Optic Nerve Decompression for Orbitofrontal Fibrous Dysplasia: Recent Development of Surgical Technique and Equipment

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ABSTRACT

Objectives: Orbitofrontal fibrous dysplasia often involves the bony orbit and optic canal. Although fibrous dysplasia reportedly produces compression of the optic nerve leading to visual disturbances, optic nerve decompression in patients without clinical signs of optic neuropathy remains controversial. We describe the recent development of surgical techniques and equipment for optic nerve decompression in orbitofrontal fibrous dysplasia. **Methods:** Optic nerve decompression was performed prophylactically for five patients and therapeutically for one patient using the transcranial extradural route. A high-speed drill and continuous suction-irrigation system has been used in five patients since 1998, and an ultrasonic bone curette in two patients since 2004. **Results:** The continuous suction-irrigation system was particularly effective for decreasing heat transfer and thus preventing thermal injury to the optic nerve from the high-speed drill. The ultrasonic bone curette was also effective, allowing bone removal with minimal pressure from the tip of the handpiece and without catching cotton pledgets or damaging surrounding tissues. Orbital dystopias and craniofacial deformities induced by fibrous dysplasia were also successfully corrected. Postoperatively, disturbance in visual function was present in only two patients. Mean follow-up period was 4.9 years. **Conclusions:** This equipment may contribute to the development of new modalities for optic nerve decompression in orbitofrontal fibrous dysplasia.

KEYWORDS: Fibrous dysplasia, optic nerve decompression, suction-irrigation system, ultrasonic bone curette

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Skull Base 2006;16:145-156. Copyright © 2006 by Thieme Medical Publishers, Inc., 333 Seventh Avenue, New York, NY 10001, USA. Tel: +1(212) 584-4662.

Received: October 10, 2005. Accepted after revision: December 10, 2005. Published online: August 21, 2006.
DOI 10.1055/s-2006-949517. ISSN 1531-5010.

Fibrous dysplastic lesions frequently involve the anterior skull base and result in encasement of the optic nerve canal. Orbitofrontal fibrous dysplasia may develop within bone adjacent to the optic canal, grow gradually, and compress the optic nerve leading to visual disturbances.¹⁻¹² It has recently been suggested that prophylactic decompression of the optic nerve is not indicated as the presence of fibrous dysplasia does not correlate with visual loss.¹³ However, when visual impairment begins it tends to be progressive^{1-3,5,9-12,14} and may continue after puberty.^{4,6,15,16} Malignant or sarcomatous degeneration is reported to happen in 0.5% of patients.¹⁷ The cause of both acute and gradual loss of vision in patients with fibrous dysplasia remains unclear and the pathological course is unknown. Optic nerve decompression in patients without clinical signs of optic neuropathy thus remains very controversial. The controversy focuses on whether surgical relief should be instituted before moderate-to-severe loss of vision has been established or delayed until visual loss must be followed by emergency decompression.^{1,3,12,14,18,19} New surgical equipment such as the high-speed drill, continuous suction-irrigation system,^{20,21} and ultrasonic bone curette^{22,23} have recently been developed. We have already reported the usefulness of combined methods with the high-speed drill and continuous suction-irrigation system for optic nerve decompression in orbitofrontal fibrous dysplasia.²⁴ We recently performed optic nerve decompression using ultrasonic bone curette in two patients with orbitofrontal fibrous dysplasia. The present article describes patients with orbitofrontal fibrous dysplasia who underwent optic nerve decompression. We discuss characteristics of the new equipment.

PATIENTS AND METHODS

Patients

Surgical correction of orbital dystopias and craniofacial deformities has been undertaken on

11 patients with fibrous dysplasia over the past 20 years. We reviewed the outcomes of our last 6 patients (3 men, 3 women) with orbitofrontal fibrous dysplasia who underwent optic nerve decompression. Table 1 documents our patients' clinical details. Mean age at the time of surgery was 25 years (range, 16 to 55 years). The underlying pathology was polyostotic fibrous dysplasia in 4 patients and McCune-Albright syndrome in 2 patients. One patient with McCune-Albright syndrome also had a growth hormone-secreting pituitary adenoma (Table 1, Patient 5). Neuro-ophthalmologic examination revealed normal visual function in all patients except for 1 patient with McCune-Albright syndrome (Table 1, Patient 2: visual acuity, 0.5 in the affected eye). Radiological examination showed fibrous dysplasia affecting the optic nerve canal in all patients. Mean duration of follow-up was 4.9 years (range, 6 months to 10 years). Descriptions of optic nerve decompression in 3 patients (Patients 2 to 4) have been published previously.²⁴⁻³¹

Surgical Equipment

High-speed drills (ANSPACH[®], The Anspach Effort, West Palm Beach, USA, or Midas Rex[®], Medtronic, Fort Worth, USA) have been used since 1998 so that any heat resulting from friction at the bone edge is removed quickly, minimizing conduction to surrounding tissue. High-speed drilling produces significantly less heat than low-speed drilling.

