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TECHNICAL NOTE

One-stage frame-guided resection and reconstruction with PEEK custom-made prostheses for predominantly intraosseous meningiomas: technical notes and a case series

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Abstract

Treatment of intraosseous meningiomas is a two-staged procedure including tumor resection and cranial reconstruction. Several are the potential pitfall of this kind of surgery such as the conspicuous dimensions usually reached by the tumor and the peculiar involvement of irregular and deep bony structures. For these reasons, a surgical accurate virtual planning and a careful removal followed by tailored reconstruction are mandatory to achieve satisfactory results. We analyzed six patients operated on for intraosseous meningiomas between September 2014 and June 2018. Resection strategy was planned beforehand and shared with the manufacturer who provided the PEEK cranioplasty used in the reconstructive phase. Between September 2014 and April 2018, six patients affected by intraosseous meningioma were operated on. Female/male ratio was 5:1 and mean age was 54 ± 10.8 years. Mean FU was 20.3 ± 16.4 months. Mean dimension was 73.9 ± 24.8 mm × 69.2 ± 16.2 mm. Mean surgical time was 5.1 ± 1.1 h. The resection of intraosseous meningiomas requires the earliest and finest reconstructive phase. Custom-made implants should be considered the gold-standard for cranioplasty, especially in large skull and in frontal or hairless areas of the skull. The described technique is simple, accurate, and effective in achieving good results in disease control as well as cosmetic and functional restoration.

Keywords Prostheses · Intraosseous meningiomas · Cranial reconstruction

Introduction

Surgical excision of the tumor along with its dural origin represents the treatment of choice for symptomatic meningiomas and provides opportunity for long-term remission [1]. Treatment of intraosseous meningiomas entails complete tumor resection including the bony involvement followed by cranial reconstruction. From an anatomopathological point of view, two entities can be encountered in the clinical practice: primitive intraosseous meningiomas (PIMs), rare entities involving mainly the fronto-sphenoidal region arising from the bone and secondarily involving the dural sheet; en-plaque/nodular meningiomas which arise from the arachnoid membrane and secondary extensive bone involvement.

Several issues increase the complexity of the surgical management of these lesions. First, the conspicuous dimensions usually reached and the peculiar involvement of irregular and deep bony structures as the sphenoid bone, the orbit, and the paranasal sinuses make surgical radicality difficult; furthermore, the aforementioned anatomical complexity of the fronto-spheno-orbital bones pose objective difficulties also during the reconstruction step, which usually is a straightforward procedure. Indeed, the restoration of a good degree of symmetry is crucial to provide good functional and cosmetic outcomes in such cases. For these reasons, an accurate virtual planning of the resection and reconstruction are mandatory in order to achieve satisfactory results. The repair of the bony defect with a pre-prepared patient specific implants (PSI) in a one-stage procedure has shown to reduce operative time and morbidity [6, 19].

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We herein report on our surgical series consisting of six consecutive patients harboring an intraosseous meningioma operated on at our Institution between September 2014 and June 2018. An in-depth description of the surgical technique of pre-planned one-stage resection and polyether ether ketone (PEEK) custom-made prostheses reconstruction along with a discussion of the pertinent literature is provided. To the best of our knowledge, ours is the largest series reporting one-stage frame-guided resection and reconstruction with PEEK custom-made prostheses for predominantly intraosseous meningiomas.

Technical aspects

Patient characteristics

We retrospectively analyzed our series consisting of six patients (5 female and 1 male) harboring predominantly intraosseous meningioma operated on at our Institution between September 2014 and June 2018 (Table 1). Demographical, clinical, and radiological data were collected. Mean age at presentation was 54 ± 10.83 years. In four cases, the lesion was located in the fronto-sphenoid region and in two patients on the frontal convexity (one case with frontal sinus involvement). Mean surgical time was 305.8 ± 62.88 min. All cases resulted grade I meningiomas accordingly to WHO classification [13]. Simpson grade I resection was achieved in all cases but one (Simpson grade III). At last follow-up (mean 20.33 ± 16.39 months), all patients had good functional and cosmetic outcome according to the Sloan classification [18]. In all cases, we used the custom-made prostheses made of PEEK, a light semi-crystalline thermoplastic biocompatible material, which has quite similar stiffness and strength to the bone (Synthes® PSI).

