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CONTENTS

E D I T O R I A L

It is with great honor that I inform you that this issue of the *Brazilian Journal of Craniomaxillofacial Surgery* marks a special anniversary: the accomplishment of the first five years of publication of the Journal. Since 1998, the Journal has served as a forum for scientific debate in the field of craniofacial surgery.

All participants in a scientific community – authors, editors, counselors, and readers – are aware of the difficulties faced by new publications and of the importance of contributions. The Brazilian Society of Craniomaxillofacial Surgery has been able to overcome these challenges since 1998. Therefore, once more I would like to thank all who sent manuscripts for publication and helped in any other way along these five years.

Also, I would like to invite all of you to the VIII Brazilian Congress on Craniomaxillofacial Surgery, to be held in Rio de Janeiro, Brazil, from June 10 to 12, 2004. Meetings like this one foster scientific knowledge and are therefore of paramount importance for the development and establishment of the craniofacial specialty.

The present issue of the Journal brings some very relevant papers. In the first one, Alonso et al. present the results obtained with the use of rigid external distractors in two cases (Crouzon and Apert syndromes) undergoing distraction osteogenesis of the midface. The second paper, by Oliveira et al., describes two cases of amniotic band syndrome and the procedures used to treat the patients. The third paper, by Costa & Nunes, provides a thorough review of oral and maxillofacial prostheses and their indications. Then, Collares et al. offer a literature review and describe their experience with a case of basilar impression treated with transmaxillary approach. Finally, Sofia et al. describe a range of orbital complications that can result from mucoceles of the maxillary sinus, also based on the analysis of a case.

We hope you enjoy this issue of the Journal.

Marcus Vinicius Martins Collares, MD, PhD
Editor

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DISTRACTION OSTEOGENESIS OF THE MIDFACE WITH RIGID EXTERNAL DISTRACTORS: PRELIMINARY EXPERIENCE IN TWO CASES

Nivaldo Alonso, MD, PhD[1]; Dov Charles Goldenberg, MD, MSc[2]; Daniel Santos Corrêa Lima, MD[3]; Paulo Roberto Pelúcio Câmara, DDS[4]; Hamilton Matushita, MD, PhD[5]; Marcus Castro Ferreira, MD, PhD[6]

Distraction osteogenesis has been used to advance the midface of patients presenting syndromic craniosynostosis. In these cases, the resistance offered by the underlying soft tissues may hinder the stability of results, mainly when a major degree of advancement is necessary. Therefore, distraction is a perfect solution, since both bone and soft tissues may be lengthened gradually. The aim of the present study was to describe the preliminary experience and results obtained with the use of rigid external distraction in the treatment of severe hypoplasia of the midface in patients with syndromic craniosynostosis. Two patients (Crouzon and Apert syndromes) were assessed. Le Fort type III osteotomy and fronto-facial monobloc advancement were carried out, with gradual bone segment advance, using a rigid external distraction device. The Crouzon patient was submitted to Le Fort III; 15-mm monobloc advancement was achieved. In the Apert syndrome case, 12.5-mm advancement was achieved with fronto-facial monobloc osteotomy. There was no morbidity associated with the device or with the distraction process. Treatment objectives were achieved. However, the long-term results of this procedure are not established, and further research must be carried out.

KEY WORDS: *Osteogenesis, distraction; fronto-orbital advancement; midface.*

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Distraction osteogenesis has become a widely used procedure for the treatment of several cranial and facial anomalies, and represents an important advancement in the field of craniofacial surgery. It has been defined as the regeneration of bone tissue between two vascularized bone surfaces, which are progressively separated by gradual lengthening (1).

This study was carried out by the Group of Craniofacial Surgery, Division of Burns and Plastic Surgery, Hospital das Clínicas, School of Medicine, Universidade de São Paulo, Brazil.

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The distraction osteogenesis technique has become accepted after the studies published by Ilizarov, who established the principles of the method through the development of experimental and clinical trials where endochondral bones of the limbs were lengthened, thus avoiding the use of bone grafts (2). In the field of craniofacial surgery, the first experimental study was published by Snyder et al. in 1973; the authors reported the distraction of a canine mandible with the use of an external device (3). In 1992, McCarthy et al. published the first report on lengthening of the mandibular ramus in humans through the use of extraoral devices (4). In the same year, Remmler et al. carried out a successful distraction of the skull and midface in rabbits (5). Rachmiel et al., in 1993, published the results of an experimental work on midface distraction in adult sheep (6). The first report of the use of distraction osteogenesis for the correction of midface deformities in humans was made by Cohen et al., in 1995 (7). Those authors presented their preliminary experience with unilateral distraction of the

midface in a patient suffering from hemifacial microsomia.

Since then, several authors have used distraction osteogenesis techniques for the correction of maxillary deficiencies in patients presenting cleft lip and palate (8,9), as well as for the treatment of severe midface hypoplasia in patients presenting syndromic craniosynostosis, through the use of internal distraction devices (10-21).

Polley & Figueroa, in 1997, were the first investigators to use a rigid and adjustable external distraction system, or rigid external device (RED) for the lengthening of the maxilla in the treatment of severe maxillary deficiency in cleft patients (8).

In published literature, the use of distraction osteogenesis in the midface has shown promising results, which are frequently better than those observed with conventional methods of facial advancement. With this new method, it has been possible to improve the treatment of these patients, reduce surgery duration, avoid the use of bone grafts and improve the stability of results even in cases where a major degree of advancement is necessary, not to mention the lower relapse index. The use of distraction osteogenesis has brought another great benefit: it allowed the use of monobloc osteotomies, but without the great morbidity usually associated with the conventional method, when it is performed directly on the bone segment. However, most of the authors performed distraction osteogenesis of the midface through the use of internal devices, a method that presents certain technical difficulties that can be countered by the use of external devices.

In this study, we present our preliminary experience of the use of distraction osteogenesis with an RED for the treatment of patients suffering from syndromic craniosynostosis with severe hypoplasia of the midface.

The aim of this study was to introduce the use of distraction osteogenesis with an RED as a method for treating midface and orbital hypoplasia in patients presenting syndromic craniosynostosis. We report our preliminary experience in two clinical cases and discuss the indications and results obtained with the method.

PATIENTS AND METHODS

Two patients, both with syndromic craniosynostosis, aged 10 and 5 years and presenting Crouzon and Apert syndromes, respectively, were submitted to assessment by the clinical team of the Department of Craniofacial Surgery, Division of Burns and Plastic Surgery, Hospital das Clínicas, School of Medicine, Universidade de São Paulo, Brazil. Both presented severe hypoplasia of the midface, with class III malocclusion and indications for surgical advancement of the midface. Distraction osteogenesis of the midface was indicated with the use of an RED (KLS Martin, Germany) (figure 1).

Parents were informed about the method being used, including information on distraction process duration and the period for which the distractor would have to be worn after the activation period. Alternative treatment methods, the potential morbidity associated with device failure and with the performance of major surgery were explained to parents. The possible need for additional surgical procedures in the future was also pointed out.

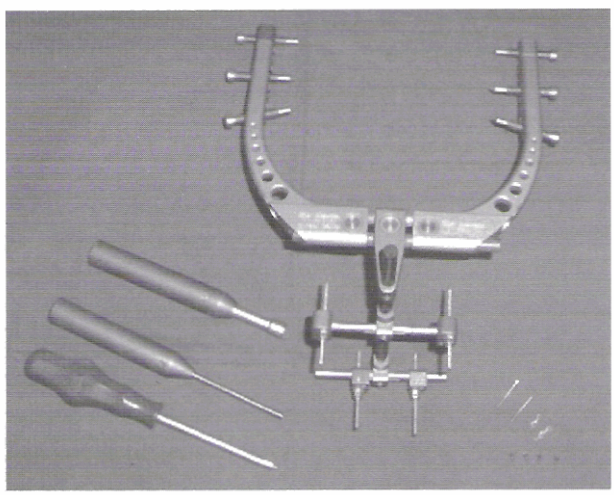

Figure 1. Rigid external distraction system (KLS Martin, Germany).

Patient 1

J.R.L., the 10-year old male patient carrying Crouzon syndrome, presented significant midface hypoplasia and class III malocclusion, in addition to mild proptosis (figure 2A) and mild obstructive sleep disorder. Mental development was normal. His father presents a mild form of Crouzon syndrome, which has never been treated surgically.

At 3 years of age, the patient was submitted to frontal advancement and developed a high-output cerebrospinal fluid (CSF) fistula requiring drainage via lumbar puncture. At 5 years, a further surgical operation was performed, consisting of a Le Fort type III osteotomy with the fitting of an internal midface distractor. There was a good initial response, however the syndrome relapsed progressively as the patient developed.

