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Single Step Resection-Reconstruction Using Precurved Titanium Mesh of a Giant Intradiploic Meningioma Mimicking Bone Malignancy: Technical Note

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ABSTRACT

Intradiploic meningiomas are rare neoplasms, often mistaken for metastases or malignant bone tumors. Surgical management can be challenging, considering their diffusive bony invasion. Two main critical decisions need to be taken: the timing for cranial vault reconstruction and the choice of the adequate material for cranioplasty. We believe that this case underscores the complexity of such lesions, the importance of a prompt devascularization, and the pivotal role of an immediate reconstruction to avoid the additional morbidity of a re-do surgery.

Here, we report a case of 68-year-old men who presented with slow growing right parietal bone swelling he noted many years before, but for which he didn't seek medical attentions, associated with mild contralateral hemiparesis. Neuroradiological examinations revealed a giant extradural intradiploic tumor affecting the right temporo-parietal bone and conditioning significant compression of the underlying brain. We planned a surgical strategy to deafferent the tumor and to reduce the intraoperative bleeding. At first, a circumferential craniectomy centered upon the lesion was performed, then it was devascularized by means of surgical ligation of the ipsilateral superficial temporal artery (STA) and middle meningeal artery (MMA); these steps allowed a subsequent en block tumor excision, despite its large size, without significant blood loss and respecting the oncological principles. At the end, a contextual calvarial reconstruction was performed using a precurved titanium mesh. The patient was discharged seven days after surgery with complete recovery of the left-sided motor deficit. Thereafter, he underwent scheduled outpatient evaluations and radiological examinations. At 1-year follow-up, the Modified Rankin Scale (MRS) was 1, with no evidence of recurrent disease.

To conclude, surgical complications can be reduced adopting an optimal preoperative work-up and a tailored surgical strategy focused on early tumor deafferentation. Moreover, an immediate cranial vault reconstruction avoids the risks related to a second procedure.

KEYWORDS: Brain, Neoplasm, Intraosseus, Intradiploic, Meningioma, Primary extradural meningioma

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INTRODUCTION

Intradiploic meningiomas are rare primary extradural meningiomas (PEM), accounting for about 1-2% of overall intracranial meningiomas. This amount could be underestimated since they are often misdiagnosed as primary bone tumors (1). Intradiploic meningiomas appear to be slow growing histologically benign lesions. Only few of them, classified as malignant, exhibit aggressive behavior.

The current body of literature lacks specific guidelines for medical and surgical management of such pathological entities, since only few experiences about PEM have been reported so far (11).

We illustrate herein the case of a giant osteolytic intradiploic meningioma, which was completely resected in a single procedure, featuring a contextual calvarial reconstruction.

CASE REPORT

We report a case of 68-year-old men who presented with slow growing right parietal bone swelling he noted many years before, but for which he didn't seek medical attentions.

For the later occurrence of left-sided hemiparesis, the patient underwent Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) scans that revealed a large intradiploic extradural mass embedded in the right temporo- parietal bone measuring 8x5 cm, conditioning significant parenchymal compression, with midline shift and subfalcine herniation. On MRI, the lesion was slightly iso-hyperintense to brain on T2-weighted images (WI) and presented intense enhancement after gadolinium administration (Figure 1).

Differential diagnosis included Intradiploic Meningioma, Giant Cell Tumor, Osteogenic Sarcoma, Eosinophilic Granuloma, Aneurysmatic Bone Cyst, and bone metastases.

The patient was admitted to our Neurosurgery Department. Neurological examination highlighted a left mild-grade brachiocrural hemiparesis (MRC=4-/5). On physical examination, the swelling appeared globe-shaped, hard, and painless on palpation; the overlying skin was intact and freely movable.

Preoperative Planning and Surgical Intervention

Three-Dimentional reconstructions of preoperative MRI (Figure 2) showed a marked hypertrophy of the right