A suction-irrigation system with continuous irrigation and suction is mandatory to decrease heat transfer and to prevent thermal injury to the optic nerve. The irrigation system, which can be manipulated with one hand, effectively cleans the operative field and decreases heat transfer with a rapid flow of saline. A suction-irrigation system designed for trans-sphenoidal surgical removal of pituitary tumors by Lüdecke and Treige²⁰ (Waldemar Link, Hamburg, Germany) was adopted in 1998. Since 2004, a new type of suction-irrigation system developed by Kamiyama and colleagues²¹

Table 1 Characteristics of the Six Patients with Orbitofrontal Fibrous Dysplasia Who Underwent Optic Nerve Decompression

Patient	Age*/ Sex	Diagnosis	Eye	Indication	Surgery (year)	Equipment	Postoperative Vision†		Follow-up (years)
							Visual Acuity	Visual Field	
1	23/M	Polyostotic fibrous dysplasia	Right	Prophylaxis	1990	LSD	1.2	Lower quadrant hemianopia	10
2	55/F	McCune-Albright syndrome	Left	Gradual disturbance	1998	SIS, HSD	1.5	Normal	7
3	16/F	Polyostotic fibrous dysplasia	Left	Prophylaxis	1999	SIS, HSD	0.03 (1.0)‡	Normal	6
4	18/M	Polyostotic fibrous dysplasia	Right	Prophylaxis	2000	SIS, HSD	1.2	Normal	5
5	16/M	McCune-Albright syndrome Gigantism	Left	Prophylaxis	2004	SIS, UBC, HSD	0.15 (1.0)	Normal	1
6	24/F	Polyostotic fibrous dysplasia	LEFT	Prophylaxis	2005	SIS, UBC, HSD	0.4	Lower half hemianopia	0.5

*At time of surgery.

†In affected eye.

‡Visual acuity 0.03, correctable to 1.0 with lenses.

LSD, low-speed drill; SIS, suction-irrigation system; UBC, ultrasonic bone curette; HSD, high-speed drill.

Figure 1 (A) Lateral view of handpiece and tip of suction-irrigation system. (B) Upper view of handpiece. (C) Flexible tips.

(Ohwa, Tokyo, Japan) has been used for optic nerve decompression. This system comprises a handpiece with a suction-irrigation tube and fluid pressure-generating system (Fig. 1A). The light weight of the handpiece promotes stability and easy handling for transcranial surgery. The pressures for suction and irrigation can be adjusted using a thumb control in the handpiece (Fig. 1B). The tips of the handpiece are extremely flexible (Fig. 1C).

The ultrasonic surgical device (Sonopet UST-2001; Miwatec, Kawasaki, Japan) comprises a power supply unit, footswitch, and handpiece. The handpiece weighs 110 g, is 210 mm long, and is 22 mm in diameter (Fig. 2A). The tip is 1.9 mm wide (Figs. 2B, C). Longitudinal-torsional vibration amplitude is 320 μ m at an ultrasonic frequency of 25 kHz. The adjusted cool-controlled irrigation fluid (20°C) emerges near the tip of the handpiece²² through the sheath, but no suction equipment is attached. The surgeon holds the handpiece with his or her dominant hand and a suction-irrigation system in the other hand to evacuate blood, irrigation fluid, and bone dust. The ultrasonic bone curette is able to remove bone without catching on cotton pledgets.

