Cavernous Angioma of the Cerebral Aqueduct

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Key words
- Cavernoma
- Cavernous angioma
- Cerebral aqueduct
- ETV
- Hydrocephalus
- Intraventricular

Abbreviations and Acronyms
CSF: Cerebrospinal fluid
ETV: Endoscopic third ventriculostomy
IVC: Intraventricular cavernoma
MRI: Magnetic resonance imaging

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BACKGROUND: **Among the rare intraventricular cavernomas, purely intraaqueductal cavernomas are exceptionally rare.**

CASE DESCRIPTION: **A 62-year-old patient presented with progressive headache, memory loss, gait instability, and urinary incontinence. Magnetic resonance imaging showed the presence of a mass lesion located in the lumen of the cerebral aqueduct, associated with triventricular hydrocephalus.**

CONCLUSIONS: **We discuss the rationale that led us to treat hydrocephalus with neuroendoscopy, which offered the possibility to directly inspect the intraaqueductal lesion, make the diagnosis of cavernoma, and treat symptoms resulting from hydrocephalus without increasing the risk of bleeding.**

INTRODUCTION

Cavernous angiomas (or cavernomas) represent 5%–10% of the vascular malformations of the central nervous system. They are usually diagnosed between the second and fifth decade of life and can occur sporadically or as part of a familial syndrome. Owing to their histologic structure, small hemorrhages are frequent, although often subclinical.[1,2]

Intraventricular cavernomas are rare (2.5%–10.8% of cerebral cavernomas), with 104 cases reported in the literature so far.[3] Aqueductal localization is exceptionally rare. Cavernomas growing inside the ventricles pose a therapeutic challenge, and the strategy needs to be tailored according to the precise location. Neuroendoscopy offers the possibility to treat hydrocephalus, which might be the primary cause of hospitalization; directly inspect the lesion; and remove the cavernoma in selected cases. We report the case of a patient with a cavernoma located in the cerebral aqueduct lumen who presented with symptoms resulting from hydrocephalus and provide a review of the literature on intraventricular cavernomas. The issues of aqueductal localization require a specific management of the disease.

CASE REPORT

A 62-year-old woman was admitted to our neurosurgical unit with an 8-month history of progressive headache, cognitive impairment (memory loss), gait instability, and urinary incontinence. Magnetic resonance imaging (MRI) showed the presence of a cavernous angioma (9 × 11 × 15 mm) in the lumen of the cerebral aqueduct, associated with a triventricular hydrocephalus (Figure 1). A neuroendoscopic procedure was scheduled to perform an endoscopic third ventriculostomy (ETV). A flexible endoscope (KARL STORZ GmbH & Co. KG, Tuttlingen, Germany) was introduced into the right lateral ventricle through a precoronal burr hole. Immediately below the aditus, the cerebral aqueduct was completely obstructed by a brown lobular mass, which was blocking cerebrospinal fluid (CSF) flow (Figure 1). The vascular malformation was visually identified as a cavernous angioma. To prevent damage to the midbrain with consequent neurologic deficits, we avoided any attempt of radical removal of the lesion. A standard ETV was performed. The postoperative course was uneventful, and computed tomography scan ruled out any complication. Immediately after surgery, urinary incontinence was no longer present. Follow-up MRI performed at 3 and 12 months confirmed the ventricle reduction (Figure 1), whereas physical examination revealed significant recovery of cognitive functions and improvement of postural instability after 6 months.