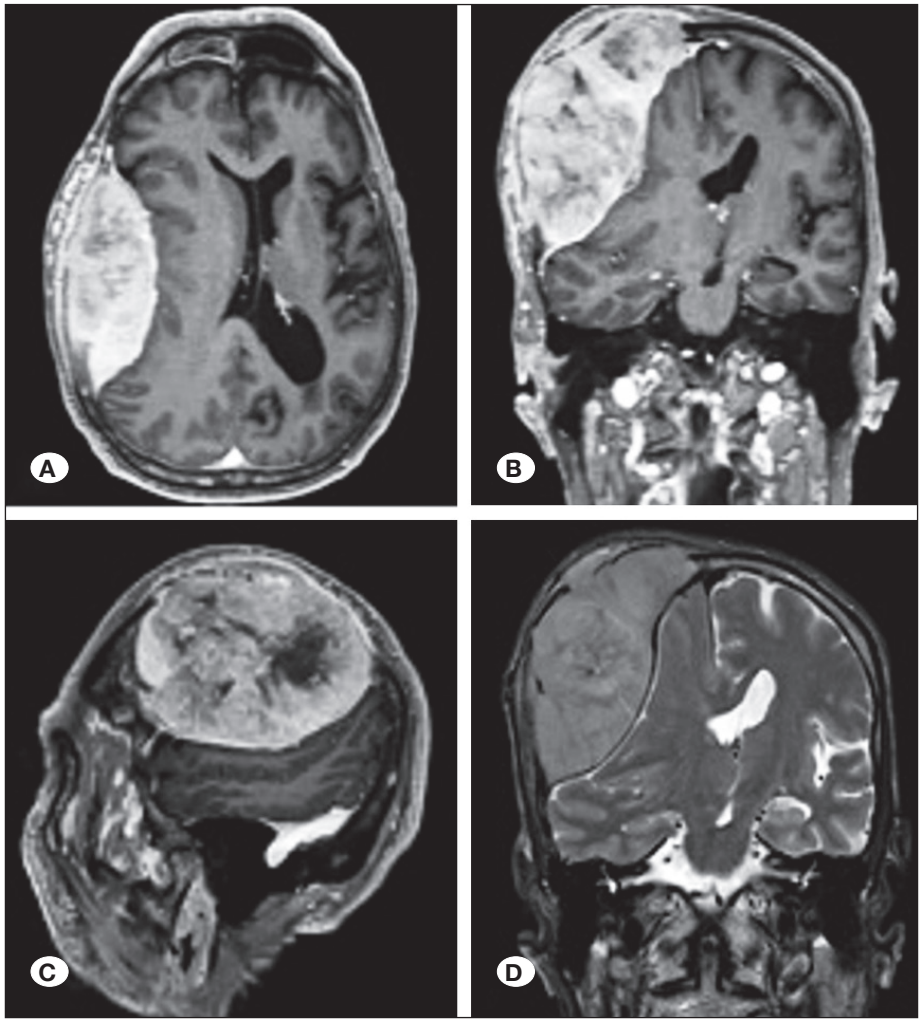

Figure 1: A) Axial brain Magnetic Resonance Imaging (MRI) demonstrated a massive fronto-temporo-parietal intradiploic meningioma with diffuse contrast-enhancement.
B, D) Coronal T2- weighted MRI showed significant parenchymal compression, midline shift and subfalcine herniation.
C) Right parasagittal sequences showed significant parenchymal compressione.

superficial temporal artery (STA) and middle meningeal artery (MMA) compared to the contralateral side (18). According to this aspect, we assumed that STA and MMA were the main feeders of the tumor. Thus, we planned a surgical strategy aimed to ligate those arteries in the first phase of the surgery, in order to deafferent the tumor and to reduce the intraoperative bleeding.

The operation was performed under general anesthesia. The patient was placed supine, with the head fixed in a Mayfield heal-holder, slightly extended, and rotated nearly 60° towards the left side. Cranial neuronavigation with SonoWand (Mison, Trondheim, Norway) was adopted throughout the operation, as illustrated in previous reports (22,23). A right fronto-parieto-temporal incision was carried on in a curvilinear fashion, to expose the whole lesion and to reach inferiorly the zygomatic root. The STA was isolated, ligated using silk thread and cut at the very first stage. A circumferential craniectomy around the lesion was performed by means of high-speed drill and craniotome, removing the entire pathological bone and exposing the unaffected dura mater, some inches beyond the tumoral boundaries. Once detected, the MMA was coagulated at foramen spinosum. The tumor was found to originate from the endosteal (or periosteal) layer of the dura mater with subsequent intradiploic osteolytic growth; once the dura mater was removed, the underneath arachnoidal plane resulted unaffected (Figure 3).