Preoperative CT imaging acquisition

Preoperative radiologic studies involved the acquisition of a thin-cut skull CT scan with preset parameters in order to achieve excellent reconstructive results: matrix 512 × 512, slice thickness 1.0 mm, feed per rotation 1.0 mm, reconstructed slice increment 1.0 mm, reconstructed algorithm bone, gantry tilt 0. CT datasets were shared and sent in Digital Imaging and Communications in Medicine (DICOM) format to the manufacturer firm (Synthes®). A brain MRI was also performed in order to better define tumor relation with brain parenchyma.

Virtual resection and manufacturing of the PSI and the frame guide

Tumor resection strategy was planned by the surgeon beforehand and shared in a web conference with the engineer of the manufacturer. The virtual resection was realized on the 3D model with simultaneous checking of the bone involvement on CT slices (Trumatch, Comprehensive Solutions in Facial Reconstruction CMF). This virtual simulation was made through the use of "The SurgiCase Viewer", a sub-program of the Trumatch CMF. This tool consents to navigate the skull of the patient after creating a 3D reconstruction of it. Several possible actions can be done on the model such as: rotation, translation, zooming, sectioning, and measuring (angle, thickness, and length). In this way a 3D imaging of the tumor and of the supposed resection can be planned in axial, coronal, and sagittal plane. Furthermore, those actions can be undone any second, and it is possible to make the model transparent to gain a better 3D view during virtual resection. The bone was considered pathological when appeared thicker, without differentiation between the cortical and spongiosa layers. A supplementary margin of at least 10 mm was considered as safety margin, except in areas near the frontal sinus (except of its major involvement in one case) and the superior sagittal sinus. As the simulation of resection was approved, the patient-specific prostheses were planned and designed to cover the bone defect by means of a computer-aided design software. Furthermore, a surgical guide was designed in the form of a frame complementary to the prosthesis. Finally, the two 4-mm thick models were manufactured and sent to the hospital for the surgical use after sterilization (Fig. 1). The entire process lasted about 4 weeks.

Table 1 Patients sample. *FO*, fronto-orbital; *FT*, fronto-temporal; *F*, frontal; *FP*, fronto-parietal

Case	Sex	Age	Location	Presentation	Surgical time (min)	Complications	KPS	Sloan scale	Follow-up (months)
No. 1	F	63	FO	Exophthalmos	230	None	100	Class 1	45
No. 2	F	70	FT	Hard swelling	360	None	100	Class 2	32
No. 3	F	41	F	Hard painful swelling	285	None	100	Class 1	14
No. 4	F	46	FT	Hard swelling	345	None	100	Class 1	23
No. 5	F	54	FP	Hard swelling, visual deficit	375	Speech disturbances	100	Class 1	1
6#	M	50	F	Hard swelling	240	None	100	Class 1	7

Operative technique

The patient was positioned supine and the head was fixed in a 3-pin head holder. In case of fronto-sphenoid localization, after a curvilinear fronto-temporal incision and temporalis muscle subfascial dissection, a fronto-temporal craniotomy was performed. The two cases at frontal convexity required neutral position and a bicoronaric flap with extensive galea pericranium preservation. The extent of bone removal was determined for each patient based on tumor location and extension during the virtual planning. The pre-planned frame was then used to check resection completeness on the antero-posterior and cranio-caudal axes following the idea of “if the flap does not fit, there is still tumor left”. The entire area of hyperostosis was removed using a series of cutting burrs and high-speed drill, resulting in a craniectomy exposing the dura. During this delicate step, bleedings coming from the bone were stopped with bone wax and using diamond drill. The superior orbital fissure and, whenever needed, the optic canal was decompressed using roungeurs. The involved dura is exposed until the border free from disease and excised.

