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REVIEW

Pure endoscopic ultrasonic removal of choroid plexus papillomas of the third ventricle: technical report of two cases

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Abstract

Background Tumors of the choroid plexus of the third ventricle are uncommon. Surgical excision is technically challenging because of the rich vascularisation, central location, and high incidence in young children. Open microsurgical resection is considered the standard treatment. However, attempts at purely endoscopic removal of choroid plexus tumors of the third ventricle have also been made in the past, with encouraging results.

Case reports We report our experience with endoscopic ultrasonic removal of two cases of tumors of the third ventricular choroid plexus. The first case was a large atypical choroid plexus papilloma (WHO grade 2) in the anterior third ventricle associated with hydrocephalus; the second case was a smaller choroid plexus papilloma (WHO grade 1) in the middle/posterior third ventricle without overt hydrocephalus requiring a more anterior neuronavigation guided approach.

Discussion and conclusion Choroid plexus papillomasof the thirdventricle can be safely treated by a purely endoscopic approach because they are usually smaller than their counterparts in the lateral ventricle and often have a recognizable vascular pedicle. Early detection and control of the vascular pedicle at the choroidal border is key to success. The use of ultrasonic aspirator facilitates and expedites endoscopic access. By alternating surface coagulation with fragmentation and aspiration with the ultrasonic aspirator, the tumor can be removed without difficult dissection maneuvers.

Keywords Endoscopic ultrasonic aspirator · Neuroendoscopy · Brain tumor · Neuronavigation · Choroid plexus papilloma · Third ventricle

Introduction

Choroid plexus tumors (CPTs) are rare and account for less than 1% of all intracranial tumors [1]. CPTs include choroid plexus carcinoma (CPC, World Health Organization; WHO grade 3), choroid plexus papilloma (CPP, WHO grade 1), which accounts for 65–75% of CPTs, and an intermediate form called atypical choroid plexus papilloma (aCPP, WHO grade 2) [2]. They originate from the epithelium of the choroid plexus, which is present in all ventricular cavities. In general, these tumors are more common in the lateral ventricles (in children) and the fourth ventricle (in adults). In contrast, the third ventricle is extremely rare [3]. In both CPP and aCPP, radical surgical resection is considered resolutive without the need for adjuvant therapies. Complete tumor resection significantly improves prognosis even in CPCs [4–6]. Surgical resection is technically challenging due to rich vascularization, central location, and high incidence in young children. Open microsurgical resection is considered the standard treatment, especially for very large tumors arising in the lateral ventricles of young children. In the past, attempts have been made at purely endoscopic resection of choroidal tumors of the third ventricle, with encouraging results [7–10]. The recent availability of modern tools, such

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as endoscopic ultrasonic saurgical aspirators, may expand the spectrum of intracranial pathologies that can be treated by modern neuroendoscopic means [11, 12]. We report our experience with endoscopic ultrasonic removal of two cases of choroid plexus tumors of the third ventricle. These cases were quite different: the first case was a large atypical CPP occupying the anterior third ventricle and associated with hydrocephalus; the second case was a smaller CPP of the middle/posterior third ventricle without overt hydrocephalus, requiring a more anterior approach.

Relevant anatomy

The third ventricle is located in the middle of the head. According to the classic anatomical description by Yamamoto et al. [13], it is a narrow, funnel-shaped midline cavity. It communicates with the lateral ventricles through the foramina of Monro and with the fourth ventricle through the aqueduct. Tumors of the choroid plexus arise from the choroid plexus, which occupies the area of the foramina of Monro and the roof of the third ventricle. This is formed by the fornix and the tela choroidea. Two parallel strands of the choroid plexus project downward from the inferior layer of the tela choroidea on either side of the midline. The tela choroidea is composed of two thin membranes with an intervening vascular layer consisting of the medial posterior choroidal arteries and their branches and the internal cerebral veins and their tributaries [13]. Normally, choroid plexus tumors of the third ventricle have one or more vascular pedicles connected to the choroid plexus and the tela choroidea. Blood supply to the CPPs is usually via the medial and lateral posterior choroidal arteries and the collicular arteries. Venous outflow is via the internal cerebral veins, the basal vein of Rosenthal, the choroidal veins (from the system of the Galen vein), and the anteromedial occipital veins [10].