After tumoral vascular deafferentation, we conducted the dissection of the inner dural surface with the assistance of flexible fiber CO2 laser (21). A Simpson grade 1 surgical resection was achieved. Duraplasty was accomplished with a

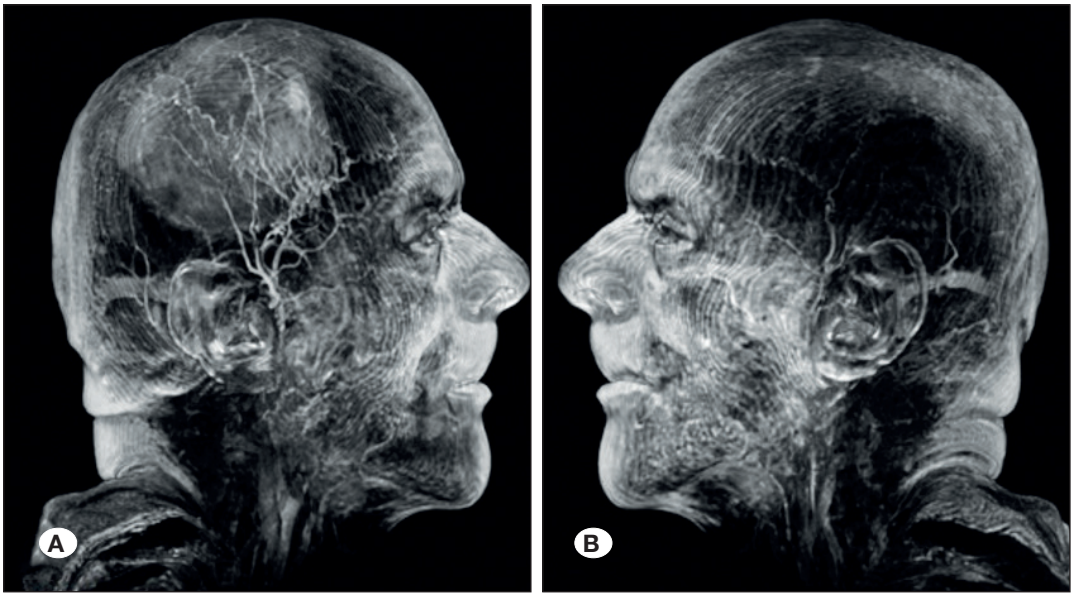

Figure 2: A) 3D reconstructions of preoperative MRI showed on the right side, a marked hypertrophy of the superficial temporal and middle meningeal artery, **B)** compared to the left contralateral side.

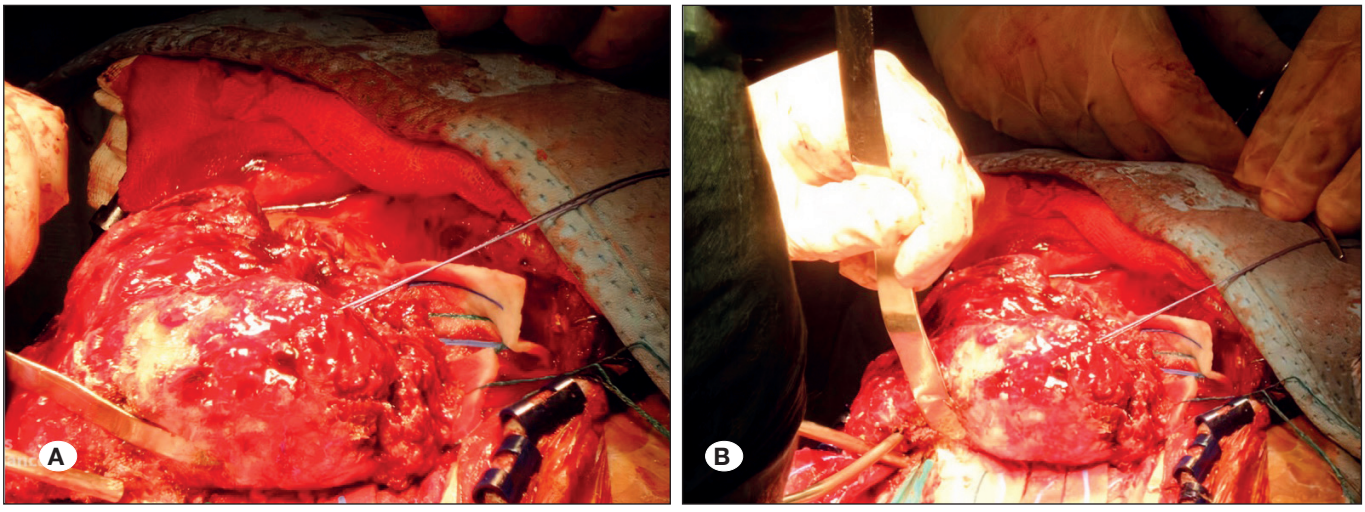

Figure 3: A) The neoplasia largely infiltrated the dural plane without exceeding the inner surface of the dura mater. **B)** Along the entire tumor surface, the arachnoidal plane respected.

dural patch, reinforced overlay by a pedicled pericranial flap harvested locally to decrease the risk of CSF leakage (19). Finally, cranial reconstruction was completed using a precurved titanium mesh (CRANIOTOP Zimmer Biomet- CL Instruments, Germany) to fill of the calvarial bony defecit (20).

