-rank, and Cox regression methods. Of all patients, 70 (85%) were men, with a median age of 62 years (mean, 58.6 years [SD, 15 years]; range, 27–85 years) at the time of diagnosis. The median follow-up time of the patients was 57 months (mean, 56.4 months [SD, 28 months]; range, 0–112 months). The 1- to 5-year overall survival rate was 91% to 62%. The tumor stage at the time of diagnosis and the treatment modality were associated with survival ($P < 0.05$) in both univariate and multivariable analyses. Patients who underwent surgery and had lower stage tumors at the time of diagnosis showed higher survival rates. No differences in patient survival were found regarding sex, age, and histopathologic type of tumors. These findings indicate that although lip tumors are curable, early detection, diagnosis, and treatment lead to even higher rates of survival. Importance of the early detection of lip cancer should be emphasized in all health care and cancer prevention campaigns directed to the public and professionals.

Key Words: Iran, lip cancer, outcome survival

Lip cancer is the most common malignancy among oral cancers in some parts of the Western world, such as Australia,[1,2] Canada,[1,3] Spain, and Finland.[1,4,5] These countries also have the highest incidence rates of lip cancer with up to 14 cases per 100,000 persons.[1,6] In contrast, in Asia, the incidence rates, ranging from 0.0% to 3%, are not as high as those reported for other oral cancers.[1,6,7] Squamous cell carcinoma (SCC) constitutes more than 90% of all lip cancers, and almost 95% of lip cancers arise on the lower lip.[8,9] The incidence rates of lip SCCs are higher in white men, and the main risk factors are chronic sun exposure and smoking.[4,5,8,10–12]

Lip SCCs are less likely to cause mortality compared with other cancers of the oral cavity.[13] Depending on the stage of tumor at the time of diagnosis, the survival rates vary from 50% to 100%.[14–16] The highest survival is for early-stage tumors.

Most lip cancers are controlled successfully by complete excision, but there exists a risk of developing recurrent tumors that can require subsequent surgical resections and reconstructive procedures.[14,15,17,18] Although lip cancer is known to be curable especially at the early stages of the disease, it has tremendous adverse individual and public health effects related to high medical costs and, in advanced or aggressive cases, lower survival rates and diminished quality of life due to devastating aesthetic and psychosocial sequel, functional impairment, and other serious consequences.[19]

Reports regarding survival after lip cancers in the developing countries are rare. To our knowledge, no published reports related to lip cancer survival in the Middle East area including Iran are available. In the current study, we determined the 1- to 5-year survival rates for patients with lip cancer in relation to age, sex, stage of the tumor, histological type, and treatment modality during 1996–2003 in 5 university hospitals in Tehran, Iran.

METHODS

Study Patients

We enrolled a retrospective cohort of patients with primary lip cancer referred to 5 university hospitals in Tehran during 1996–2003. These hospitals are the main tertiary and referral centers for the treatment of patients with oral cancer from all over the country. The eligibility criterion was pathologically confirmed lip malignant lesion.[20] Patients with incomplete medical records, tumors of hematologic origin, carcinoma in situ, recurrent tumors, metastases, and previous history of any cancer were excluded.

Design and Setting

The patients were followed up from the date of diagnosis to late part of 2005. We defined *survival* as the time from diagnosis until December 31, 2005, or until death due to cancer, whichever occurred first. Survival time was censored if a patient was alive at the end of follow-up, was lost to follow-up, or died of causes other than cancer. Ethical approval for the study was given by the ethics committee of the School of Dentistry, Shaheed Beheshti Medical University.

Data Collection

All eligible patients were identified according to the patient attendance list and information files held in the patient registration department of each hospital. No patient can be hospitalized or treated in these 5 hospitals without the prior filling out of the required forms and documents.

Information abstracted from patient records included birth date, sex, and date of diagnosis. The tumor, node, metastases (TNM) stage[21]; primary tumor site; and histopathologic type were retrieved from pathology reports. Treatment modality (surgery, preoperative or postoperative radiotherapy) and the final admittance dates to the hospital were also recorded, as were the date and the cancer or noncancer causes of death if death happened in the hospital. The distribution of our study patients by age (mean, 58.6 years [SD, 15.2 years]) and sex (85% men) was similar to our previous study regarding oral cancers in Tehran, Iran.[9] Data on the stage of tumor were missing for 12% of all lip cancer cases. Patients with missing data were excluded from the corresponding analyses.

Vital status was ascertained using a combination of information from patient records, telephone calls, and death register files of the Iran Ministry of Health and was considered as uncensored if the death had been due to cancer and censored if the patient was alive at the end of follow-up or had died of other causes. Of all patients with lip cancer, 2 (2%) were lost to follow-up, which was recorded for the following conditions:

- There was no record in the death certificate file.
- There were no data on patient survival in the medical record.

From the *Department of Oral Public Health, Institute of Dentistry, University of Helsinki, Helsinki, Finland; †Department of Community Oral Health and ‡Iran Centre for Dental Research, School of Dentistry, Shaheed Beheshti Medical University, Tehran, Iran; and §Institute of Dentistry, University of Helsinki, Helsinki, Finland.
Received July 4, 2007.
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Address correspondence and reprint requests to Katayoun Sargeran, DDS, PhD, Institute of Dentistry, University of Helsinki, PO Box 41, FI-00014 Helsinki, Finland; E-mail: katayoun.sargeran@helsinki.fi
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- The person who answered the telephone did not know the patient or the date of death.
- There was no answer to 5 telephone calls at different times of the day during 1 week.

Any change in the phone numbers was checked from the telephone information center. The time of death and final follow-up were recorded to an accuracy of 1 month.

Statistical Analysis

Survival curves were generated using Kaplan-Meier methods. After initial descriptive statistics, univariable analyses of the relationships between survival status and age, sex, primary tumor site, stage, and histopathologic type were evaluated using the log-rank test. Multivariable analysis was done using Cox regression model to calculate the hazard ratios. Statistical significance was determined for $P < 0.05$.

RESULTS

The median age of our 82 patients with lip cancer was 62 years (mean, 58.6 years [SD, 15.2 years]; range, 27-85 years) at the time of diagnosis. There were 70 (85%) men in our series, and only 2 (2%) of the tumors were on the upper lip. To date, 31 study patients (38%) died of oral cancer, and 46 patients (56%) survived, whereas in 3 patients (4%), death was due to other causes, and 2 patients (2%) were lost to follow-up.

Table 1. Survival Rates for Patients With Lip Cancer (N = 82) by Age, Sex, Stage of Tumor at Diagnosis, Treatment Modality, and Histological Type

		Survival Rates*			
Variable	**n (%)**	**12 mo, %**	**24 mo, %**	**60 mo, %**	***P*†**
Sex					0.32
Women	**12 (15)**	—‡	—‡	—‡	
Men	**70 (85)**	91	86	65	
Age, y					0.35
≤40	**13 (16)**	85	77	36	
41–64	**35 (43)**	89	83	66	
≥65	**34 (41)**	94	88	65	
Stage of tumor					<0.001
I	**35 (43)**	97	91	81	
II	**17 (21)**	93	87	75	
III	**11 (13)**	81	73	45	
IV	**9 (11)**	67	—‡	—‡	
Missing	**10 (12)**				
Treatment					<0.001
Surgery	**34 (42)**	97	91	69	
Surgery + radiotherapy	**35 (43)**	89	86	75	
Radiotherapy	**6 (7)**	—‡	—‡	—‡	
Missing	**7 (8)**				
Histology					0.62
SCC	**77 (94)**	91	86	62	
Not SCC	**5 (6)**	—‡	—‡	—‡	
All	**82 (100)**	91	86	62	

*Kaplan-Meier analysis.
†Log-rank test.
‡Not estimated because of small number of participants.

Table 2. Determinants of Survival Length as Assessed by Cox Regression Analysis in Patients With Lip Cancer (N = 82)

Variable	**Estimate of Strength**	**SE**	**OR**	**95% CI**	***P***
Sex					
Men,* women	**0.5**	**0.6**	**1.6**	**0.5–5.2**	**0.40**
Age group, y					
≤40*					
41–64	**1.1**	**0.8**	**3.0**	**0.7–13.9**	**0.15**
≥65	**0.7**	**0.7**	**2.0**	**0.5–7.9**	**0.33**
Stage					
I*		**1.0**			
II	**0.6**	**0.6**	**1.9**	**0.6–6.2**	**0.31**
III	**1.0**	**0.7**	**2.8**	**0.7–11.0**	**0.15**
IV	**2.0**	**0.7**	**7.6**	**1.9–29.6**	**<0.001**
Treatment					
Surgery*		**1.0**			
Surgery + radiotherapy	**0.1**	**0.5**	**1.1**	**0.4–3.1**	**0.91**
Radiotherapy	**2.0**	**0.8**	**7.7**	**1.5–39.7**	**0.01**
Histology					
SCC,* non-SCC	**1.0**	**0.9**	**2.9**	**0.5–16.1**	**0.23**

*Reference category.

At the time of diagnosis, 43% of all the tumors were at stage I, 21% were at stage II, 13% were at stage III, and 11% were at stage IV. For 10 patients (12%), data regarding the stage were missing. Squamous cell carcinoma was the most predominant histological type (94%).

Of all patients with known treatment status (n = 75), 92% underwent surgery (45% had surgery alone, and 47% had surgery with adjuvant radiotherapy), and 8% were treated with radiotherapy only. The basis for the decision to treat nonsurgically (radiotherapy alone), as indicated in patient records, was the patients' poor general health condition or very advanced stage of cancer at the time of diagnosis. The rationale for postoperative radiotherapy for our patients was (1) positive neck lymphadenopathy; (2) unclear surgical margins; (3) perineural, perivascular, or osseous invasion; (4) extranodal spread; and (5) regional recurrence.

