Computer-Based Surgical Planning and Custom-Made Titanium Implants for Cranial Fibrous Dysplasia

BACKGROUND: The procedure of reconstruction after the removal of cranial fibrous dysplasia (FD) must be precise to achieve good functional and aesthetic results. Intraoperative modeling of implants is difficult and may cause cosmetic disturbances.

OBJECTIVE: To present our experience with the treatment of cranial FD using preoperative computer-based surgical planning of tumor removal with reconstruction of the cranium with custom-made titanium implants.

METHODS: Four patients underwent surgical treatment for cranial FD over a 2-year period. All patients were male with a mean age of 25.25 years and had monostotic-type FD. Computed tomography (CT) with 0.5-mm slices was obtained preoperatively. Computer-based planning of the tumor removal was performed, and a template was created by the computer to determine the margins of tumor removal. After this procedure, the preoperative computer-based construction of the titanium implant was performed. The patients underwent surgical treatment, and the tumor was removed with the use of this template. Then, the titanium implant was inserted onto the bone defect and fixed with mini-screws. Patients were followed up by periodic CT scans.

RESULTS: The histological diagnosis of all patients was FD. No intraoperative or postoperative complications have occurred. Postoperative CT scans showed complete tumor removal and confirmed appropriate cosmetic reconstruction. The mean follow-up period was 15.25 months.

CONCLUSION: Computer-based surgical planning associated with the production of custom-made titanium implants is a highly promising method for the treatment of cranial FD. Better radiological and cosmetic outcomes could be obtained by this technique with interdisciplinary work with medical designers.

KEY WORDS: Cranium, Computer-based planning, Custom-made cranioplasty, Fibrous dysplasia

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ibrous dysplasia (FD) is a benign bone tumor that most commonly involves the cranial and facial region.¹ It is rare and makes up 2.5% of all bone tumors.² Replacement of normal bone with immature bone marrow stromal cells is the possible pathophysiological mechanism of FD.³ The symptoms associated with FD are cosmetic deformities and visual disturbances, including blurred vision, diplopia, and epiphora.^{4,5} Loss of vision secondary to optic nerve compression is the most important consequence of cranial FD.⁶

ABBREVIATIONS: DICOM, digital imaging and communications in medicine; FD, fibrous dysplasia

Surgery is the main treatment method of cranial FD with cosmetic and visual problems. The surgical technique depends on the site and volume of the lesion and the compression of the cranial nerves. Surgical treatment has required complex and technically demanding methods for reconstruction owing to extension of the tumor. Craniectomy, optic canal decompression (if required), and cranial reconstruction are the parts of the surgical treatment for cranial FD. However, different neurosurgical techniques specific to the different indications may be used, including a transsphenoidal approach for resection of sphenoidal sinus FD, a transmaxillary approach to decompress the maxillary branch of the trigeminal nerve with widening of the

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Ozkan Tehli, MD* Ahmet Murat Dursun‡ Caglar Temiz, MD* Ilker Solmaz, MD* Cahit Kural, MD* Murat Kutlay, MD* Yunus Kacar, MD* Mehmet Can Ezgu, MD* Erbil Oguz, MD‡ Mehmet K. Daneyemez, MD* Yusuf Izci, MD*

*Department of Neurosurgery and ‡Medical Design and Production Center, Gulhane Military Medical Academy, Ankara, Turkey

Correspondence:

Yusuf Izci, MD, Department of Neurosurgery, Gulhane Military Medical Academy, 06010 Etlik, Ankara, Turkey. E-mail: yusufizci@yahoo.com

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foramen rotundum, and complete calvarial craniectomy with cranioplasty reconstruction.⁸

The use of computer-based surgery systems is becoming more and more common, especially in neurological surgery. Most of these systems are aimed at ensuring that during the operation the surgeon is able to achieve the implant position previously planned with specific computer tools. On the other hand, there are many controversies concerning the material of choice for cranioplasty after removal of cranial bone tumors. Some authors advocate autogenous materials for reconstruction,⁹ and others suggest methyl methacrylate, titanium, polyetheretherketone, and other alloplastic materials.¹⁰⁻¹² Autologous bone has some disadvantages: The amounts of donor bone are limited; the need to remodel the harvested bone into complex shapes may complicate the operation; the bone may resorb; and there is associated donor-site morbidity.^{9,10,13} Alternatively, alloplastic materials not only replace missing bone but also act as a scaffold for bony regrowth.^{10,11} Titanium alloy is ideal as alloplastic material because of its high tensile strength, light weight, and good resistance to corrosion. There is no associated donor-site morbidity, and both operating time and duration of hospital stay are short.

