Case Report

Reconstruction of the Occipital and Parietal Congenital Defect with 3D Custom-Made Titanium Prosthesis: A Case Report with Four and a Half Years of Follow-Up and a Brief Review of Literature

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Management of patients with congenital skull defects requires a multidisciplinary approach. Considering the defect’s location and size, brain protection, and the cosmetic outcome makes such reconstructions challenging [1].

Autogenous bone grafts are assumed to be the gold standard for osseous reconstructions due to their immunocompatibility and low infection rates; however, donor site morbidity and lack of resemblance to skull contour are important drawbacks that limit their usage for the reconstruction of complex skull defects [2].

Due to these limitations, alloplastic materials that are biocompatible, inert, sterilizable, lightweight, noncarcinogenic, and cost-effective can be the option of choice for these reconstructions [3].

Whether prefabricated or custom-made, alloplastic materials are mainly made from titanium or polymethacrylate (PMMA) [3]. PMMA is economically affordable [3, 4]; however, it is not as strong as titanium [5]. Moreover, PMMA prostheses are more prone to infection, local

1. Introduction

Skull defects are mainly due to congenital deformities, trauma, infection, or malignancy. Considering the location and size of the defect, along with brain protection and the cosmetic outcome, it makes such reconstructions challenging [1].

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irritation, foreign body reaction, and thermal damage during polymerization in the case being used as a cement for prosthesis adaptation. Furthermore, due to the native tissue ingrowth, prosthesis removal may be challenging [6].

Titanium alloys are amongst the most popular metals used in reconstructive procedures due to their high strength, biocompatibility, excellent contour, rigidity, low infection rate, favorable osseointegration property [7, 8], and excellent marginal adaptability, which can precisely fit into the complex geometry of skull defects, especially when 3D printed with CAD/CAM. These 3D-printed prostheses also have fewer radiographic artifacts in comparison to titanium meshes [9]. However, they are more expensive to design and manufacture [10].

Hence, the authors present a case of congenital skull defect with a history of several unsuccessful surgical treatments aimed at restoring the contour of skull. She was successfully treated with a 3D custom-made CAD/CAM titanium implant. For this reconstruction, the total surgery time was 90 minutes, which is 70% less than that of conventional cranioplasty procedures. Moreover, on 4.5 years of follow-up, excellent contour and the aesthetic outcome were evident.

2. Presentation of Case

The patient was a 13-year-old female with congenital skull defects at the superior third of occipital bone along with the posterior thirds of the bilateral parietal bones. Upon physical examination, the neurologic status was intact, and she did not have growth retardation. The genetic analysis showed no hereditary syndromes. She had her first

Figure 1: 3D analysis. (a) CT scan shows the exact size and shape of the congenital defect. (b) The 3D virtual model of the prosthesis. (c) The 3D virtual model of the prosthesis adapted into the defect.

Figure 2: Intraoperative photograph showing the prosthesis fitness to the defect.

Figure 3: Four and a half years of follow-up. Note the perfect contour and healthy intact soft tissue at the age of 17.
reconstructive surgery in May 2008, at the age of 4 years, when the defect was bridged with a titanium mesh plate. Ever since that surgical treatment, she developed relentless petit mal seizures which were refractory to medications. The plate became exposed three months later, and she underwent several debridements in the following year, till the mesh was totally removed at the age of 5 leading to the resolution of seizures. She had not received any other medical treatment until the age of 13 when she was referred to our institution.

Because no further skull growth was expected [11], the patient was a candidate to receive the definitive prosthesis. A preoperative CT scan was obtained to determine the exact size and shape of the defect (Figure 1(a)). With CAD/CAM, a 3D virtual model of the prosthesis was designed and manufactured to match the borders of defect and provide ideal contour. The titanium alloy (TiAl6V4) was used to manufacture the prosthesis by implementing 3D printing via additive manufacturing method (Figures 1(b) and 1(c)).

For placing the prosthesis, bicoronal approach with subperiosteal dissection on the surrounding bony areas was performed. Extreme caution was exercised not to penetrate the dura during dissection between the galea and dura. The defect was totally exposed, and the dura remained intact. Then, the prosthesis was placed over the dura and fixed to the surrounding bone with titanium 2.0 mm miniplates. The ideal marginal adaptability and contour are shown in Figure 2. The incision was closed in three layers, taking care to approximate the tissues in a tension-free manner. The patient received intensive care for 24 hours and was discharged in the following week. On 4.5 years of follow-up (Figure 3), the skin over the defect remained intact with no signs of inflammation or neurologic deficits.

3. Discussion

Reconstructing complex skull defects is a challenge in craniomaxillofacial surgery. Major skull defects cannot be addressed with autografts since the contour of the skull is not similar to the common osseous donor sites such as the fibula, iliac, or ribs [2]. Therefore, these defects may be best managed with a customized prosthesis. The 3D-printed PMMA or titanium prostheses were reported to have favorable outcomes. PMMA acrylic prostheses are more susceptible to infection [12] and tissue necrosis, which not only necessitates prostheses removal [13] but also jeopardizes the soft tissue over the defect, making further reconstructions more challenging [4]. Jaberi et al. reconstructed 70 patients with PMMA custom-made implants and reported a 24% complication rate [14].