Histopathological Findings

Histopathological examination highlighted the proliferation of meningothelial cells arranged in bundles or in vorticed pattern, characterized by round and tapered nuclei with inclusions and eosinophilic cytoplasm. These characteristics were suggestive of meningotheliomatous meningioma Grade I (Ki67<1%) according to the 2016 World Health Organization Classification of Tumors of the Central Nervous System (2016 CNS WHO).

The patient was discharged seven days after surgery with complete recovery of the left-sided motor deficit. Thereafter, he underwent scheduled outpatient evaluations and radiological exams. After 1 year, the Modified Rankin Scale (MRS) was 1, with no evidence of recurrent disease (Figure 4).

The patient enrolled has provided its written consent for anonymous data collection and inclusion in the present study.

DISCUSSION

Primary Extradural Meningioma

Meningiomas represent the most common benign primary intracranial neoplasms (24). They are generally slow- growing tumors composed of abnormal arachnoidal cells (27). Most of the time, meningiomas arise from arachnoid cap cells and show intracranial growth pattern. Some of them may cause hyperostosis of the nearer cranial vault or skull base regions.

Primary Extradural Meningioma (PEM) are those meningiomas arising outside the intracranial compartment. PEMs are rare tumors, classified by Lang et al. as purely extracalvarial (Type I), purely calvarial (Type II) and calvarial with extracalvarial extension (Type III) (13). Extracalvarial extension refers to an extraosseous origin of the tumor, whereas calvarial meningiomas originate solely from the bone (inner table, diploe and/or outer table). Type II and III tumors can be further classified as convexity (C) or skull base (B) forms (13). According to literature, most of PEM belong to Type IIIC (6,28) The tumor herein review belongs to Type IIC PEM.

The origin hypothesis of these tumors is still controversial, and several hypotheses have been made. The location of PEM might be connected to the entrapment of arachnoid cells across the cranial sutures during the embryogenesis. Nevertheless, it was found that only 8.3% of calvarial meningiomas had an origin correlated to this hypothesis (13).

Shuangshoti et al. suggested that multipotential mesenchymal cells could exhibit neoplastic transformation into meningioma cells, which might explain the extracranial location (26). Cushing and Eisenhardt have proposed a post- traumatic etiopathogenesis (7). According to this hypothesis, some arachnoidal cells could be entrapped in the fracture line due to head injury, resulting in the subsequent development of meningioma at that level. Nevertheless, only about 0.2-4% of patients with PEM had a history of significant head trauma (13).

In our case, considering patient's medical history, tumor pathogenesis is more likely to be related to the first hypothesis concerning the entrapment of arachnoid cells during embryogenesis.

These tumors can be diagnosed with a contrast-enhanced MRI: they appear to be typically hypointense on T1-WI and hyperintense on T2-WI and present intense and homogeneous enhancement after gadolinium administration. However, a basal CT scan with bone window is often necessary to exclude sclerotic/osteolytic margins which characterize these types of