Surgical Technique

Our surgical technique has already been described in detail elsewhere.²⁴ Surgical approaches to the optic canal must adequately expose the deep orbital apex and optic nerve to allow ablation and decompression. All optic nerve decompressions at our institution have been performed by the same surgeon (TA) except for the first patient in 1990. Optic nerve decompression was performed prophylactically for five patients and therapeutically for one patient with McCune-Albright syndrome through the transcranial extradural route. Optic nerve decompression has been performed together with correction of orbital dystopias and craniofacial deformities induced by fibrous dysplasia: surgery has never been performed for optic nerve decompression alone. Optic nerve decompression for the first patient in 1990 (Table 1, Patient 1) was performed using a low-speed drill. A high-speed drill and continuous suction-irrigation system has been used in five patients since 1998, and the ultrasonic bone curette in two patients since 2004. Thus, in the most recent two patients, all of these instruments have been used.

Figure 2 (A) Handpiece and tip of the Sonopet UST-2001 (110 g). (B) Frontal view of tip (width, 1.9 mm). (C) Lateral view of angled tip for resecting bone using a scratching motion.

First, lumbar drainage is established to prevent injury when the brain is later retracted. A frontoparietal craniotomy was then performed 1.5 to 2.0 cm above the superior orbital rim. The supraorbital rim on the side is removed along with a frontoparietal cranial bone flap. The superior orbital roof can then be resected carefully using a high-speed drill with a diamond burr using a microscope. A suction-irrigation system with continuous irrigation and suction at the site of drilling is mandatory to decrease heat transfer and prevent thermal injury to the optic nerve. The combined method using the high-speed drill and continuous suction-irrigation system was used for resection of large amounts of abnormal bone distant from the optic nerve in the two most recent patients. The ultrasonic bone curette was used for resection of abnormal bone around the optic nerve canal. Drilling or curettage directly over the optic canal should be avoided as vibratory and thermal energy can be transferred through the bone and injure the optic nerve.

Dura mater underlying the optic canal was protected using cotton pledgets. The tip of the handpiece was inserted between the bone edge of the optic canal and underlying cotton pledgets and was used to remove abnormal bone in the same manner as scratching with a curette. Use of the ultrasonic bone curette allows the surgeon to remove fibrous dysplasia rapidly with minimal pressure on the tip of the handpiece and without catching cotton pledgets. Once the affected bone had been thinned at the posterior orbital cone, a sphenoid punch was used to remove the roof of the optic canal in pieces. Extreme care was needed to avoid compression of the optic nerve. The entire roof of the optic canal was removed to decompress the nerve completely. The orbital roof and lateral wall were reconstructed from iliac bone and/or split cranial bone and secured using microplate or titanium wires to ensure structural integrity. The supraorbital rim can be reconstructed from calvarial bone or ribs and secured with microplates.

SURGICAL RESULTS

The suction-irrigation system was particularly effective in decreasing heat transfer and preventing thermal injury to the optic nerve from the high-speed drill. Fibrous dysplastic bone was resected easily with minimal pressure from the tip of the ultrasonic bone curette against the bone surface through simple scratching motions toward the surgeon, as if with a curette, without catching cotton pledgets or damaging surrounding tissues. Orbital dystopias and craniofacial deformities induced by fibrous dysplasia were also successfully corrected in all our patients. After surgery, none of the patients displayed brain contusion, cerebrospinal fluid leakage, or infection. Disturbance of visual function was identified only in the first and most recent patients (Table 1, Patients 1 and 6, respectively). Patient 1 displayed lower quadrantic hemianopia in the affected eye after surgery. In the most recent two patients who underwent optic nerve decompression with the ultrasonic bone curette, although one patient displayed no change to visual function, the other patient showed lower-half hemianopia in the affected eye after surgery. No patients exhibited disturbance of visual acuity in the affected eye after surgery (Table 1). In one patient with gradual loss of visual function preoperatively (Table 1, Patient 2), visual acuity in the affected eye improved markedly from 0.5 to 1.5. Subsequent orbital reconstruction achieved a satisfactory cosmetic result. While the use of a high-speed drill is better suited for resecting large amounts of bone due to rapid drilling, ultrasonic bone curettage provides much less risk of injury to fragile tissues like the optic nerve.

Illustrative Cases

Patient 5 (Fig. 3) was a 16-year-old boy with McCune-Albright syndrome and gigantism caused by a growth hormone-secreting pituitary adenoma. The pituitary adenoma was subtotally resected using a right-sided transcranial approach followed by

stereotactic radiosurgery. Computed tomography (CT) demonstrated extensive orbitofrontal fibrous dysplasia surrounding and narrowing the left optic canal (Figs. 3A to C). Visual function was normal. The fibrous dysplasia was largely resected and a left-sided optic nerve decompression was performed through the transcranial extradural route using a high-speed drill, suction-irrigation system, and ultrasonic bone curette without damaging surrounding tissues (Fig. 3D). The orbital roof was reconstructed using split cranial bone grafts and two ribs (Fig. 3E). Follow-up CT confirmed an enlarged optic canal (Figs. 3F, G). The post-operative clinical course was uneventful, and both the visual acuity and visual fields were normal 1 year after surgery (Fig. 3H).