In this article, we describe our computer-based technique in the surgical treatment of cranial FD and report the radiological and surgical characteristics of our cases. We also discuss our results with the current literature.

METHODS

Patient Population

Four patients with the diagnosis of cranial FD underwent surgical treatment in our department. The patients consisted of 4 men ranging in age from 21 to 36 years (mean, 25.25 years). The main clinical manifestation was swelling (cranial mass lesion) in all patients. All patients underwent computed tomography (CT) and magnetic resonance imaging studies to reach a preoperative radiological diagnosis. Vision test and visual field analysis were done in 1 patient who had a mass lesion in the fronto-orbital region.

Surgical Technique

A preoperative CT scan with 0.5-mm slices was obtained from a helical scanner for each patient to make preoperative computer-based surgical planning and to produce patient-specific titanium implants. The planning of surgery and production of titanium implants were performed in the Medical Design and Production Center of our institution. The CT data were digitally transferred from the scanner console to the planning work station with the use of a DICOM (digital imaging and communications in medicine) interface. The pictures were opened with Mimics Innovation Suite software (Materialise, Leuven, Belgium). The surgeon and medical designer (engineer) performed the surgical planning using planning and image processing software. This software allows the surgeon to delineate the borders of the bone tumors and to "navigate" the titanium implant into the 3-dimensional (3-D) space of the CT data to the proper position. The surgeon can dynamically change the borders of tumor resection and the size and shape of the titanium implant among those available data. After this planning, a titanium mesh template was produced by a 3-D printer (Concept Laser, Hofmann Innovation

Group, Lichtenfels, Germany) to determine the boundaries of bone tumor during the surgery. A cranial model was also created by Z-Corp 650 (3D Systems, Rock Hill, South Carolina) from the data of the CT scan. Surgical resection of the tumor was first performed on this model, and a computer-assisted titanium implant was also virtually inserted into the bone defect. Then, the template and titanium implant were placed in a sterile bag for use during the surgery. A standard skin incision was performed around the mass lesion, and the tumor borders were exposed. The resection margins were determined with the template, and complete resection of the tumor was performed with a high-speed drill. Because none of the patients have optic neuropathy, the optic canal was not unroofed in all patients. The titanium implant, which was created preoperatively, was inserted onto the cranial defect and fixed with mini screws. The length of these mini-screws was also determined by preoperative computer-based planning using 3-matic software (Materialise). The cost of the whole production procedure and titanium implants was approximately US \$2500, depending on the sizes of the tumor and custom-made implant. The duration of surgical planning and implant production was 4 weeks for each patient. Patient satisfaction level was evaluated by use of the Odom criteria (excellent, good, fair, poor) during the follow-up period after surgery.

RESULTS

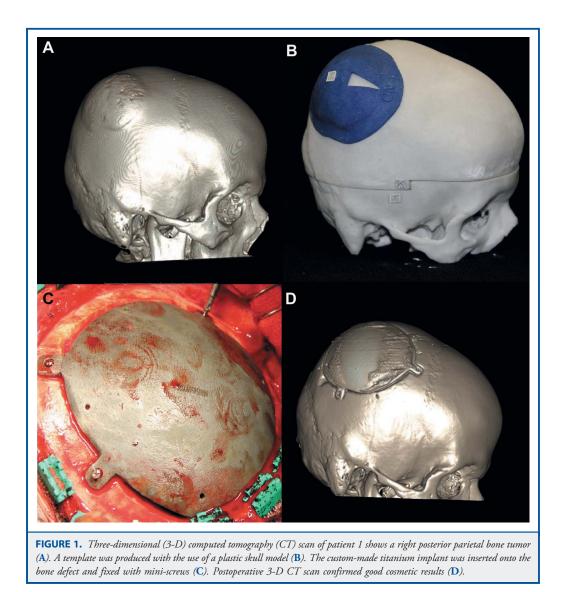
A good cosmetic result was obtained in all patients. The mean duration of surgery was 132.5 minutes (range, 110-160 minutes), and the mean follow-up period was 15.25 months (range, 5-26 months). There were no complications after the surgeries. No implant-related infection was observed in any patient. Histological examination of the tumor specimens confirmed the diagnosis of FD in all patients. Postoperative periodical CT scans of the patients revealed total removal of the tumors. These scans also demonstrated the accuracy of the preformed titanium implants and confirmed appropriate cosmetic reconstruction. Overall satisfaction was excellent (n = 3) or good (n = 1) in all patients. No tumor recurrence was observed in any patient during the follow-up period. The clinical, radiological, and surgical features of 4 patients are summarized below.