Indications

Small tumors of the third ventricle can be completely resected by a minimally invasive transventricular endoscopic approach. Prerequisites traditionally required for this type of surgery are that the tumor is small (> 3 cm in diameter), lacks hard consistency, and is poorly vascularized [11]. Endoscopy can also be considered for small remnants of large tumors. Optical and electromagnetic neuronavigation may obviate the need for ventricular dilatation, which in the past was considered a prerequisite for ventricular neuroendoscopy. The use of modern neuroendoscopic tools like endoscopic ultrasonic surgical aspirators may obviate to some of these limitations.

Surgical technique (videos 1–2)

Depending on the location of the tumor, two main approaches can be used: a coronal burr hole provides access to the anterior third ventricle (case 1—video 1), while a more anterior burr hole allows access to the middle and posterior part of the third ventricle (case 2—video 2).

Almost 80% of CPP of the third ventricle arises in its anterior part and is associated with hydrocephalus; therefore, a standard coronal burr hole can be used for surgery (as in our case 1). Neuronavigation is useful in these cases but not mandatory. On the other hand, for a tumor arising in the middle or posterior third ventricle (as in our case 2), navigation is mandatory to plan the correct entry point and trajectory. We usually use electromagnetic neuronavigation (StealthStation S8, AxiEM, Neuronavigation, System, Medtronic, Minneapolis, MN, USA), which can be easily coupled to the endoscope by inserting the electromagnetic stylet into the working channel. The surgical equipment is supported by a rigid endoscope (Gaab Endoskop-Karl Storz, GmbH, Tuttlingen, Germany, or the Minop® InVent Endoscope—BBraun Aesculap AG, Tuttlingen, Germany), an endoscopic ultrasonic aspirator (Sonoca, Soering-GmbH, Quickborn, Germany), biopsy scissors, forceps, monopolar/bipolar coagulation, and thulium laser (Revo-Lix Jr, LISA Laser Germany).

Patients are positioned supine with the head in a neutral position and slightly flexed (30°) only for coronal approach (case 1). The access side is chosen according to the asymmetric dilation of the lateral ventricles (left in case 1, right in case 2). For the coronal approach, a linear parasagittal incision can be used; for the more anterior, precoronal approach, a transverse incision behind the hairline (which can easily be converted to a bicoronal incision if needed) can be used (the incision should be centered on the planned entry point). The correct entry point is determined using neuronavigation, extending a line down to the skin connecting the target (the center of the tumor) to the center of the foramen of Monro. The burr hole is performed, the dura mater is coagulated, and the lateral ventricle is pierced with the sheath of the endoscope. After the removal of the stylet, the endoscope is inserted into the sheath. The anatomic landmarks of the lateral ventricle (choroid plexus, thalamostriate and septal veins, and Monro's foramen) are identified. In our first case, the tumor with its characteristic red cauliflower aspect occupied the entire foramen of Monro. In the second case, the tumor was visible through the foramen of Monro in the middle part of the third ventricle, where it attached to the tela choroidea of the roof in continuity with the choroid plexus. Mammillary body

and tuber cinereum were free of tumor. The tumor is detached from the choroid plexus by laser coagulation. The upper pole of the tumor is then coagulated and small fragments removed by biopsy forceps, to obtain material for histological purposes. The endoscopic ultrasonic aspirator is then inserted through the operative channel of the endoscope, and tumor aspiration and fragmentation are started. To avoid ventricular collapse during aspiration, continuous irrigation with 36 °C Ringer's lactate solution is maintained, and an assistant is always ready to clamp the aspiration tubing when necessary. The tip of the ultrasonic aspirator is always kept in direct contact with the tumor surface, to remove as much blood and debris as possible, avoiding cloudiness of the cerebro-spinal fluid (CSF) and reducing at the same time the amount of CSF aspirated to limit the risks of excessive drop in intraventricular pressure. Aspiration is alternated with coagulation of the surface of the tumor (in larger tumors). The floating part of the tumor is easily aspirated by the cavitron and moved into the lateral ventricle, where it can be more easily manipulated and removed. The vascular pedicle is recognized and coagulated. The third ventricle is then inspected to check for residual tumors. Once hemostasis is achieved, the endoscope is pulled out from the ventricular system, and the planes are closed in a watertight fashion.

