

## Case Report

# Single Stage Cranioplasty with Customized Polyetheretherketone Implant After Removal of Eosinophilic Granuloma skull bone: Case Report

Islam MR<sup>1</sup>, Rahman MM<sup>2</sup>, Hasan MM<sup>3</sup>, Mostafa AH<sup>4</sup>, Singh BG<sup>5</sup>, Barua S<sup>6</sup>, Abbas MA<sup>7</sup>, Chaurasia B<sup>8</sup>, Das S<sup>9</sup>

**Conflict of interest:** There is no Conflict of interest relevant to this paper to disclose.

**Funding Agency:** Was not funded by any institute or any group.

**Contribution of Authors:** Principal Investigator- Prof.

**Manuscript preparation-** Dr. Md. Rokibul Islam, Dr. Md. Moshir Rahman

**Data collection-** Dr. Md. Mahbub Hasan, Dr. Abu Hena Mostafa, Dr. Bibek Gaurab Singh, Dr. Shamit Barua, Dr. Mohammad Abed Abbas, Dr. Bipin Chaurasia

**Editorial formatting -** Prof. Sukriti Das

**Copyright:** @2022bang. BJNS published by BSNS. This article is published under the creative commons CC-BY-NC license. This license permits use distribution (<https://creativecommons.org/licenses/by-nc/4-0/>) reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes.

**Received:** October 10, 2023

**Accepted:** November 09, 2023

### Abstract

*Eosinophilic granuloma is a localized form of Langerhans cell histiocytosis, most commonly involving the skeletal system. Their origin from the dura is rare with only a handful of cases on record. We present one such rare case of an eosinophilic granuloma originating from the Extradural with secondary osseous invasion in a 20 year old male who presented with a swelling in the right fronto orbital region.*

*A complete resection was scheduled with single stage reconstruction of the fronto-orbital bone by a polyetheretherketone (PEEK) specific implant. This implant was computer-assisted designed and manufactured and verified by the surgeon before the intervention.*

*Bang. J Neurosurgery 2024; 13(2): 152-156*

### Introduction

Langerhans cell histiocytosis (LCH) is a group of idiopathic disorders which are characterized by an abnormal proliferation of histiocytes. Eosinophilic granuloma is an older term for unifocal LCH. It is a

slowly progressing disease characterized by an expanding proliferation of Langerhans cells most commonly involving the skeletal system. [1] Many reports of eosinophilic granuloma of the skull and the brain have been reported. However, there are only a

1. Dr. Md. Rokibul Islam, Assistant Professor, Dept. of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
2. Dr. Md. Moshir Rahman, Assistant Professor, Holy Family Red Crescent Medical College, Dhaka, Bangladesh
3. Dr. Md. Mahbub Hasan, Resident, Phase-B, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
4. Dr. Abu Hena Mostafa, Resident, Phase-B, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
5. Dr. Bibek Gaurab Singh, Resident, Phase-B, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
6. Dr. Shamit Barua, Medical Officer, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
7. Dr. Mohammad Abed Abbas, Medical Officer, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh
8. Dr. Bipin Chaurasia, Department of Neurosurgery, Birgunj Clinic, Birgunj, nepal
9. Prof. Sukriti Das, Professor, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh

**Address of Correspondence:** Dr. Md. Rokibul Islam, Assistant Professor, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Shahbag, Dhaka 1000, Bangladesh. Mobile: +8801711452526, E-mail: mail2rokib@gmail.com

few reports of intradural LCH in literature. [2,3] We report a rare case of Extradural eosinophilic granuloma with osseous invasion. . As a consequence, cranioplasty with good cosmetic results can be challenging for the surgical team. Historically, many techniques with different materials have been used for cranioplasty, including autologous bone, titanium mesh, porous polyethylene, polymethylmethacrylate (PMMA) or hydroxyapatite. [9,10]. Polyetheretherketone (PEEK) implants are now widely used for cranioplasty [5,6], craniofacial defects repair [4], or others parts of skeleton reconstruction[8]. PEEK prosthesis advantages are radiolucency, chemical inertness, robustness and comfort. It does not create artifacts on imaging and does not conduct temperature [5]. The recent advances in the era of computer-assisted design and computer-assisted manufacturing (CAD/CAM) prosthesis allow surgeons to consider the resection of large intraosseous meningioma with one-step reconstruction with a patient specific implant (PSI) designed using a preoperative three dimensional computerized tomography (3D CT) scan[11].

This report describes the case of 20 year old male suffering from fronto Orbital Eosinophilic granuloma treated by surgical resection using a 3D printed resection template modelled on the 3D printed skull of the patient with a one-step reconstruction of the skull defect using a computer-assisted designed PEEK PSI.