The overall 1-, 2-, and 5-year survival rates were 91%, 86%, and 62%, respectively, in our patients. The results of the univariate analysis between survival and independent variables of age, sex, tumor stage at the time of diagnosis, histological type, and treatment modality are listed in Table 1. Sex, age, and histological type were not associated with survival status. Treatment modality and stage of tumor at the time of diagnosis were related to survival. Patients treated with surgery were more likely to have a longer survival ($P < 0.05$). Patients at stage III or IV had lower survival rates than those at early stages ($P < 0.05$). No difference in survival rates appeared with regard to the histological types of tumors.

The multivariate analysis using Cox regression method showed that treatment modality and stage of tumor were the most important

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determinants of survival compared with age, sex, and histopathologic type of tumor in lip cancers (Table 2). The overall survival was longer in patients with lip cancer at stage I or II than in those at stage III (odds ratio [OR], 2.8; 95% confidence interval [CI], 0.7–11.0) or IV (OR, 7.6; 95% CI, 1.9–29.6) at the time of diagnosis. Patients treated with radiotherapy alone had lower survival rates than those who had undergone surgery and radiotherapy (OR, 7.7; 95% CI, 1.7–39.7).

DISCUSSION

Lip cancers are a form of oral cancer with a totally different tumor behavior and distinct epidemiology that warrants a separate study.[1] Although these cancers are not common in Asian populations, in contrast to other oral cancers, deeper assessments are needed because they are usually considered to be easily detectable and thus curable.[15,17]

Our findings on the survival of patients with lip cancer and its relationship with sex, age, tumor stage, treatment modality, and histopathologic type are in line with previous reports mentioning 60s as the decade of age during which the most frequent presentation of lip tumors occurs.[1,9]

We also found that these cancers occurred mostly in men (85%), on the lower lip (98%), and as SCCs (94%), which is also comparable with other available reports.[1,8,9]

In our patients with lip cancer, the overall survival rates for 1, 2, and 5 years after diagnosis were 91%, 86%, and 62%, respectively. These figures are lower than those reported for the Western countries,[14–16] possibly because all study patients came from tertiary centers. This accounts for the much higher percentage of advanced or aggressive SCCs than generally would be seen in community practice. Nonetheless, advanced cases do occur in the community, albeit less frequently, and so community practices can benefit from the findings reported here.

Our findings that sex, age, and histopathologic type of tumors were insignificant predictors of survival, both in univariate and multivariable analyses, are in line with previous reports.[15,18]

The stage at which lip cancer is diagnosed certainly has a direct impact on the survival of the patient. Previous reports show higher survival rates for patients at earlier stages compared with those diagnosed at later stages, after radiotherapy alone or surgical resection with or without adjuvant radiotherapy.[14–16,18] The survival of our patients certainly was higher in cases diagnosed at the earlier stages of tumors than those diagnosed at the advanced stages.

In the current study, univariate and multivariable analyses showed that patients who were treated with radiotherapy alone had lower survival rates than those who were treated surgically or who had undergone surgery together with adjuvant radiotherapy. Patients receiving radiotherapy alone had a more advanced disease (50% at stage III or IV at the time of diagnosis) and/or might be of poor general health. Our findings support previous studies regarding the relationship between treatment modality and survival, which show higher rates of recurrence and lower disease-free survival in patients with lip cancer initially treated with radiotherapy.[15,22]

In this study, we investigated the survival rate of patients with lip cancer and its relation with age, sex, tumor site, stage at diagnosis, treatment modality, and tumor histology. Assessing the quality of surgical techniques and the other treatment details such as the clearance of surgical margins and tumor details such as differentiation would be fundamental issues for further studies.

Considering possible future trends in important risk factors such as smoking and chronic sun exposure in our society, the data reported here are of fundamental importance to be used as a baseline for comparing any changes in the survival rates of patients with lip cancer. This will enable health care providers to organize more comprehensive cancer prevention programs in Iran. Further population-based studies on lip cancers and advances in their risk assessment, surveillance, and prevention will be crucial for controlling the substantial threat of lip cancers to the public health.

The authors thank the Iran Cancer Institute and all hospital personnel for their cooperation.

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Giant Lymphangioma of the Tongue

Joon Pio Hong, MD, PhD, MMM, Moo Young Lee, MD, Eun Key Kim, MD, Dong Hwan Seo, MD

Abstract: This report presents the treatment of an extensive lymphatic malformation of the tongue. Sclerosing agents are now widely used as the first-line treatment of lymphatic malformation. However, treatment of lymphatic malformation involving the face and the vital structures such as the airway remains to be challenging. A 4-year-old boy underwent a total of 15 OK-432 injection sclerotherapy treatments over a 2-year period, having slow progress until sudden enlargement of the tongue was noted shortly after the last injection. Partial excision of the lesion was performed. This case demonstrates the risk in treating large microcystic lymphatic malformation of the tongue with sclerotherapy and provides an insight in the management protocol.

Key Words: Lymphatic malformation, OK-432, tongue

Lymphatic malformation is an uncommon, harmatomatous, benign tumor involving the skin and subcutaneous tissue. It can be acquired from trauma or surgery, but congenital forms are more frequently seen. It occurs anywhere in the skin and mucous membrane, but the common sites are the head and neck. It may regress spontaneously, but it usually persists throughout the craniofacial region and remains to be annoying.[1,2] Lymphatic malformation rarely involves any mortality and is approached in a more cosmetic manner. However, total surgical excision is warranted when it involves the airway, which may lead to respiratory compromise, aspiration, and infection of the respiratory system.

Currently, various sclerosants such as bleomycin, tetracycline, inactivated OK-432 (Picibanil, Choongwae Pharm Corporation, Japan), and 100% ethanol are used as first-line treatments through an intralesional percutaneous route.[3,4] It has many advantages in overcoming the complications that may occur from surgery, such as damage to surrounding structures including nerves and vessels, scarring, and recurrence due to incomplete excision. However, despite its advantages, lesions in the airway still remain to be difficult with any means. This is a report on such a lymphatic malformation involving the tongue.

From the Department of Plastic Surgery, Asan Medical Center, University of Ulsan College of Medicine, Seoul, Korea.
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Address correspondence and reprint requests to Joon Pio Hong, MD, PhD, MMM, Department of Plastic and Reconstructive Surgery, Asan Medical Center, Songpa-gu, Poongnap2-dong, 388-1, Seoul, 138-736, Korea; E-mail: joonphong@amc.seoul.kr
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CLINICAL REPORT

A 4-year-old boy was referred to Asan Medical Center for evaluation and management of congenital macroglossia. The mass was initially diagnosed as cystic hygroma at a local facility, and surgical resection was attempted 2 times at the age of 2 and 4 months. Temporary regression of the mass was observed but was soon followed by slow regrowth over the years. At admission, he had no difficulty breathing despite the enlarged tongue. The mass was diagnosed as microcystic lymphatic malformation, and OK-432 was injected in an attempt to shrink the mass. Improvement was noted by a decrease in the size of the mass and an increase in the mobility of the tongue. However, the improved state was brief; it was followed by partial recurrence but not to the extent of the previous state. Over the 2-year period, the patient was treated with OK-432, and 15 injections were made, achieving a steady decrease in size. After the last sclerotherapy, the tongue relapsed into a state similar to the initial size before sclerotherapy, and the patient consulted our department for a resection (Fig. 1). In the magnetic resonance imaging finding, the mass had irregular margin, and intralesional hemorrhage was observed (Fig. 2). Tracheostomy was performed to maintain airway, and surgical resection was made involving the central and anterior wedges of the tongue. The patient was cared in the intensive care unit, with careful monitoring of vital signs overnight. Postoperative course remained uncomplicated, and L-tube feeding was begun after 2 days of parenteral nutrition. However, on the fifth day after surgery, he experienced respiratory arrest due to blockage of the tracheostomy site. After prompt change of the tracheostomy cannula and oxygen supplement, the vital signs were stabilized. The T-cannula was removed after ensuring the patent airway, and the patient was discharged. During the 9-month follow-up, the lymphatic malformation of the tongue showed no signs of recurrence.

DISCUSSION

The lymphatic malformation is a congenital abnormality of the lymphatic system. It can be morphologically classified into 3 groups: the macrocystic lymphatic malformation, tissue-infiltrating microcystic lymphangioma that leads to the formation of an infiltrated layer with scattered clear or hematic vesicles at the surface, and the mixed type frequently occurring in facial or buccal regions. Most lesions are either present at birth or detected before 2 years of age. The surgery has been the mainstream treatment of most lymphangiomas but with various success rates. Since the introduction of sclerotherapy, especially OK-432, it is now widely accepted as primary approach to lymphatic malformations. It is much easier than surgery, leaves no scar, and has minimal risk of nerve damage.