Distraction osteogenesis of the midface was then planned using an RED (KLS Martin, Germany). The patient underwent an orthodontic procedure to fit an intraoral molding anchored to the first upper molars the day before the operation. Nasotracheal intubation was performed with the aid of an endoscope. The incision followed the existing coronal scar that had resulted from previous surgery. A coronal approach was then performed, which, together with bilateral infraciliary incisions and an incision along the upper gingival labial sulcus, allowed access to the orbits and midface.

The Le Fort type III osteotomy was performed in the conventional manner and craniofacial disjunction was achieved with no intraoperative bone advancement. Fitting of the RED was performed, anchoring to the upper dental arch via the intraoral molding and at two other points, bilaterally, at the lower orbital border.

The operation lasted 360 minutes. The patient remained on mechanical ventilation for 1 day after the operation, was discharged from intensive care 2 days postoperatively, and discharged from hospital 17 days after the operation.

The distraction device was activated after a 5-day latency period and was extended by 0.5 mm per day for 30 days, reaching a total bone elongation of 15 mm. The device remained inactivated for 8 weeks after the last activation, when it was removed.

In terms of occlusion, there was an overcorrection. Class III malocclusion became class II malocclusion, and now the patient is being treated orthodontically.

Patient 2

M.R.S., 5 years old, female, suffering from Apert syndrome, presenting significant hypoplasia of the midface and class III malocclusion, a moderate degree of proptosis (figure 3A) and moderate obstructive sleep disturbance. She showed a mild degree of neuropsychomotor development compromise.

At 4 years, the patient underwent sequential correction of syndactyly between the second, third, fourth and fifth fingers of both hands, with good functional response.

Distraction osteogenesis of the midface was then planned using an RED (KLS Martin, Germany). The patient underwent an orthodontic procedure to fit an intraoral molding anchored to the first upper molars the day before the operation. Nasotracheal intubation was performed with the aid of an endoscope. A coronal approach was then performed, which, together with bilateral infraciliary incisions and an incision along the upper gingival labial sulcus, allowed access to the orbits and midface.

The frontal orbital maxillary monobloc osteotomy was performed, and disjunction achieved, with no intraoperative bone advancement. Fitting of the RED was performed, anchoring to the upper dental arch via the intraoral molding and at two other points, bilaterally, at the lower orbital border.

The operation lasted 390 minutes and two units of concentrated erythrocytes were used. The patient remained on mechanical ventilation for 2 days after the operation, was discharged from intensive care 3 days postoperatively, and discharged from hospital 10 days after the operation.

The distraction device was activated after a 5-day latency period and was extended by 0.5 mm per day for 25 days, reaching a total bone elongation of 12.5 mm. The device remained inactivated for 8 weeks after the last activation, when it was removed.

As with the earlier case, there was an overcorrection, with a change from class III to class II malocclusion, and the patient is now being treated orthodontically.

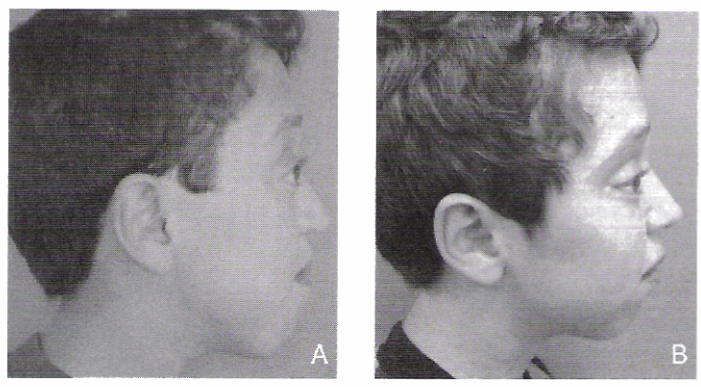

Figure 2. Patient 1, Crouzon syndrome. A) Preoperative view. B) Postoperative view, 9 months after completion of the distraction process.

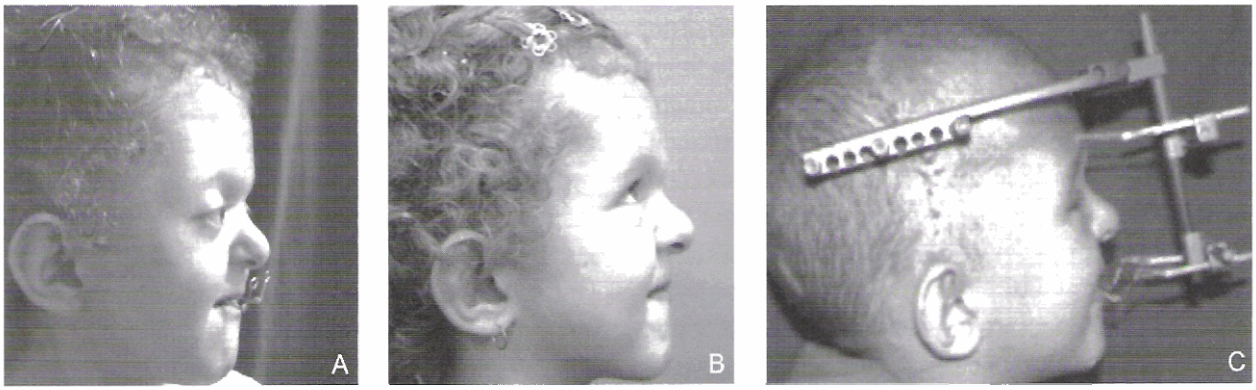

Figure 3. Patient 2, Apert syndrome. A) Preoperative view. B) Postoperative view, 6 months after completion of the distraction process. C) Patient in the postoperative period, during activation of the distraction device.

RESULTS

There was no morbidity associated with the activation process or during the 8 weeks that the device remained as retention and fixation apparatus.

The activation process began after a 5-day latency period and was performed at 0.5 mm/day. There was no pain associated with the use of the device or the distraction process. No signs of infection were observed.

The length of time in surgery was significantly reduced in comparison with usual orthognathic procedures, in which bone grafts are used to meet the bone shortfall resulting from the advance of the osteotomized segment and rigid internal fixation.

For patient 1, distraction was effective in resolving proptosis and improving the facial profile (figure 2B). The total distraction was 15 mm. There was considerable improvement in quality of sleep.

Improvements in facial profile and proptosis were even more obvious in patient 2 (figure 3B), since midface hypoplasia and ocular displacement were more conspicuous than in the other case. Total distraction was 12.5 mm. Sleep disturbances, which were also more pronounced in this case, were markedly improved after distraction.

DISCUSSION

Distraction osteogenesis is nowadays a procedure of great value for treating the craniofacial deformities found with syndromic craniosynostosis. These abnormalities are characterized by severe midface hypoplasia with class III malocclusion.

Reduced orbital volume is another characteristic finding, leading to proptosis and frequent ocular displacement, which can result in permanent loss of sight. Airway obstruction is common with severe syndromic craniosynostosis patients, and there is a predisposition to respiratory infections, sleep apnea, *cor pulmonale*, neurological dysfunction and brain damage (16).

The objective of treating craniosynostosis facial abnormalities is to achieve a result that restores both form and function. Patients suffering from syndromic craniosynostosis have been subjected to their first fronto-orbital advancement at 4-9 months of age. A large number of patients require a secondary surgical operation to treat increased intracranial pressure or inadequate skull curvature (16).

The majority of protocols envisage treating midface hypoplasia at between 4 and 7 years of age. Le Fort type III osteotomies have successfully increased orbital volume by increasing antero-posterior and vertical orbit diameters (16). However, when patients have severe exophthalmia, monobloc advancement, associated with fronto-orbital advancement by Le Fort type III osteotomy, has been the treatment of choice.

Distraction osteogenesis presents advantages in comparison with more usual orthognathic surgery, in which midface advancement is supported by bone grafts and rigid internal fixation. There are no age-based limitations, as it is not necessary to wait for skeletal maturity to perform the procedure (22).

Standard techniques present great limitations in terms of the extent of bone advancement possible, because of the resistance offered by the enveloping soft tissues. Cases treated show anterior facial advancement varying from 6 to 17 mm on average, with a majority of cases at around 10 mm (16). However, the average orbital and midface deficiency in patients suffering from syndromic craniosynostosis is 24 mm (14), therefore exceeding the bone displacement magnitude possible with conventional procedures, without risk of treatment failure. Often, large bone advances may technically be obtained, but the instability of large advances greatly increases the risks of losing the results achieved. The

resistance offered by subjacent tissues and reabsorption of bone grafts due to lack of contact or intense compression are causes of relapse which characterize failure with this treatment.