Case 1

A 22-year-old male patient presented with a 4-month history of swelling on the right side of the skull. Swelling was progressive and caused cosmetic disturbance for the patient. CT scan revealed a right parietal solid mass lesion (60×22 mm in size) on the cranium. The patient underwent surgical treatment after the computer-based planning and production of patient-specific titanium implant. The tumor was removed en bloc, and cranioplasty was performed with this implant. The duration of surgery was 130 minutes. Postoperative CT scan showed complete removal of the bone tumor (Figure 1). The histological diagnosis was FD. The follow-up period was 26 months, and no complication or recurrence was observed during the follow-up period.

Case 2

A 36-year-old male patient presented with an 18-year history of swelling in the left supraorbital region. The swelling was



progressive in the last 2 years. Cranial CT of the patient showed a left fronto-orbital bony lesion $(45 \times 45 \times 30 \text{ mm} \text{ in size})$ on the cranium. Preoperative computer-based surgical planning was performed with the use of helical CT data, and a titanium implant was created by the computer. Then, surgical tumor removal was performed by drilling the tumor. A CT-based navigation system was used in this patient to confirm the resection limits of tumor. The titanium implant was inserted on the bone defect after tumor removal (Figure 2). The duration of surgery was 160 minutes. The histological diagnosis was FD. The follow-up period of this patient was 16 months, and no complication or recurrence was observed within the follow-up period.

Case 3

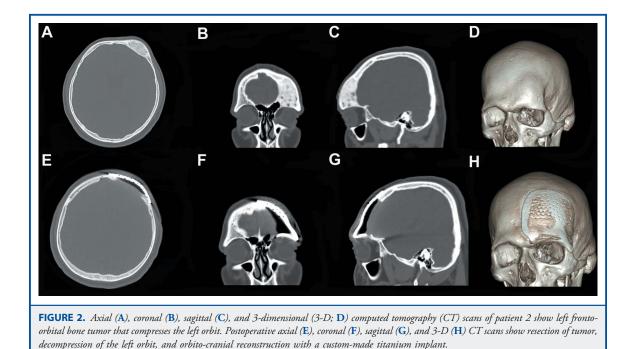
A 22-year-old male patient presented with a 6-month history of swelling on the right side of the cranium. CT scan revealed a right

parietal bone lesion (42×35 mm in size), and the preoperative computer-based surgical planning associated with the production of custom-made titanium implant was performed. Bone tumor was removed en bloc, and the titanium implant was inserted (Figure 3). The duration of surgery was 110 minutes, and the histological diagnosis was FD. The follow-up period of this patient was 14 months. No complication or recurrence was observed within the follow-up period.

Case 4

A 21-year-old male patient presented with a 4-year history of swelling at the back part of the head. CT scan revealed a bony mass lesion (70×22 mm in size) in the occipital part of the cranium. The tumor was removed en bloc after preoperative computer-based planning, and the custom-made titanium implant was inserted onto the bone defect (Figure 4). The duration of surgery

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was 130 minutes, and the histological diagnosis was FD. The follow-up period of this patient was 5 months, and no complication was observed within the follow-up period.

DISCUSSION

Surgical results of 4 cases with cranial FD were presented. Patients underwent surgical treatment using preoperative computer-based surgical planning and custom-made titanium implants. A titanium template was created before the surgery and used during the operation to determine the limits of tumor resection. Good cosmetic results and satisfaction levels were obtained in all patients with this technique. Thanks to computer technology, preoperative resection planning and defect size estimation are possible in association with implant molding before the tumor resection.

