Case Report

Decompressive Cranioplasty Using Titanium Mesh in a Patient with Osteopetrosis: Case-based Reviews

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ABSTRACT

Background and Aim: Osteopetrosis is an inherited disease that causes the thickening of the cranial bone. The need for both cerebral decompression and intracranial volume-enlarging cranioplasty in the same surgical session in a patient suffering from rising intracranial pressure is critical and a very rare decision in neurosurgical practice. The only indication that can lead to this surgery is osteopetrosis.

Case Presentation: A 24-year-old female patient with osteopetrosis was admitted with the complaint of severe headache that did not respond to medical treatment, and surgery was decided. After decompressive craniectomy was performed with difficulties, cranioplasty with titanium mesh was performed in the same session to protect the cerebral tissue, close the defect area, and give more space to the cerebral tissue. It was observed that the complaints of increased intracranial pressure were completely resolved in the early postoperative period and the one-year clinical follow-up. The surgical intervention technique and the radiological findings obtained during the follow-up are presented.

Conclusion: Performing cranioplasty with titanium mesh, which is used to protect and save cerebral tissue in patients with osteopetrosis, seems the most appropriate choice.

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Highlights

- Osteopetrosis causes increased intracranial pressure in cases in their twenties.
- The fact that titanium is a thin but solid material enabled it to take its place when the thickened bone was resected.
- The titanium mesh is the most suitable material for performing cranioplasty in the same session together with decompressive craniectomy for patients with osteopetrosis.

Plain Language Summary

Thickening of the skull bones and compression of the brain tissue cause clinical complaints in these patients. In this patient, we aimed to increase the available intracranial space for brain tissue. For this reason, we removed a section of the skull that was 3 cm thick with 8×10 cm dimension. We covered the removed area with a thin metal cover so that the brain tissue fills it easily. Thus, in the same surgery, we both opened a space for the brain tissue and closed the brain tissue with a protective titanium cage.

1. Background and Importance

Intracranial hypertension is a critical event frequently occurring after traumatic brain injury (TBI) as a delayed secondary pathologic process initiated at the moment of injury. Due to the rigid nature of the skull and the dura, brain edema, expanding hematomas, or blossoming of contusions can rapidly exhaust the compensation mechanisms leading to the maintenance of a controlled intracranial pressure (ICP). Following the failure of medical management, decompressive craniectomy (DC), a procedure consisting of the removal of part of the skull and opening of the underlying dura, can be used as a last-tier therapy to mitigate ICP elevation. DC used as a last-tier therapy for patients with severe, sustained, and refractory posttraumatic intracranial hypertension leads to a substantial mortality reduction. In patients with osteopetrosis with increased intracranial pressure, the need for decompressive craniectomy becomes essential as reported in the literature [1, 2].

However, since the increased intracranial pressure decreases when the cranial volume expands, it is necessary to close the cranium again in the same session. Both bone flap removal and cranioplasty are accomplished at the same time. The procedure of decompressive bone flap replacement (DBFR) is capable of preserving skull integrity and achieving a decompressive effect. The increase in intracranial pressure in osteopetrosis is not very high. In the same surgical session, the cranial defect can be closed again either with its own thinned cranial flap or with a titanium mesh graft. There are different opinions about which one is more suitable for cranioplasty in osteopetrosis. However, we have obtained findings supporting the literature that cranioplasty with titanium mesh is a more accurate choice [3]. After the acute phase, the intracranial pressure gradually returns to the normal range, and the implanted titanium mesh graft can protect brain tissue from swings caused by atmospheric pressure or gravity [4]. In addition, DBFR itself does not induce new neurological dysfunction. We can state that the most important surgical indication of DBFR is osteopetrosis.