DISCUSSION

Although cavernomas are relatively common, cavernomas growing inside the ventricular system need to be considered separately because of their peculiar characteristics. Their natural history remains undefined to some extent, although selected studies and reviews defined some features of this uncommon pathologic entity. Intraventricular cavernomas (IVCs) have shown more rapid growth than intraparenchymal cavernomas, probably because they are not surrounded by brain tissue, although onset of symptoms is usually delayed.[4] It is unknown whether waiting after acute hemorrhage before operating on an IVC is the correct strategy or if it unnecessarily increases the risk of harmful bleeding. Although intralesional bleeding can frequently occur, intraventricular bleeding is rare; it was reported in 14%–27% of the cases described in literature.[3,5,6] Nevertheless, in a more recent series, Kivelev et al.[3] found a high tendency of IVCs to rebleed

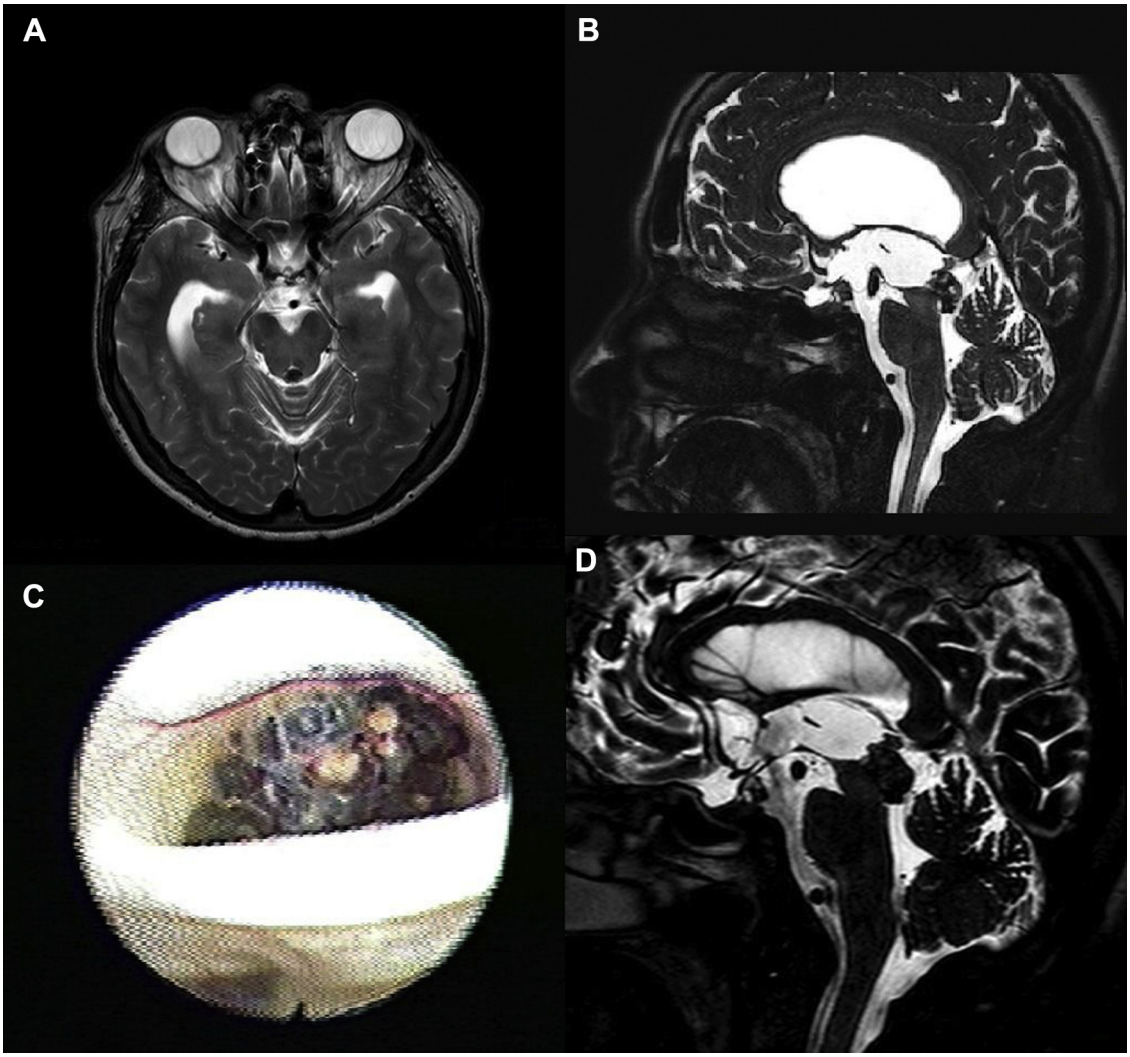

Figure 1. Preoperative (**A**) axial and (**B**) sagittal T2-weighted magnetic resonance imaging showing the cavernous angioma located in the lumen of the cerebral aqueduct. (**C**) Intraoperative neuroendoscopic view of the lesion. (**D**) Postoperative sagittal T2-weighted magnetic resonance imaging follow-up scan after 1 year, showing ventricle reduction.

