Orbitocranial Fibrous Dysplasia: Outcome of Radical Resection and Immediate Reconstruction With Titanium Mesh and Pericranial Flap

Khalid Nasser Fadle, MD, Ahmed Gaber Hassanein, MD,† and Abdin K. Kasim, MD**

Introduction: Fibrous dysplasia (FD) is a non-neoplastic developmental fibro-osseous disease. It represents 2.5% of all bone tumors and 5% to 7% of the benign bone tumors. Orbitocranial region is involved in about 20% of the patients. The main presentations are craniofacial deformity and headache. Loss of vision is the most devastating result of this disease. There is no medical treatment to cure or prevent FD. Radiation therapy is contraindicated. Surgery for the orbitocranial FD is often challenging because of the proximity of neurovascular and ocular structures. Conservative surgical shaving and recontouring is always associated with suboptimal results. Radical excision is potentially curative with no extra morbidity. Orbital hypertelorism, dystopia, or proptosis can be corrected only by radical excision and reconstruction.

Aim: The aim of the study was to evaluate the outcome of radical excision of the orbitocranial FD and immediate reconstruction using titanium mesh and pericranial flap.

Patients and methods: This prospective study had been conducted on 22 patients with orbitocranial FD with age range from 17 to 52 years (mean 29.5). Radical excision of the lesions was done for all patients through transcranial approach. Immediate reconstruction was achieved using titanium mesh and pericranial flap.

Results: Intraoperative dural tears and cerebrospinal fluid leak were reported in 2 patients and repaired with galeal graft. Supraorbital anesthesia occurred in 6 patients. Of these, 2 patients were transient, while the remaining 4 patients were permanent. Wound infection was noticed in 1 patient who improved by medical treatment. Temporary postoperative diplopia occurred in 1 patient and temporary postoperative impaired vision in 1 other patient. In all patients, acceptable or good aesthetic results were observed. No recurrence was detected in our series during the follow-up period that ranged from 24 to 58 months (mean 37.5 months).

Conclusion: Radical excision of orbitocranial FD is potentially curative with no extra morbidity. It can achieve good aesthetic and functional results with no recurrence.

From the *Neurosurgery Department; and †Maxillofacial Surgery Unit, Faculty of Medicine, Sohag University, Sohag, Egypt.

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Address correspondence and reprint requests to Ahmed Gaber Hassanein, MD, Maxillofacial Surgery Unit, Sohag University Hospital, Sohag, Egypt; E-mail: ahmedgaber_74@yahoo.com

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Key Words: Fibrous dysplasia, orbitocranial, pericranial flap, radical excision, reconstruction, titanium mesh

Fibrous dysplasia (FD) is a benign fibro-osseous developmental disorder of the growing bone in which weak fibrous and osseous tissue replaces the normal medullary bone.[1] It represents 2.5% of all bone tumors and 5% to 7% of the benign bone tumors. Craniofacial involvement with FD has been variably described in the literature as craniomaxillofacial, orbital, fronto-orbital, cranio-orbital, or complex FD.[2–4] It occurs in 25% of all patients with FD. FD is most commonly observed between 3 and 15 years of age, and the majority of the patients are diagnosed before the age of 30 years. Males and females are equally affected.[5–7]

Four clinical types of FD are reported. They are the monostotic FD, the polystotic FD, McCune–Albright syndrome, and Mazabraud syndrome.[8]

The most affected bones in the craniofacial region are maxilla, mandible, sphenoid, ethmoidal, and frontal bones, while temporal and occipital bones are affected less frequently. The main presentation of cranio-orbital FD is craniofacial asymmetry and deformity. Headache is the most common symptom. Other symptoms due to mass effect and compression emerge as the lesion grows. Orbital and periorbital bone involvement leads to hypertelorism.[9] The most devastating result of this disease is loss of vision, which is caused by progressive compression of the venous drainage of the optic nerve and decreased retinal perfusion.[10] Visual impairment may be attributed to traction on the optic nerve, spontaneous hemorrhage, or sinus mucocele formation with subsequent pressure placed on the optic canal contents.[11–13] Once visual impairment is noted, it is generally progressive, with periods of waxing and waning.[14]

