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Neurosurgical Techniques

A multimodal staged approach for the resection of a Sylvian aqueduct rosette-forming glioneuronal tumor: A case report and literature review

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A B S T R A C T

Background and importance: The rosette-forming glioneuronal tumor (RGNT) is a rare central nervous system tumor which often arises intraventricularly. We report the first surgical case of an RGNT arising from the Sylvian aqueduct treated through a double approach.

Clinical presentation: A 25-year-old female presented with triventricular hydrocephalus on MRI secondary to a 2 cm Sylvian aqueduct mass. Emergent endoscopic third ventriculostomy with biopsy confirmed the diagnosis of RGNT. She was first followed up and due to the rapid tumor's growth a double surgical approach was proposed. The first was a telo-velar approach to the lower third of the aqueduct. The second stage was an endoscopic ultrasound aspirator aided transfrontal transforaminal approach; last postoperative MRI shows a 6 mm residual tumor. Patient leads an active working and social life.

Conclusion: Choosing a two stages approach for this rare and complex Sylvian aqueduct RGNT resulted in a positive clinical and radiological outcome.

1. Introduction

The rosette-forming glioneuronal tumor (RGNT) is a central nervous system lesion that has been considered an independent entity since 2007 and is included as a “neuronal and mixed neuronal-glial tumor” in the World Health Organization (WHO) classification system of 2016. RGNT shows a relatively well-defined tumor parenchyma interface and is composed of biphasic neurocytic and glial components. The tumor can arise in various sites of the brain but an intraventricular origin is most frequent [1]. Only one non-operative case has been previously reported originating from the Sylvian aqueduct [2]. Here, we describe the rare case of RGNT arising in the Sylvian aqueduct in whom we performed a biportal approach. Along with the surgical technique description and the clinical and radiological peculiarities, we also report a detailed literature review on the topic.

2. Clinical presentation

A 25-year-old female came to our attention in January 2012 with a three-month history of blurred vision. The neuro-ophthalmological examination demonstrated papilledema, and an emergent MRI showed a triventricular hydrocephalus due to the presence of a Sylvian aqueduct mass (Fig. 1A and B).

The patient underwent emergent endoscopic third ventriculostomy (ETV) with biopsy. Histology documented a low grade glioneuronal proliferation with immunophenotypic positivity for GFAP and synaptophysins, confirming the diagnosis of a RGNT (Fig. 2). Watchful waiting was then performed to determine the tumor growth rate. At 6 and 12 months, no signal changes or volume increases were detected (Fig. 3). The patient was then lost to follow-up until she returned to our Institution in September 2015. At that time, an MRI performed in another institution demonstrated a clear increase in the tumor volume (Fig. 4).

Considering the fusiform morphology and the complex location of the lesion a biportal-combined approach was planned. In December 2016, the patient underwent the first surgical step through a telo-velar approach to the aqueduct. Postoperatively, the patient suffered intense nausea and occipital headaches for two weeks. A new MRI demonstrated the residue and obstructive hydrocephalus due to basal adhesive meningitis (Fig. 5A and B). The patient underwent placement of a ventriculoperitoneal shunt (VPS). CSF samples excluded bacterial infection. Subsequently, she recovered very well and regained an independent life.

The second surgical step consisted in an endoscopic transfrontal transforaminal approach to the Sylvian aqueduct. The post-operative MRI in May 2017 showed an estimated 6 mm of residual tissue (Fig. 6).