and advocated for surgery when hemorrhages are frequent and the mass effect causes progressive neurologic deficits. They reported a 19% rate of intraventricular bleeding risk. Similarly, our review of published cases revealed that intraventricular hemorrhage occurs in approximately 21.4% of cases (Table 1). The symptoms related to their growth and the management of these lesions differ based on the specific location. It has been shown that cavernomas in the third ventricle are different in biologic nature and need more aggressive therapy. They grow very rapidly, leading to significant morbidity.[68] In particular, cavernomas occurring at the narrowest passages of the ventricular system such as the foramen of Monro and the cerebral aqueduct are of great interest because of their rarity and because they can lead to hydrocephalus, which may sometimes develop acutely.

Although microsurgical removal of IVCs is usually safe, it can carry increased risk in the fourth ventricle. The 63 patients surgically treated with either total or partial IVC resection showed neurologic improvement; mortality was seen in only 9 cases, and the remaining patients presented with various postoperative symptoms, such as hemianopsia, memory loss, and hemiparesis.

Bailey and Woodard in 1959[69] and Courville in 1961[13] reported 2 cases of cavernoma obstructing the upper end of the cerebral aqueduct. To our knowledge, our case is the first reported cavernoma of the cerebral aqueduct lumen. The aqueduct location carries the main risk of CSF obstruction, similar to the foramen of Monro.[70] Our patient as well as Courville's patient presented with symptoms related to hydrocephalus. It is

Table 1. Intraventricular Cavernomas: Literature Review

Reference	Age (years)/Sex	Clinical Presentation	Location	Treatment	Outcome
Finkelburg, 1905[7]	2/male	Mass effect	Fourth ventricle	Partial resection	Died
Dandy, 1928[8]	31/male	Mass effect	Fourth ventricle	Total removal	Improved
Merritt, 1940[9]	16/female	Mass effect	Lateral ventricle	Total removal	Comatose
Arnstein et al., 1951[10]	2 days/male	Mass effect	Lateral ventricle	No operation	Died
Lattermann, 1952[11]	68/female	Mass effect	Third ventricle	No operation	Died
Schneider and Liss, 1958[12]	33/female	Mass effect	Lateral ventricle	Total removal	Homonymous hemianopsia
Courville, 1961[13]	44/female	Hydrocephalus	Cerebral aqueduct	VC shunt	Died
Jain, 1966[14]	15/male	Mass effect	Lateral ventricle	Total removal	Improved
Coin et al., 1977[15]	36/female	Seizures	Lateral ventricle	Total removal	Hemianopsia
Numaguchi et al., 1977[16]	43/male	Mass effect	Lateral ventricle	Total removal	Hemiplegia and hemianopsia
Giombini and Morello, 1978[17]	27/male	Mass effect	Fourth ventricle	Partial resection	Died
Terao et al., 1979[18]	29/female	IVH	Fourth ventricle	Total removal	Improved
Pau and Orunesu, 1979[19]	56/ND	IVH	Lateral ventricle	No operation	Died
Namba et al., 1979[20]	45/female	IVH	Lateral ventricle	Partial resection	Improved
Vaquero et al., 1980[21]	18/female	Mass effect	Third ventricle	Total removal	Improved
Pozzati et al., 1981[22]	31/female	Mass effect	Third ventricle	Total removal	Improved
Iwasa et al., 1983[23]	8 days/female	Mass effect	Lateral ventricle	Total removal	Improved
Kendall et al., 1983[24]	60/female	Mass effect	Fourth ventricle	Partial resection	Symptom recurrence
Lavyne and Patterson, 1983[25]	48/female	Mass effect	Third ventricle	Partial resection	Hydrocephalus, bleeding
Amagasa et al., 1984[26]	40/male	Mass effect	Third ventricle	Total removal	Improved
Harbaugh et al., 1984[27]	44/female	IVH	Third ventricle	Total removal	Improved
Chadduck et al., 1985[28]	21/female	Seizures	Lateral ventricle	Total removal	Hemianopsia
	29/female	Mass effect	Lateral ventricle	Total removal	Improved
	4 months/female	Seizures	Lateral ventricle	Total removal	Improved
Simard et al., 1986[29]	22/male	Mass effect	Lateral ventricle	Not registered	Not registered
	13/female	Mass effect	Lateral ventricle	Not registered	Not registered
Yamasaki et al., 1986[30]	73/male	Mass effect	Lateral ventricle	Total removal	Improved
	9/male	Mass effect	Third ventricle	Partial resection	Improved
	36/male	Mass effect	Third ventricle	Total removal	Improved
	47/male	Mass effect	Fourth ventricle	Total removal	Improved
Suzuki, 1988[31]	40/male	Mass effect	Lateral ventricle	Total removal	Improved
Sabatier et al., 1989[32]	9 months/male	IVH	Lateral ventricle	No operation	Cerebellar dysfunction
Voci et al., 1989[33]	19/female	IVH	Third ventricle	Total removal	Improved
Ogawa et al., 1990[5]	16/male	Mass effect	Third ventricle	Total removal, VP shunt	Improved
	40/male	Mass effect	Third ventricle	Total removal	Transient DI, homonymous hemianopsia
Andoh et al., 1990[34]	62/female	Mass effect	Lateral ventricle	Total removal	Homonymous quadrantanopia