Reconstruction and outcome

After duraplasty with galea or synthetic substitutes, PSIs were positioned and secured to the bone with plates and screws. Eventually, they can be also gently remodeled using the drill during the surgery to adapt it and let it fit better to the bone margins [3, 7]. The dura was then suspended in order to avoid epidural bleeding.

Personal experience (6 cases)

Between September 2014 and April 2018, we operated on six patients affected by predominantly intraosseous meningioma at our Institution. Five patients were female (gender ratio 5:1) and mean age was 54 ± 10.8 years. Mean FU was 20.3 ± 16.4 months. Mean maximum AP diameter in axial CT was 73.9 ± 24.8 mm and maximum CC in coronal CT was 69.2 ± 16.2 mm. Mean surgical time for the one step procedure was 5.1 ± 1.1 h (305.8 ± 62.9 min). In Simpson III patient, resection was stopped due to profuse bleeding; hence, to adapt the prepared cranioplasty, some drilling was done to make it fit. Patients’ characteristics are summarized in Table 1.

Case 1 (Figs. 2 and 3)

A 70-year-old female with short history of dizziness and balance disturbances underwent MR imaging which showed a left side en-plaque meningioma with extensive hyperostosis involving pterional region, great sphenoid wing, and temporal squama. The lesion displayed compressive effect on the brain with shift of the midline. Another similar but smaller lesion involving the right anterior clinoid process and tentorial edge was found out. The patient showed no signs at neurological examination and a large painless left pterional lump at local examination. The lesion was 71.5 × 81.4 mm in maximum diameter. After preoperative imaging for the prosthesis production, the patient underwent the one-stage procedure; interfascial dissection of the temporalis muscle allowed to preserve its innervation and vasculature in order to avoid atrophy with functional and esthetic problems. During the craniotomy, the large frontal sinus was opened; then, it was cranialized with muscle and pericranium. The operative time was 360 min and the postoperative CT scan showed no complications. Histological diagnosis showed transitional meningioma, WHO grade I. Postoperative course was uneventful. At 33.9 months, esthetic and functional outcome was excellent.

Case 2 (Fig. 4)

A 54-year-old female during hormonal therapy for menopause showed slowly increasing left frontal lump associated with ipsilateral visual deficit and exophthalmos. CT and MR imaging ruled out extensive hyperostosis involving the pterional region, the temporal squama, and the parietal convexity. A huge prosthesis was planned for the large defect originated from the 120.5 × 95.9 mm lesion. A craniectomy was performed using drill on a large left fronto-temporo-parietal flap. Pterional region drilling and orbital lateral wall and roof removal were necessary, and dural removal showed aggressive invasion of the arachnoidal and pial plane. The one-stage procedure lasted 375 min. Multiple dural suspensions worked to avoid epidural collection but at postoperative CT scan, multiple hemorrhagic foci and clinical course were characterized by aphasia that recovered in 3 months. At last follow-up (3 months), radiological and clinical outcomes were good, excellent, and esthetic.

Discussion

PIMs are tumors arising from the arachnoid cap cells at the skull vault and the skull base with major involvement of the bone and related hyperostosis [17]. These are rare entities while intraosseous extension of meningioma is quite common especially in fronto-orbital regions. This type of meningiomas usually affects the lesser sphenoid wing, the orbital roof, the lateral orbital wall and the middle fossa, sometimes with bony proliferation of the anterior clinoid process, the superior orbital fissure, the optic canal, and the sphenoid or ethmoidal sinuses. These tumors are typically slow-growing thus allowing careful surgical planning [5]. Failure to accomplish a good