Radiographically, the lesions of FD have classically been described as having a ground-glass appearance.[15,16] Biopsy and histopathology examination are necessary for a definitive diagnosis.[17]

No medical treatment is available to cure or prevent FD. Bisphosphonates were reported to stop the progression of the lesion.[18] Calcitonin, mithramycin, and etidronate have also been tried in a few patients with little success.[19] Systemic steroids are used for acute visual loss attributable to optic canal involvement as temporary therapy until the patient can undergo optic nerve decompression.[20,21] Radiotherapy has no role in treatment of FD and should be avoided because it has a high risk of malignant change.[22,23]

Conservative surgical therapy (curettage, contouring, or remodeling) is always associated with suboptimal results and may be repeated while radical excision is potentially curative.[24] Early radical surgery has been advocated both as a means for prophylaxis and for reversing the compressive forces.[3,21] Immediate reconstruction after resection of the involved fronto-orbital bone is usually performed to restore contour, symmetry, orbital volume, an intact cranial base, and barrier.[2,21,25] Different materials have been used to reconstruct the cranio-orbital defects, such as autogenous bone grafts, chondrocostal grafts, titanium meshes, and other alloplastic material.[26,27] Extensive portions of resected bone may be recontoured and treated by autoclaving or cryotherapy, and then reimplanted.[28–30] Synthetic materials such as polyethelene (Medpor) and titanium mesh have an increasing role in reconstruction of skull base defects with excellent craniofacial symmetry and stability.[31] Titanium mesh has many advantages: it is solid, easily molded, and easily fastened.[32]

AIM OF THE WORK

The extent of resection of orbitocranial FD lesions is a matter of debate. Conservative shaving is widely used in the treatment of FD;

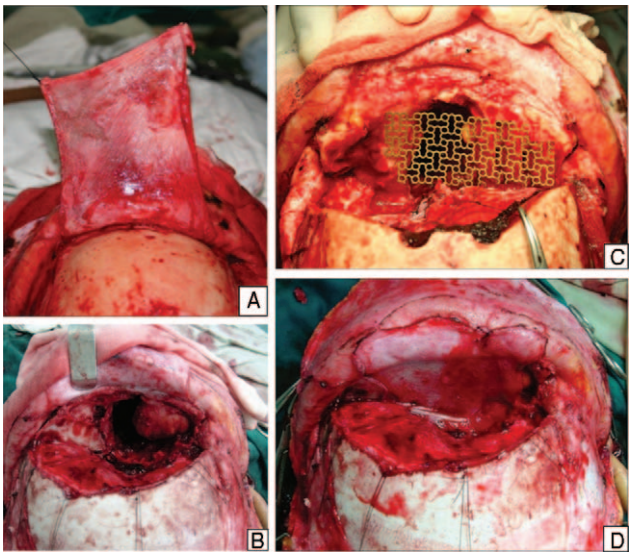

FIGURE 1. The technique of reconstruction of the defect using titanium mesh and pericranial flap. (A) Harvesting the vascularized pericranial flap, (B) the orbitocranial defect after radical resection of the lesion, (C) reconstruction of the defect with titanium mesh, and (D) separation of the anterior cranial base from the sinonasal cavities by vascularized pericranial flap.