Stable results, even with large bone movements, became possible with the advent of distraction osteogenesis, which also gradually lengthens soft tissues, surmounting the resistance they offer. The formation of new, orthotopic bone tissue, of higher quality when compared to bone grafts, is another factor that yields better results. Additional advantages relate to the lower rate of morbidity associated with the procedure, with reduced surgery times because there is no need for rigid internal fixation or bone grafting (14).

Gradual distraction is also responsible for less morbidity than monobloc osteotomies. In the conventional procedure, the bone segment is advanced immediately, resulting in a retro-frontal dead space which is immediately filled by blood, occasioning the possibility of infection due to opening of the nasofrontal region. The risk of epidural abscess is therefore reduced when distraction osteogenesis is associated with monobloc osteotomy, since bone advancement is performed gradually from the fifth postoperative day, with no retrofrontal dead space being immediately formed (16), which also reduces the possibility of CSF fistula.

The type of device used for midface distraction varies across the different published series. The use of internal distractors is recommended by authors such as Cohen et al. (8,13,15), Chin and Toth (10), and others (11,12,14,16,19,20). Cohen et al. (13,15) developed an internal modular distraction system (MID, Modular Internal Distraction System, Howmedica Leibinger, Inc., Rutherford, NJ, USA) and have described their experiences with this device in many papers, listing as advantages the fact that it is worn in a less visible position and has a smaller volume than external devices, which facilitates handling, particularly with young patients, and that it is not fixed to the upper dental arch, which is underdeveloped in patients with severe hypoplasia of the midface. These devices, however, have the disadvantages of making distraction possible along just one vector and of requiring a second operation, with a coronal approach, to remove the device (21).

Authors such as Cedars et al. (12) and Gosain et al. (21) used internal distractors custom-fabricated from three-dimensional computerized tomography reconstructions, in an attempt to make the device more suitable to the patient and to control distraction vectors more precisely. Custom-made devices employing biodegradable bone anchorage plates, as used by Cohen et al. (17,18), dispense with the need for a second coronal approach to remove the device. However, the high cost of such devices and their limited availability restrict their use.

Worth noting is the fact that the magnitude of distraction is not directly correlated with the magnitude of advance achieved, probably due to rotation of the advanced segment, causing the maxilla and zygoma to move forward in relation to the incisors (18). Furthermore, the resistance offered by enveloping soft tissues is greater in the antero-posterior direction, which may force the bone segment downwards during distraction.

The occurrence of infection, making it necessary to remove the device, is another cause of internal distraction failure. These distractors are fixed to the temporal region, the body of the zygoma, and to the lateral and anterior orbital borders, with the majority of the force exerted against the zygoma body; this makes internal distractors difficult to use for certain midface hypoplasia patients as a weak union between the body of the zygoma and the maxilla is characteristic and fractures at the level of the maxilla-zygoma transition during distraction are common. These factors all mean that the use of internal distractors is especially problematic with children under 5 years of age suffering from syndromic craniosynostosis and make the method inappropriate for this age group. Gosain et al. (21) presented a series of eight patients in which, even though they advocated the use of MID, MID was initially planned for 7 patients. In all cases there were problems related to fracture or instability of the zygoma-maxilla junction after osteotomy or during the distraction process, forcing the use of the RED in two cases.

Experience with external midface distraction began with the works by Polley and Figueroa (22-24). They were responsible for the development of the RED used in the two cases described above.

The authors defend the use of RED because they offer greater control over the distraction process and make possible adjustments to and control of the gradual mobilization of bone segments in horizontal, vertical and transverse directions, since vectors can be changed at any point. This being the case, the segments that are mobilized are placed in the desired locations, producing more predictable and satisfactory results (22). They can even be used with bone that is extremely hypoplastic, because these bones are not used to support the traction forces, which also means that osteotomy is performed according to the esthetic and functional needs of each patient and does not have to be based on the necessary quantity of sufficiently stable bone on either side of the osteotomy line for distractor fixation, as is the case with

internal devices (21). RED are fixed to the upper dental arch, offering the chance of better occlusal results, although this has been seen as a disadvantage as it makes adequate teeth indispensable to intraoral molding fixation (22). This may be especially problematic with very young craniosynostosis patients due to their characteristically underdeveloped upper dental arch, with missing elements or dental hypoplasia (21). In such cases, however, skeletal anchorage points or osseointegrated implants may be the solution. Hierl and Hempricht (13) recently developed a modular retention system (Martin Medzin-Technik, Tuttlingen, Germany) that can be employed in these cases, guaranteeing easy control of the torque applied to the maxilla.

Additional advantages to the RED are no need for a second large-scale surgical operation to remove the device and the fact that any infection that may occur does not make it necessary to remove the device, and it remains possible to continue distraction with no prejudice to the final result.

Midface distraction osteogenesis employing RED has proved an effective method for dealing with the facial abnormalities presented by patients suffering from syndromic craniosynostosis. They offer stable results with satisfactory degrees of bone advancement.

Initial experience with this method has been satisfactory and suggests it should continue to be indicated, thus increasing patient sample and making it possible to assess long-term results.

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AMNIOTIC BAND SYNDROME

Andrea Fernandes de Oliveira, MD[1]; Max Domingues Pereira, PhD[2]; Sérgio Cavalheiro, PhD[3]; Lydia Masako Ferreira, PhD[4]

Amniotic band syndrome has a number of different synonyms: amniotic band disruption complex or sequence, annular constriction bands and aberrant tissue bands. The incidence of amniotic band syndrome is unknown and its etiology is also unclear. Clinical manifestations are extremely varied, and abnormalities may be isolated, such as the presence of a single constriction ring, or multiple, as in the cases herein described. Limb constrictions are the most common findings, and craniofacial anomalies are the most serious ones, due to the high-level functions of the organs involved. This paper reports on two cases of patients with amniotic band syndrome. One patient was male and the other female. Both presented encephalocele, facial clefts and constriction rings on upper and lower limbs. The diagnosis of this syndrome is based on clinical findings that include, in addition to the patient's anatomical anomalies, an examination of the placenta and the amniotic membranes. Prognosis is poor when the central nervous system is affected and, in the rare cases in which such children survive, sequelae are severe, which makes the study of this disease important.

KEY WORDS: *Amniotic bands; amniotic band syndrome; congenital defects; abnormalities; neonatal diseases.*

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Amniotic band syndrome is a relatively rare fetal abnormality involving varied clinical alterations that were first described by Chaussier (1812) and Watkison (1824). However, it was not until 1832 that Montgomery recognized the relationship between fibrous strings and amniotic bands (1). The etiological factors that precipitate this disease are yet unknown. The theory that has gained widest acceptance is that of Torpin (2), who proposed that the condition is the result of amniotic sac rupture, leading to chorion and amnion becoming separated, with amniotic fluid entering the chorionic cavity (2). These fibrous chords that originate in the chorion impede the normal development of parts of the fetus, resulting in varying anomalies (2,3).

There are a number of different synonyms for this condition: congenital annular constriction, amniogenic bands, aberrant tissue bands, ADAM complex (amniotic deformity, adhesions, mutilations), amniotic band disruption sequence, intrauterine amputation, and Streeter's dysplasia. Almost all cases are sporadic, but published literature does record some family cases. There is no predominance according to sex and no racial predisposition (4).

DESCRIPTION OF CASES

Patient A.A.S. (figure 1A), female, presented at birth (January 20, 2000) frontal meningoencephalocele (figure 1B), incomplete cleft lip on the left, left-side choanal atresia, upper right limb agenesis, amniotic band on the fourth finger of the left hand, amputation of the second and fifth fingers of the same hand (figure 1C) and of the second, third, fourth and fifth toes of the right foot (figure 1D). No visceral abnormalities. Meningoencephalocele was corrected on the sixth day of life, cheiloplasty and correction of the fourth finger of the left hand were performed on the sixteenth day of life. On the twentieth day of life, a right-side ventriculoperitoneal shunt became necessary. Choanal atresia was corrected by surgery the following day.

Patient V.S.A.A. (figure 2A), male, presented at birth (December 14, 2000) frontal meningoencephalocele (figure 2B), craniofacial clefts numbers 1-13, cleft number 11 on the left, amniotic bands on the second, third and fourth fingers, and amputation of the distal phalanx of the fifth finger of the left hand (figure 2C), and amputation of the hallux, second and fifth toes of the left foot (figure 2D). No visceral injuries were present. During the first month of

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life, a ventriculoperitoneal shunt was performed and meningoencephalocele was corrected. During the fifth moth of life, coloboma and the craniofacial cleft were corrected. At 2 years of age, cranioplasty was performed and the upper, left eyelid was grafted (figures 2E and 2F).

The patients are under clinical observation, progressing satisfactorily and being prepared for future operations.