### Case Description

A 20-year-old man was admitted to our unit with a swelling above his right eye that had been gradually



**Figure 1:** Right supra orbital mass

increasing in size over three months. He reported no history of trauma, pain, redness, discharge, or vision problems associated with the swelling. Upon examination, the swelling measured approximately 2x3 cm and felt firm to the touch, with slight tenderness. There was a bony gap above supra orbital ridge, but no pulsation, bruit, or protrusion of the eyeball was noted.



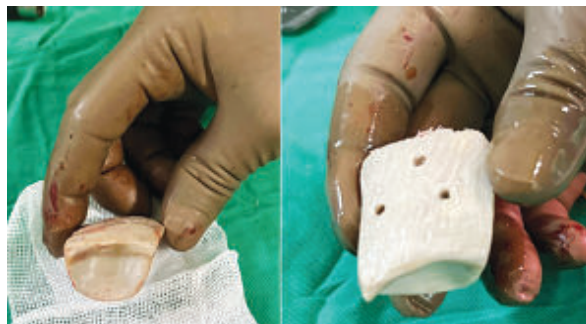
**Figure 2:** X-ray of skull shows Osteolytic lesion



**Figure 3:** 3D CT shows involvement of orbital roof

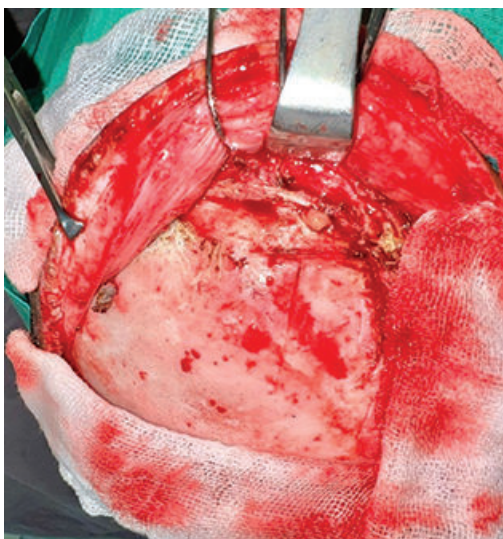


**Figure 4:** Contrast MRI shows intracranial extension

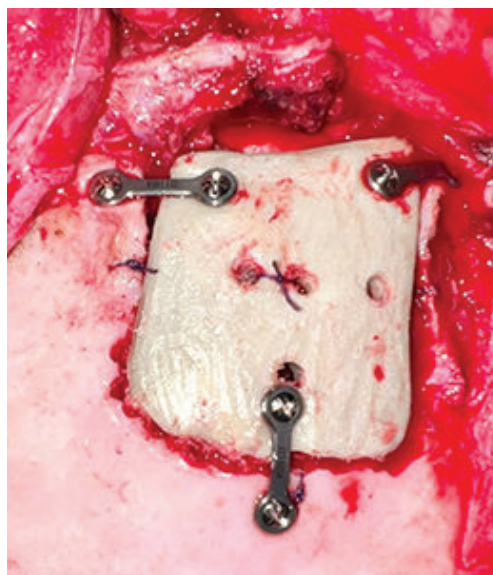


**Figure 7:** PEEK material ready for Cranioplasty

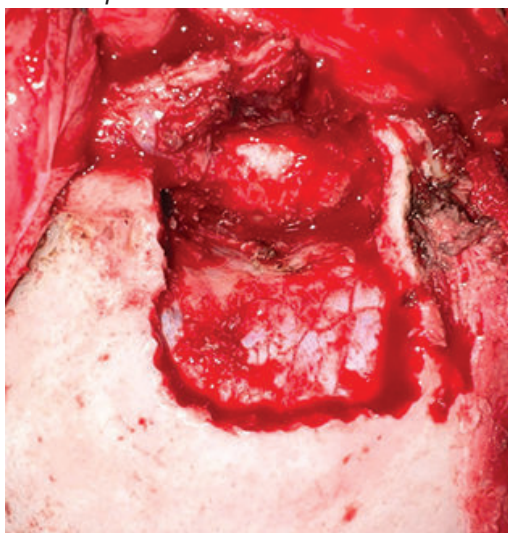
Neurologically, he was intact. Plain X-ray and CT scan revealed a round to oval osteolytic lesion in the right lateral supraorbital region, involving the roof of the orbit. MRI showed a non-enhancing soft tissue mass above the right eye, extended intracranially into the frontal



**Figure 5:** Exposure of lesion



**Figure 8:** Cranioplasty done



**Figure 6:** Removal of lesion & completing bony work



**Figure 9:** 1<sup>st</sup> Post-operative day

base. We diagnosed him as a case of Eosinophilic Granuloma involving the right orbit with intracranial extension to right frontal lobe. Surgical removal of the tumor and affected bone was performed, followed by reconstruction of orbital roof and Frontal bone by one step Customised PEEK Cranioplasty. Histological examination confirmed infiltration of Langerhans histiocytes with areas of necrosis and the presence of tumor giant cells, confirming the diagnosis of Eosinophilic Granuloma of the skull.