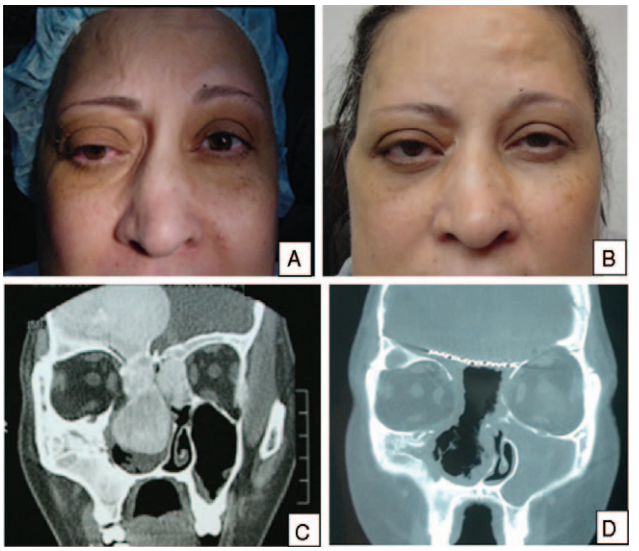

FIGURE 2. A 42-year-old female patient with right orbitocranial fibrous dysplasia treated with radical resection and reconstruction by titanium mesh and pericranial flap. (A) The patient before surgery, (B) the patient after surgery, (C) preoperative CT scan, and (D) postoperative CT scan.

however, its role in orbitocranial is limited. Radical resection is the preferred treatment. Its outcome has not been evaluated before in our patients. Hence, the aim of this study is to evaluate the outcome of radical excision of the orbitocranial FD and immediate reconstruction using titanium mesh and pericranial flap.

PATIENTS AND METHODS

This prospective study had been conducted in the Maxillofacial Surgery Department in cooperation with the Neurosurgery Department, Sohag University Hospital, Sohag, Egypt, in the period from June 2011 to April 2016.

This study included all newly diagnosed patients with FD involving the orbitocranial region. All patients signed an informed consent form and the study was approved by the local institutional research and ethics committee. The exclusion criteria included patients with recurrent disease, optic canal involvement, and patients with other concurrent neurosurgical and visual problems not related to this disease and those who refused to consent to be enrolled in this study.

All patients were subjected to thorough clinical evaluation and assessment of ocular function and neurosurgical evaluation. Essential investigations were done including CT scan for all patients and MRI in selected patients with visual impairment.

The indications for surgery were craniofacial deformity, visual affection, headache, nasal obstruction, anosmia, and/or proptosis. Radical excision of the lesion was done for all patients through transcranial approach. Reconstruction of the orbitocranial defect was done by titanium mesh. The anterior cranial base was separated from the sinonasal cavities by vascularized pericranial flap (Figs. 1–2).

The titanium mesh for the orbital roof and medial and lateral orbital walls was prefabricated using a standard skull model. Intraoperative and postoperative antibiotics and steroids were used for 5 days.

All patients were followed up and evaluated at first postoperative day, and 1, 2, and 4 weeks postoperatively, and then every 3 months during the first year and every 6 months in the remaining follow-up period. Craniofacial CT was performed 12 months postoperatively.

All patients were evaluated during surgery and postoperatively as regarding cerebrospinal fluid leak, infection, visual impairment, diplopia, supraorbital anesthesia, cosmetic appearance, and recurrence.

RESULTS

This study included 22 patients; 10 (45.5%) were males and 12 (54.5%) were females. Their age was from 17 to 52 years with a mean of 29.5 years. The commonest age group affected at presentation was in the third decade (Table 1).

The most common presentations were craniofacial deformity, proptosis, and headache (Table 2). Bone scan revealed 16 monostotic and 6 polystotic patients. The most commonly affected orbitocranial areas were the orbital walls, cribriform plate, and sphenoidal and ethmoidal bones (Table 2). Radical excision was done for all patients through transcranial approach.

Cerebrospinal fluid leak was identified in 2 patients who had basal dural tears during surgery and were managed by direct repair and galeal grafts. Six patients developed supraorbital anesthesia, which was temporary in 4 patients while was permanent in 2 patients. Wound infection occurred in 1 patient and resolved by medical treatment. Temporary postoperative diplopia was reported in 1 patient and improved by time after 2 months. Another patient had temporary postoperative impaired vision, which improved after 2 weeks of corticosteroid therapy.