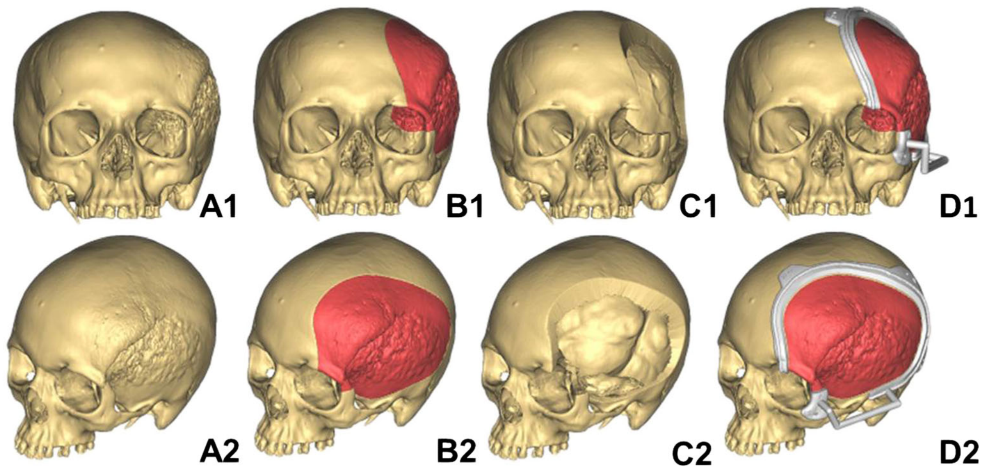

Fig. 1 Figure PSI. Surgical plan: Preoperative situation (A1, A2); Osteotomies (B1, B2); Planned outcome (C1, C2); PSI resection guide (D1, D2)

reconstruction can bring to esthetic and functional issues that can need reintervention [2, 10].

One-stage procedure represents the treatment of choice for this type of lesions. Surgical treatment for intraosseous meningiomas is a challenging task as it combines three difficult procedures: meticulous surgical planning, tumor resection, and cranial reconstruction. The first step can be complicated by the complex three-dimensional configuration of the involved bone; the second one by the thickness of the bone as well as by the depth of some of the involved areas that overall make a total resection harder to achieve.

The most interesting published series are those in which the one-stage surgical procedure was aided either by neuronavigation [3, 15, 21] or by using a frame [3, 7] to guide resection and reconstruction. These experiences are those majorly comparable with our series in which a frame-guided resection is proposed. Being considered nowadays, one-stage procedure, the gold standard in the treatment of