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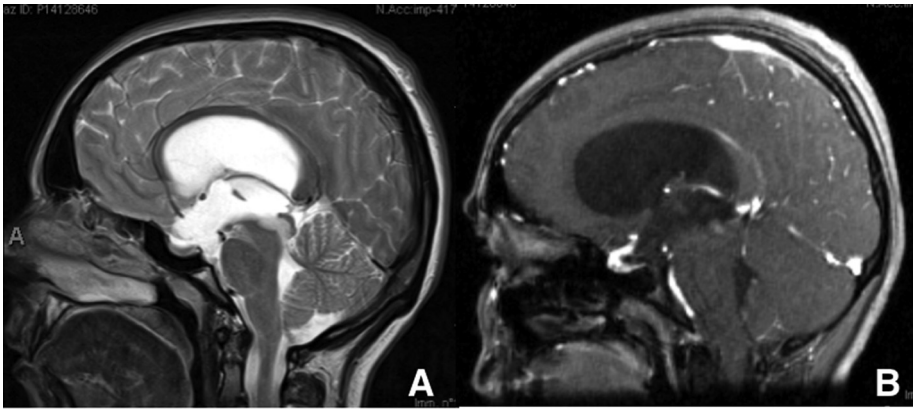

Fig. 1. A) T2 TSE MRI at admission showing a triventricular hydrocephalus associated to a hyperintense mass completely occluding the Sylvian aqueduct. B) Contrast enhanced T1 displays a faint circular spot of gadolinium in the upper pole of the mass.