DISCUSSION

Amniotic band syndrome is relatively rare. In this disease, fibrous bands originating from the amnion adhere to different parts of the fetus, causing the three basic types of anomaly: disruptions, malformations and deformities. Disruptions originate in the adherence and strangulation caused by the amniotic bands. If the bands are already present during the embryonic

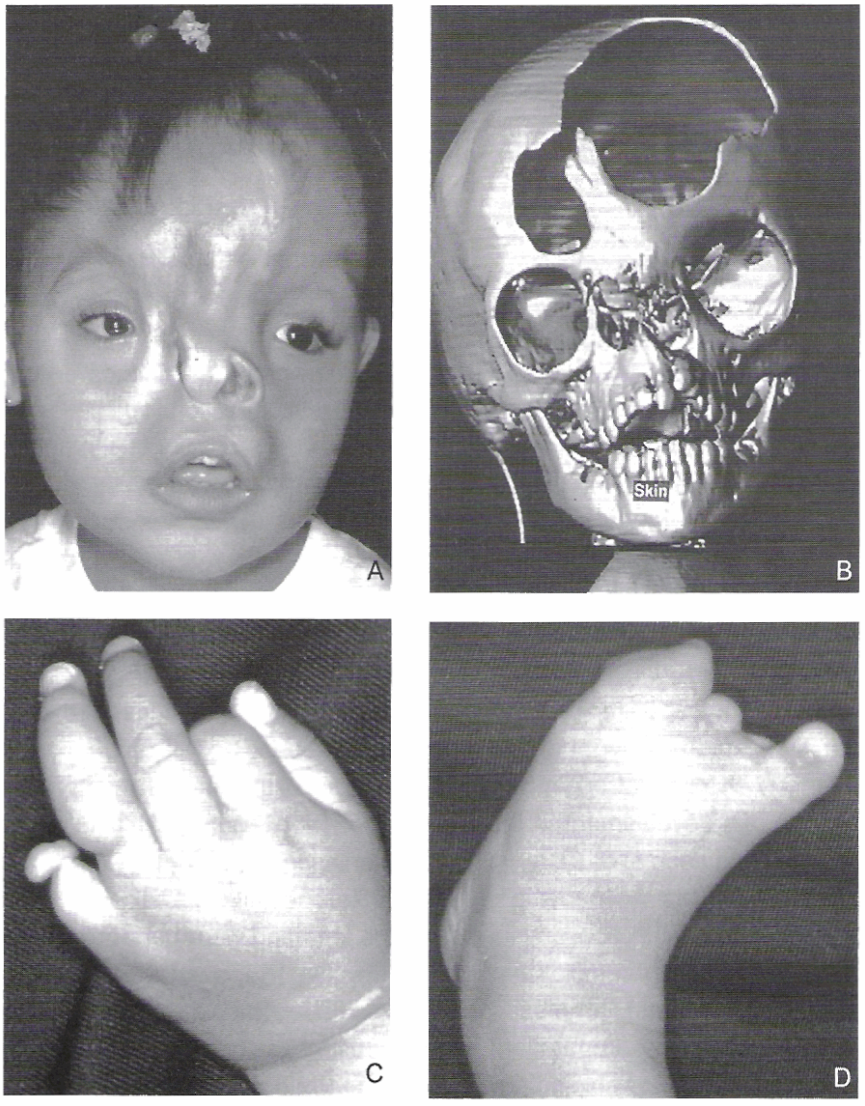

Figure 1. A) Female patient with amniotic band syndrome showing frontal meningoencephalocele, hyperteleorbitism and scarring from surgical correction of the left-side incomplete cleft lip. Also presenting Tessier facial cleft number 3 on right and number 1 on left. B) Computerized tomography of the skull (tridimensional reconstruction) showing bone failure in the frontoparietal region, the presence of hyperteleorbitism and hard palate cleft between the left central and lateral incisors. C) Amniotic band on the fourth finger and amputation of the second and fifth fingers of the left hand. D) Amputation of the second, third, fourth and fifth toes of the right foot.

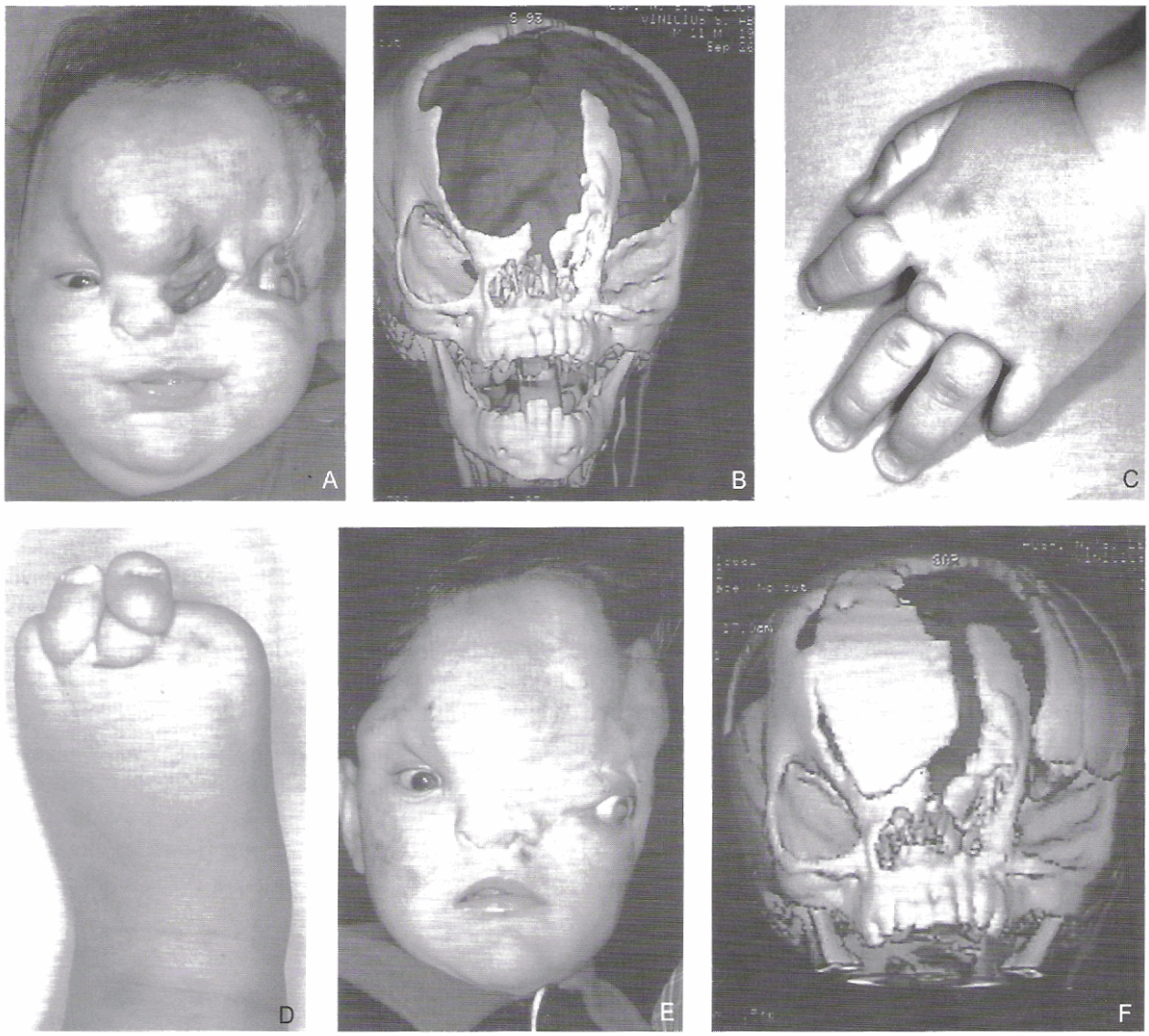

Figure 2. A) Male patient showing frontal meningoencephalocele, craniofacial cleft numbers 1-13, Tessier cleft number 13 and hyperteleorbitism at birth. B) Computerized tomography of the skull (tridimensional reconstruction) showing large bone defect in the frontoparietal region. C) Amniotic bands on the second, third and fourth fingers and amputation of the distal phalanx of the left hand. D) Amputation of the hallux and second and fifth toes of the left foot. E) Postoperative period of meningoencephalocele, facial cleft and coloboma corrective surgery. F) Tridimensional computerized tomography of the skull in immediate postoperative period.

period, they can interfere with normal embryogenesis, resulting in malformation. Deformities result from oligohydramnios, which leads to constriction and compaction of parts of the fetus. Severe compression leads to vascular engorgement, hemorrhage, edema and necrotic tissue, resulting in severe disruption, such as wall or limb defects. Finally, certain malformations cannot be explained by the bands – constriction or compressions that could result in the amniotic rupture sequence (3).