To date, titanium is the most biocompatible material available in which the rigidity, anti-inflammatory, and antibacterial nature of this metal has made it the option of choice.
| Num. | Author/year         | Patients | Gender Age | Location | Titanium implant type | Follow-up | Post op complication                                                                 |
|------|---------------------|----------|------------|----------|-----------------------|-----------|--------------------------------------------------------------------------------------|
| 1    | 1998/E. Heissler [16] | 15       | 12 male/3 female 21-35 years | Not mentioned | Solid | Mean 16.6 months | 1 infection, 1 convulsion upon drainage removal                                      |
| 2    | 1999/J.JOFFE [17]   | 141      | Not mentioned 6-67 years | Not mentioned | Solid | 6 weeks to 1 year | 1 failure                                                                             |
| 3    | 2009/Mario Cabraja [13] | 26       | Not mentioned 35.6 | 16 (frontotemporoparietal), 4(bifrontal), 3(frontal), 2(temporal), 1(frontoparietal) | Solid | Median 8.1 years | A transient palsy of the frontal ramus of the facial nerve postoperatively just in 1 case |
| 4    | 2010/Jules Poukens [18] | 1        | Not mentioned 6 weeks to 1 year | Unilateral frontotemporoparietal | Solid | NM | No                                                                                   |
| 5    | 2011/H. Sudhoff [9] | 1        | Male 48 | Unilateral frontotemporoparietal, sphenoid, frontal, orbital rim, ethmoid, maxillary, zygomatic, nasal bones | Solid | 1 week | No                                                                                   |
| 6    | 2011/Juan Felipe Isaza Saldarriaga [15] | 1        | Male 13 | Unilateral frontoparietal area | Solid | 4 months | No                                                                                   |
|      | 2014/S.A. Eolchiyan [19] | 4       | Female 24 | Left fronto-orbital region | Solid | Mean 4.4 years | No                                                                                   |
|      |                     |          | Male 44 | Right parietal-temporal region | Solid | Mean 4.4 years | No                                                                                   |
|      |                     |          | Female 23 | Left fronto-orbital region | Solid | Mean 4.4 years | No                                                                                   |
|      |                     |          | Female 23 | Both side frontal region | Solid | Mean 4.4 years | No                                                                                   |
| 7    | 2015/Hyung Rok Cho [10] | 3       | Female 41 | Left parieto-occipital area extended to right parietal area | Porous | 6 months | No                                                                                   |
|      |                     |          | Female 32 | Right frontotemporoparietal area | Porous | 6 months | No                                                                                   |
|      |                     |          | Female 21 | Left parietotemporal area | Porous | 2 months | No                                                                                   |
| 8    | 2018/Jae Yoon Kim [20] | 1       | Male 36 | Unilateral forehead and glabella area | Solid | 6 months | No                                                                                   |
| 9    | 2019/Seong Hwan Kim [21] | 2       | Male 55 | Right parietotemporal area | Solid | 1 year | No                                                                                   |
|      |                     |          | Male 33 | Right temporal area | Solid | NM | No                                                                                   |
| 10   | 2019/Champeaux C [22] | 19      | 12 male/7 female 47.4 ± 11.3 | 6: right side, 9: frontal side, 4: left side | Solid | Mean 1.2 year | 31.6%: early postoperative complications, 1 lost to follow-up, 1 prosthesis removal, 63.1% no complication |
Conventional cranioplasty surgeries with prefabricated prostheses or titanium meshes usually take 4-5 hours due to the trial-and-error procedures to achieve a good fitness of the implant. By CADs, the complete procedure of the surgery is preplanned, and a customized prosthesis with excellent contour and fitness can be produced before the surgery. Therefore, not only there is no need for prosthesis adjustment during surgery but also the surgery time decreases considerably, and the outcome improves remarkably. By such a decrease in surgical time, the risk of postoperative complications such as severe pain, infection, and wound dehiscence decreases substantially. Our entire surgery lasted 1:30 hours which is approximately 70% less than conventional cranioplasty surgeries. Moreover, Saldarriaga reported the same, 85% reduction in surgery time with the similar method [15].

To get a deeper understanding, a literature review was performed. Two authors (N.N. and H.Z.) independently conducted the electronic searches using Ovid MEDLINE/PubMed, Scopus, EMBASE, Google Scholar (first 200 results), and Cochrane Database, including articles published in March 2021. Alloplastic, titanium, and cranioplasty results were used as keywords. The bibliographies of included studies were also searched for relevant articles. Preclinical studies, papers not describing cranioplasty techniques, and studies not reporting follow-up durations were excluded. Figure 4 shows the PRISMA flowchart of selection criteria for this review. Finally, 11 studies with 214 reported cases were included. Table 1 shows the extracted data, and Table 2 reports the analyses of data obtained from the included studies.

According to the results, the complication rate for titanium prosthesis reconstruction is relatively low. Among 215 cases with follow-up periods from 1 week to 8 years, 12 (5.6%) post-op complications and 2 (0.9%) prosthesis failures were reported. All studies reported excellent contour, acceptable rigidity, and high patient satisfaction rates. This rate of complications is by far more acceptable than that of PMMA prostheses.

The expenses of custom-made titanium implants are relatively high; however, the benefits definitely outweigh, such as decreased surgical time, shorter recovery and hospitalization, fewer postoperative complications, and less need to further corrective surgeries.

### Table 2: Summary of the demographic data and complication rates of the included patients.

| N     | 214 |
|-------|-----|
| Sex   |     |
| Female | 7.47% |
| Male   | 14.02% |
| Not mentioned | 78.50% |
| Age    | 38.93 ± 8.27 |
| Follow-up | 45.85 ± 40.01 |
| Complication rate | 5.61% |

4. Conclusions

In conclusion, custom-made titanium implants can be considered the treatment of choice, which offers predictable aesthetic outcomes for the reconstruction of complex skull defects, with the low rate of complications and high aesthetics and patient satisfaction outcomes.

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Acknowledgments

The authors wish to acknowledge Mr. Ahmad Bereimipour for his assistance in journal selection.

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