Historical background

The procedure of DC was first described by Kocher in the treatment of TBI by the removal of an area of the skull to expand the potential cranial volume. In the following years, from the lessons learned watching Kocher in Bern, the US neurosurgeon Cushing proposed DC for the treatment of other brain disorders [5, 6]. Cushing described, for the first time, the decompressive craniectomy performed in severe head traumas with 250 cases during World War I [7]. In 1976, Cooper et al. established the end of DC as a standard practice to limit intracranial hypertension linked to cerebral edema. They reported a 10% total and a 4% functional survival rate in 50 patients with TBI. In 1999, Guerra et al. conducted a prospective clinical study on the effect of bilateral or front temporal craniectomy in patients with refractory intracranial hypertension not responsive to medical therapy. In summary, at the end of the 20th century, the indications for DC were as follows: ICP >30–35 mmHg or CPP <45–70 mmHg, age <50 years, GCS >4, CT signs of brain swelling, associated masses, and GCS 3 plus bilateral fixed dilated pupils excluded [8]. DC is a life-saving procedure [9, 10], which may also lead to a series of complications because of the pathological and physiological changes induced by removing the cranium bone flap [3]; for example, skull defect, subdural hygroma, hydrocephalus, cortical herniation, paradoxical herniation, encephalocele, and seizures [11]. Thus, whether or not to remove the cranium bone flap is controversial. In some cases, removing the bone flap could be overtreatment. After cranium bone flap removal, the secondary cranioplasty can ensure adequate biomechanical protection for the underlying brain, keeps table intracranial pressure, and re-establish cerebrospinal fluid dynamics and cerebral blood flow [12, 13]. Cranioplasty presentations and cosmetic and protective benefits for patients after the procedure of DC contribute to the improvement of neurological functions [14].

The necessity of performing cranioplasty in the same surgical session in a patient who has undergone decompressive craniectomy is a very rare need. The important indication for this condition is osteopetrosis, a hereditary pathology that is rarely encountered in neurosurgery practice and affects the cranium. It presents with increased bone density due to dysfunction of osteoclasts. The biological explanation of the pathogenesis is attributed to the dysregulated ATPase pump activity. Dysfunction of chloride channels, transmembrane proteins, and carbonic anhydrases leads to deficient cell polarization, resulting in a lack of hydrochloric acid production and dysfunction of osteoclasts [15]. Although it is generally known as bone disease, the most serious and dramatic results are seen in the central nervous system. The disease results in increased intracranial pressure due to abnormal bone growth and various symptoms related to pinched cranial nerves, vascular structures, and neuronal structures in the narrowed skull base foramen [16, 17]. No definitive treatment for the disease has yet been found.

Clinical presentation

Osteopetrosis is an inherited disorder that results from the abnormal development of bone and excessive increase in density as a result of decreased activity of osteoclasts. It was first described by Albers-Schönberg in 1904 [18, 19]. Clinically, macrocephaly due to abnormal bone formation, recurrent pathological bone fractures, anemia, hepatosplenomegaly due to extramedullary hematopoiesis, susceptibility to infections, and neurological findings have been reported [20]. Narrowing of the neural foramen due to the increase in bone tissue may result in neurological symptoms, such as hydrocephalus, hearing loss, vision loss, headache, and compression of large vascular structures [19]. It is divided into two forms autosomal dominant osteopetrosis and autosomal recessive osteopetrosis. The former occurs in approximately one in 20000 births [21]. There is usually recurrent persistent headache and accompanying evidence of papilledema supporting increased intracranial pressure at the base of the eye [4].

Diagnosis

The disease usually remains asymptomatic and is diagnosed incidentally. Symptoms are observed in late childhood and adulthood. Occurring in approximately one in 200,000-300,000 births, the autosomal recessive form has a poor prognosis and is also known as the infantile form [21]. The diagnosis of osteopetrosis is usually made with the combination of progressive clinical and radiological findings. Cranial computed tomography (CT) imaging revealed thickening of all cranial bones and decreased intracranial volume. Cranial magnetic resonance imaging (MRI) revealed that the lateral ventricles were depressed, the subarachnoid spaces were narrowed, the bilateral temporal lobe uncus was closer to the tentorium, and there was compression in the brain parenchyma in both cerebral hemispheres.