Incidence is unknown, but prevalence is estimated as being around 1.17/10,000 live births.

Etiology is unclear and clinical manifestations are extremely variable. Single abnormalities, such as a discrete scar, can occur in isolation, and multiple anomalies are also possible. While there is a report of family history in published literature, the majority of cases are sporadic (5).

Diagnosis is based on clinical findings that include, in addition the anatomical defects, an examination of the placenta and amniotic membranes, which will always present abnormalities. Limb constrictions are the most common findings, but craniofacial abnormalities are the most serious

ones because of the function of the organs involved. These abnormalities are both frequent and varied, including encephalocele of varying sizes, generally associated with missing skull bones and located forward. Severe microcephalia can occur and even anencephaly and facial deformities such as cleft lips and palates and rare facial clefts. A number of different ocular conditions can also occur, such as microphthalmia, anophthalmia, ectropion, eyelid coloboma and obstructed tear ducts. Visceral anomalies are rare, with gastroschisis being the most frequent. Omphalocele, genital exstrophy and ambiguous genitalia can also occur (3,6,7).

Treatment of the most complex cases in which the skull is involved is performed in a number of different stages and requires family members' understanding.

CONCLUSIONS

Amniotic band syndrome causes incapacitating and anti-aesthetic anomalies, and, in cases where the skull is compromised and sequelae are severe, mortality rates are high, making early diagnosis and treatment even more important to attempt to rehabilitate such patients.

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ORAL AND MAXILLOFACIAL PROSTHESES

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The authors describe oral and maxillofacial prostheses (ocular, nasal, auricular prostheses and palatal obturators) and their current indications. They comment on the role of prosthetists and their areas of competence and demonstrate how, with the use of oral and maxillofacial prostheses, this specialty is capable of reintegrating facially mutilated patients into society.

KEY WORDS: *Maxillofacial prostheses; esthetics; reconstructive surgical procedures.*

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As a result of accidents and urban violence, physical risk has an elevated incidence nowadays, and people are exposed to a larger number of traumas and physical damages. Other causes, such as congenital and hereditary factors, result in physical deformities, as does oncological surgery, which is responsible for significant mutilation and sequelae. Our objective in this paper is to demonstrate that by means of oral and maxillofacial prostheses we can reintegrate facially mutilated patients into society.

REVIEW

Human beings' attempts to restore parts of the oral and maxillofacial regions through alloplasty are as ancient as civilization itself. As mentioned in an earlier publication (1), the Chinese, Romans, Hindus, Incas and Aztecs made ocular, nasal and auricular prostheses. Ambroisé Paré is accorded the honor of being the first to write on the subject, describing several types of oral and facial prosthesis (2). Other authors consider Pierre Fauchard to be the father of dentistry because of his large contribution to oral and facial prosthetics. He left much writing, including a report on the "silver mask" used for a soldier mutilated in battle (3). Another author, Delabarre, published work on the mechanics of dentistry, innovating in retention techniques for metallic palatal obturators. Claude Martin should also be mentioned, since he recognized oral and maxillofacial prosthetics as a specialty within the scientific and didactic standard; he created several prosthetic devices and developed the use of prostheses in reconstructive surgery, leaving for prosperity noteworthy work on the use of prostheses immediately after maxillary resection (3). Other contributors to the area who should also be cited are Snell, Goodyear, Suersen, Kingsley and Tettamore.

In Brazil, the pioneer in this specialty was the dentist Monteiro de Barros. Souza Cunha was the first professor of oral and maxillofacial prostheses. Viana Novaes defended the first thesis in the specialty, and Brito Viana became a full professor at Universidade de São Paulo (4). Also worthy of mention are the contributions of domestic authors to the manufacture of oral and maxillofacial prostheses (1,3,5-8) and of others who have attempted to further the specialty's development. Oral and maxillofacial prostheses can be fabricated within specialty subareas, such as the following:

- ***Ocular or ophthalmic:*** here the objective is to recover facial aesthetics, prevent eyelids from collapsing or becoming deformed and to restore the direction of tear secretion, in addition to protecting the sensitive anophthalmic socket against external aggression such as dust, smoke and other pollutants.

- ***Facial prosthesis or epithesis:*** these become necessary when there has been extensive loss of facial muscular and cutaneous covering and of the supporting skeleton. These structures are restored artificially or alloplastically, recovering function and appearance in addition to protecting exposed tissues. They may be nasal, orbital, labial or auricular.

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- ***Extensive facial prosthesis:*** used when there has been extensive facial loss with soft tissue or bone eliminated as a result of mutilating surgery or accidental traumas.

- ***Mandibular prosthesis:*** these may be internal or external. They restore facial contours, avoiding mandibular distortion, and restore aesthetics, in addition to serving as orthognathic devices guiding the opening and closing of the mouth.

- ***Maxillary prosthesis or obturator:*** manufactured to be used at points of communication between the sinuses and the oral cavity; they are aimed at aesthetic repairs.

- ***Prosthesis for malformed lips and palates:*** used for lip and palate malformations, this category includes obturators for newborns, orthognathic reducers, palatal protectors, cover-up prostheses and pharyngeal obturators.

- ***Devices:*** these are adjuncts to surgery, and their aim is to aid plastic surgery treatment, such as dental droppers and sagittal guides. The device used for actinotherapy (radiotherapy prosthesis) allows more efficient endobronchial radiotherapy or actinotherapy by external contact administration.

- ***Implants:*** this technique is based on the introduction of small cylinders of titanium into bone. Once the necessary osseointegration period has passed, these intraoral anchors are used as a treatment option for patients with cleft lips and palates. Extraoral implants have been used to support prosthetic eyes, noses, ears and others, including feet and hands, and even for fitting semi-implanted sound amplifiers in cases where auditory conduction is absent.

- ***CAD/CAM biomodeling:*** this is a combination of two technologies, prototyping and image-based diagnosis. Images are manipulated with medical imaging software and, based on a computer-generated model, we can fabricate a rapid prototype; biomodels are made by either stereolithography or selective laser sintering. Biomodels are employed in the treatment of patients with facial deformities.

DISCUSSION

Physical losses are primarily caused by accidents involving traffic or at work (industrial injuries), violent or radical sports and interpersonal mishaps (9,10). Oncological surgery is an aggressive treatment. When head and neck surgery is radical and there is significant loss of function, the patient suffers both physiological and psychological problems, due to secondary mutilations and deformities (11). There is consensus that reconstruction, whenever possible, should be carried out via plastic surgery (8). There are, however, serious limitations, both local and general, when dealing with mutilations from surgery for malignant tumors. The general condition of the patient, the prognosis of the case, age and a psychological disposition to undergo autoplastic treatment involving consecutive touch-up sessions are factors that can make this method of treatment either difficult or impossible. The extent of tissue loss, including tegument and bone support, together with the condition of irradiated tissues, reduce the chances of a successful outcome in terms of transplant and graft integration; the biological foundations of plastic and reconstructive surgery itself allow for oral and maxillofacial prosthesis indication.

In cleft lip and/or palate treatment, surgery meets the expectations of most patients. In Brazil, there are many centers and groups of surgeons that operate on people with cleft lips and palates. However, in some cases, patients are left with sequelae because the cleft is simply closed, with no concern for craniofacial growth or the other peculiarities involved in this condition, such as associated anomalies which are sometimes left untreated. The patient and their family must be made aware of the fact that, in addition to surgery, it is of fundamental importance to execute prosthetic-orthopedic treatment in such a manner as to respect facial growth, since facial structure is made up of bones and teeth and the rehabilitation process therefore involves the disciplines of oral and maxillofacial prosthetics, orthopedics, preventive and functional orthodontics, and goes well beyond the aesthetic aspects.

Oral and maxillofacial prosthetics is a recognized specialty and should be carried out by prosthetists (1,3-5,7,8). Integration is its primary characteristic, in that the prosthetist should be in contact with a team of doctors, speech therapists, psychologists, physicists, social workers and other professionals. The experience of the interdisciplinary and multidisciplinary team facilitates advances in care and makes it possible to extend treatment to other deficiencies associated with congenital malformations, such as syndromes whose frequency has been increasing with time. If non-specialists find it strange that the fabrication of prosthetic eyes, ears and noses falls within the sphere of dentistry, how much more perplexed they become on discovering that the prosthetist is also the competent professional responsible for making artificial hands and feet, known

as somatic prostheses. While this is the case, production of feet and hands by prosthetists is not yet performed systematically. With advances in osseointegrated implants, however, there has been a significant increase in this type of prosthesis.