Case presentations

Case 1

A 7-month-old infant presented with macrocrania and bulging fontanel. His head circumference was at the 95th percentile and had exceeded 2 percentile ranks within 3 weeks. Fundoscopic observation revealed no papilledema. Cerebral ultrasonography revealed hydrocephalus. Magnetic resonance imaging (MRI) of the head showed a multilobular contrast-enhanced mass in the anterior third ventricle measuring 20×20×28 mm with resultant obstructive hydrocephalus (Fig. 1a, b, c). MRI of the spine revealed no evidence of metastatic disease. On admission, the patient was awake, active, and hemodynamically stable, with no apparent neurologic deficits. The patient underwent endoscopic ultrasonic removal of the tumor through a left coronal approach (Fig. 2, video 1). The histologic diagnosis was atypical choroid plexus papilloma. Postoperative MRI confirmed complete removal, with the presence of a small clot in the third ventricle, which disappeared along the follow-up. On postoperative day 5, a subcutaneous CSF collection occurred at the level of the burr hole. This regressed after two lumbar punctures. No adjuvant therapy was administered. At the 3-year follow-up examination, neither recurrence nor residual tumor was visible on MRI (Fig. 1d, e, f), and the patient achieved normal neurodevelopmental milestones.

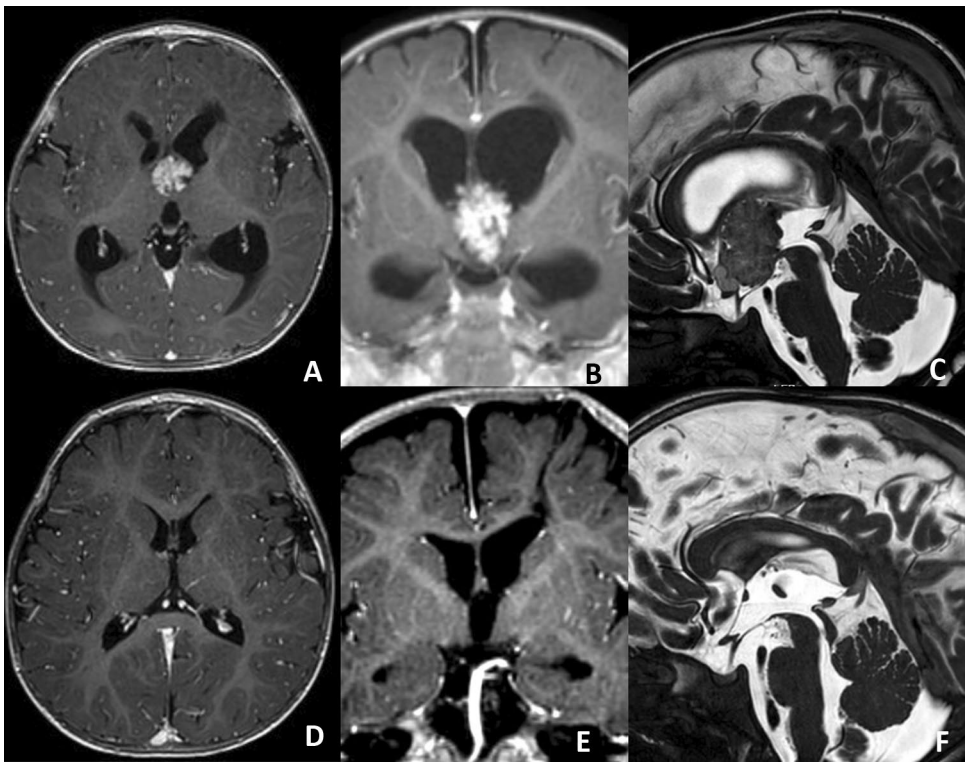

Fig. 1 Preoperative (**A**, **B**, **C**) and post-operative (**D**, **E**, **F**) magnetic resonance images of case 1. Axial (**A**), coronal (**B**) contrast-enhanced T1, and sagittal high-resolution T2 (**C**)-weighted images showing the tumor at presentation, occupying the anterior portion of the third ventricle, occluding both Monro foramina, with biventricular hydrocephalus. Axial (**D**), coronal (**E**) contrast-enhanced T1 and sagittal T2-weighted images showing complete tumor removal at 1-year follow-up