Management

A multidisciplinary approach is absolutely necessary in the follow-up and treatment of clinically symptomatic osteopetrosis cases [17]. Acetazolamide is known as an alternative in medical treatment in cases with increased intracranial pressure. However, it is usually not sufficient. Decompressive craniectomy plays a significant role in treating refractory intracranial hypertension [3]. Neuronal decompression should be considered in symptomatic cases with insufficient response to medical treatment as well as severe calvarial and foraminal stenosis [17].

Al-Mefty et al. mentioned visual recovery after optic nerve decompression with the supraorbital bilateral approach in five out of six patients with osteopetrosis in 1988 [22]. Hwang et al. shared the results of visual recovery after optic nerve decompression in their case report and underlined the importance of visual findings in the follow-up and management of osteopetrosis [23]. In our patient, visual complaints improved after unilateral decompressive cranioplasty without optic nerve decompression. We believe that the improvement in

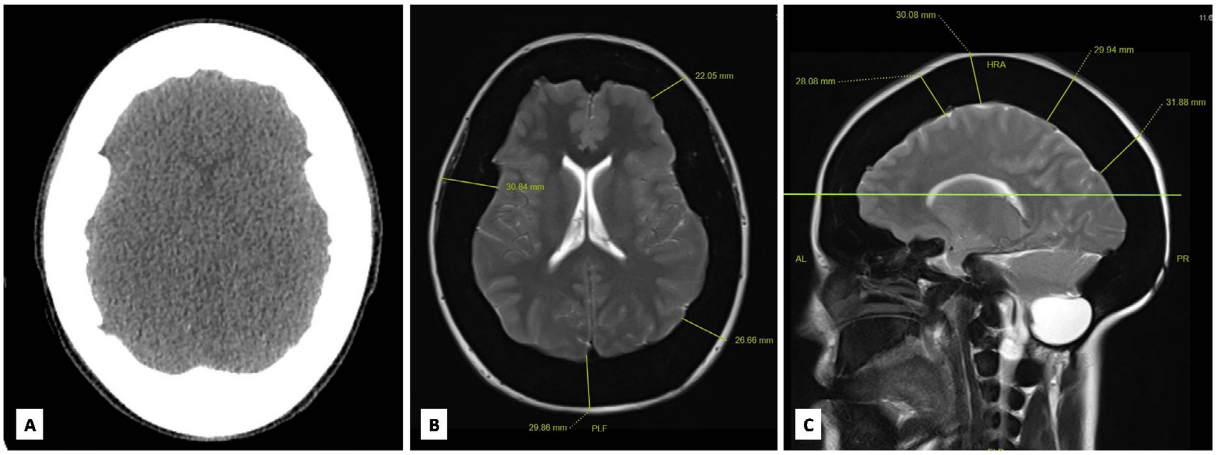

Figure 1. A): Preoperative CT image. B and C): Preoperative MRI images.

The lateral ventricles are reduced. Calvarial bone thickness is increased and pseudomeningocele sac is observed in the suboccipital decompression area on the sagittal section.

Author/Date	Age	Gender	Presentation	Treatment	Outcome
Kulkarni et al., 2007	15	M	Headache, Hoarseness	Not Reported (NR)	NR
Al Tmimi et al. 2008	11	F	Neck Pain	NR	NR
	5	F	Neck Pain	NR	NR
	1.5	M	Cranial Nerve Involvement	Posterior Fossa Decompression	Died Sepsis Postop 3. Month
Jamjoom et al. 2009	4	M	Increased ICP signs and Symptoms	Hypertrophied Bone Resection With Bifrontal Craniotomy	Peroperative Cardiac Arrest
Dlouhy et al. 2011	25	F	Swallowing Difficulty, Headache	Posterior Fossa Decompression	Symptoms Resolved
Mahmoud et al. 2013	9 months	M	Irritability,Seizure After Falling	Ventriculoperitoneal Shunting	NR
Ekici et al. 2015	25	F	Headache, Numbness	Posterior Fossa Decompression	Relieved Symptoms
Alsahlawi et al. 2017	26	F	Decrease in Visual Acuity	Optic Nerve Decompression + Unilateral Cranial Decompression + Cranioplasty with Thinning Bones	Stabilized Vision
Isler at al. 2018	7	M	Acute Tetraparesis	Posterior Fossa Decompression + Ventriculoperitoneal Shunting	Relieved Signs, Symptoms and Radiological Findings
Present Case	24	F	Headache, Agitation	Unilateral Cranial Decompression + Cranioplasty with Titanium Mesh	Resolved Symptoms