Despite the complexity and the social character of the specialty, few health institutions in Brazil have services set up specifically to care for facially mutilated patients who need reparative prosthesis. In the public service and its competent authorities, there is not, in fact, any policy for the complete rehabilitation of those in need. The Brazilian National Health System (SUS - Sistema Único de Saúde), in the majority of cases, does not cover prostheses or ortheses, classifying them as cosmetic. The National Health System should consider the possibility of including oral and maxillofacial prostheses and biomodels among diagnostic and treatment procedures subsidized by SUS, since the results achieved are decisive to complete recovery of the patient. It is important to point out that treatment with oral and maxillofacial prostheses is not elective but the last chance for these patients.

CONCLUSIONS

Now that the constitutional principles of universal access, of the integration of health activities and of equality have been won, care activities (treatment) must be as resolutive as possible and aimed at all lifecycles. In terms of both aesthetics and health, reconstructing a person's face has come to mean saving that individual's identity, thus making their reintegration into society possible.

Prostheses and ortheses are used to substitute structures whose loss may be congenital or acquired. Treatment in this area is personalized and is based on dentistry, with medicine as its fulcrum, all integrated with the remaining specialties. The primary objective is, by means of alloplastic rehabilitation of missing or compromised regions of the face, to care for all the patient's physical, functional and aesthetic needs.

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TRANSMAXILLARY APPROACH FOR BASILAR IMPRESSION TREATMENT: CASE HISTORY AND LITERATURE REVIEW

Marcus Vinicius Martins Collares, MD, PhD[1]; Albert Brazil, MD[2]; Rinaldo De Angeli Pinto, MD[3]; Luis Carlos Acevedo-Rangel, MD[4]; Ciro Paz Portinho, MD[5]; Rafael Marques de Souza, MD[6]

The authors present the case of a patient with basilar impression (BI). An 8-year old Caucasian male began with upper-limb coordination deficit and disphony. Magnetic resonance (MR) demonstrated basilar impression and syringomyelia. Two months after symptoms had started, the child underwent surgery with double approach. During the anterior approach, a maxillotomy (Le Fort I) and an odontoidectomy were performed. After that, a posterior approach was created through a posterior craniectomy, in order to provide cervical spine arthrodesis. The patient had a good outcome, with complete resolution of neural and behavioral symptoms. BI is a cranial-vertebral junction deformity caused by migration of cervical spine into the cranium. It can be either a primary or a secondary condition, the latter being a consequence of bone thinning disorders. Clinical presentation has signs and symptoms related to direct neural compression, liquor flow obstruction and vascular involvement. Almost all of these patients have headache. IB can lead to secondary syringomyelia. MR is the imaging exam of choice for diagnosis. Nowadays, it is a consensus that anterior neuroaxial compression should be treated with decompression through an anterior approach (usually a maxillotomy). Anterior approach for odontoidectomy is an adequate procedure. It should be performed by experienced surgeons in transfacial accesses, with care to prevent tooth bud damage in children (a high Le Fort I should be made), and reconstructing palate in order to avoid velopharyngeal sphincter alterations.

KEY WORDS: *Basilar impression; odontoid process; spine; cervical vertebrae; maxilla; surgery.*

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A white, male, 8-year-old patient began to suffer from a lack of coordination of the upper limbs and dysphonia 2 months before surgery. A neurologist was seen and work-up exams were requested. Magnetic resonance imaging (MR) showed a malformation at the craniocervical junction (odontoid process) and syringomyelia (figure 1). The patient was referred to the neurosurgery department, which indicated surgical intervention.

In August 2002, the patient underwent transoral odontoidectomy (figures 2-4), posterior craniectomy and arthrodesis of the cervical spine. Tracheostomy was performed at the start of this procedure.

After securing the skull with a halo, a transmaxillary, anterior approach was made, and a high Le Fort I osteotomy was performed in such a way as to preserve the tooth buds. During the anterior approach, dissection of C1, C2 and clivus was performed under microscope. The C1 arch and odontoid process were also drilled under microscope. During this same approach, the posterior ligament was removed. Once the transoral odontoidectomy was complete, the maxillary area was closed up and secured with titanium miniplates and screws, which were removed 3 months later in a separate surgery.

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The second-phase surgery consisted of the posterior approach. A bicoronal incision was made, in addition to a suboccipital craniectomy with C2 laminectomy. The dura mater was opened and the cerebellar tonsils removed. The dura mater was reconstituted with Beriplast®. Arthrodesis performed during the posterior approach was occipital of C5. A lumbar puncture was performed at the end of the second phase of the procedure.

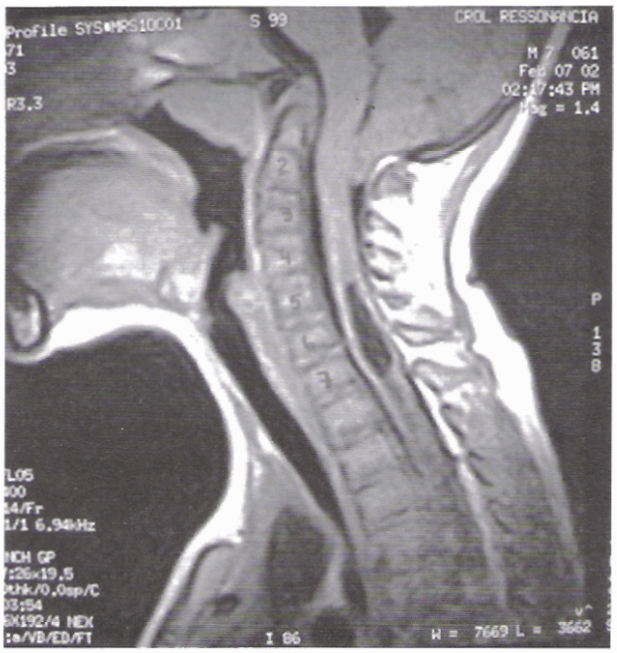

Figure 1. Magnetic resonance imaging showing basilar impression. Note the C2 odontoid process located above the Chamberlain line (a line traced between the posterior border of the hard palate and the posterior border of the foramen magnum).

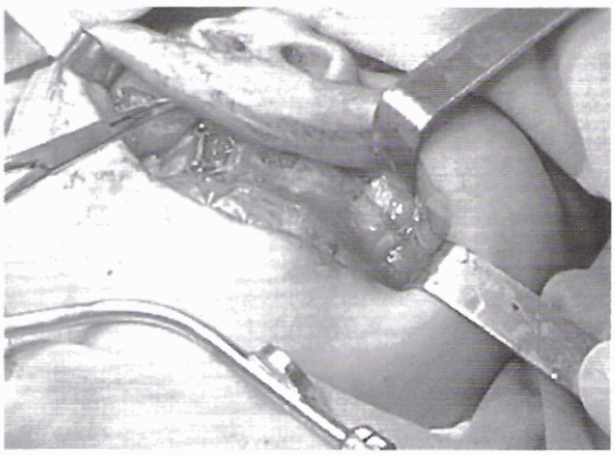

Figure 2. Intraoral access for Le Fort type I maxillary osteotomy. Note the unfixed miniplate, so far with only the holes marking its correct position for the end of surgery.

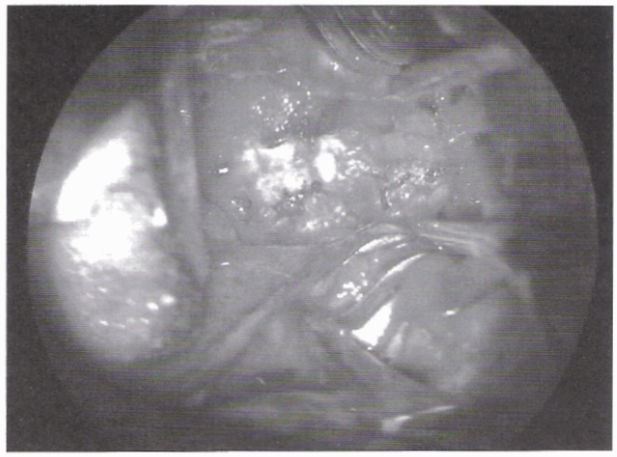

Figure 3. View of the operating field for an anterior approach to the cervical spine.

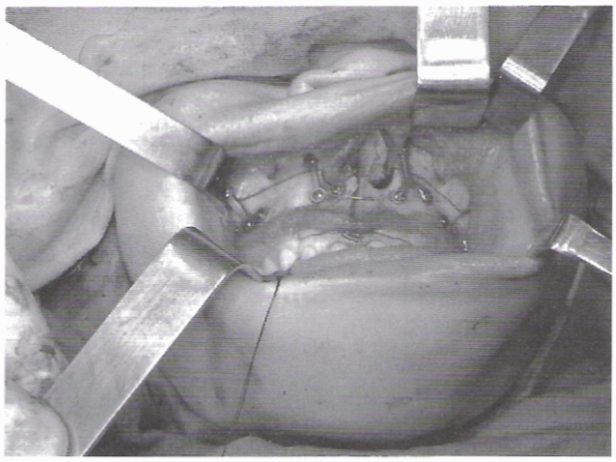

Figure 4. Closure of the transmaxillary approach

The patient progressed most satisfactorily during the postoperative period. A cerebrospinal fluid (CSF) fistula was observed on the fourth day after surgery, which closed spontaneously. Both agitation and motor function abnormalities improved.