TABLE 1. Sociodemographic Characteristics of the Patients of the Study

Item	No. (%)
Gender	
Male	10 (45.5%)
Female	12 (54.5%)
Age group, y	
11–20	4 (18.2%)
21–30	10 (45.5%)
31–40	6 (27.3%)
>41	2 (9%)

TABLE 2. Clinical Characteristics and Site of Affection

Item	Frequency
Clinical characteristics	
Craniofacial deformity	15 (68.2%)
Proptosis	6 (27.3%)
Headache	5 (22.7%)
Nasal obstruction	5 (22.7%)
Orbital dystopia	4 (18.2%)
Anosmia	3 (13.6%)
Site of affection*	
Orbital walls	17 (77.2%)
Cribriform plate	16 (72.7%)
Sphenoid	14 (63.6%)
Ethmoid	13 (59.1%)
Frontal bone	5 (22.7%)
Frontal sinus	4 (18.2%)
Maxilla	4 (18.2%)
Extra-axial mass	4 (18.2%)

NB: >1 bony area can be affected in 1 patient.
*Detected by radiological evaluation.

TABLE 3. Complications and Outcome

Complications and Outcome	No. of Patients (%)
Dural tears	2 (9.1%)
Supraorbital anesthesia	
Temporary	4 (18.2%)
Permanent	2 (9.1%)
Infection (resolved by medical treatment)	1 (4.5%)
Temporary postoperative visual impairment	1 (4.5%)
Temporary postoperative diplopia	1 (4.5%)
Aesthetic results	
Good or acceptable	20 (90.9%)
Fair	2 (9.1%)
Recurrence	0 (0%)

In all patients, acceptable or good aesthetic results were observed as described by the patients or their relatives. No recurrence was detected in our series during the follow-up period, which ranged from 24 to 58 months (mean 37.5 months) (Table 3).

DISCUSSION

The management of cranio-orbital FD represents a dilemma for the surgeons because of the anatomic confines of the orbit and cranial base. The aim of the treatment of FD is to correct or prevent functional problems and to achieve an aesthetic improvement.[3,33]

The surgical treatment of FD can be executed through 2 different approaches, conservative or radical. Some authors divided the craniofacial skeleton into 4 zones on the basis of surgical, aesthetic, and functional considerations. They adopted conservative treatment for the alveolar part of the maxilla and mandible and for the cranial base, and the radical approach for the fronto-orbital area and the maxillozygomatic complex.[33,34]

The most important functional area or zone, according to Chen and Noordhoff's[34] scheme, is that of the cranial base and orbit. Surgical access is limited and, if there is functional compromise, conservative surgery may not offer complete or long-term maintenance of function if there is extensive disease. This was the condition of most of our patients who had disease involving the orbit and anterior cranial base and may be extending to the contralateral side. Therefore, early radical surgery that uses craniofacial and microsurgical techniques with immediate reconstruction may offer optimal results in terms of ultimate control of the disease as well as improved aesthetics.

In our study, complete radical resection was done for all patients. In agreement with our protocol, most authors are in favor of radical surgical therapy, which permits the complete removal of the lesion followed by immediate reconstruction.[2,3] It also prevents the occurrence of relapses, eliminates the illness, and has a positive psychological effect for the patient. This agrees with Posnick[35] who states that the treatment of choice should be radical as it allows for the complete resolution of the illness with immediate functional and aesthetic recovery. In accordance with Ozek et al,[24] we found that orbital hypertelorism, dystopia, or proptosis can be corrected only by radical excision and reconstruction (Fig. 3).

One patient of our series experienced temporary postoperative visual impairment where CT scan of the orbit (optic canal) was done and no compression on the orbital cone was detected. This was attributed to edema[36] and it improved after 3 weeks with corticosteroid administration. Brusati et al[37] noticed that temporary postoperative visual impairment after optic canal decompression is often noted; however, it improves with corticosteroid administration and time as the edema subsides.

Resection of tumors arising in the anterior skull base may create extensive skull base defects and produce a free conduit between the paranasal sinuses and the intracranial space. These defects require precise and durable reconstruction to form a watertight dural seal, provide a barrier between the contaminated sinonasal space and the sterile subdural compartment, prevent airflow into the intracranial space, maintain a functional sinonasal system, and provide a good cosmetic outcome.[38]

We used titanium mesh and pericranial flap for immediate reconstruction of the defect to separate the sinonasal cavity from cranial cavity. This procedure can maintain the original anatomy and eliminate donor-site morbidity from the grafting of large amounts of autogenous bone.