Case 2

A 3-year-old boy underwent MRI because he had headaches for 1 month. MRI showed the presence of a multilobular mass in the third ventricle, posterior to the foramen Monro, with intense contrast enhancement (dimensions: 11 × 14 × 15 mm) (Fig 3a, b, c). The ventricular system was mildly dilated. On admission, neurologic examination and fundoscopy were normal. The patient underwent endoscopic ultrasonic excision of the tumor through a right anterior frontal (precoronal) approach (Fig. 4, video 2). The histologic diagnosis was choroid plexus papilloma. The patient recovered without problems and the headache disappeared completely. Postoperative MRI confirmed the complete removal of the tumor. During the 5-year follow-up period, there was no evidence of tumor recurrence (Figs. 3d, e, f, 4).

Discussion

Choroid plexus tumors are vascularized lesions with a high potential for intraoperative hemorrhage. Therefore, few attempts at endoscopic removal of these tumors have been reported in the literature. However, CPPs of the third ventricle represent a unique feature because they are usually smaller than their counterparts in the lateral ventricle and often have a recognizable vascular pedicle. Santos and Souweidane [9] recommended a high-resolution T2-weighted MRI to visualize the focal attachment of the tumor to the tela choroidea. Early detection and control of vascular tributary at the choroidal interface during endoscopic surgery should be the key to success. Meng et al. [7] reported the first successful endoscopic removal of cystic CPP of the third ventricle in an 18-year-old patient. Reddy et al. [8] described a combined (endoscopic and microsurgical) approach to third ventricle CPP in a 6-month-old patient with secondary hydrocephalus. Santos and Suweidane [9] and Sufianov et al. [10] reported a purely endoscopic surgical approach for the successful removal of solid third ventricle CPP in two infants. All these authors used conventional endoscopic instruments to dissect, coagulate, cut, and remove tumors. Bimanual work using biopsy forceps, endoscopic scissors, and a bipolar coagulation electrode was often required to dissect, isolate, cauterize, and resect the vascular pedicle. Because of the mismatch between the papilloma and the endoscopic corridor, the tumor should be fragmented, and the fragments removed along with the endoscope. In one case,