LITERATURE REVIEW

Definition and etiopathogenesis

Basilar impression or basilar invagination is a deformity of the craniovertebral junction characterized by the migration of the cervical spine into the skull (1). It is generally associated with platybasia (abnormal angle of the skull base), atlanto-occipital fusion (total or partial fusion of the first vertebrae with the occipital one), and often with deformities of the foramen magnum. This may present small, deformed or eccentric.

Basilar impression may be either a primary or a secondary condition, the latter being the result of bone thinning disorders (1). Causes of secondary basilar impression include: rheumatoid arthritis, Paget's disease, osteogenesis imperfecta, osteomalacia, hyperparathyroidism, type 1 neurofibromatosis, Down syndrome and hemangiomas of the base of the skull (1-7). Basilar impression has even been described in Goldenhar syndrome (8). Cases of basilar impression secondary to trauma are very rare (9). With inflammatory conditions such as rheumatoid arthritis, neurological abnormalities may occur, not just because of osteoarticular involvement, but also due to the presence of granulomatous tissue (*pannus*) compressing the spinal marrow.

Severe basilar impression leads to the upper cervical spine and clivus being displaced cranially into the foramen magnum (1). Arnold-Chiari malformation and basilar impression are part of a group of "osteo-neural growth pathologies", which includes other dysplastic disorders of the axial or appendicular skeleton, such as platyspondyly scoliosis, Scheuermann kyphosis, achondroplasia-like conditions, congenital dysplasia of the hips, etc. (10).

Clinical status

Clinical status depends on symptoms related to direct compression of the neuroaxis, obstruction of CSF flow and vascular involvement (1,11). Almost all patients present with headaches. There are difficulties with walking, paresis of extremities, hyperreflexia that is generally bilateral and affects both upper and lower limbs, while the Babinski reflex and clonus may also be manifest (12). Basilar impression due to bone abnormalities at the craniovertebral junction is a rare, but treatable cause of ataxia in children (13), and calls for differential diagnosis in what concerns other types of ataxia.

In some cases, nuchal pain and vertigo are associated (14). This manifestation, indeed, calls for differential diagnosis to rule out Ménière's disease (15,16). The most commonly affected cranial nerve pairs when compression occurs are the fifth and eighth. Occasionally there is paralysis of the abducens nerve (17).

Basilar impression may also involve secondary syringomyelia (1). Syringomyelia is a cavity extending throughout a number of different marrow segments, exhibiting preference for the cervical region and possibly extending upwards (within the brainstem, where it is defined as syringobulbia). Syringomyelia is characterized by motor abnormalities, painful thermal sensitivity in the chest, cervical and occipital pain, lesions of the hands, spontaneous fractures and abnormal elbow and shoulder articulation. Signs and symptoms are bilateral and generally asymmetrical. Syringomyelia can also be secondary to conditions other than basilar impression, such as medullary tumors, type I Arnold-Chiari malformation, and post-traumatic medullary scarring.

Silva (18) describes a Brazilian sample of 209 cases of craniovertebral anomalies. According to that author, the prevalence of basilar impression in the Brazilian northeast region is notorious: 13.3% had basilar impression in isolation, 4.7% had Arnold-Chiari malformation in isolation, and 81.8% had both conditions.

Diagnosis by imaging

Diagnosis may be made using computerized tomography (CT) or MR imaging (1). MR is preferable to CT according to a number of different authors (19). If the odontoid process is located above a line between the posterior border of the hard palate and the posterior border of the foramen magnum (Chamberlain's line), then basilar impression can be diagnosed.

A comparative study of a number of radiological measurements taken of 100 normal individuals and 10 individuals with basilar impression demonstrated significant differences between the groups only in terms of the position of the odontoid process (1.2±2.28 mm below the baseline in controls against 9.0±2.7 mm above the baseline in patients) and of the nasion-basion-opisthion angle (162±4 degrees in controls against 178 ± 5 degrees in patients) (20).

Treatment

Treatment depends on the exact nature of the abnormality found, but there is consensus that patients with anterior neuroaxis compression should undergo anterior decompression, normally accessed by maxillotomy (1,21-24). Young Su et al. (25) described a case where an anterior approach was used to treat basilar impression with a mandibulotomy instead of the transmaxillary approach, with good postoperative results. Whichever anterior approach is preferred, it must allow the resection of the odontoid process, the anterior arch of the atlas and the lower clivus with least risk possible (26). After performing transfacial access, the whole of the surgical process should ideally be

performed with the aid of a microscope, in order to increase the safety and efficacy of the procedure (27).

Basilar impression may occur as a well-known complication of osteogenesis imperfecta, which affects up to 25% of these patients and 70% of those who have dentinogenesis imperfecta (1,7,12,22,28). In such cases, the disease has a progressive nature. The syndrome begins with asymptomatic ventricular dilation, passes through a compressed foramen magnum syndrome, and death is by brain stem constriction (7). Patients with osteogenesis imperfecta exhibiting headaches when coughing and trigeminal neuralgia merit detailed investigation. The progressive nature of these cases has led to the development of a more specific surgical technique, "open door maxillotomy", combined with a "contoured loop fixation". It is recommendable that family members and patients with osteogenesis imperfecta be assessed with the intention of preventing severe neurological complications (29).

The anterior approach should be followed with a posterior approach for rigid fixation (22), thus transferring the weight of the head to the thoracic spine and avoiding renewed invagination (or basilar impression).

DISCUSSION

This patient showed an atypical clinical presentation, illustrating the heterogeneous nature of basilar impression syndromes. There were no complaints of headaches or dizziness. The patient sought treatment while less than 10 years old, but there was no hereditary disease which would explain secondary basilar impression or any previous family history.

The anterior approach is highly appropriate to odontoidectomy. It should be performed by surgeons with experience of such access routes. However, there are certain important factors that should be taken into consideration with a maxillotomy approach. The Le Fort type I osteotomy should be high in order to avoid damaging the teeth buds. Closure includes palatoplasty when treating a child, and this should be done in such a way as to avoid causing functional damage to the velopharyngeal sphincter (which is the reason why the craniofacial surgeon should be experienced in cleft palate patients). While there are descriptions of mandibulotomy access, treatment performed by maxillotomy appears to be more suitable and is preferred by a majority of authors.

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SEVERE ORBITAL COMPLICATION RELATED TO MUCOCELES OF THE MAXILLARY SINUS: CASE REPORT

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The following article is a report of the case history of a patient who sought treatment at the ophthalmology service complaining of red eye and diplopia. Based on physical examination, the ophthalmologist diagnosed exophthalmia and, suspecting an expansion, referred the patient to our craniomaxillofacial surgery service. We asked for a CT scan that showed a lesion on the left maxillary sinus, obliterating the anterior bone wall structure and bone of the lower wall of the eye floor. Progress to date is satisfactory.

KEY WORDS: *Maxilla; mucocele; exophthalmos.*

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Mucoceles are expansile cystic lesions which affect the paranasal sinuses. The majority affect the frontal sinus (60% of cases), ethmoidal sinus (30%), maxillary (10%), and it is seldom found in the sphenoid sinus (1-3). It usually occurs in adults, especially those between 40 and 60 years old, and does not seem to have a gender preference.

Mucocele can originate from many causes: chronic infection, allergic rhinosinusitis, facial trauma, previous surgery and a small percentage are considered idiopathic.

The symptoms and clinical course can vary according to stage and evolution. As long as it remains restricted to the maxillary sinus, it is asymptomatic, but as it starts growing it can erode the bony wall, resulting in local pain and edema as well as posterior rhinorrhea. As the disease advances, it can cause orbital complications such as exophthalmoses, reduced visual acuity, red eye and diplopia (3-5).

Diagnosis is based on clinical history, detailed physical examination and imaging exams, of which computerized tomography is the most important. Pathological anatomy is the final and definitive diagnosis.

Treatment is conducted by surgical excision using different techniques depending on the size of the lesion.

CASE REPORT

A.N., a 71-year old man, arrived at our department of ophthalmology complaining about an irritation on his left eye (red eye) (figure 1), decreasing sight and diplopia in the same eye. He was then directed to our clinic, where we could perceive asymmetry between his eyeballs and exophthalmos of the left eye (figures 2 and 3). Otorhinolaryngological examination revealed no significant alterations, except pain as we palpated his left malar region. His personal history did not include chronic nasal infection, previous surgery or local trauma.