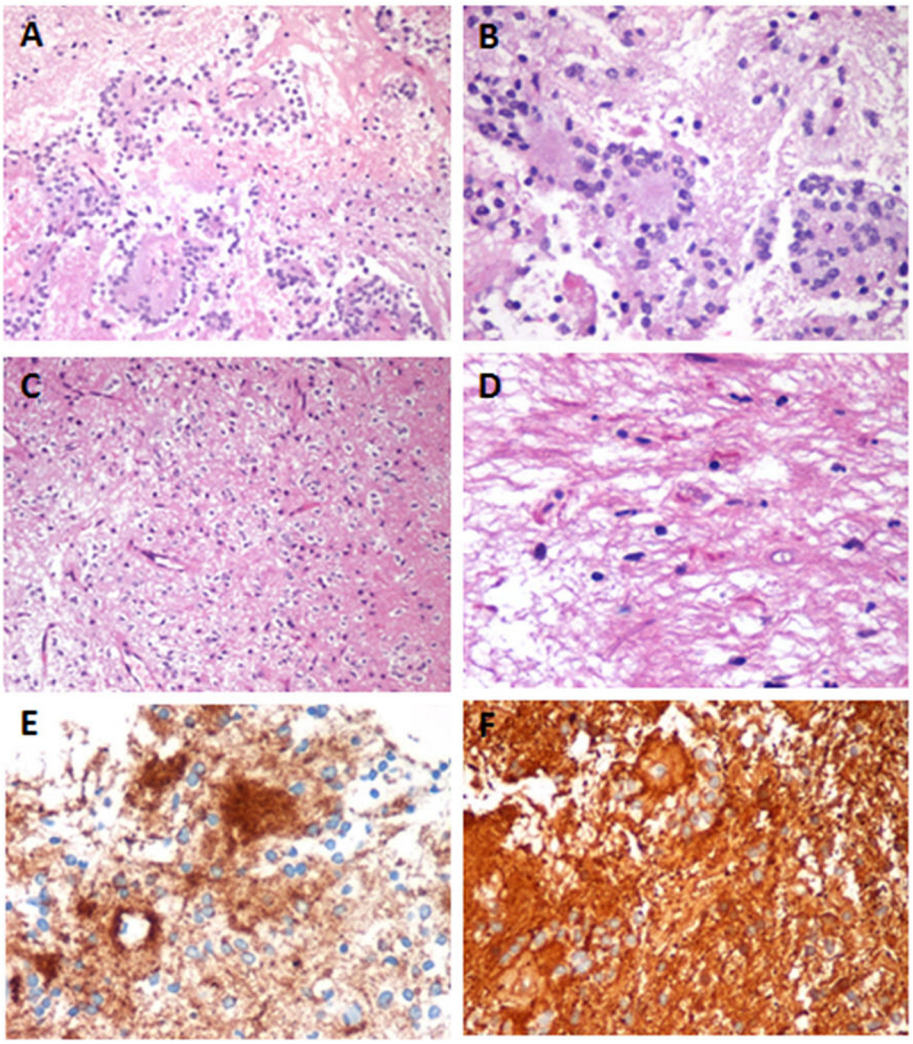

Fig. 2. Histologic features of the Rosette Forming Glioneuronal Tumor of the IV ventricle. At low power magnification the tumor shows typical well-formed rosettes and perivascular pseudo-rosettes (A) composed of cell with neurocytic features and dispersed in a fibrillary background. At higher magnification (B) tumor shows also true rosettes with a neuropil core. Area of rounded oligodendrocyte-like cells (C) and an astrocytic component reminiscent of pilocytic astrocytoma with Rosenthal fibers are also present (D).
A–D, H&E, 20× and 40× original magnification; E–F, synaptophysin and GFAP immunostaining, 40× original magnification.

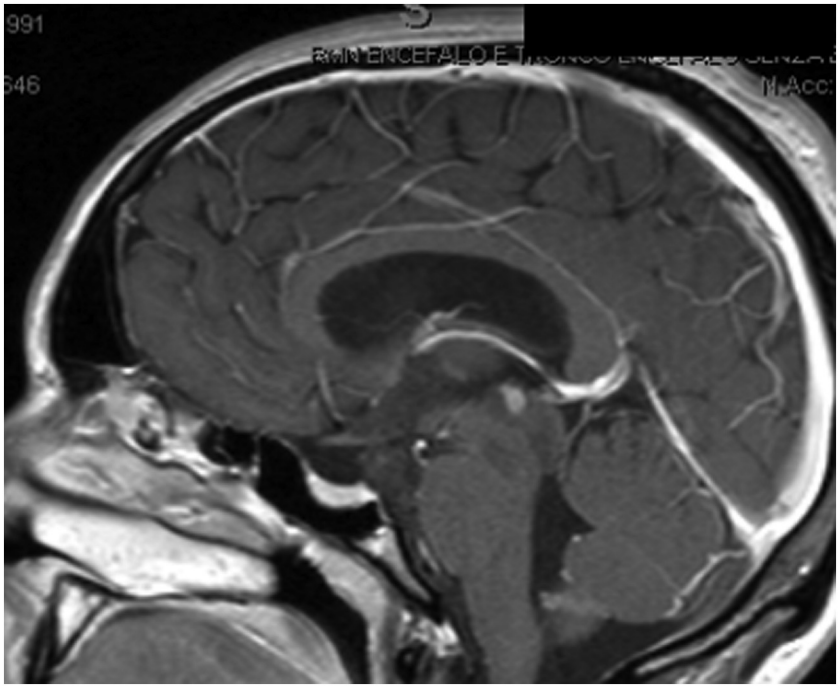

Fig. 3. 12 months postoperative MRI showing stable volume and signal with complete resolution of the hydrocephalus.

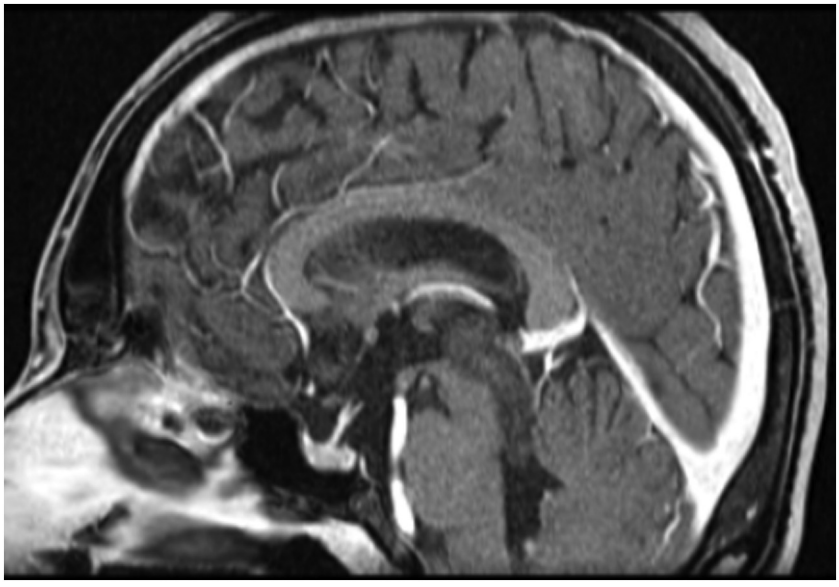

Fig. 4. Three year later MRI demonstrated a more then evident volume increase (from 20 mm to 37 mm in longer diameter). Interestingly contrast enhancement was no longer visible.