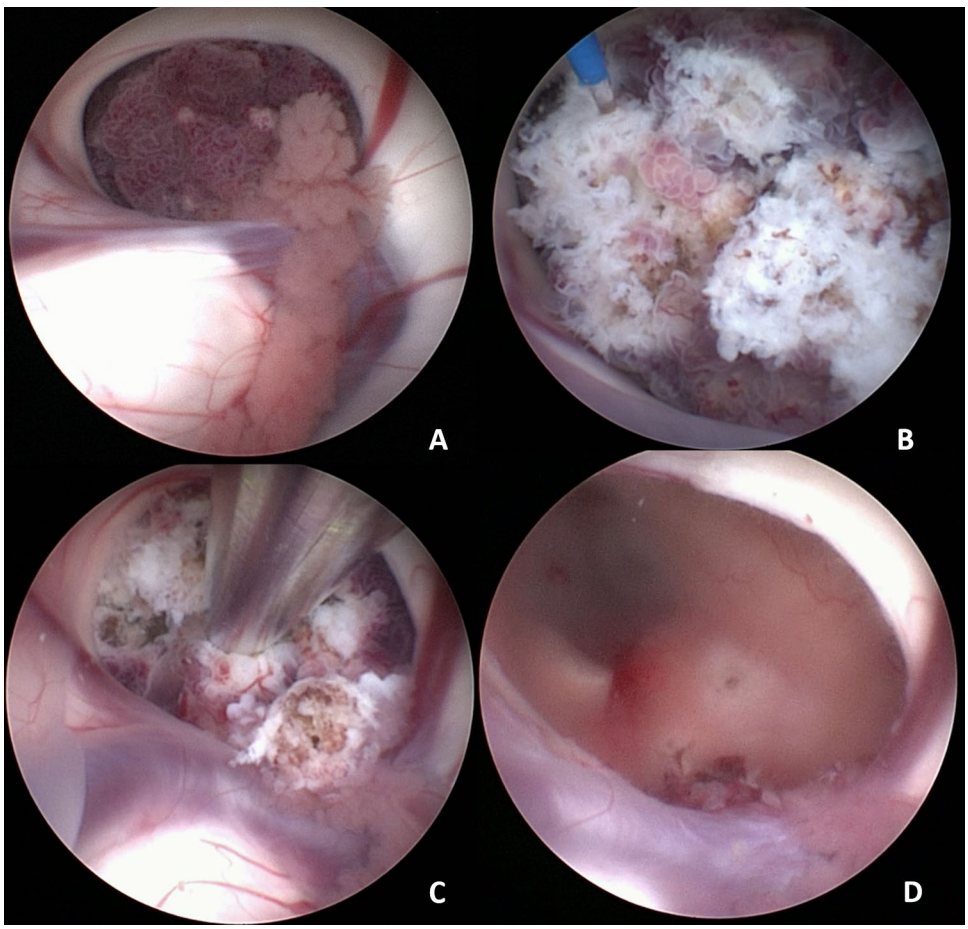

Fig. 2 Intraoperative images of case 1. **A** Appearance of the tumor occluding the left foramen of Monro, with its typical cauliflower appearance. **B** Coagulation of the surface of the tumor. **C** Ultrasonic debulking of the tumor. **D** Complete removal of the tumor

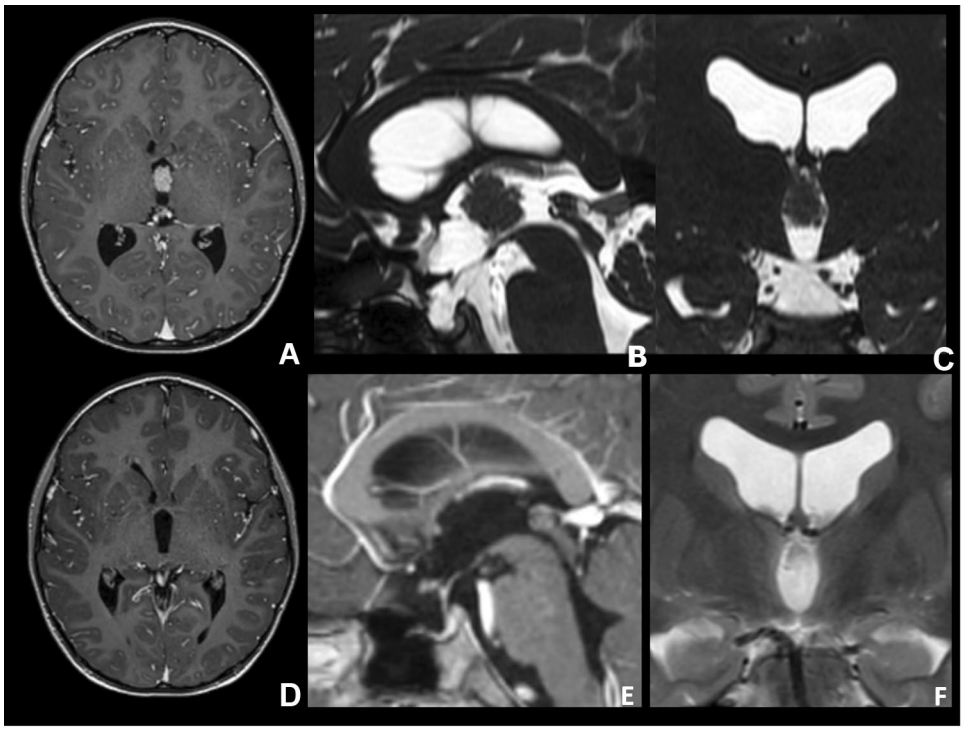

Fig. 3 Preoperative (**A**, **B**, **C**) and post-operative (**D**, **E**, **F**) magnetic resonance images of case 2. Axial contrast-enhanced T1 (**A**), sagittal (**B**), and coronal high-resolution T2 (**C**)-weighted images showing the tumor at presentation, occupying the middle portion of the third ventricle just behind the foramen of Monro. Axial (**D**), sagittal (**E**) contrast-enhanced T1 and coronal T2-weighted images showing complete tumor removal