There are many reports regarding the success of OK-432 injection sclerotherapy for the treatment of lymphangiomas in children.[1,3,4–7] Some may recommend that use should be limited to larger cysts, and there is an overall consensus for less favorable outcomes in microcystic and mixed cases.[6,7] However, a report by Luzzatto et al[4] notes that about half of his cases with microcystic or mixed type shows good response to OK-432 sclerotherapy and recommends its use as a primary approach to all lymphatic malformations. In our report, there was a definite response to OK-432 despite of a recurrence after 2 early operations. Once there is failure to respond after 3 injections, it has been shown that there is no benefit in insisting sclerotherapy treatment.[4] In our report, what made the decision to proceed with the difficult surgery was the fact that responses have been shown after 1 or 2 injections despite being a microcystic type. Surgery also poses the risk of having nerve damage, scarring, and recurring after surgery. The local recurrences are common when lymphangiomas are partially resected, as seen in

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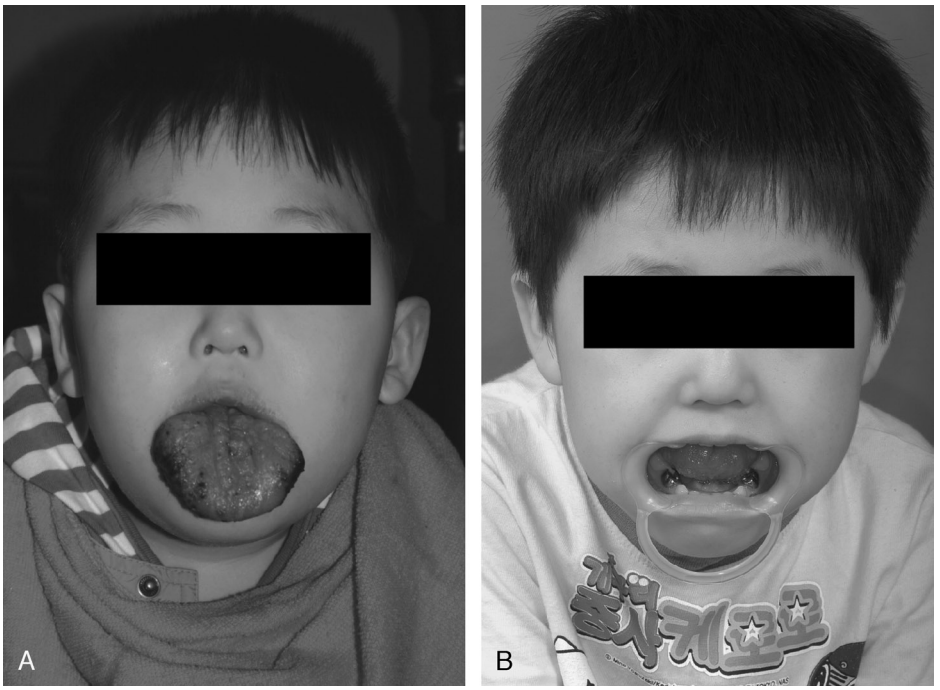

FIGURE 1. A 6-year-old boy with extensive lymphatic malformation of the tongue. A, Sudden enlargement was noted after the last injection of the 15 attempts with OK-432 (Picibanil) sclerotherapy. B, At 9 months after central-anterior resection.

our report. Adequate excision is very difficult and frequently unfeasible in the head and neck. Another reason for reluctance in making the surgical approach was the extensive involvement of the tongue, leaving only partial resection as choice.

Lymphatic infiltration of the tongue presents difficult problems. It results in macroglossia, which may impair speech and eating or may be complicated by swelling or bleeding. In addition, the enlarged tongue may lead to complications such as excessive salivation, halitosis, lingual protrusion, and numerous dental cavities. Further enlargement of the tongue could also lead to upper airway obstruction, dysphagia, and cosmetic deformity. In addition, the excessively large tongue makes open bite and dehydrated tongue prone to injury and bleeding. In our report, sudden enlargement of the tongue after the last injection revealed a small pocket of internal bleeding of the lesion with inflammation leading to obstruction of the airway. A report by Hall et al[8] notes that intralesional injection of lymphangioma surrounding the airway may be hazardous because of the increase in size of lesion after injection. However, we cannot conclude whether this response was from a swelling after OK-432, a complication after injection, an infection, or a combination of the aforementioned. Clearly, the enlarged tongue was indicated for surgery. Greinwald et al[5] reported that the number of treatments, total dose for sclerotherapy, appears to have no influence on the response to surgery and does not hinder the surgery.

When operating on the tongue, the surgeon must consider the possible involvement of neural and vascular structures and try to

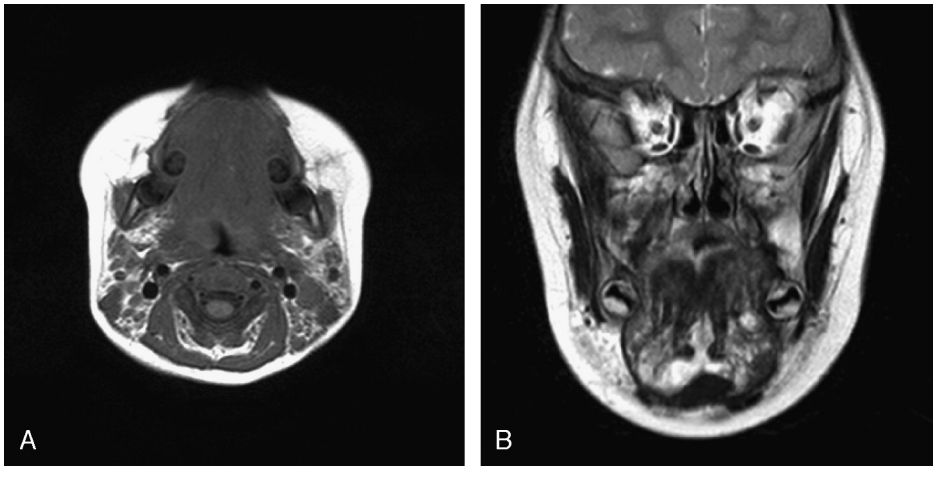

FIGURE 2. Magnetic resonance imaging scans revealing the oral cavity full of the giant lymphangioma and the mass having irregular margin with intralesional hemorrhage.

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limit blood loss. Many surgical incisions have been proposed to accomplish tongue reduction.[9,10] The tongue base neurovascular bundle maintains a close relationship to the superior aspect of the hyoid bone and lies midway between the tongue midline and lateral tongue and inner mandible margin.[9] Anterior wedge resection and central resection can be safe because of the anatomic location of neurovascular bundle and will allow further resection if the lymphangioma recurs. The central-anterior resection is cosmetically feasible, achieving both narrowing and shortening of the tongue.

As seen in our report, the maintenance of airway is critical in any surgery involving the structure along the airway. The possibility of airway obstruction requires preventive tracheostomy and preservation in a prompt manner.

The lymphatic malformation involving the tongue presents multiple challenges. Efforts should be made to maintain airway while under treatment, and functional and cosmetic outcomes should be considered. Despite the advantages of OK-432 as first-line treatment, patients with microcystic or mixed-type lymphangioma of the tongue should not undergo sclerotherapy because of lack of predictability and because of the risk of swelling after injection. Furthermore, as seen in our report, the response can be slow and may increase the risk of sclerotherapy complications.

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Carpenter Syndrome

Pip Hidestrand, MD, Henry Vasconez, MD,† Carol Cottrill, MD‡*

Abstract: Carpenter syndrome is a rare autosomal recessive disorder that belongs to a group known as Acrocephalopolysyndactyly syndromes. Carpenter syndrome is one of the rarest in the group. There are 2 common features in all of these syndromes: Craniosynostosis, early fusion of different sutures with skull base abnormalities and musculoskeletal abnormalities, primarily in the hands and feet. Clinical features of Carpenter syndrome include peculiar faces, asymmetry of the skull, brachy-polysndactyly, obesity, hypogenitalism, congenital heart disease, and variable psychomotor delay. The abnormal head shape is caused by early fusion of some or all of in the metopic, coronal, sagittal, and lamboidal sutures. Most of the affected patients have a surgical procedure between 3 and 9 months of age to open the cranial vault to make space for the brain to grow. We present a patient with Carpenter syndrome who is unusual in that she is an adult who has never had any surgical intervention.

Key Words: Carpenter syndrome, craniofacial surgery, acrocephalopolysyndactyly, craniosynostosis

Since the advent of surgery for craniosynostosis, the lives of these children have been enhanced. Craniosynostosis has varying etiologies, mostly genetic. The influence of the defective genes is seen at different times during embryogenesis, and this timing is syndrome specific. The defective gene that produces characteristics of Apert syndrome expresses between the 30th and 32nd day of gestation.[1] This defective gene encodes a fibroblast growth factor receptor, which is thought to regulate sutural fusion.[2] The defective gene is also involved in cranial base growth abnormalities.[3] On the other hand, Laurence-Moon Biedel-Bardet results from a defective gene that expresses later (between the 42nd and 49th day). Although the genetic etiology is not well known for Carpenter syndrome, it is believed to be a result of a defective gene that expresses somewhere between the 30th and 49th day, as Carpenter syndrome has features of both these syndromes.[1] This report documents the natural history of a child without surgery for craniosynostosis.

CLINICAL REPORT

A 26-year-old woman was diagnosed with Carpenter syndrome at 3 months of age. The patient weighed 11 lb at birth (>95th percentile), whereas head circumference was less than the fifth percentile. She was scheduled to have cranial surgery but upon induction of anesthesia experienced a cardiac arrest. Resuscitation was successful, but surgery was cancelled, and she recovered in the intensive care unit. Her mother refused any further surgery because of the complication occurring with the first attempt. She has always shown developmental delay. At 2 years of age, she could pull herself up and stand with support. She could say several words at age 2 years. At age 7 years, she was placed in a special educational program. By 10 years of age, she was able to feed herself and go to the bathroom by herself. At 19 years of age, she was released from special aid high school. She is dependent on adult help and during the day attends an adult day-care program. Her activities are limited because of her blindness and at home can only answer the telephone and take messages.

From the *Department of Pediatrics, †Division of Plastic and Reconstructive Surgery, and ‡Division of Community Pediatric Cardiology, University of Kentucky, Lexington, KY.