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Table 1. Continued

Reference	Age (years)/Sex	Clinical Presentation	Location	Treatment	Outcome
Tatagiba et al., 1991[35]	33/male	IVH	Lateral ventricle	Total removal	Improved
	35/male	Seizures	Lateral ventricle	Total removal	Died
	24/female	Mass effect	Lateral ventricle	Total removal	Improved
Itoh and Usui, 1991[36]	44/female	IVH, mass effect	Fourth ventricle	Total removal	Improved
Miyagi et al., 1993[37]	3/female	IVH, mass effect	Lateral ventricle	Total removal	Mild hemiparesis
Lynch et al., 1994[38]	39/female	Seizures	Lateral ventricle	Partial resection	Improved
	5/male	Seizures	Lateral ventricle	Partial resection	Improved
	10/female	Mass effect	Lateral ventricle	Total removal	Improved
Katayama et al., 1994[4]	9/female	Seizures	Third ventricle	Partial resection, VP shunt	Died
	50/female	Mass effect	Third ventricle	Not registered	Not registered
	45/female	IVH	Third ventricle	Not registered	Not registered
	49/male	Mass effect	Third ventricle	Not registered	Not registered
	49/male	Mass effect	Third ventricle	Total removal	Transient DI
Sinson et al., 1995[39]	43/female	Mass effect	Third ventricle	Total removal	Died
	36/female	Mass effect	Third ventricle	Total removal	Hemiparesis, hypothyroidism, hydrocephalus
	52/female	Mass effect	Third ventricle	Total removal	Improved
	32/female	Mass effect	Third ventricle	Total removal, VP shunt	Improved
Hashimoto et al., 1997[40]	2 days/male	Mass effect	Lateral ventricle	Total removal, VP shunt	Mild mental retardation
Kaim et al., 1997[41]	64/male	Mass effect	Third ventricle	Total removal	Not registered
Gaab and Shroeder, 1999[42]	44/female	Mass effect	Lateral ventricle	Total removal	Permanent memory loss
Reyns et al., 1999[43]	16/female	Mass effect	Lateral ventricle	Total removal	Improved
	36/male	Seizures	Lateral ventricle	Total removal	Right hemihypertonia
	42/male	Asymptomatic	Third ventricle	Partial resection	Improved
Fagundes-Pereyra et al., 2000[44]	15/female	Mass effect	Lateral ventricle	Total removal	Improved
Suess et al., 2002[45]	36/female	Mass effect	Third ventricle	Total removal	Improved
Crivelli et al., 2002[46]	38/male	Mass effect	Third ventricle	Total removal	Improved
Nieto et al., 2003[47]	11/female	Seizures	Lateral ventricle	Total removal	Homonymous hemianopsia
Tatsui et al., 2003[48]	17/female	Seizures	Lateral ventricle	Total removal	Improved
	52/male	IVH	Lateral ventricle	Total removal	Improved
Wang et al., 2003[49]	62/female	Mass effect	Third ventricle	Total removal	Improved
Anderson et al., 2003[50]	45/female	Mass effect	Septum pellucidum, lateral ventricle	Total removal	Improved
Darwish et al., 2005[51]	47/female	Incidental	Third ventricle	Total removal, shunt	Improved
Milenkovic, 2005[52]	56/male	Mass effect	Third ventricle	Total removal	Improved
Chen et al., 2006[53]	51/female	Mass effect	Third ventricle	Total removal	Improved