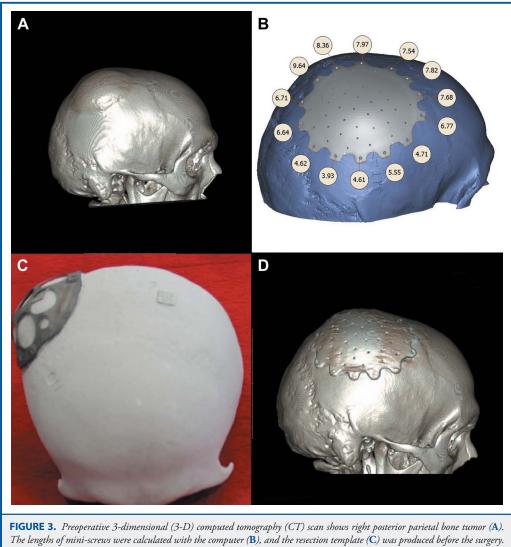
FD is a slow-growing, benign lesion of the cranium. It was first introduced by Lichtenstein¹⁴ in 1938, referring to fibrous tissue replacing normal bone tissue. It represents about 2.5% of all primary bone tumors and 7% of benign bone mass lesions.¹⁵ This disease usually occurs in children and teenagers, but it may also be seen in adults.⁷ For preadolescent patients, the lesion tends to stop progressing with the onset of puberty, but not for all patients. It may involve all parts of the cranium, most commonly the ethmoid and frontal bones.¹⁶ The cause of the disease is still not well understood, but several possible factors based on embryology, genetics, and molecular biology have been proposed.¹⁶ It is believed that a defect in the osteoblastic differentiation and maturation results in the developmental

anomaly of the mesenchymal precursor of bone.¹⁷ Although many patients had trauma history, it seems that trauma is not directly relevant to the disease.¹⁶ It has long been believed that the disease is a benign course of bone malformation, and malignancy is rare, with about 1% of patients having malignant transformation.^{16,18}

The indication for surgery depends on different situations such as the degree of cranial nerve involvement, the severity of symptoms, and the cosmetic demands of the patients. The main purposes of surgical treatment are to correct or prevent neurological problems and to achieve a normal cranial appearance.¹⁶ Cranial reconstruction is necessary for the closure of bone defect secondary to tumor resection. However, the outcome of surgical correction often depends on surgeon experience and the approach to preoperative planning. Today, customized surgical implants are in use, and the benefits of computer-assisted planning and surgery in reconstruction of the cranium have been extensively investigated. Preformed titanium implants are used especially for the reconstruction of cranium. The osteoneogenetic potency of titanium is low but can be increased by industrial modifications of the titanium surface.¹⁹ Fitting the prefabricated titanium implant requires an adequately perfused and vital soft-tissue environment to prevent perforations through the skin or in the sinuses with a possible consequent superinfection.²⁰ We also preferred titanium implants for the closure of bone defects after tumor removal. We have not observed any complications after these operations.

Since the 1960s, several groups have applied computer-aided techniques to design and manufacture custom titanium cranioplasty plates to repair a range of cranial defects.²¹ The techniques for

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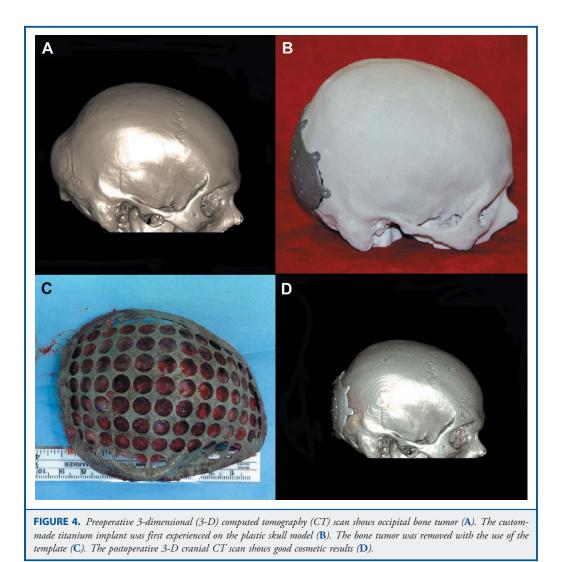
Postoperative 3-D CT scan shows good cosmetic results (D).

preparing customized titanium cranioplasty plates start with making a model of the defect from CT data and designing a surface to mimic the missing cranial segment. Once the surface has been generated, a plate is then designed from this surface to repair the defect.²² Recently, Williams et al²³ reported the largest series on custom-made titanium implants in 149 patients with 151 cranioplasties. They concluded that custom-made, patient-specific titanium cranioplasty has clear advantages over other biomaterials and techniques. This study also confirmed that custom-made titanium cranioplasty is a good option for reconstruction of all sizes of cranial defects with low long-term failure rate.