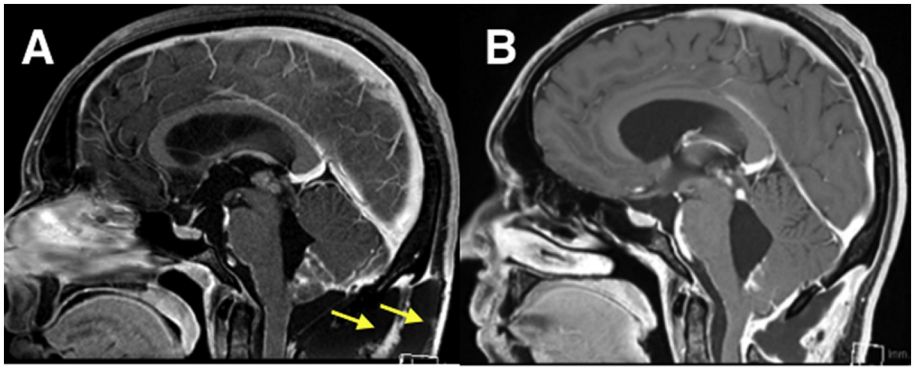

Fig. 5. A) Post contrast T1 MRI showing resection of the inferior half of the tumor. Note the large pseudomeningocele (yellow arrows) and the contrast uptake at the level of the basal cisterns as for an adhesive meningitis. B) Three months MRI control demonstrates the partial resolution of the pseudomeningocele. Of note, the residual tumor displayed modification of contrast uptake. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

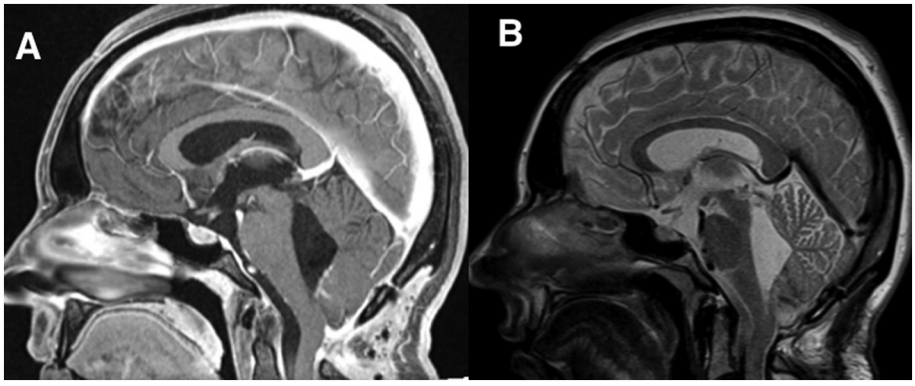

Fig. 6. Post contrast MRI 3 months postoperatively T1-WI with Gadolinium (A) and T2-WI (B). The upper third of the tumor popping up into the third ventricle has been removed. Residue occluding the aqueduct was estimated about 6 mm in maximum diameter.

The patient totally recovered her daily activities life, even though she reports having double vision.