Like the majority of authors,[39,40] we feel that the pediculate pericranial flap is the best system in our patients because it is simple, readily available, easily harvested, and powerful and has guaranteed vascularization.

Titanium mesh systems present several advantages, such as excellent biocompatibility, easy handling and shaping, reasonable

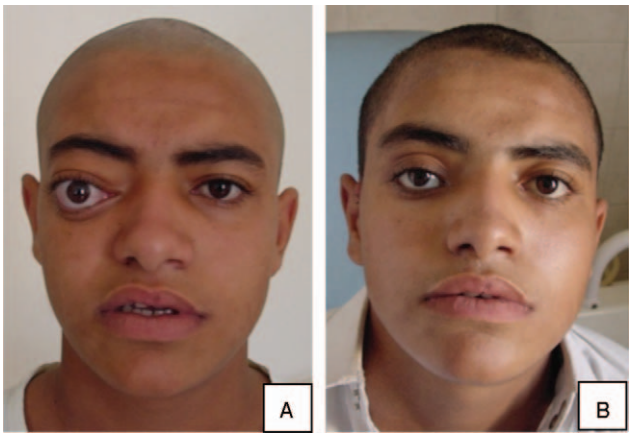

FIGURE 3. A 27-year-old male patient treated with our technique. (A) Before surgery, hypertelorism, and orbital dystopia; (B) after surgery, hypertelorism, and orbital dystopia are corrected.

stability, and versatility.[41,42] Moreover, a minimal imaging artifact can be observed on magnetic resonance and CT imaging.[43,44] The titanium mesh is an ideal material for frontal sinus restoration because it has low susceptibility to infection, even when it is in direct contact with paranasal sinuses.[42]

Reconstruction of the roof and/or lateral orbital wall has been considered unnecessary by some authors; Maroon et al[45] argue that they had no pulsating exophthalmos in >200 patients treated with orbital decompression; Schick et al[46] reconstruct the lateral orbit wall, but they consider further reconstruction of the orbital roof unnecessary. However, radical resection of the involved bone without proper reconstruction may lead to postoperative aesthetic and functional complications such as meningocele formation, diplopia from extraocular muscle fibrosis, orbital pain, pulsating enophthalmos, and restrictive ptosis.[47]

In our study, orbitocranial reconstruction was achieved by titanium mesh. We used a standard skull model preoperatively to shape and cut the mesh, a technique described by Andrades et al[48] for the treatment of floor and medial orbital wall fractures. This was found as an easy, simple, and inexpensive technique with excellent results.

Regarding the outcome, we found that radical resection of orbitocranial FD is potentially curative with no extra morbidity. It can achieve good aesthetic and functional results with most of the encountered complications being temporary and improved by either time or medical treatment. No major or disabling long-term morbidities were recorded. No recurrence was detected.

CONCLUSION

Radical excision of orbitocranial FD is potentially curative with no extra morbidity. Immediate reconstruction using titanium mesh and pericranial flap can achieve good aesthetic and functional results.

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Tympanic Membrane Perforation Caused by Thunderbolt Strike

Nazim Bozan, MD, Ahmet Faruk Kiroglu, MD,* Muzaffer Ari, MD,† Mahfuz Turan, MD,* and Hakan Cankaya, MD**

Abstract: Thunderbolt strike is a life-threatening entity. In the patients presenting with thunderbolt strike, the central nervous system and the cardiovascular system should be primarily evaluated, followed by tympanic membrane lesions. The most important symptom connected with ear is the loss of hearing. In this report, a 43-year-old male patient with unilateral tympanic membrane perforation, tinnitus, and bilateral sensorineural hearing loss caused by thunderbolt strike is presented through a review of the literature.