Using the advanced techniques, surgeons can precisely diagnose the defect in a 3-D environment, predesign a surgical plan, and even perform the plan in the clinic with the help of customized tools such as a surgical guide, thereby significantly improving the quality and efficiency of the surgery.²⁴ Similar to this technique, resection of FD can be performed in a computer-assisted procedure under navigation conditions. With this technology, the resection boundaries were transferred preoperatively to the computer and a resection template, which precisely reflects the contours of the actual implant that was manufactured. Thanks to computer-assisted planning, it is possible to precisely create a custom-made titanium implant before the resection of tumor. Similar to descriptions in the literature, we manufactured a titanium template for the precise identification of the boundaries of tumor during the surgery. This template also facilitated the appropriate insertion of the titanium implant on the bone defect. In 1 case, we used a CT-based navigation system for resection template could not provide enough information on the boundaries of tumor.

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Eufinger and Wehmöller²⁵ stated that direct computer-aided design and computer-aided manufacturing prefabrication of individual cranial and craniofacial implants based on helical CT data fulfill geometric requirements by 3-D modeling and avoid indirect manual fabrication on life-sized models, which always increases expenses, decreases precision, and does not use the advantages of geometric design in computer-aided manufacturing. Höhne et al²⁶ evaluated many implants and noted that computerized, virtually designed implants have found increasingly wider use. In their study, a thin-slice CT scan was obtained for each patient through the use of a standardized imaging protocol (helical CT; length of acquisition, 1 mm; Gantry tilt, 0°). The DICOM data were transmitted to the manufacturer. A 1:1 scale model of the skull, including the craniectomy, was customized after 3-D data reconstruction and rapid prototyping with a 3-D plotter. The implant consisted solely of titanium with a minimal thickness of 0.5 mm. The delay between image acquisition and shipment of the cranioplasty was approximately

14 days. For custom-made titanium cranioplasty, a small volume of implant material is needed. The plate is simply laid over the defect and fixed with mini-screws. Thus, it is not necessary to dissect the dura and the osseous rims. Wei et al¹⁶ performed a study of the FD of skulls that included a larger number of patients than previous studies, and they further analyzed the clinical characteristics of craniofacial FD. They suggested that children be screened more strictly for surgical treatment; the decision should always balance the benefits of each choice and the risks that the children and parents are willing to take. For acceleration of the lesion growth after the slow progression, surgery can also be considered. Wei et al showed that the farther the cut is from the lesion, the smaller the possibility of recurrence is. In our series, we planned bone cut on a skull model in the preoperative period, and then we produced a template to delineate the borders of the tumor. We removed the tumors using this template and did not observe any tumor recurrence during the follow-up period.

After removal of the tumor, immediate bone reconstruction is important if necessary. Kusano et al²⁷ suggested reconstructing the skull after the tumor stops growing. They performed a study with 50 patients who underwent decompressive craniectomy and closed the bone defect with custom-made titanium implants. They concluded that the custom-made titanium cranioplasty is safe and feasible for large cranial bone defects. However, complications caused by exposure of the brain are far worse than those in first-stage repair. Thus, in our 4 patients, we performed immediate cranial reconstruction using preformed titanium implants.

Computer-aided design and computer-aided manufacturing of the implants have improved the cosmetic results and shortened the operation time. However, this technique is associated with long production times and high costs.²⁸ To avoid the high expenses of computer-aided manufacturing/computer-aided design implants, various preoperative cast- and template-based reconstruction methods have been implemented to create similar patient-specific implants.²⁹ In our 4 patients, the design and production of the skull models, templates, and titanium implants were performed over 4 weeks in the Medical Design and Production Center of our institution. The mean duration of surgery was 132.5 minutes, which was shorter than the other surgeries without predesigned implants. However, we did not perform statistical analysis for comparison of operation times. The main advantage of our technique was safe and effective surgery with good cosmetic results. The total cost of our procedure, including surgical planning and production of implants, was approximately US \$2500. Because the patients had no malignant tumors, they waited for the production of custom-made implants to achieve good cosmetic results.

Limitations

There are 2 limitations of this study. First, the number of patients is too low for a precise conclusion. The second limitation is the short follow-up period for the observation of tumor recurrence.

CONCLUSION

FD is a benign tumor, and, unlike other malignant tumors, its resection margin can be clearly estimated by preoperative CT. As a result of these disease characteristics, preoperative computerbased resection planning and defect size estimation were possible, and implant molding could be done before resection of tumor. Better cosmetic results could be obtained with use of the advanced computer technology.

Disclosure

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

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