Address correspondence and reprint requests to Pip Hidestrand MD, Department of Pediatrics, University of Kentucky, 1760 Nicholasville Rd, Ste #602, Lexington, KY 40503; E-mail: pip.huang@uky.edu

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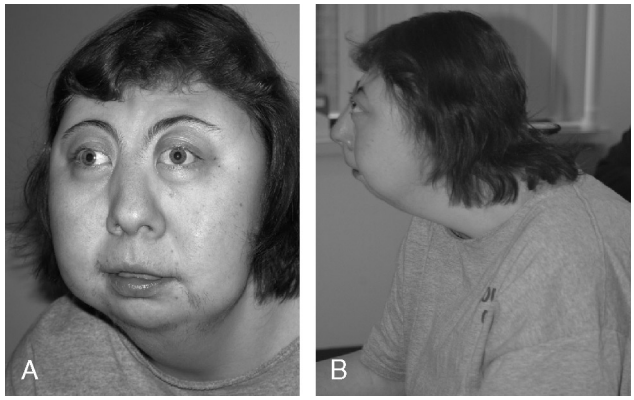

FIGURE 1. Anteroposterior (A) and lateral (B) views of the face. A, She has brachycephaly and shallow orbits with protrusion of the globes of her eyes. Note hair on her upper lip and chin. B, Brachycephaly with marked craniofacial disproportion. Kyphosis is evident by the position of her head.

During early childhood, growth was slow (weight was at the 50th percentile at age 12 years). Rapid weight gain then commenced, and by 17 years of age, weight was in the 90th percentile (height was in the 75th percentile). As an adult, she is 65 inches tall and 200 lb (body mass index is 33.3 kg/m^2). Her occipital frontal circumference at age 2 years (42.75 inches, <5th percentile) has not changed appreciably over many years. She has typical facies for Carpenter syndrome (Fig. 1). She has ear pits and a high-arched palate. She has brachysyndactyly of her fingers and toes, with post-axial polydactyly of her toes (Fig. 2). She also has a severe pectus carinatum deformity.

At age 6 years, because of congestive heart failure, secondary to a large ventricular septal defect associated with a bicuspid aortic valve and mild aortic stenosis, digoxin and furosemide (Lasix) were started, with potassium added later. Cardiac catheterization/surgery was refused. Intermittent abdominal distension continued into adulthood. She developed Eisenmenger physiology and is now inoperable from a cardiac standpoint.

Progressive loss of vision secondary to shallow orbits and optic nerve compression continues, and cranial surgery was again suggested, but the mother declined. After an inability to negotiate obstacles and find doorways, ophthalmologic examination indicated a high degree of hyperopia. A prescription for glasses was given, but the patient had difficulty wearing them as it was hard to get them fit to the shape of her head. Optic nerve atrophy eventually led to complete blindness. There was no evidence for glaucoma. Chronic epiphora, due to abnormal naso-lacrimal anatomy, prevented normal drainage of tears into the nose. A dacryocystorhinostomy was declined. Many sinus infections developed with nosebleeds, and she was hospitalized for recurrent bouts of pneumonia.

Marked kyphosis has been present since 13 years of age. Dental problems arose in her teens due to abnormal maxillo-mandibular growth. No dental work has been done.

She had breast development bilaterally, and menarche occurred at 14 years. She has irregular cycles, experiencing only 1 to 2 menstrual cycles per year. At age 19 years, she became very hirsute on her chest, abdomen, and legs.

DISCUSSION

Carpenter syndrome is a form of Acrocephalopolysnyndactyly Type II. In 1993, there were 45 patients documented since Carpenter[4] reported a family with these characteristics in 1909.[5] We are unaware of any patients in adulthood who have not had surgery. Patients with craniosynostosis usually undergo surgical release of the fused sutures between 3 and 9 months of age as the intracranial volume rapidly increasing due to brain growth.[6] Due to the rarity of this disease and the frequent repair of craniosynostosis early in childhood, little is known about the prognosis of syndromic patients without craniofacial correction.

With early fusion of the sutures, many have postulated that brain growth is inhibited, causing impaired cognitive and neurodevelopmental function. Cerebral spinal fluid resorption can be impaired, leading to increased intracranial pressure. Surgical treatment can prevent hydrocephalus and premature death.[7] Craniofacial reconstruction can improve facial features so that patients do not develop poor self-esteem and the psychological and behavioral problems associated with it.[8] It was previously thought that surgery would allow for normal cranial growth and development, thus preventing mental retardation,[7] but studies have shown that some patients with either nonsyndromic or syndromic craniosynostosis may continue to have cognitive, psychological, speech, and behavioral problems.[9] It is unknown what effect surgery could have achieved in our patient with regard to neurodevelopment.

Although brain structures can compress vascular supply to the optic nerve, as well as the nerve itself, leading to degradation of the nerves resulting in permanent vision loss, other causes of vision loss are well known. Apert and Crouzon syndromes are also subject to visual impairment, which may be due to increased intracranial pressure,[10] amblyopia, horizontal strabismus, optic atrophy, corneal damage, and others. Astigmatism, seen in these patients, is most likely a result of corneal distortion, ptosis, and the shape of the orbits.[11] Surgical treatment can preserve vision.[10,12]

Craniofacial reconstruction would allow for maxillary and mandibular growth. Patients with craniosynostosis have smaller

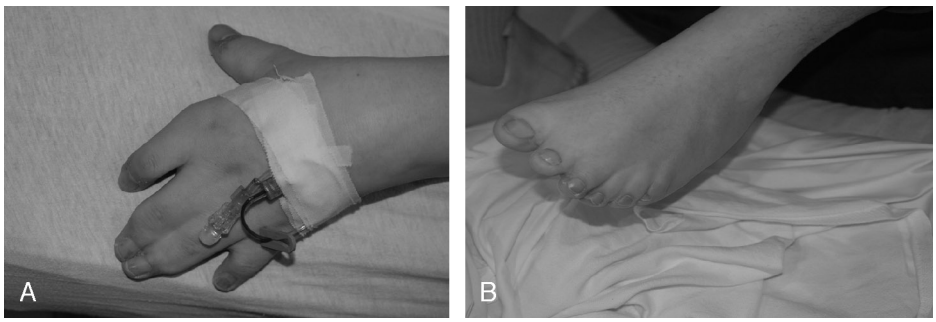

FIGURE 2. Hands (A) and feet (B). A, Short finger length and syndactyly of the second to fourth fingers. B, Post-axial polysyndactyly.

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maxillary volumes at birth, but tend to normalize after surgery. The smaller maxillary size affects the anterior and posterior superior dental nerves.[13] Thus, had our patient had craniofacial surgery, her dental complications may have been minimized. With surgical correction, her other facial bones could have grown to allow proper expansion of her sinuses; thus, the sinus infections would not have been as severe or as frequent. With cardiac surgery to repair the ventricular septal defect, this patient would have avoided pulmonary hypertension and resultant Eisenmenger physiology, which is inoperable.

Two siblings with Carpenter syndrome and an empty sella turcica[14] had an increase in growth hormone when given an injection of both Thyrotropin-releasing hormone (TRH) and Luteinizing-releasing hormone (LRH); they also had an increase in prolactin when given LRH. Our patient's endocrine dysfunction could be explained by pituitary compression. Pituitary dysfunction could explain her menstrual irregularity and excessive weight gain, but excessive weight can be a clinical feature of Carpenter syndrome itself. Another explanation for her irregular menarche could be from her obesity causing polycystic ovarian disease. This would lead to excess androgen secretion and, in turn, result in hirsutism and irregular menses. To determine the exact reason, more studies including hormone levels, imaging studies of her brain, and pituitary gland would have to be performed.

It is very important to diagnose Carpenter and other craniosynostosis syndromes early to allow for optimal surgical treatment. Diagnosis of craniosynostosis has been based on a combination of physical examination and confirmation with radiographic studies including computed tomographic scan and conventional radiographs.[6] Advances in molecular genetics are permitting the identification of defective genes and their pathways in describing these congenital anomalies. Recent studies have demonstrated mutations in RAB23,[15] a negative regulator of hedgehog signaling, associated with Carpenter syndrome. The variable phenotypic presentation of craniofacial syndromes such as Carpenter,[4] make genetic testing, keen clinical observation and physician observation very important. In this way early surgical intervention can be recommended in order to try and prevent the ravages of ongoing disease.

CONCLUSION

Many of the complications seen in this patient probably could have been avoided. This young lady graphically illustrates the natural history of Carpenter syndrome, and her story speaks to the necessity for early accurate diagnosis and appropriate surgery for this and all of the craniosynostosis syndromes.

Addendum: She recently passed away suddenly at the completion of this manuscript at the beginning of October 2007. She was 26 years of age when she died.

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Frontal Bandeau Reconstruction With a Fibula Flap in a Patient With Freeman-Sheldon Syndrome

Rachel Bluebond-Langner, MD,† Amir Zamani, BS,* Eduardo D. Rodriguez, DDS, MD*†*

Abstract: A 29-year-old woman with Freeman-Sheldon syndrome had a history of recurrent frontal sinus infections for which she underwent a 1-stage frontal sinus obliteration and cranioplasty using a free fibula osteocutaneous flap. This case is unique in that a free fibula flap had never been used to obliterate the frontal sinus in a patient with Freeman-Sheldon syndrome, nor had it been harvested from a limb with a clubfoot.