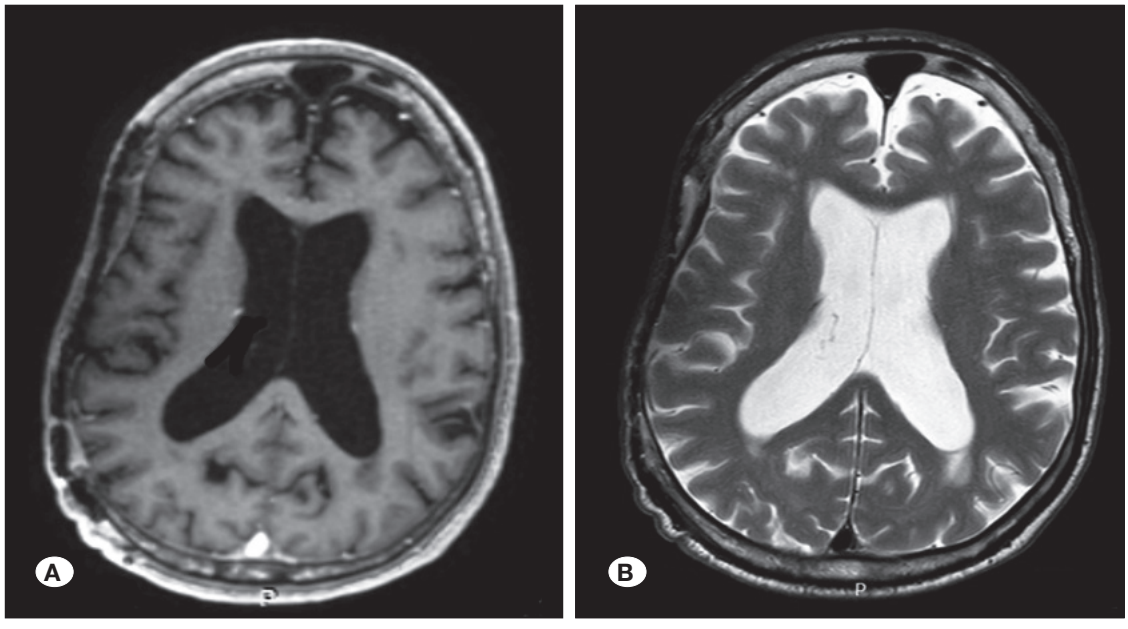

Figure 4: A) T1-weighted and **B)** T2-weighted post-operative Magnetic Resonance images demonstrate the complete resection of the tumor, the recovery of the midline shift and the optimal silhouette of right cranial vault obtained after the proper reconstruction of the bony defect.

Table I: Review of 13 Reports Dealing with Intradiploic Meningioma: Clinical Data

ID	Authors, Year	Age/Sex	Bony location	Type (Lang's classification)	Treatment	Dura invasion	Recurrence	Follow-up (months)
I	Kwon et al., 2019 (12)	80/M	Petrous	III B	GTR	None	None	12
II	Parington et al., 1995 (17)	84/F	Fronto-Temporal	IIIC	GTR + RT	Whole dura	Yes (double)	24
III	Bassiouni et al., 2006 (2)	62/F	Frontal	IIC	GTR	Whole dura	None	/
IV	Kim et al., 2012 (10)	68/M	Parietal	IIIC	GTR	Outer dural layer	None	12
V	Yun et al., 2014 (28)	64/F	Frontal	IIIC	GTR	Whole dura	None	12
VI	Bohara et al., 2016 (4)	38/M	Parietal	IIIC	STR + RT	Only dural adherence	None	6
VII	Lang et al., 2000 (13)	59/M	Sphenoid Wing	/	GTR	Whole dura	None	24
VIII	Cheng et al., 2012 (5)	68/F	Frontal	IIIC	GTR	Dural and brain	Yes (Single)	3
IX	Sakakibara et al., 2018 (25)	80/F	Parietal	IIIC	GTR	None	None	8
X	Liu et al., 2015 (14)	/	/	/	STR	/	/	/
XI	Nakae et al., 2017 (16)	30/M	Parietal	IIIC	GTR	Outer dural layer	None	/
XII	Kumar et al., 2021 (11)	40/M	Parietal	IIIC	GTR	None	None	12
XIII	**Di Pellegrini et al., 2022 (18) (*Present Case)**	**68/M**	**Fronto-Temporo-Parietal**	**IIC**	**GTR**	**None**	**None**	**12**

GTR: *Gross total resection,* ***STR:*** *Sub total resection,* ***RT:*** *Radiotherapy,* ***/:*** *Data not available,* ***M:*** *Male,* ***F:*** *Female.*

lesions. Intradiploic meningiomas could exhibit hyperostosis with osteoblastic and/or osteolytic features. It is important to note that osteolytic meningiomas which infiltrate soft tissues have a greater tendency to be atypical or malignant; thus, they should be considered as malignant tumors until proven otherwise.

Differential Diagnosis

Generally, intradiploic meningiomas are mistaken at diagnosis with skull bone tumors or fibrous dysplasia. Differential diagnosis includes cerebral metastases, plasmacytoma, giant cell tumor, osteogenic sarcoma, eosinophilic granuloma and aneurysmatic bone cyst. Most of the PEM are WHO grade I tumors. Meningothelial and psammomatous meningiomas are the most commonly identified subtypes (6). This case, according to the last WHO classification, was a grade I meningothelial meningioma. Atypical intraosseous meningiomas are rarely reported in the current body of literature.