Figure 2. Cases reported in the literature

F: Female; M: Male; NR: Not reported; ICP: Intracranial pressure

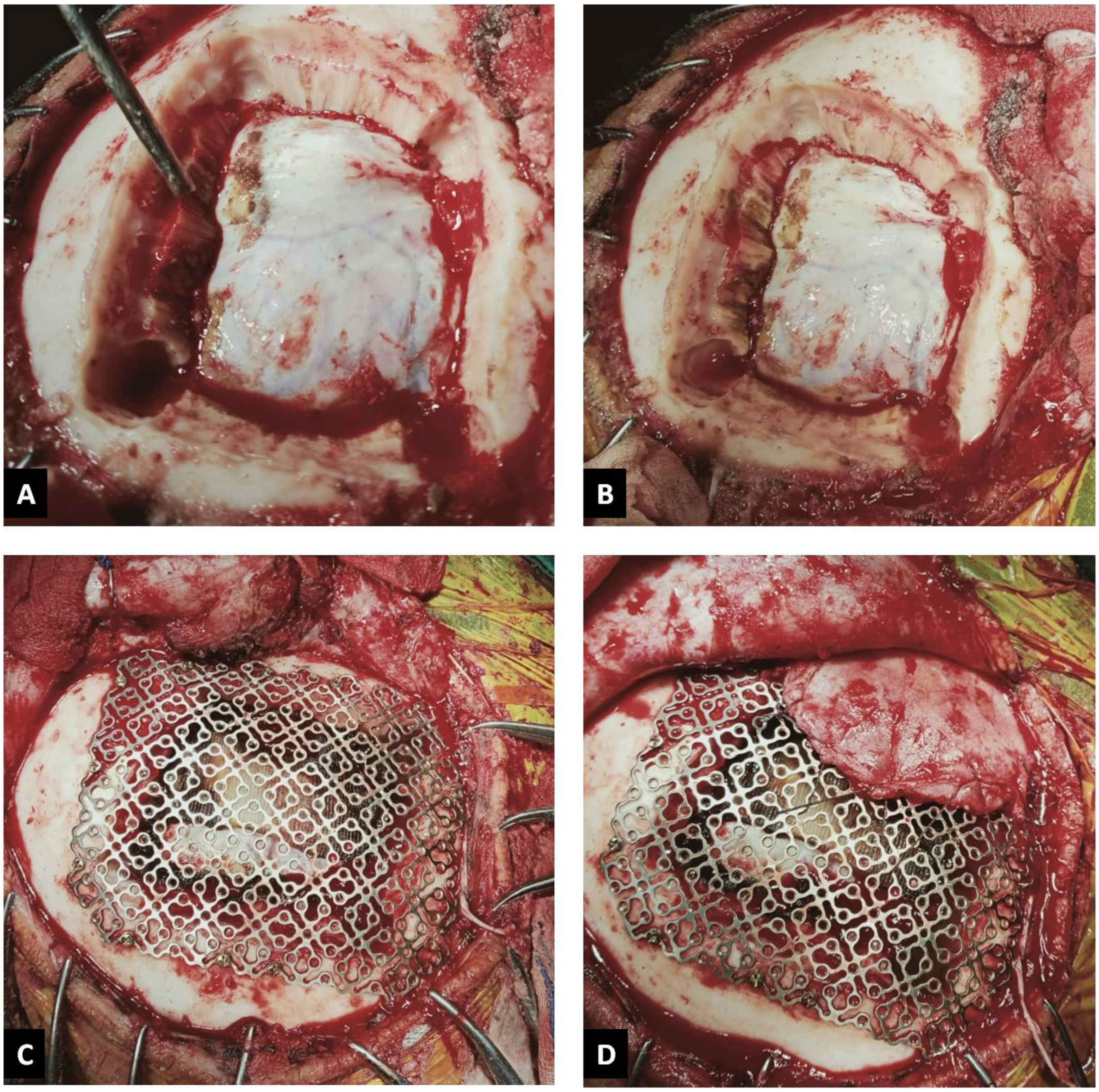

Figure 3. Perioperative images. A and B; Cranial thickness is seen. It is understood that the craniotomy is first thinned at the free edge of the bone and then it can be cut. Two separate layers are visible on the wall at the edge of the craniotomy. C and D: After the titanium mesh is fixed with mini-screws and temporal muscle hanging on the titanium mesh.