Key Words: Frontal bandeau, free fibula flap, Freeman-Sheldon syndrome

From the Johns Hopkins University School of Medicine and Division of Plastic, Reconstructive and Maxillofacial Surgery, R. Adams Cowley Shock Trauma Center, Baltimore, Maryland.
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Address correspondence and reprint requests to Eduardo D. Rodriguez, DDS, MD, Plastic and Reconstructive Surgery, University of Maryland School of Medicine and Johns Hopkins Schoolof Medicine, 22 South Greene St, Baltimore, Maryland 21201; E-mail: erodriguez@umm.edu
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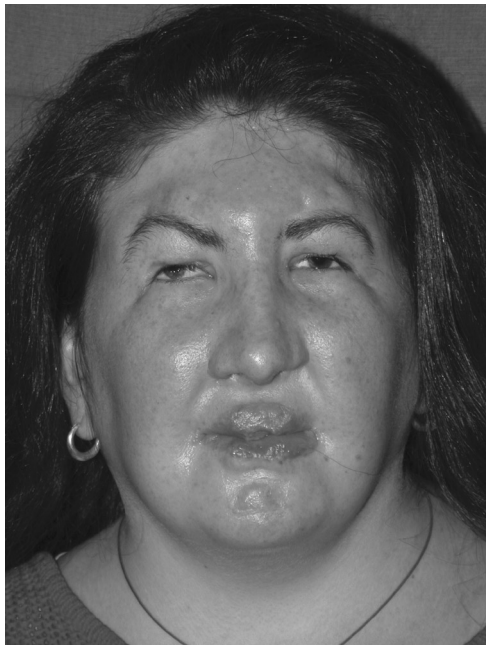

FIGURE 1. Preoperative clinical photograph.

Freeman-Sheldon syndrome (FSS) is characterized by both limb and facial abnormalities, including camptodactyly, talipes equinovarus, microstomia, microglossia, a short nose, long philtrum, sunken eyes, prominent supraorbital ridge, mask-like facies, and H-shaped cutaneous dimpling of the chin.[1–3]

Frontal headaches are a known clinical manifestation of FSS, with 36% of patients reporting frontal headaches in 1 series.[3] However, the association on these headaches with frontal sinus infection or osteomyelitis in this population has not been reported. The treatment of persistent frontal sinus infection includes debridement followed by obliteration or ablation. Both autologous tissue and alloplastic materials have been used to obliterate the sinus, but autologous tissue, particularly cancellous bone, and local flaps are preferred. We present the unique case of a woman with FSS and recurrent frontal sinus infections treated with a free fibula osteocutaneous flap from a clubfoot.

CLINICAL REPORT

This 29-year-old woman with FSS had undergone multiple facial reconstructive procedures (Fig. 1). Most notably was a chronic history of headaches associated with frontal sinusitis and osteomyelitis. Despite attempts at debridement and obliteration of the frontal sinus using cancellous bone, she had persistent pain, swelling, and drainage. She was subsequently referred for definitive treatment. The frontal sinus was sharply debrided, all previously placed cancellous bone was removed, a galeal flap was rotated for temporary coverage, and antibiotics were intravenously administered to her for 1 week. After resolution of the infection, a free fibula osteoseptocutaneous flap was harvested. The surgical dissection was challenging because the anatomic planes were fibrotic with ill-defined musculature and neurovascular pedicles as well as narrowing of the interosseous space between the tibia-fibula. The bone measured 7 cm, and the skin paddle 15 × 7 cm (Fig. 2). The fibula was contoured with 1 central osteotomy. A portion of the flexor hallucis longus muscle along with the subcutaneous tissue of the skin paddle was used to obliterate the frontal sinus and nasofrontal ducts. An end-to-end microvascular anastomosis was performed to the right superficial temporal vessels. At 8 months, there is no evidence of infection, the donor site is healed, and range of motion at the ankle and knee and her gait have returned to baseline.

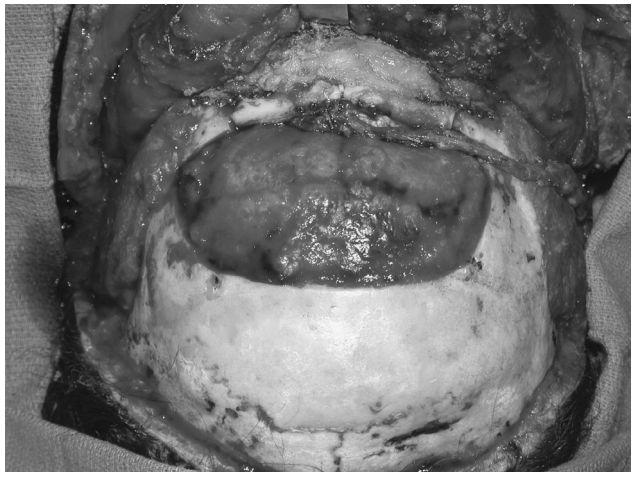

FIGURE 2. Intraoperative photograph depicting the fibula inset as the supraorbital bar, the skin paddle de-epithelialized and used to obliterate the frontal sinus.

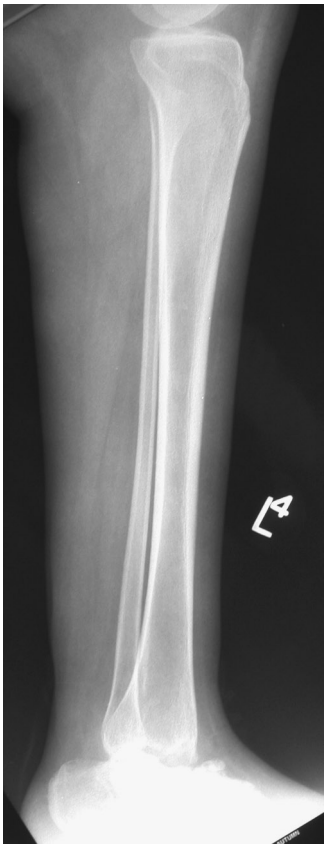

FIGURE 3. Preoperative lateral radiograph of the leg.

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DISCUSSION

Successful frontal sinus obliteration depends on complete removal of sinus mucosa, obliteration of the sinus, and occlusion of the nasofrontal duct. The ideal material for obliteration continues to be debated. Autologous fat and cancellous bone are the most commonly used graft materials; however, both are associated with mucocele formation and significant resorption.[4,5] In addition, nonvascularized grafts are not ideal in an infected area.

The free fibula osteocutaneous flap provides both soft tissue to obliterate the sinus and bone to reconstruct the frontal bar. Because it is vascularized, it is a good option for an infected sinus, and its inherent length allows for simultaneous reconstruction of craniofacial defects, which in our case included reconstruction of the right supraorbital rim. Thus, the benefits of simultaneous craniofacial reconstruction must be weighed against the drawbacks of donor-site morbidity, particularly in a patient with clubfeet.

This is the first report of frontal sinus obliteration with a fibula free flap in a patient with FSS (Figs. 3, 4). Harvest of a free fibula flap in a patient with a clubfoot has not been previously described and presents unique challenges. Preoperative evaluation revealed palpable dorsalis pedis and posterior tibial pulses, obviating the need for an angiogram. The anatomic planes were fibrotic with ill-defined posterior and lateral compartment musculature, anomalous peroneal muscle tendon insertions, and narrowing of the interosseous space between the tibia and fibula. The neurovascular pedicles were therefore difficult to identify. However, sizable skin perforators were visualized, and tibial vessels (anterior and posterior) were identified and protected before division of the peroneal vessels.

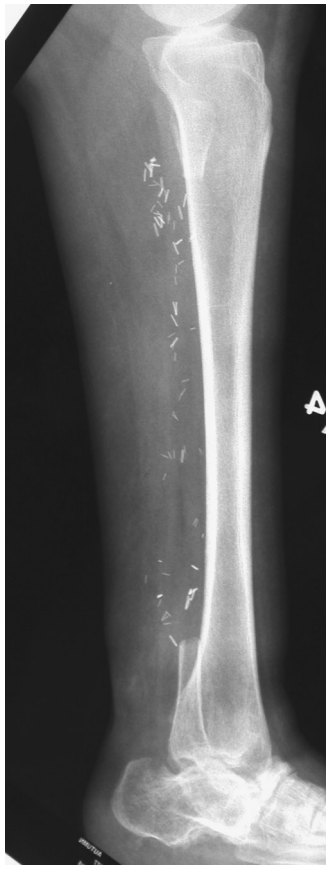

FIGURE 4. Postoperative radiograph of leg after fibula flap harvest.

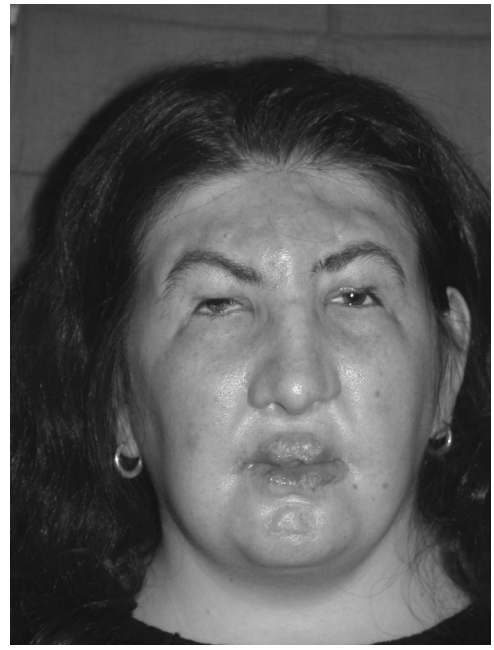

FIGURE 5. Postoperative clinical photograph.

Eight months postoperatively, the patient has not had recurrence of her frontal sinus infections (Fig. 5). In addition, this is the first report of a fibula free flap harvest from a clubfoot. Thus far, donor-site morbidity has been minimal, and we will continue to follow the patient closely in the coming years. The pros and cons of the various options for frontal sinus obliteration were carefully weighed, and we believe that a free fibula osteocutaneous flap has a role when simultaneous craniofacial reconstruction is needed.