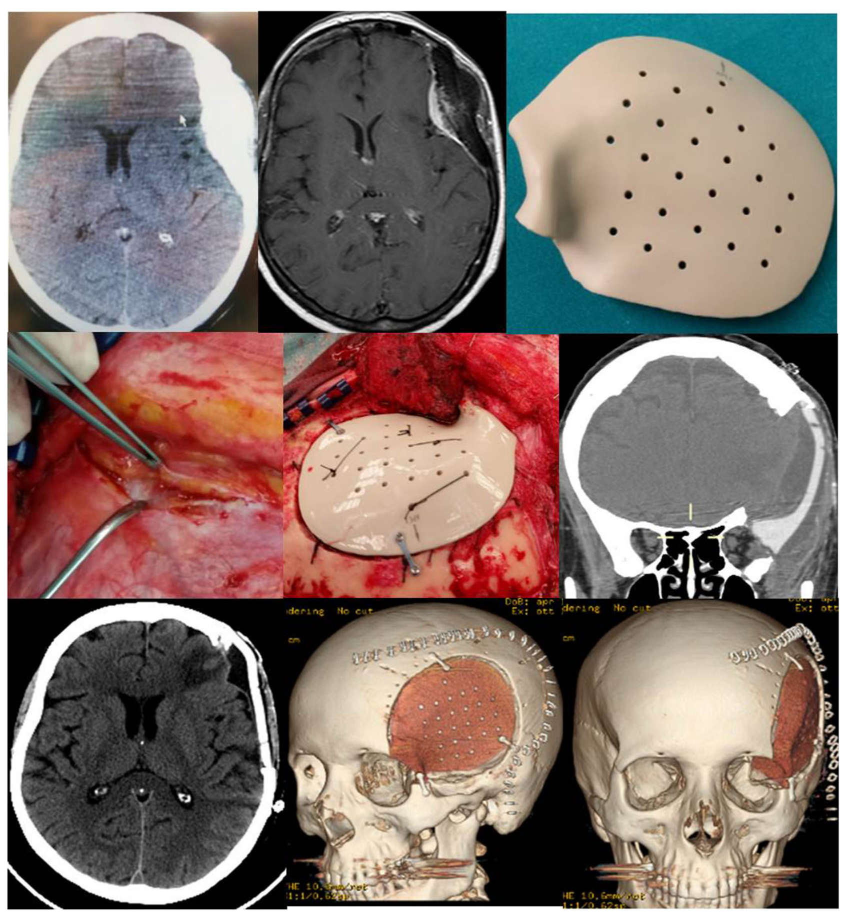

Fig. 2 MR imaging showing a left side en-plaque meningioma with extensive hyperostosis involving pterional region, great sphenoid wing, and temporal squama

Fig. 3 A 70-year-old female with short history of dizziness and balance disturbances underwent MR imaging

predominantly intraosseous meningiomas, no comparison was made with two-staged procedures not being considered as an option in a benign pathology. Furthermore, no comparison with hand-made cranioplasty was made in consideration of the higher infection risk, of the non-comparable esthetic results, and of the risk of decubitus in the more frequent spiky edge of the hand-made cranioplasty. Nonetheless, a review of the literature on the matter was carried on. Only a few papers consider hand-molded cranioplasty as a way to complete reconstruction in intraosseous meningioma. Two of them deal with vault meningiomas while only one is referred to craniofacial reconstruction recognizing the esthetic limits of the technique [11, 14, 22]. As for the double-staged procedure, only a paper mentions it in a case of complex tumor with intracranial and extracranial extension with superior sagittal sinus involvement. In this case, two surgeries were needed to complete resection, and only in the second, one cranioplasty was performed [20].

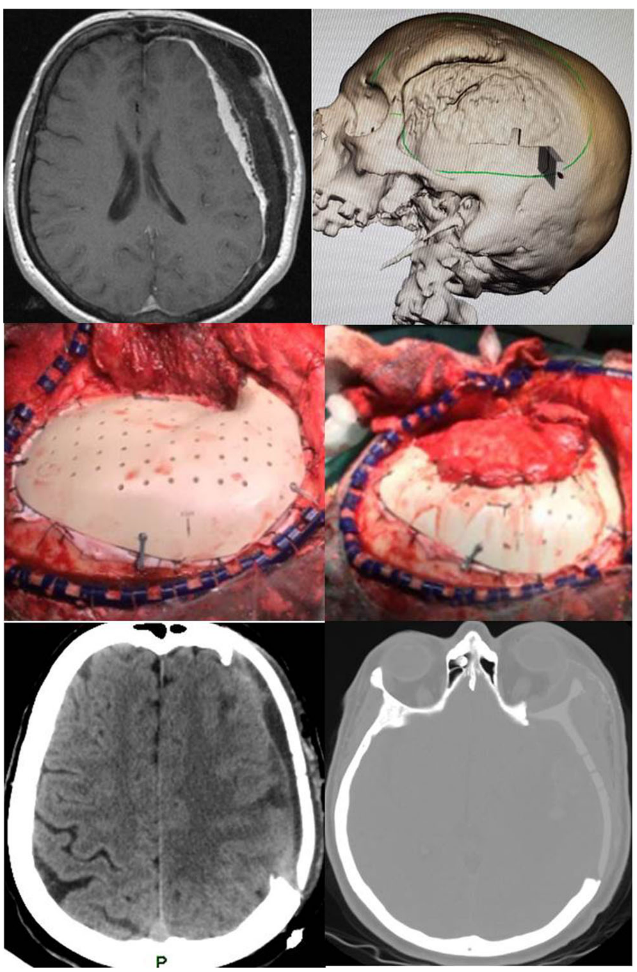

Fig. 4 CT and MR imaging ruled out extensive hyperostosis involving the pterional region, the temporal squama, and the parietal convexity

Similarly, to the experience of Broeckx and Carolus [3, 7], we used a frame mimicking the cranioplasty flap as a guide for resection and as a reliable indicator of tumor removal following the statement "if the flap does not fits, there is still tumor left". In other words, assuming that complete resection is the most important aspect to reach cure in this setting [1], planning resection during virtual surgery can help the surgeon not to miss hidden portion of the tumors in deep areas Other authors suggested navigation-assisted procedures [15, 21] while Broeckx et al. combined the two procedures [3] order to maximize resection.