visual complaints is due to the decreased ICP after decompression.

In the literature, no patient with a diagnosis of osteopetrosis, who had previously undergone suboccipital decompression, had both unilateral decompressive craniectomy to reduce intracranial pressure, and cranioplasty with a titanium mesh to protect the cerebral tissue in the same session has not been identified. It has been observed that there is no clear decision in the literature indicating the most appropriate cranioplasty material for these patients. The surgical technique steps and the patient's follow-up findings are presented in light of the literature.

The operation for both cerebral decompression and cranioplasty in the same session in a patient whose intracranial pressure increases is a very rare decision in neurosurgery practice. According to a study published in 2017, Allahsawi et al. used thinned autologous bone as cranioplasty material after cranial decompression in a case of osteopetrosis presenting with decreased visual acuity, coughing, and sneezing, and progressively increasing headache [4]. Although autogenous grafts,

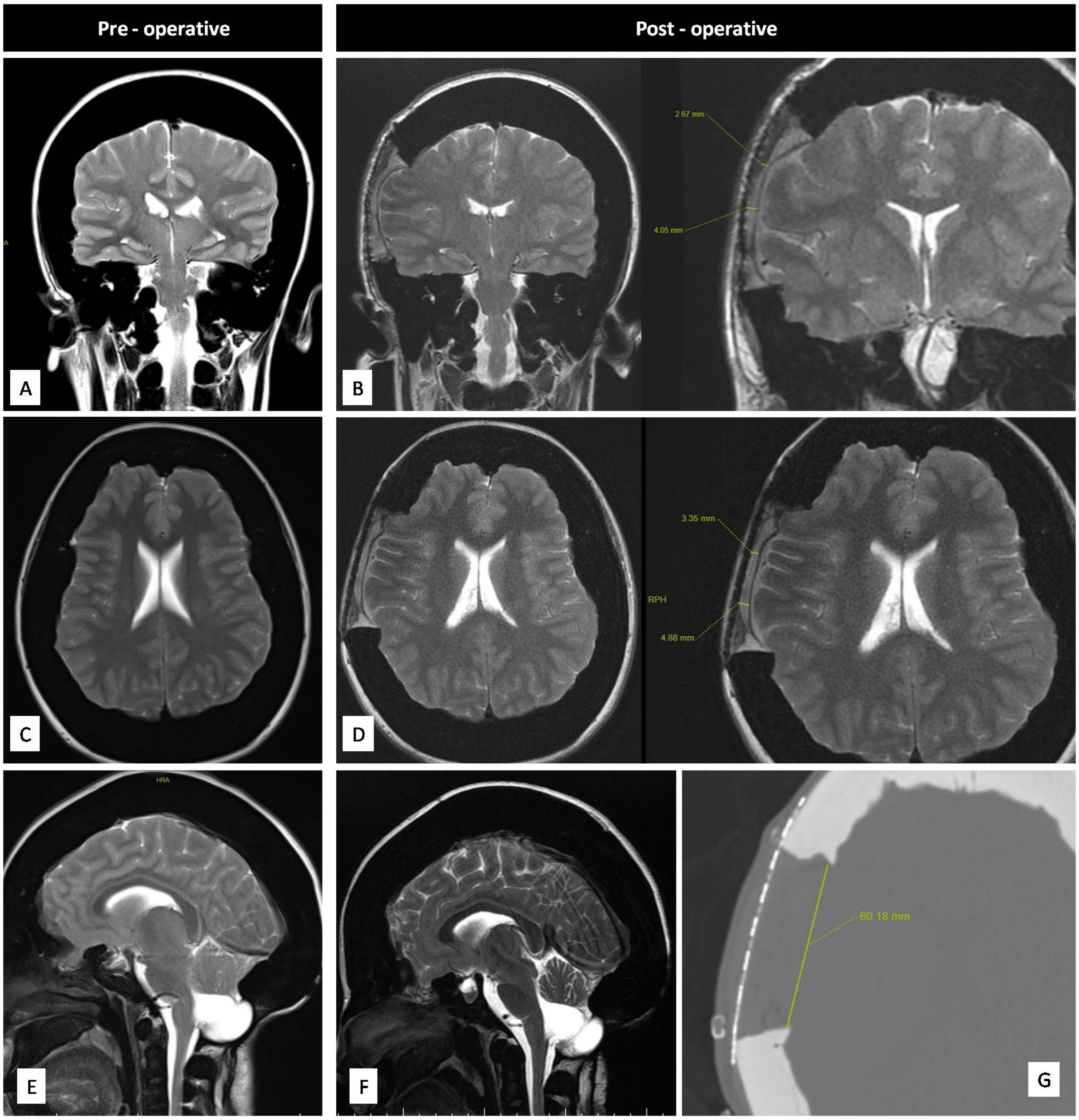

Figure 4. A, C, and E: Preoperatively. B, D, F, and G: Postoperatively. After the cranial volume expands, postoperative change is observed with MRI in the third month. It is observed that the cerebral cortex fills the space gained on the coronal and axial planes. Since the dura mater is not opened, no damage is observed on the temporal lobe cortex surface and slight enlargement of the lateral ventricles is observed.

allogeneic implants, or alloplastic materials can be used for the reconstruction of craniofacial defects, the most appropriate material is still controversial [4]. The aim of decompression in osteopetrosis patients should be to provide more volume to the neuronal tissue. In this case, considering that the young age of the patient may cause bone re-growth due to osteopetrosis and eventually the re-occurrence of symptoms as the result of increased intracranial pressure, titanium mesh was preferred. This preference is supported by the results obtained in the study by Ye et al. [3].

The surgical techniques and results for decompression in patients with osteopetrosis are presented in Figure 2 [4, 16, 24–30]. Titanium mesh has been increasingly used in reconstructive surgery in recent years due to its many advantages, including ease of application, the possibility of personalized preoperative three-dimensional reconstruction, suitability for radiological imaging, and

Figure 5. A-D: Early appearance of the patient's wound after z-plasty and after sutures are removed.D; Appearance of scalp wound at the end of oneyear.