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Combined Treatment Protocol for Temporomandibular Joint Ankylosis: An Observation on a Clinical Problem

*Cornelia Katharina Mueller, DMS, MS,**
Michael Wolf Thorwarth, MD, DMD, Andrea Mueller, DMD,†*
*Stefan Schultze Mosgau, MD, DMD, PhD**

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Abstract: Various techniques have been defined for the treatment of temporomandibular joint ankylosis. However, in some cases, they are unsuccessful, resulting in continuing pain and limitation in interincisal distance after surgery. This report describes the case of a 32-year-old woman who has been experiencing temporomandibular joint ankylosis for a period of 8 years. Several surgical procedures failed. A treatment approach combining auricular cartilage interposition arthroplasty with postsurgical functional treatment using a spring activator is presented. Using this approach, pain settled and maximal interincisal distance was raised from 22 to 35 mm after 4 months and remained stable for further 10 months.

Key Words: Ankylosis, arthroplasty, auricular cartilage, spring activator

Choice of surgical technique and postoperative functional exercises is of utmost importance to ensure therapeutic success in the treatment of temporomandibular joint (TMJ) ankylosis.

Interposition arthroplasty seems to be superior to operation without insertion of materials separating the articular surfaces. These spacers not only reduce the risk of postoperative ankylosis[1] but also prevent the articular surfaces from degenerative changes.[2,3] Alloplastic[4] and autogenous[5,6] materials have been introduced for interposition until now. Auricular cartilage is a readily available tissue adjacent to the operative field. The low donor site morbidity and natural graft shape, which corresponds well to the joint surface, contribute to its excellent suitability.[7,8] Furthermore, the grafted tissue seems to remain viable after implantation.[9] Common postoperative treatment strategies aim at the enhancement of neuromuscular activity[10] and vertical distraction of the ramus from the glenoid fossa.[11]

PATIENT

A 32-year-old woman presented with recurrent pain and swelling of the right TMJ. She had already undergone interposition arthroplasty using the temporalis fascia flap and a Silastic sheet (Dow Corning, Midland, MI), which had to be removed.

The patient reported a severe, constant, highly limiting pain. Joint palpation was painful as well. A crepitation of the right joint could be recognized. Furthermore, a maximal interincisal distance of 17 mm with a deflection to the right was observed (Fig. 1). Computed tomography showed that the right condyle was in a severe postinflammatory state of defect with deformation and sclerosis. Fibrous adhesion between the condyle and the glenoid fossa could be visualized by magnetic resonance imaging.

From the *Department of Oral- and Cranio-Maxillofacial Surgery/Plastic Surgery, University of Jena; and †KFO-Suhl, Private Practice of Orthodontics, Suhl, Germany.
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Address correspondence and reprint requests to Cornelia Katharina Mueller, DMS, MS, Department of Oral- and Cranio-Maxillofacial Surgery/Plastic Surgery, Friedrich-Schiller-University, Erlanger Allee 101, 07747 Jena, Germany; E-mail: cornelia.mueller1@med.uni-jena.de
Dr Andrea Mueller is engaged in the private practice of orthodontics in Suhl, Germany.
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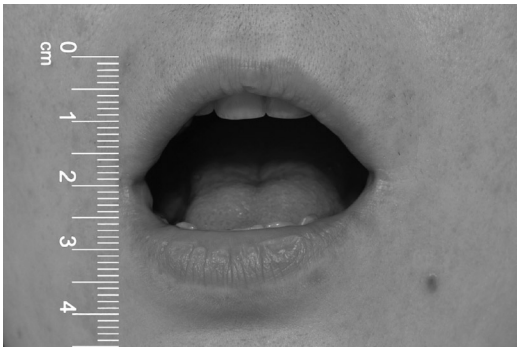

FIGURE 1. Maximal interincisal distance of 17 mm with deflection to the right measured before interdisciplinary treatment.

Open joint surgery was performed under general anesthesia. At the beginning, a cartilage graft was harvested from the ear using a retroauricular approach. It was elevated with perichondrium attached to its convex site, measuring 2 × 1.5 cm^2. The perichondrium covering the concave site was left in place to ensure the regenerative process at the donor site. Wound closure was performed in 1 layer.

A modified preauricular approach gave access to the joint. Subsequently, the present fibrotic mass was removed. At that point, an interincisal distance of 35 mm was noted. Condylar shaving was carried out to remove sclerotic bone substance.

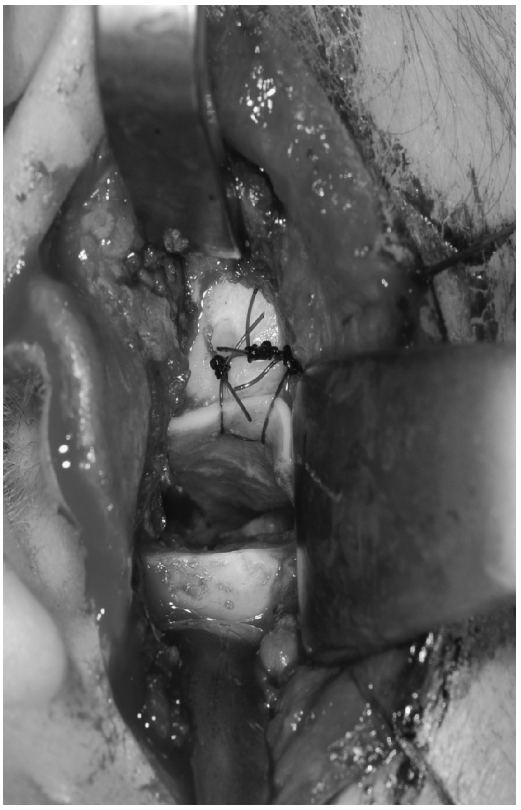

FIGURE 2. Intraoperative picture showing the auricular cartilage placed on the condyle secured with 3–0 sutures.

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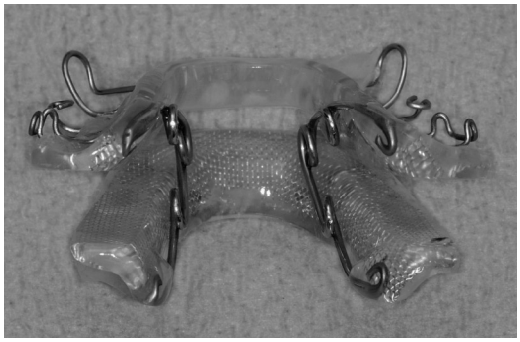

FIGURE 3. Dorsal aspect of the spring activator used for the treatment of that patient.

The previously harvested auricular cartilage graft was placed in the appropriate position between condyle and fossa, with the perichondral site facing the fossa. It was secured in that position using 3-0 slow resorbing sutures (Fig. 2). The temporoparietal fascial flap was finally used to form a new capsule. In the end, the wound was closed in 2 layers, and a suction drain was left in the joint for 1 day.

One week after surgery, a spring activator (Fig. 3) was incorporated and applied as often as possible, especially during day time. The springs were reactivated every 4 weeks to a point where the patient noted a distracting force in the joint.

Four months after surgery, the patient was pain-free, and joint palpation was without any pathologic reaction. No crepitation was detected anymore, and interincisal distance improved to 35 mm (Fig. 4). This situation remained stable until now (10 months of follow-up).

DISCUSSION/CONCLUSION

Sandler et al[12] described auricular cartilage as an interpositional material that generally achieves favorable results. However, in difficult cases of previously operated joints or joints with significant inflammatory destruction, they suggested the use of another interpositional material.

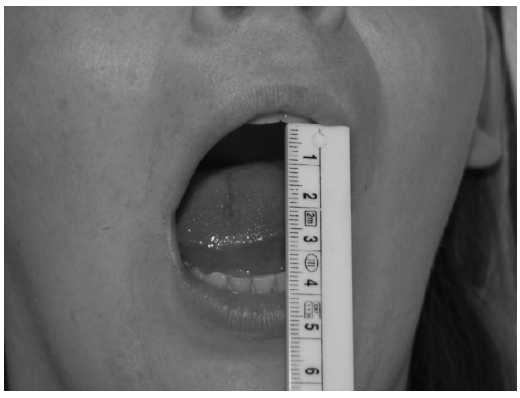

FIGURE 4. Maximal interincisal distance of 35 mm with slight deflection to the right after operation and 3 months of functional treatment.

The patient presented in this report was multiply operated, and the joint was in a postinflammatory state. Nevertheless, short-term follow-up reveals significant improvement regarding pain and mouth opening.

Chidzonga[13] postulated that the main cause of complications is failure to carry-out postoperative treatment. Therefore, postoperative exercises play a crucial role in ensuring lasting success.[14] In our case, the spring activator[15] has been proven to be an alternative to the standard treatment procedures that is especially well working in risk patients.

Elasticity and arrangement of the springs in the spring activator lead not only to the distraction between condyle and fossa as reported for other techniques but also to an increase in neuromuscular activity.[16]

Consequently, this treatment protocol is worthy to be validated in future studies using a larger amount of patients.

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Multiple Fractures Involving the Orbit and Incidental Finding of Large Fourth Ventricular Epidermoid

Amit Agrawal, MCh, Rajeev M. Borle, MDS,†*
Nitin Bhola, MDS,† Akshay Daga, MDS,† Smriti Bora, BDS,† and Sachin Sachdeva, MDS†

Abstract: Epidermoid tumors located in the fourth ventricle are exceedingly rare, and the patients usually present with headaches, vertigo, and/or disequilibrium. We discuss the management of orbital floor and lateral wall fracture in a patient who was also incidentally diagnosed to have a large fourth ventricular epidermoid cyst. Although because of their availability and ease of use many allografts have been described, when available, the use of similar membranous bone of equivalent thickness makes the bone graft ideally suited for reconstruction of the orbital floor.