Patient 6 (Fig. 4) was a 24-year-old woman with facial asymmetry due to a left-sided protuberant periorbital region and frontal bossing. CT revealed left-sided orbitofrontal fibrous dysplasia and narrowing of the optic canal (Figs. 4A to C). Visual function was normal. Her fibrous dysplasia was resected and a prophylactic left-sided optic nerve decompression was performed via the transcranial extradural route using a high-speed drill, suction-irrigation system, and ultrasonic bone curette (Figs. 4D, E). The orbital roof was reconstructed using split cranial bone graft. Follow-up CT showed an enlarged optic canal (Figs. 4F to H). Lower-half hemianopia was noted in the affected eye after surgery (Fig. 4I).

DISCUSSION

The causes of both acute and gradual loss of vision associated with fibrous dysplasia remains unclear. Acute visual loss has been reported in association with mucocoeles,^{2,4,5,18} hemorrhage,^{5,25} and hemorrhagic cysts⁵ and with fibrous dysplasia when the optic canal is involved.^{1,3,4,19,26,27} Chronic, gradual visual decline is also associated with fibrous dysplasia,^{1,4} although this phenomenon is commonly thought to reflect optic nerve compression within

Figure 3 Patient 5. (A) Preoperative three-dimensional CT. (B) Preoperative axial bone CT. (C) Preoperative coronal bone CT. (D) Optic nerve decompression. White lines: optic nerve canal. (E) Reconstruction of orbital roof. (F) Postoperative axial bone CT. (G) Postoperative coronal bone CT. (H) Postoperative visual field illustration for left eye demonstrating normal visual field. CT, computed tomographic scan.

a stenotic foramen.^{1,4,5,28,29} Optic nerve decompression is indicated for patients with visual deterioration. However, appropriate management of fibrous dysplasia around the optic nerve in patients with normal vision remains controversial. Prophylactic decompression of the optic nerve represents one option,^{1,4,8,24,28,30} while regular ophthalmo-

logic examination in patients with asymptomatic encasement is another.¹³ Two studies have strongly advocated prophylactic decompression of the optic nerve for treating fibrous dysplasia.^{1,28} Chen and associates¹ recommended prophylactic decompression in two circumstances: (1) with radiological involvement of the optic canal, which is associated

Figure 4 Patient 6. (A) Preoperative three-dimensional CT. (B,C) Preoperative axial bone CT. (D) Bone resection using ultrasonic bone curette. Black asterisk: continuous suction-irrigation system. White asterisk: ultrasonic bone curette. Arrow: optic nerve canal. (E) Optic nerve decompression. Arrow: decompressed optic nerve canal. (F,G) Postoperative axial bone CT. (H) Postoperative coronal bone CT. (I) Postoperative visual field illustration for left eye showing lower-half hemianopia. CT, computed tomographic scan.

with a reasonably high incidence of visual loss in the affected eye and a very high incidence of some visual disturbance; and (2) if the speed at which visual deterioration develops is rapid, because any defect may not be reversible. Recently, Lee and others¹³ stated that prophylactic decompression of the optic nerve should not be undertaken on the basis of diagnostic imaging alone, as CT findings of encasement or constriction of the optic canal did not correlate with loss of vision, and fibrous dysplasia was not progressive in the region of the optic nerve. They stressed that the risks associated with optic nerve decompression include lack of visual improvement and postoperative blindness.^{1,6,8,10,19,28,31} These findings cannot be confirmed without long-term studies of appropriate patient populations. In our series, optic nerve decompression has always been performed together with correction of orbital dystopias and craniofacial deformities induced by fibrous dysplasia. No patients underwent optic nerve decompression alone. The efficacy of prophylactic decompression of the optic nerve is difficult to assess as by definition patients display no preoperative visual loss. Since predicting the arrest of pericanal fibrous dysplasia is difficult, the growth of fibrous dysplastic bone should be assumed likely to continue and optic nerve compression should be assumed to be relatively imminent.³² Prophylactic decompression of the optic nerve is likely to be beneficial on the theoretical grounds that once vision has been impaired, chances of restoring vision are greatly reduced.