Reconstruction of the skull after resection of an intraosseous meningioma is another challenging step. The skull, in particular in periorbital region, has a moderate-to-high inter-individual anatomic variability and also varies with age and ethnicity [8]. For many years, the gold standard graft for calvarial reconstruction in small and medium-sized defects has been the autologous bone due to its bio- and genetic compatibility, and the unlikelihood of rejection. However, in the last 20 years, the progress of bioengineering has led to a numerous array of alternative compatible materials such as from metals (titanium and tantalum), alloplastic material (polymethylmethacrylate, PEEK), and hydroxyapatite (HA) [12]. The evolution in biomaterials and the possibility to pre-prepare cranioplasty flaps using symmetry principles have dramatically changed this task moving from intraoperative hand molding of the implant to computer-assisted preoperative fabrication. It is well known, in fact, how PSI (patient specific implants) designed on CT scan through mirroring the contralateral unaffected side are used in cranioplasty after trauma with excellent results [16]. The same procedure has been used for reconstruction after resection of bone lesions, primary as osteomas or secondary as meningiomas [9]. The main difficulty in performing computer-assisted prosthesis reconstruction after skull resection is that the expected defect is not visible on the preoperative CT scan, thereby preventing any direct prosthesis modeling based on mirroring of the unaffected side [4]. In our experience, the computer-assisted virtual resection of a tumor defect followed by the placement of a well-matched ready-to-implant prosthesis, is a simpler and faster method that allows at the same time an accurate bone resection and good esthetic results. Among different materials for PSI, over the last years, polyetheretherketone (PEEK) has proved to be the most reliable in our Institution and is considered the material of choice. PEEK displays numerous advantageous properties: it has excellent biocompatibility, good mechanical strength, radiographic translucency with consequent high quality of follow-up imaging; it is resistant to thermal and ionizing radiation (compatible with radiotherapy) with no changes in mechanical behavior (repeatedly sterilizable). The few disadvantages of PEEK are the following: it is quite expensive and it lacks osteointegrative properties; actually, PEEK is a hydrophobic material. As such, it does not actively bond to tissue, creating potential issues as regards to anatomical integration as bone cells will not bond to the implants. Finally, the PEEK mirroring technique gives the possibility, in case of recurrence, to either substitute the whole cranioplasty after a “second virtual removal” or to customize a smaller new cranioplasty fitting the recurrence removal in order not to touch the older surgical field and to connect the two cranioplasty. Usually, the second option is the most used in consideration of the higher cost of creating a new bigger cranioplasty.

Limitations of the technique

The effective transposition of a virtual treatment plan into the intraoperative setting strongly depends on the accuracy of the planning itself and on the experience of the operating surgeon. Nevertheless, advances in hardware and software development are making these systems increasingly user-friendly and more cost-effective, although costs remain high and there is still a need for considerable technical expertise.

Conclusions

The resection of predominantly intraosseous meningiomas, particularly in the fronto-orbital region, results in a significant cranial defects and deformity with functional issues and requires the earliest and finest reconstructive phase. Custom-made implants should be considered the gold-standard for cranioplasty, especially in skull defects greater than 25 cm^2, and in frontal or hairless areas of the skull. CAD/CAM fabrication of custom implants produces satisfactory cosmetic results, is technically simple, and can reduce surgery times and surgical blood loss without donor site morbidity, with faster recovery and lower indirect costs. To the best of our knowledge, our series represents the largest series of consecutive cases of predominantly intraosseous meningiomas treated with one-stage surgery with PEEK PSI produced with virtual planning of resection and reconstruction. The described technique is simple and accurate and has allowed us to achieve optimal tumor resection and cranial reconstruction with good results in terms of disease control and cosmetic and functional restoration without early or delayed infections, not even in the frontal region.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers’ bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements) or non-financial interest (such as personal or professional relationships, affiliations, knowledge, or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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