Key Words: Epidermoid tumor, fourth ventricle, orbit, orbital floor, fracture

We discuss the management of orbital floor and lateral wall fracture in a patient who was also incidentally diagnosed to have a large fourth ventricular epidermoid cyst.

PATIENT

A 26-year-old man presented with history of fall from a tree. He had a transient loss of consciousness. There was no history of vomiting, ear or nasal bleeding, or convulsion. He was well aware of the episode. No abnormalities were seen in his general and systemic examination. On examination, he sustained injury to the right eye and palpable deformity of right inferior orbital rim and lateral wall that was associated with overlying lacerated wounds. Radiographic paranasal sinus view showed fracture of the right orbital lateral and inferior rim as well as the floor (Fig. 1). Computed tomography scan of the brain in addition to the orbital fractures showed large hypodense mass and abnormal dilatation of the fourth ventricle without hydrocephalus. All lacerated wounds were sutured primarily. Further investigations using magnetic resonance imaging (MRI) revealed a large mass occupying the whole of the fourth ventricle, which appeared as a homogeneous signal area similar to cerebrospinal fluid (CSF) on both T1- and T2-weighted images, without enhancement after contrast administration. The tumor was hypointense on fluid attenuation inversion recovery (FLAIR) images. Despite the enormous tumor occupying the fourth ventricle, there were no findings of hydrocephalus (Fig. 2). Magnetic resonance imaging also showed herniation of the orbital contents into the maxillary sinus on the right side (Fig. 1, right arrow). A diagnosis of fourth ventricular epidermoid with orbital floor and lateral wall injury was made. The patient was planned for surgical resection of the tumor and at the same time repair of the orbital floor and lateral wall of the orbit. First, he underwent midline suboccipital craniotomy with a C1 laminectomy. Immediately after opening the dura mater just beneath the arachnoid membrane, a "pearly" tumor which filled the fourth ventricle and cisterna magna was identified (Fig. 3). The tumor was removed piecemeal, and then the capsule containing some fragments of the tumor, which had firmly adhered to the floor of the fourth ventricle especially in the portion of the obex, was excised. After closing the occipital wound, the patient was put in supine position, and orbital fractures were exposed (Figs. 4 and 5). Plating for lateral wall fracture was performed. The calvarial graft harvested from the outer table of the occipital bone at the time of occipital craniotomy was used to repair the orbital floor defect (Fig. 5). Postoperative outcome was excellent. He was discharged on the 10th postoperative day without neurologic sequelae and doing well at follow-up.

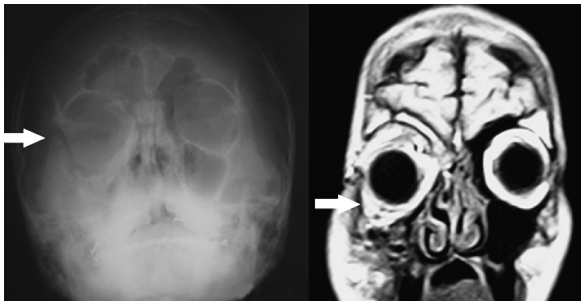

FIGURE 1. Radiographic paranasal sinus view shows fracture of the right orbital lateral and inferior rim as well as floor (left); MRI showing herniation of the orbital contents into the maxillary sinus (right arrow).

DISCUSSION

Epidermoid tumors located in the fourth ventricle are exceedingly rare and account for between 5% and 31.4% of all intracranial epidermoid tumors.[1–5] Patients with epidermoid cyst

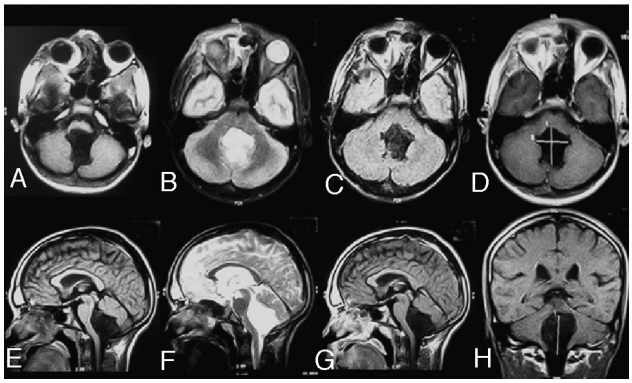

FIGURE 2. MRI showing the details of tumor extension: (A) T1 axial, (B) T2 axial, (C) FLAIR axial, (D) postcontrast axial, pboard2 (E) T1 sagittal, (F) T2 sagittal, (G) postcontrast sagittal, (H) postcontrast coronal.

From the *Department of Surgery, Datta Meghe Institute of Medical Sciences, and †Department of Oral and Maxillofacial Surgery, Sharad Pawar Dental College, Wardha, India.
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Address correspondence and reprint requests to Amit Agrawal, MCh, Division of Neurosurgery, Datta Meghe Institute of Medical Sciences, Sawangi (Meghe), Wardha- 442004, Maharashtra, India; E-mail: dramitagrawal@gmail.com; dramit_in@yahoo.com
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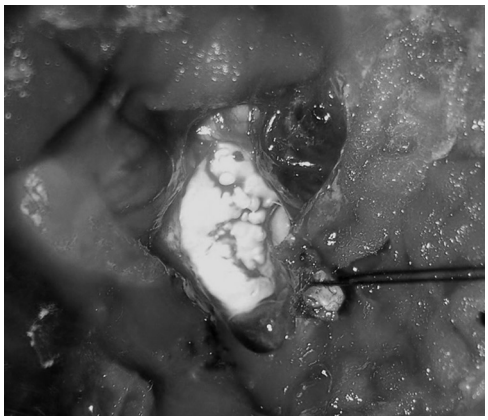

FIGURE 3. Intraoperative photograph showing pearly white tumor in the fourth ventricle.

of the fourth ventricle usually present with headaches, vertigo, and/or disequilibrium.[1–5] As in the present case, increased intracranial pressure and hydrocephalus are rare even when the tumors are large in size and filled up the fourth ventricle.[1,2,5] Our case was unique because despite the massive size of the lesion, he was not symptomatic for the tumor, and these lesions could be diagnosed only after trauma.[6] Computed tomography scan is the main diagnostic technique for these lesions and typically shows a highly hypodense, round area within the fourth ventricle.[4] Epidermoid tumors have similar radiologic characteristics with arachnoid cysts on routine imaging techniques.[7] These cysts are characterized by a focal lesion that is nearly isodense to CSF in computed tomography scan. On MRI, epidermoids are nearly isointense to CSF on T1- and T2-weighted images. Magnetic resonance imaging using FLAIR and diffusion-weighted images, as well as three-dimensional constructive interference in steady state acquisitions, is useful to better characterize the lesions and their relation with the vermis, foramen magnum, and cerebellopontine angle cisterns. Diffusion-weighted images are also useful for postsurgical evaluation of residual tumors.[1,7–9] Although total removal of the neoplasm is the theoretical aim of operative treatment, this purpose may be harmful when the ventricular floor is involved by the capsule.[1,2,5] When total resection is not possible, subtotal surgical removal of the cysts produced excellent results in most cases.[1,2,4,5] In our patient, the presenting problem was multiple fractures involving right side. All these fractures needed reconstruction (large orbital floor defect [>1 cm^2], enophthalmos, significant hypoglobus) to restore the continuity of the orbital floor, provide support of orbital contents,

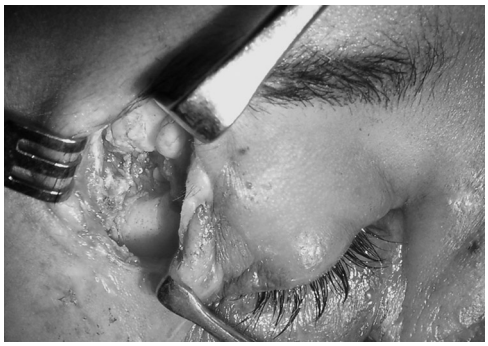

FIGURE 4. Intraoperative photograph showing the fracture of lateral orbital wall.

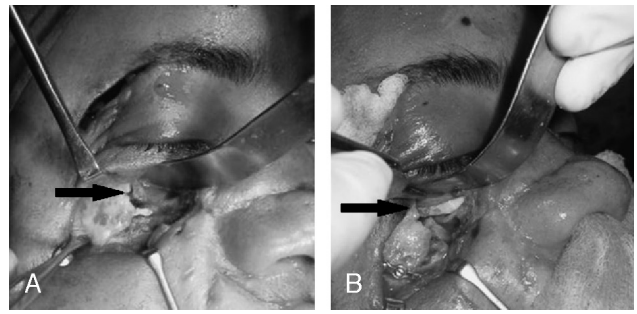

FIGURE 5. A, Intraoperative photograph showing fracture of the orbital floor and (B) note the positioning of split calvarial graft to reconstruct the floor (arrow).

prevent soft tissue fibrosis, and restore aesthetics.[10,11] As the fractures involved the anterior half of the orbit, anterior approach was used.[12] Although because of their availability and ease of use many allografts have been described, traditionally, autogenous grafts have been used as the material of choice.[10,12–14] Also, when available, the use of similar membranous bone of equivalent thickness makes the bone graft ideally suited for reconstruction of the orbital floor.[15,16] As the patient underwent occipital craniotomy at first instance, split calvarial graft harvested fulfilled all these criteria.