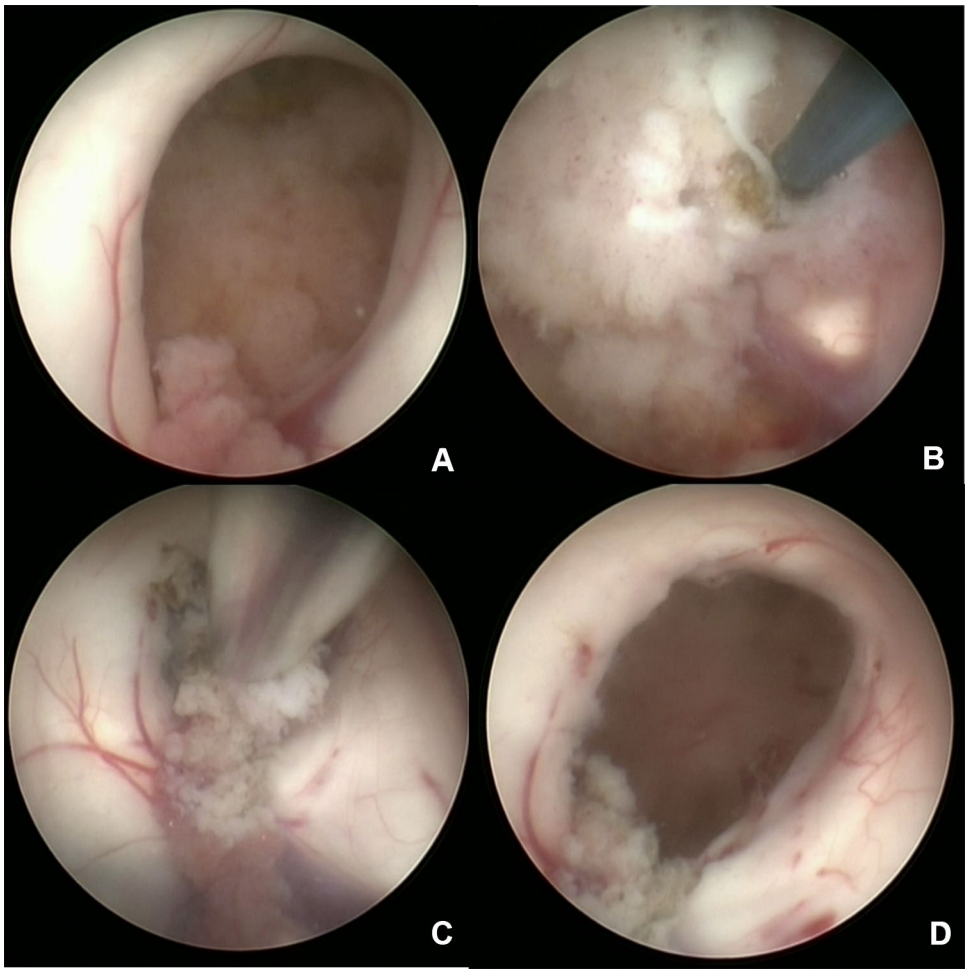

Fig. 4 Intraoperative images of case 2. **A** Appearance of the papilloma behind the foramen of Monro, with its typical cauliflower appearance. **B** Coagulation of the surface of the tumor. **C** Ultrasonic debulking of the tumor. **D** Complete removal of the tumor

a combined approach was required to remove the tumor, first using endoscopy to biopsy and mobilize the tumor from the third ventricle into the right lateral ventricle and then removing the lesion openly via a transcortical approach [8].

The use of an ultrasonic aspirator makes endoscopic access easier and faster. By alternating surface coagulation with fragmentation and aspiration with the ultrasonic aspirator, the tumor can be removed without difficult dissection maneuvers. The vascular pedicle is easier to handle after initial debulking. Hemorrhage, which obstructs endoscopic vision, remains a major problem. This can be reduced by continuous irrigation and keeping the tip of the ultrasound aspirator very close to the tumor to aspirate the blood as well. In case of excessive bleeding, all CSF can be aspirated, and resection can be continued in a "dry field technique" as described by Oertel et al. [14].

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s00381-023-05979-0.

Author contribution Primary surgeon video 1: Mirone. Primary surgeon video 2: Spennato. Assistant surgeon/supervisor: Cinalli G. Editing and drafting the video and manuscript: Spennato, Onorini, Vitulli, Cinalli M. A., Mirone. Critically revising the work: Cinalli G., Spennato. Approved the final version of the work on behalf of all authors: Spennato. Supervision: Cinalli G. All authors reviewed the manuscript.