Surgical Treatment

The treatment of choice for meningioma is Gross Total Resection (GTR). When suspecting an atypical/malignant tumor with extracalvarial component, a wide *en bloc* resection of the lesion with a 1 cm of safe bone supramarginal resection should be performed (28). In the case of large tumors, the risk of intraoperative blood loss is very high. Therefore, an appropriate surgical planning is mandatory to identify the main afferences to the tumor and to achieve an early devascularization.

Modern MRI systems with contrast enhancement, Angio-MRI, and 3D reconstructions (15,18) are able to identify successfully abnormal or hypertrophic vessels. In other cases, however, it is necessary to use the classical digital subtraction angiography (DSA) which remains the gold-standard for neurovascular assessment; it also delivers the priceless advantage of pre-operative tumoral embolization. Early devascularization (surgical or endovascular) reduces blood losses and simplifies the procedure. After complete tumor resection, the surgeon has to deal with the issue of bone reconstruction. Reconstruction can be achieved in two ways: a delayed cranioplasty (delivered in a second surgical intervention) or a contextual cranioplasty (single-step surgery with tumor resection and calvarial reconstruction). Regarding delayed cranioplasty, custom-made prosthesis can be shaped using different materials such as hydroxyapatite, titanium, polimethylmethacrylate (PMMA), polyetheretherketone (PEEK) (9,20). Compared with standard precurved mesh, custom-made prosthesis decreases early and late postoperative complications and increases cosmetic results (20). Delayed cranioplasty carries the drawback of subjecting patients to two different anaesthetic procedures and possibly delaying the administration of planned adjuvant therapies (RT, chemotherapy). During single-step surgery (resection–reconstruction), the reconstruction is carried out using standard malleable pre- curved mesh, adapting the prosthesis to the patient's cranial conformation as best as possible. While this strategy guarantees an immediate result avoiding lengthy waiting times for any subsequent therapies and the potential complications of a second procedure, the aesthetic and functional result may be less satisfactory (20).

Recently, however, technological evolution has allowed to prepare the construction of the prosthesis before surgery through the use of neuro-navigation. In selected cases of neoplasms, the use of planning procedures for bone removal in a preoperative phase can allow the production of 3D-printed custom-made prostheses to be used with a single step surgery (3). The planning of the prosthesis involves the creation of a cut-out mask of the skull that forms a bone defect complementary to the prosthesis itself. The bony cutting lines are planned in detail on the 3D reconstructions of the patient's skull considering the extent of infiltration of the skull cavity and the neoplastic mass to be removed. This allows immediate reconstruction with considerable benefits for the patient (3). Della Puppa et al. have shown how the preoperative design of the prosthesis is feasible by defining the cutting lines of the skull on the three-dimensional image of the patient's images (8). These strategies both allow to obtain optimal cranial reconstructions in a single surgical step.

Adjuvant Radiotherapy and Systemic Therapy

According to literature, most of these tumors are treated with Gross Total Resection [GRT] with or without adjuvant radiotherapy.

Regardless the pathological grading, in case of subtotal resections, the residual tumor should undergo radiotherapy (28). However, there is still disagreement about the need of adjuvant radiotherapy in atypical and malignant PEMs, despite complete surgical resection (13,28). Finally, systemic therapy can only be carried out when surgery and radiotherapy strategies are no longer available. Chemotherapy and molecular targeted drugs should be reserved for non-resectable PEMs, for recurrent WHO grade I meningiomas, and atypical/malignant grade II or III meningiomas (28). Targeted therapy, and immunotherapy for meningiomas are also under investigation.

CONCLUSION

Intradiploic PEM are relatively rare neoplasms. A complete neuroradiological evaluation is mandatory to make a optimal preoperative work-up and a tailored surgical strategy focused on early tumor deafferentation. Another issue is about choosing the appropriate surgical procedure for skull reconstruction (immediate or delayed). Both single- step surgery (resection–reconstruction) and delayed cranioplasty come with some pros and cons that should be carefully evaluated and openly discussed with the patient.

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Consent for publication: Written informed consent was taken to publish this manuscript and any accompanying images.

AUTHORSHIP CONTRIBUTION

Study conception and design: GD
Data collection: GN
Analysis and interpretation of results: RB
Draft manuscript preparation: AM, ES
Critical revision of the article: FVS
Other (study supervision, fundings, materials, etc...): DP
All authors (GD, RB, AM, GN, FVS, ES, PV, DP) reviewed the results and approved the final version of the manuscript.

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