low infection rates [8]. In our study, the titanium mesh was easily shaped in accordance with the anatomy and fixed to the cranium with a miniplate screw. In this way, both a wider area and a solid cover have been provided to the neuronal tissue. ICP has decreased with the increase in intracranial volume. Another advantage of using titanium mesh instead of thinning cranial bone is the elimination of the risk of re-operation due to bone development.

Prognosis and outcome

In patients with symptomatic osteopetrosis, cranioplasty can be considered as an option to reduce increased ICP and it is reported that their complaints decreased of the cranial nerve palsies together [4].

2. Case Presentation

A 24-year-old female patient with a diagnosis of osteopetrosis underwent suboccipital decompression due to severe headaches as a result of Chiari malformation seven years ago. She was admitted to our outpatient clinic with complaints of headaches especially provoked by coughing and sneezing, aggressive personality changes, and loss of vision and hearing over the past year. There was an increase in the complaints of nausea and dizziness. No motor defects were reported in the extremities in the neurological examination. Cranial nerves were evaluated. A decrease in visual acuity was detected. Finger counting from approximately 5 meters was decreased in both eyes (Figure 1). An ophthalmological examination was performed. Bilateral first-degree papilledema was detected. In Pure-tone audiometry, mild hearing loss was detected with an average of 32 dB in both ears. Pseudomeningocele appearance, which is a sign of increased ICP, was observed in the posterior wall of the posterior fossa, especially in the area of previous suboccipital decompression (Figure 1). Abnormal bone growth due to osteopetrosis has resulted in the growth of the calvarium and decreased cranial volume, and eventually presented with clinical manifestations due to increased intracranial pressure. In our patient, bilateral decompressive cranioplasty was considered in the first place, but due to the cranial thickness ranging from 2.2-3.1 cm, we first decided to perform the surgery unilaterally to see the clinical results. It was decided to perform right-sided temporoparietal decompressive cranioplasty due to the left dominant hemisphere. Our patient underwent unilateral decompressive cranioplasty by using titanium mesh. The dura was not opened considering the risk of herniation to the outside. The sphenoid wing on the proximal of the Sylvian sulcus was thoroughly defeated with a high-speed drill. No intraoperative complications were encountered. Rapid improvement in clinical symptoms was observed in the early postoperative period.