3. Surgical notes (see video for details)

For the telovelar approach, the patient was placed in the prone position with the head flexed in a three pin-headrest. An incision was made in the midline, 2 to 3 cm above the external occipital protuberance, and extending to C3. The occipital bone, the posterior arch of the atlas, the upper cervical lamina and the spinous process of C2 were exposed. Then, a suboccipital craniotomy and a posterior laminectomy of C1 were performed. After wide dissection of the arachnoid of cisterna magna, the tela choroidea was exposed and cut bilaterally; tonsils and vermis were carefully retracted to expose the floor of the fourth ventricle and the caudal part of the tumor arising from the aqueduct. The most exophytic portion was removed with grasping forceps. Using an endoscope-assisted technique and an endoscopic ultrasound aspirator, the most intra-aqueductal part of the tumor corresponding to its inferior third was removed.

For the transfrontal-transforaminal approach, a neuronavigation system was used to access the right lateral ventricle and identify the foramen of Monro through an anterior frontal burr hole. At the most posterior border of the third ventricle, the vegetating mass completely hidden the aqueduct. Resection was pursued by both endoscopic grasping forceps and the ultrasound aspirator. Total tumor resection and aqueductoplasty was tried and ultimately abandoned due to neoplastic infiltration and the tissue's consistency.

4. Discussion

RGNT is a rare central nervous system tumor that was first included as a grade I independent entity by the WHO in 2007. It affects predominantly older children and young adults with a female prevalence. Histologically, this slow-growing lesion is comprised of a glial component, whose morphology similar to a pilocytic astrocytoma, and a neurocytic component, which forms neurocytic rosettes and/or perivascular pseudorosettes. There are cases of mixed RGNT/DNET as well [3]. Molecular aspects of RGNT have been investigated, and genetic mutations in PIK3CA and FGFR1 genes and an association with neurofibromatosis type 1 have been found [4]. Radiologically, these lesions are heterogeneous, showing calcified, nodular and cystic components. They appear hypointense on T1- and iso/hyperintense on T2-weighted MRI sequences; contrast enhancement is variable. Peculiar biological features underlying neuroradiological changes with RGNTs have already been observed by Mattogno et al., who documented spontaneous regression likely due to intervening ischemia [5].

The first of approximately one-hundred cases in the literature, was described by Komori et al. in 1998 [6]. Thirty-four RGNTs originated from the fourth ventricle and only one from the Sylvian aqueduct and reported only post-mortem information [7]. The current case is, to the best of our knowledge, the only such surgical case [8–56].

In cases of Sylvian aqueduct tumors, the primary goal of surgery is to treat the obstructive hydrocephalus, perform a biopsy or, when feasible, remove the tumor. Different techniques have been used to approach the aqueduct and all depend upon the volume and location of the tumor. The endoscopic transfrontal approach is one technique that has proven to be successful, especially in the case of lesions that extend into the upper part of the aqueduct. This technique presents some disadvantages however, such as the suboptimal trajectory to the aqueduct and the necessity of crossing the frontal cerebral parenchyma [57,58]. In our case, we planned to create a communication through the aqueduct, but we soon realized that penetration through the tumor carried a high risk of causing midbrain damage. An alternative is represented by the trans-fourth ventricle route through a telo-velar approach. This technique favors a straight trajectory to the aqueduct, which lies parallel to the floor of the fourth ventricle, and a direct visualization of lesions located in or extending into the lower part of the aqueduct [59]. The application of endoscope and endoscopic ultrasound aspirator in both surgical procedures speeded up the tumor resection; however, the tightness of the aqueduct and the surrounding critical structures, made too risky to push forward the endoscopic ultrasound aspirator.

5. Conclusions

The RGNT is a rare and distinct tumor of the glioneuronal family, classified as WHO grade I. When symptomatic tumors are located in the Sylvian aqueduct, the optimal therapy should be surgery, but it is important to provide the patient with the best and safest approach. Surgery should not only have the goal of confirming diagnosis and removing as much tumor as possible, but also treating hydrocephalus. Certain locations require carefully planning of the surgical strategy, specifically regarding the route to the tumors that may need to be multiple, as in the current case. To the best of our knowledge, this is the first documented case of resection through a biportal approach, which allowed a positive clinical and radiological outcome.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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Nothing to declare.

Conflict of interest

None.

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