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Reconstruction of Complicated Scalp Defect Via Skin Traction

Gürsel Turgut, MD, Arzu Özcan, MD, Onur Sümer, MD, Nebil Yeşiloğlu, MD, Lütfü Baş, MD

Abstract: Management of an open wound is a problem frequently faced, with skin defects that cannot be closed primarily. Functional and aesthetic outcome could be improved if primary approximation of skin in these large defects were possible. Primary closure may be assisted using the viscoelastic properties of the skin. The viscoelastic properties of mechanical creep and stress relaxation in the skin were described more than 40 years ago. If skin is stretched with a constant force, it will expand with time as long as it is kept under tension, a phenomenon known as *mechanical creep*. In contrast, if the skin is stretched to a constant distance, it will expand and lead to a decrease in the force or tension on the skin with time, a phenomenon known as *stress relaxation*. We have recently applied these stretching properties to close the scalp because of a defect, which previously would have undergone pericranial flap and split-thickness graft. Because of the partial necrosis of the pericranial flap, skin grafting failed. In the second stage, we inserted only the wrist part of elastic latex gloves to the wound edges in full thickness. The skin margins were advanced slowly and gently.

Key Words: Mechanical creep, skin traction, stress relaxation

Management of an open wound is a problem frequently faced, with skin defects that cannot be closed primarily. Various techniques for closure of such large wounds have been developed, including wide undermining; tissue expanders; split skin grafts; and local, distant, or free revascularized flaps. However, functional and aesthetic outcome could be improved if primary approximation of skin in these large defects were possible. Primary closure may be assisted using the viscoelastic properties of the skin. The viscoelastic properties of mechanical creep and stress relaxation in the skin were described more than 40 years ago.[1,2] If skin is stretched with a constant force, it will expand with time as long as it is kept under tension, a phenomenon known as *mechanical creep*. In contrast, if the skin is stretched to a constant distance, it will expand and lead to a decrease in the force or tension on the skin with time, a phenomenon known as *stress relaxation*. We have recently applied these stretching properties to close the scalp because of a defect, which previously would have undergone pericranial flap and split-thickness graft. Because of the partial necrosis of the pericranial flap, skin grafting failed. In the second stage, we inserted only the wrist part of elastic latex gloves to wound edges in full thickness. The skin margins were advanced slowly and gently.

From the Department of Plastic and Reconstructive Surgery, Sisli Etfal Research and Educational Hospital, Istanbul, Turkey.
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Address correspondence and reprint requests to Arzu Özcan, Department of Plastic and Reconstructive Surgery, Sisli Etfal Research and Educational Hospital, Istanbul, Turkey; E-mail: ozcanarzu79@yahoo.com
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CLINICAL REPORT

A 2-year-old girl presented to the emergency department after a car accident wound on the temporofrontal region of her hair-bearing area in the scalp. Defect size was 4 × 4 cm in diameter. Radiographs did not reveal any frontal or temporal fractures. The patient was taken to the operating room and underwent debridement. Skin edges could not be approximated easily. First, we had tried to cover the defect with pericranial flap and split-thickness skin graft, but partial necrosis was seen in the pericranial flap, and grafting was failed. In the second procedure, we have planned to use skin stretching to close this wound. After the end of the 1 week that the elastic glove bands were applied to skin edges, the defect closed primarily. No suture detachment was observed during the follow-up. The patient was followed up for 1 year.

SURGICAL TECHNIQUE

Our technique requires only the wrist strip of elastic latex gloves. The elastic glove strips were provided from the wrist part of the glove, adapted to straight needle, and were inserted through the full thickness of the scalp like horizontal mattress suture technique at approximately 1 cm from the wound margins (Fig. 1). The skin margins were advanced gently without undermining. At the end of the operation, skin margin circulation was checked, and capiller refill was normal. These strips of gloves were kept in place for 1 week for approximation of the edges; we also did not perform more traction. In the follow-up period, any circulation compromise was not seen at the skin edges. After 1 week, the subcutaneous

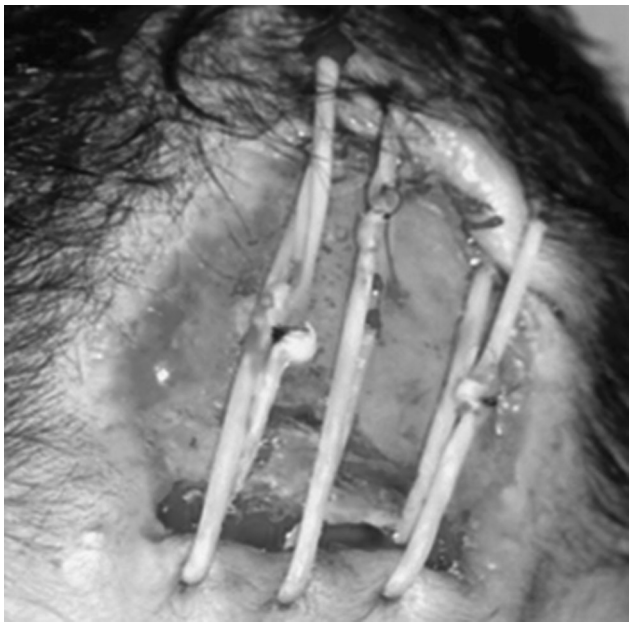

FIGURE 1. The elastic glove strips were inserted through the full thickness of the scalp like horizontal matress suture technique at approximately 1 cm away from the wound margins.

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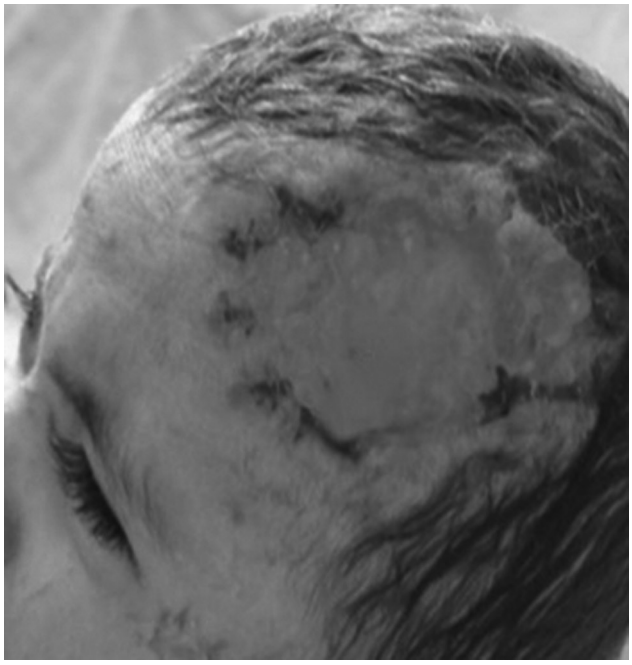

FIGURE 2. At the end of the one week, the defect closed the primarily.

tissues were loosely approximated with 4-0 vicryl sutures, and the skin was closed with 4-0 nylon sutures (Fig. 2).

DISCUSSION

One of the earliest reports on the use of a skin-stretching device for wound closure was published in a book about war injuries by Sir Harold Gillies[3] in 1920. This technique was not revisited until 1993 when Hisrhowitz et al[3] reported the use of the Sure-Closure skin-stretching device (The MedTech Group, Inc., South Plainfield, NJ) to achieve primary wound closure of surprisingly large defects with good aesthetic results.

Hirshowitz et al[3] described a technique that used 2 pins embedded intradermally and attached to a skin-stretching device.[4] This technique distributes tension more evenly along the wound edge. The investigators discovered that stretching occurred within 20 to 30 minutes when both subcutaneous and cutaneous tissues were normal. However, stretching was considerably slowed by chronic edema and fibrosis secondary to the pace at which collagen fibers realign.[3] The Sure-Closure skin-stretching system is relatively expensive ($450.00 per device), whereas gloves cost approximately $1.

Abramson et al[5] described an alternative to the Sure-Closure skin-stretching system in a different setting that mimics the technique presented here. The method used a rib approximator and 18-gauge spinal needles to allow for primary closure in large cutaneous wounds. The rib approximator was progressively tightened until the skin edges aligned, and vicryl sutures were used to close the wound. Abramson et al[5] placed the spinal needles 3 cm from the wound edge. There is no built-in monitoring system, transducer, force-limiting applicator, or other safety mechanism. Capillary refill does provide information about the blood supply, but it does not give any indication as to the architecture of the dermis itself.

Almekinders[6] described a technique in which staples were placed along the skin edges, and half of each staple was embedded in the skin, whereas the other half acted as an eyelet for a large nylon suture. The suture was tightened daily, and primary closure was obtained after 5 to 10 days. In our technique, which uses the principle of skin relaxation, we did not perform any tightening daily; however, we did not observe any circulation problem on the skin edges.

We thought that expander protheses would not be the ideal choice to cover bone exposed due to temporoparietal defect because of the our patient's young age. She was only 2 years old, and she would not bear the expander discomfort and pain. Also, expander protheses, which would be placed under the galea, would affect her skull and cranial developments. Actually, treatment with expander protheses is a multistage procedure, so it cannot be used for defects that should be treated in a short time, such as our bone-exposed scalp defect.

We did not plan to perform any transpositional, rotational scalp flaps to cover the defect because the defect was already large, 4×4 cm in diameters; also, the redundant scalp tissue would not able to cover defect without using skin graft for donor area.

Our technique described uses dermatotraction via inserted latex gloves bands to wound edges to stretch the skin in a cost-effective way to achieve primary closure of large wounds with supplies that are readily available in every operating room. However, functional and aesthetic outcome could be improved if primary approximation of skin in this large defect were possible. Skin stretching decreased the wound-closing tension and allowed primary closure of large defect. Our technique eliminated donor-site defects and related morbidity.

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