Key Words: Hearing loss, lightning injuries, sensorineural, tympanic membrane perforation

Lightning flashes leap from cloud to cloud, from cloud to earth, or from earth to cloud in high-structured areas. It is called lightning if the visible discharge of the electricity occurs between 2 clouds, whereas it is defined as a discharge of lightning accompanied by thunder when it occurs between the earth and the cloud, which is abrupt and vehement discharge of electricity.[1,2]

The electrical power in only 1 single flash of lightning is approximately 200,000 ampere and can heat its route up to 28,000°C in a period of a millisecond or less. This explosive heating causes dilation of the weather that generates a tumultuous energy which is called the "clap of thunder." Lightning and thunderbolt track the route that has the least electrical resistance.[1,3,4]

The impact of electric current on body differs through 6 factors. These factors include the route that the current tracks, the resistance of the tissues, voltage, amperage, the contact period, and the current as being alternating or direct current.[3] The current in the thunderbolt flows toward lower resistance points.[4] Cartilage and bone have a high level of resistance since they have low electrolyte and liquid content. The resistance of muscle and tendon tissues is low but their conductivity is better. There are various impacts of the electrical energy on the living tissues, such as affecting the permeability of cell membranes. Additionally, the risen heat causes denaturation of the tissue proteins, and the neuromuscular, cardiovascular, and central nervous systems are also affected.[5]

Clinical Report

A 43-year-old patient who had been struck by a thunderbolt was admitted to our emergency department with the complaints of hearing loss, earache, and tinnitus on both ears. The patient had 2 lacerations measuring 2 cm in size on the skin. There were also 10% first-degree and 5% second-degree burns on his neck, back, the inner side of the right arm, chest, and scrotum.

An automicroscopic examination revealed 2 sites of perforation with hemorrhagic edges on the left tympanic membrane (Fig. 1A). The middle ear mucosa was normal. The right tympanic membrane was normal in appearance. In a computed tomography scan, the temporal bone was normal. An audiogram revealed a level of 57 dB sensorineural hearing loss (SNHL) in the right ear and 107 dB SNHL in the left ear. The patient was hospitalized in the general surgery department for his burns and was followed up by an audiometrist once in 2 days. In the meantime, the patient was given intravenous pentoxifylline 400 mg 3 × 1 and prednisolone (1 mg/kg). Approximately 48 hours after the treatment, a marked recovery was achieved in terms of tinnitus and SNHL. In the second audiogram, the SNHL was decreased to 52 dB in the right ear and to 80 dB in the left ear. The treatment was continued for 10 days. The patient was discharged from the hospital and was advised to take Ginkgo flavonglycosides 19.2 mg 3 × 1 and trimetazidine dihydrochloride 35 mg 2 × 1 perorally.

During the follow-up, the double perforation became a single perforation (Fig. 1B). Moreover, tinnitus gradually decreased but did not recover completely. The patient was followed up once a month for a period of 3 months. Since the perforation remained unhealed, tympanoplasty was performed. The last audiometry prior to the surgery showed a mixed-type hearing loss of 33 dB in the right ear and 48 dB in the left ear.

One month after the surgery, the hearing loss was reduced to 30 dB in the right ear and to 38 dB in the left ear (mixed-type

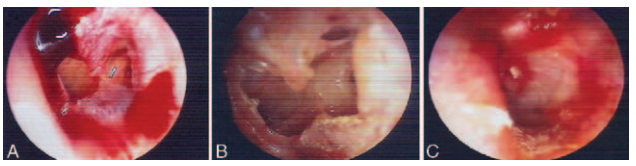

FIGURE 1. (A) Double perforation on the tympanic membrane (Day 1). (B) Tympanic membrane after 15 days (a single perforation). (C) Intact graft at postoperative month 12.

From the *Department of Otorhinolaryngology, Medical Faculty, Yuzuncu Yil University, Van; and †Department of Otorhinolaryngology, Mardin State Hospital, Mardin, Turkey.
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Address correspondence and reprint requests to Nazim Bozan, MD, Department of Otorhinolaryngology, Medical Faculty, Yuzuncu Yil University, 65080 Van, Turkey; E-mail: drnzmbozan@hotmail.com
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