Optic nerve decompression should be performed by an experienced neurosurgeon to minimize the risk of iatrogenic damage to the optic nerve. Although several approaches have been reported for optic canal decompression, the unilateral frontotemporal extradural approach may represent the best method. The extent of decompression in the optic canal is likely to be a factor in recurrence, as any residual bone is likely to contain fibrous dysplasia.¹ Extensive decompression is required and the risk associated with intracranial facial surgery may necessitate the transcranial extradural approach.

Drilling can cause temperature elevation of bone to 70° C around the drill tip,^{33,34} so continuous irrigation with cool-controlled fluid is required to prevent thermal damage to the optic nerve. High-speed drilling produces significantly less heat than low-speed drilling.³⁵ With high-speed drills, the bone edge heated as a result of friction is removed quickly, minimizing conduction of heat to surrounding tissue. The first patient who underwent optic nerve decompression by another surgeon using a low-speed drill alone developed a lower quadrant hemianopia in the affected eye after surgery (Table 1, Patient 1). Some modified systems and equipment have been proposed for continuous irrigation and suction. The suction-irrigation system developed by Kamiyama and colleagues²¹ has an extremely lightweight handpiece, facilitating stability and easy handling for transcranial surgery. Pressures for suction and irrigation are adjusted by a thumb control in the handpiece. The irrigation system, which can be manipulated with one hand, can clean the operating field effectively and decrease heat transfer by a rapid flow of saline.

Ultrasonic bone curettes are used for anterior clinoidectomy of paraclinoid aneurysm, opening of the internal auditory canal for vestibular schwannoma, and spinal surgery without damage to the surrounding normal tissues.^{22,23} The effectiveness of this tool has been reported.^{22,23} The device is so light and easy to use and has such a smooth motion that even beginners in skull base surgery can handle the equipment safely. This equipment enables easy resection of bone with minimal compression on the bone surface and the view of the operating field is not obscured. Although power adjustment is necessary during surgery, the equipment does not include any rotating parts, so the potential for damage to the surrounding tissue and optic nerve is minimized. In addition, the potential for catching cotton pledgets or sutures, as can happen with a high-speed drill, is eliminated. Placement of cotton pledgets on the site may prevent damage to the optic nerve. When resecting hard bone using a high-speed drill, the surgeon must sometimes hold the drill with both

hands to prevent damage to surrounding tissue caused by unexpected movements of the drill. The ultrasonic surgical bone curette^{22,23} can obviate the need for support from assistants and ensures that surgery is not interrupted by allowing the surgeon to use both hands while drilling.

Although heat production associated with use of the ultrasonic bone curette has not yet been investigated, Hadeishi and coworkers²² reported that temperatures in the surrounding bone would not be high, as cool-controlled irrigation fluid (20° C) is used during surgery. However, unfortunately, visual field disturbance was observed after surgery in one patient who underwent ultrasonic bone resection. We paid extremely close attention to avoid compression of the optic nerve, and we avoided drilling or curettage directly over the optic nerve canal, as vibratory and thermal energy can be transferred through the bone, injuring the optic nerve. The roof of the optic canal was removed in pieces using a thin sphenoid punch once the diseased bone was thinned at the posterior orbital cone. The cause of postoperative disturbance of visual field remains unclear. In the first patient for whom the ultrasonic bone curette was used (Table 1 Patient 5), optic nerve decompression was performed with continuous irrigation and suction. In the second patient (Table 1 Patient 6) compression sometimes happened but was eased by protective cotton pledgets and without continuous use of irrigation and suction. Protection of the dura mater using cotton pledgets may have been incomplete. Visual field disturbance may thus have resulted from heat or vibration produced by the ultrasonic equipment. Additional research is required to evaluate heat production and the effects of shock waves generated by the equipment in normal tissue, particularly in the optic nerve. However, the ultrasonic bone curette seems to represent an extremely safe and effective method for optic nerve decompression in orbitofrontal fibrous dysplasia. The equipment may contribute to the development of new modalities for optic nerve decompression in orbitofrontal fibrous dysplasia.

ACKNOWLEDGMENTS

None of the authors has any financial interest in the equipment described in this paper.

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