Availability of data and material Not applicable.

Declarations

Ethics approval and consent to participate All procedures performed in the studies involving human participants conformed to the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its subsequent amendments or comparable ethical standards. Written informed consent was obtained from the parents for publication of the details of the medical case and associated images. Because this was a retrospective case report and all procedures performed were part of routine care, the study was exempt from ethical approval by the institutional review board.

Conflict of interest The authors declare no competing interests.

References

1. Gupta P, Sodhi KS, Mohindra S, Saxena AK, Das A, Khandelwal N (2013) Choroid plexus papilloma of the third ventricle: a rare infantile brain tumor. J Pediatr Neurosci 8(3):247–249. https://doi.org/10.4103/1817-1745.123696
2. Louis DN, Perry A, Wesseling P et al (2021) The 2021 WHO classification of tumors of the central nervous system: a summary. Neuro Oncol 23(8):1231–1251. https://doi.org/10.1093/neuonc/noab106
3. Pfeifer CM, Wong K, Malireddy R, Van Tassel D, Veltkamp DL, Cornejo P (2020) Third ventricle choroid plexus papilloma: 2 cases. Radiol Case Rep 15(5):454–456. https://doi.org/10.1016/j.radcr.2020.01.021
4. Passariello A, Tufano M, Spennato P et al (2015) The role of chemotherapy and surgical removal in the treatment of choroid plexus carcinomas and atypical papillomas. Childs Nerv Syst 31(7):1079–1088. https://doi.org/10.1007/s00381-015-2697-3
5. Pencalet P, Sainte-Rose C, Lellouch-Tubiana A et al (1998) Papillomas and carcinomas of the choroid plexus in children. J Neurosurg 88(3):521–528. https://doi.org/10.3171/jns.1998.88.3.0521
6. Wolff JE, Sajedi M, Brant R, Coppes MJ, Egeler RM (2002) Choroid plexus tumours. Br J Cancer 87(10):1086–1091. https://doi.org/10.1038/sj.bjc.6600609
7. Meng H, Feng H, Zhang L, Wang J (2011) Endoscopic removal of a cystic choroid plexus papilloma of the third ventricle: a case report and review of the literature. Clin Neurol Neurosurg 113(7):582–585. https://doi.org/10.1016/j.clineuro.2011.02.010
8. Reddy D, Gunnarsson T, Scheinemann K, Provias JP, Singh SK (2011) Combined staged endoscopic and microsurgical approach of a third ventricular choroid plexus papilloma in an infant. Minim Invasive Neurosurg 54(5–6):264–267. https://doi.org/10.1055/s-0031-1287775
9. Santos MM, Souweidane MM (2015) Purely endoscopic resection of a choroid plexus papilloma of the third ventricle: case report. J Neurosurg Pediatr 16(1):54–57. https://doi.org/10.3171/2014.12.PEDS14287
10. Sufianov AA, Gaibov SS, Sufianov RA (2015) Endoscopic monoportal removal of a choroid plexus papilloma in the posterior third ventricle in a child. J Neurosurg Pediatr 16(1):107–111. https://doi.org/10.3171/2014.12.PEDS14306
11. Cinalli G, Imperato A, Mirone G et al (2017) Initial experience with endoscopic ultrasonic aspirator in purely neuroendoscopic removal of intraventricular tumors. J Neurosurg Pediatr 19(3):325–332. https://doi.org/10.3171/2016.10.PEDS16352
12. Spennato P, De Rosa A, Meccariello G et al (2022) Endoscopic ultrasonic aspiration as alternative to more invasive surgery in initial management of optic pathway gliomas in children. Childs Nerv Syst. https://doi.org/10.1007/s00381-022-05515-6
13. Yamamoto I, Rhoton AL, Peace DA (1981) Microsurgery of the third ventricle: part I. Microsurgical anatomy. Neurosurg 8(3):334–356. https://doi.org/10.1227/00006123-198103000-00006
14. Oertel J, Linsler S, Csokonay A, Schroeder HWS, Senger S (2018) Management of severe intraoperative hemorrhage during intraventricular neuroendoscopic procedures: the dry field technique. J Neurosurg 131(3):931–935. https://doi.org/10.3171/2018.4.JNS172537

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