**Figure 10:** Follow up after 4 months

### Discussion

Cranioplasty in the skull remains difficult for specialists in craniofacial and neurosurgery. Often, surgeries are done after a craniectomy, especially when it's done to relieve pressure on the brain or to treat infections. CAD/CAM technology enables surgeons to carry out single step reconstructions, particularly following extensive benign tumor removals without skin involvement, utilizing PSI. Numerous techniques have been devised over time for such resections. Some researchers have outlined the employment of a neuronavigation system fused with the virtual design of the implant to precisely demarcate resection margins, while others favors the utilization of a resection template or a blend of both approaches. CAD/CAM technology enables the creation of stereo lithographic models and 3D printed skulls. Some researchers have devised indirect methods involving molding PSI directly on the 3D printed models using PMMA, except for PEEK and PMMA materials.

Previously hydroxyapatite or titanium were the main materials utilized for computer assisted allograft procedure. PEEK implants are highly suitable for computer assisted surgery because they offer excellent confirmation potential, can be drilled when necessary, and boast mechanical strength similar to cortical bone. Additionally they are biocompatible, comfortable for patients, and radiolucent, making surgery for these implants feasible. Their thermo stability is another significant advantage. The PEEK implant can undergo sterilization multiple times without losing its shape, making it useful for reuse after infection treatment. However, drawbacks include its high cost and lack of integration with bone tissue. The expense must be weighed against benefits such as reduced surgery time or the avoidance of additional procedures compared to other methods like intraoperative molding or two step procedures. Two recent meta-analysis have found that PEEK Cranioplasty carry a lower risk of infection compared to autologous or PMMA Cranioplasty, which addresses a major concern about their cost. While titanium mesh offer similar strength and cosmetic outcomes with low infection rates, it can deform under trauma and cause significant radiological artifacts.

### Conclusion:

A single-step surgical method employing CAD/CAM technology to create a custom PEEK implant for individual patients is both safe and precise, yielding favorable cosmetic outcomes. This approach is expected to decrease surgical duration and minimize postoperative complications, notably infection.

### Referencing:

1. Yang JT, Chang CN, Lui TN, Ho YS. Eosinophilic granuloma of the skull—report of four cases. *Changgeng Yi Xue Za Zhi*. 1993 Dec 1;16(4):257-62.
2. Park SH, Park J, Hwang JH, Hwang SK, Hamm IS, Park YM. Eosinophilic granuloma of the skull: a retrospective analysis. *Pediatr Neurosurg*. 2007;43(2):97-101. doi: 10.1159/000098380. PMID: 17337919.
3. Rawlings III CE, Wilkins RH. Solitary eosinophilic granuloma of the skull. *Neurosurgery*. 1984 Aug 1;15(2):155-61.
4. Brandicourt P, Delanoé F, Roux FE, Jalbert F, Brauge D, Lauwers F: Reconstruction of cranial vault defect with polyetheretherketone implants. *World Neurosurg* 105:783-789, 2017 2.
5. Broeckx CE, Maal TJJ, Vreeken RD, Bos RRM, ter Laan M: Single-step resection of an intraosseous meningioma and cranial reconstruction: Technical note. *World Neurosurg* 108:225-229, 2017 3.

6. Guerrini F, Dallolio V, Grimod G, Cesana C, Vismara D, Franzin AB: It is time to reduce free-hand manipulation: Case report of our proposal for an innovative 1-step cranioplasty. *World Neurosurg* 107:1052.e7-1052.e10, 2017 4.
7. Jalbert F, Boetto S, Nadon F, Lauwers F, Schmidt E, Lopez R: One-step primary reconstruction for complex craniofacial resection with PEEK custom-made implants. *J Craniomaxillofac Surg* 42(2):141-148, 2014 5.
8. Jonkergouw J, van de Vijfeijken SECM, Nout E, Theys T, Van de Castele E, Folkersma H, Depauw PRAM, Becking AG: Outcome in patient-specific PEEK cranioplasty: A twocenter cohort study of 40 implants. *J Craniomaxillofac Surg* 44(9):1266-1272, 2016
9. Shah AM, Jung H, Skirboll S: Materials used in cranioplasty: A history and analysis. *Neurosurgical Focus* 36(4):E19, 2014
10. Zanotti B, Zingaretti N, Verlicchi A, Robiony M, Alfieri A, Parodi PC: Cranioplasty: Review of materials. *J Craniofacial Surg* 27(8):2061-2072, 2016
11. Barros A, Brauge D, QuÉhan R, Cavallier Z, Roux FE, Moysé E. One-Step Customized PEEK Cranioplasty After 3D Printed Resection Template Assisted Surgery for a Frontal Intraosseous Meningioma: A Case Report. *Turk Neurosurg.* 2021;31(1):142-147. doi: 10.5137/1019-5149.JTN.30192-20.2. PMID: 33372259.