Surgical approach

After surgical sterilization, in the supine position with the head turned 45 degrees to the left under general anesthesia, a right-sided wide pterional incision was performed to access the skull base. After controlling the bleeding with bipolar cautery, the temporal muscle was hung by opening the base per the procedure. Four burr holes, two on the base of the temporal bone and two on the anterior and posterior parts of the parietal bones, were carefully opened with the help of a high-speed drill. Since the cranium was extremely hard and its thickness was approximately 2.6 cm, continuous irrigation of the drill bit with saline was required, which overheated during the procedure. Each burr hole took about 20-25 minutes to open. The bone flap was released with dif-

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ficulty after the bone surface between the opened burr holes was drilled and adjusted to the depth of the cutting attachment. Then, 150 cc of mannitol was injected by slow infusion without lifting the flap. There was no dural adhesion (Figure 3A and B). The opening time of the cranium exceeded 2 hours as a result of the utmost care to avoid complications due to bone hardness and thickness. The sphenoid wing was drilled and excised by Kerrison Rongeur. Considering the risk of cerebral herniation and cortical damage due to increased intracranial pressure, duraplasty was not performed. The dura membrane was not opened. The titanium mesh was fixed to the intact bone using eight mini-plate screws to cover the craniectomy area and cranioplasty was performed. The temporal muscle was attached by stretching on titanium (Figure 3 C and D). Hemovac drain was placed on titanium mesh in the surgical field after the control of the bleeding has established. The distal end of the temporal muscle was suspended on the titanium mesh. The layers were closed in accordance with the anatomy. There were no complications. Hemovac drain was kept for two days.

Postoperative follow-up and outcomes

There was no problem with the wound care of the patient. At the end of two weeks, the stitches were removed. At the end of the 4^{th} month, there was a punctual opening at the incision site. The relationship between titanium and cranium was evaluated by computerized tomography (Figure 4). It was understood that the cause of the problem was the tension in the scalp due to the chronic enlargement of the cranium. Even though the titanium was thin and was attached to the bone under the skin with mini-screws from multiple points, it created a slight tension on the skin. The strain was increased. A suitable skin flap was planned for the patient and z-plasty was performed (Figure 5). The tension in the skin spread to other areas. Thus, healing occurred in the scalp. At the end of approximately one year of follow-up, the patient's wound was completely healed (Figure 5D) and she had no neurological complaints. She described improved hearing performance and better vision during follow-up.

3. Conclusions

We believe that titanium mesh is the most suitable material for performing cranioplasty in the same session together with decompressive craniectomy in order to protect neuronal tissue and increase the intracranial space in symptomatic patients with osteopetrosis. It was also understood that not opening the dura during the surgical procedure did not change the result, but contributed to the protection of the brain tissue.

Patient Consent

The patient consented to the submission of this case report to a medical journal. Written informed consent was obtained from the patient for publishing this article accompanied by the MRI images.

Ethical Considerations

Compliance with ethical guidelines

All procedures performed in the studies involving human participants followed the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standard.

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Authors' contributions

Conception and design: Cem Demirel; Data Collection: Vaner Köksal; Data analysis and interpretation, drafting the article, reviewing the submitted version of the manuscrip and final approval: Both authors. Critically revising the article: Vaner Köksal

Conflict of interest

The authors have no conflict of interest to declare.

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