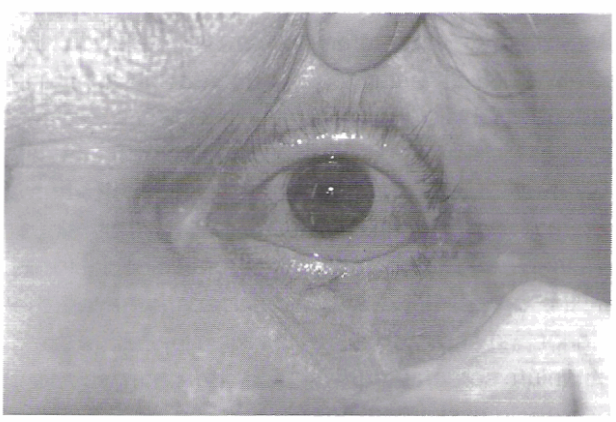

Figure 1. Left eye conjunctival irritation.

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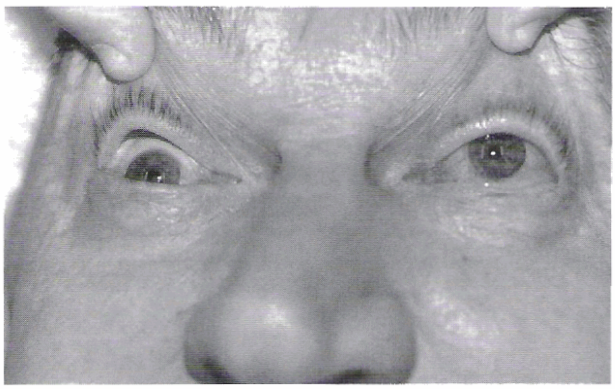

Figure 2. Eye asymmetry. Observe asymmetric irises and pupils, with the left eye positioned more superiorly than the right eye.

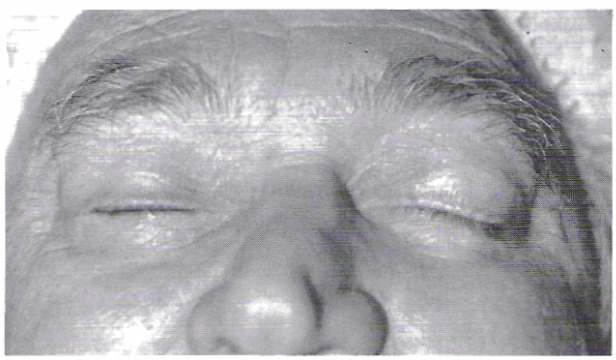

Figure 3. Proptosis of left eye.

We then requested a set of CT scans of the paranasal sinuses and nasal cavity, with fine cuts of coronal and axial films, as well as sagittal reconstruction. It revealed an expansile image with soft tissue density, regular contours in the left maxillary sinus associated with remodeling and discontinuing of the adjacent bony structures with superior extension to the orbital floor, occupying a postero-inferior area of the orbital cavity, and discontinuity of the anterior wall of the maxillary sinus, extending to the malar region and causing erosion on the postero-lateral wall of the maxillary sinus and masticator space (figure 4).

Because the lesion presented compressive and expansive characteristics, we decided to conduct surgical treatment in order to remove the lesion and obtain a definitive diagnosis. We made our approach through a subciliary incision in the left lower eyelid and another incision in the gingival sulcus also on the left (Caldwell Luc), dissecting it via subperiostium, osteotomy of the anterior wall of left maxillary sinus. We then removed the entire lesion with special focus on the orbital floor, and finally reconstructed the inferior and anterior walls of the maxillary sinus using a titanium screen (figure 5). The specimen was sent for pathological anatomic evaluation, which revealed a characteristic cystic lesion. The patient presented a very good postoperative recovery and an almost immediate improvement of his visual disturbance and diplopia, together with repositioning of the eye balls (figure 6).

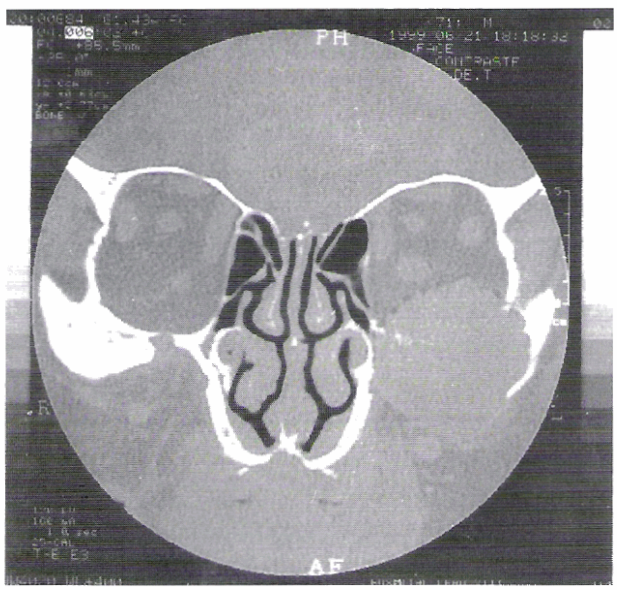

Figure 4. Coronal CT scan showing the lesion (orbit size).

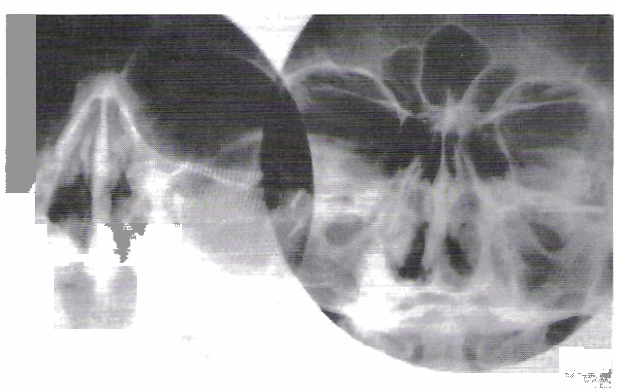

Figure 5. X ray showing reconstruction with a titanium screen.

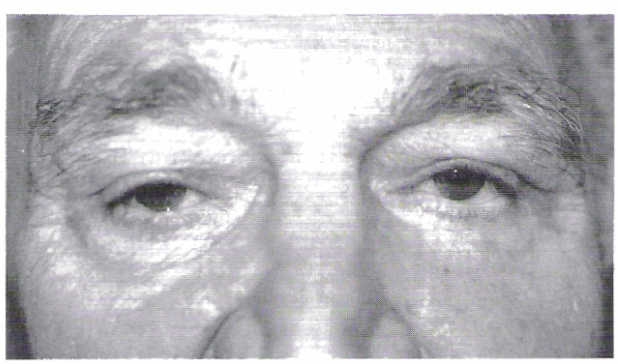

Figure 6. Postoperative follow-up showing red eye and ocular diplopia resolution.

DISCUSSION

Maxillary sinus mucocele is a benign condition, relatively rare, with a cystic aspect constituted of secretor respiratory mucosa and stratified columnar epithelium. Pathophysiology is related to the obstruction of the maxillary sinus ostium, and there are many theories that attempt to explain its origin. These include: congenital, retention, infectious, traumatic, and inflammatory theories, as well as post-surgery theories (2,4). If we considered one of these theories, our patient most probably would fall into the infectious or retention categories. Considering that the patient had never had any past history or complaint of sinus disease, the drainage ostium must somehow have become obstructed (this can be seen in the CT scan).

The diagnosis of this pathology is strictly clinical, and there is a latent period at onset. There may also be an indeterminate amount of time following an exteriorization period, during which the patient presents symptoms such as pain and edema of the maxillary region, posterior rhinorrea, and nasal obstruction – during this period, some complications may occur, such as diplopia due to compromise of the extrinsic musculature of the eye or ocular dystopia with loss of corresponding points in the retina, numbness of the infra-orbital nerve region and lowering of visual acuity. Because of the chronic inflammatory process, bone erosion may occur more often on the eye floor, resulting in exophthalmos.

Differential diagnosis should be performed to rule out benign tumors of the paranasal sinuses, such as ossifying neurofibroma, inverted papilloma, fatty cyst and some other cystic lesions.

The gold standard examination is computerized tomography, where erosion of the bonny wall trough osteolysis may be revealed. Final diagnosis is performed by anatomic pathology, which reveals a single columnar mucus producing epithelium wall that delineates a cystic area, along with another submucosal layer and cystic lymph infiltration. Treatment is surgical, and the sooner the better, so that complications such as those that occurred with our patient can be avoided.

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