VC, ventriculocisternal; IVH, intraventricular hemorrhage; ND, no data; VP, ventriculoperitoneal; DI, diabetes insipidus; N/V, nausea/vomiting; CN, cranial nerve; ICH, intracerebral hemorrhage; ETV, endoscopic third ventriculostomy.

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Table 1. Continued

Reference	Age (years)/Sex	Clinical Presentation	Location	Treatment	Outcome
Kumar et al., 2006[54]	8/male	Mass effect	Lateral ventricle trigonum	Total removal	Improved
	19/female	Seizures, mass effect	Lateral ventricle trigonum	Total removal	Improved, seizures remained
	20/male	Mass effect	Lateral ventricle trigonum	Total removal	Improved
Longatti et al., 2006[55]	35/male	Mass effect	Third ventricle	Total removal	Improved
Zakaria et al., 2006[56]	8/male	Mass effect	Third ventricle	Total removal	Improved
Sato et al., 2006[57]	47/female	Mass effect	Third ventricle	Total removal	Improved
Gonzalez-Darder et al., 2007[58]	25/male	Mass effect	Lateral ventricle trigonum	Total removal	Improved
Prat and Galeano, 2008[59]	56/ND	Mass effect	Third ventricle	Total removal	Improved
Stavrinou et al., 2009[60]	52/female	Mass effect	Lateral ventricle trigonum	Total removal	Improved
Kivelev et al., 2010[3]	66/male	Gait disturbances, mild headaches, hydrocephalus	Lateral ventricle	VP shunt	Improved
	43/female	Mild headaches, N/V	Fourth ventricle	Total removal	Improved
	65/male	IVH, severe headaches, N/V, hydrocephalus	Lateral ventricle	Total removal	Improved
	58/female	IVH, mild headaches, N/V, CN deficit	Fourth ventricle	Total removal	CN VI and VII peripheral paresis same as preoperatively
	20/male	IVH, mild headaches	Lateral ventricle	Stereotactic biopsy, partial resection	Improved
	15/male	IVH, CN deficit	Fourth ventricle	Total removal	Persistent CN VII peripheral paresis
	52/male	Headache, N/V	Third ventricle	Total removal	Improved
	49/female	CN deficit	Fourth ventricle	Total removal	CN VI and VII paresis
	35/male	IVH, acute severe headaches, N/V	Lateral ventricle	Conservative	Improved
	49/female	IVH, acute severe headaches, hydrocephalus	Fourth ventricle	Total removal	Improved
	65/male	IVH, acute headaches, N/V, hydrocephalus	Lateral ventricle	Conservative	Improved
	53/male	IVH and ICH, headaches, hemiparesis	Lateral ventricle	Total removal	Minor memory disturbances
Prada et al., 2010[61]	32/female	Headaches, gait disturbances, N/V	Lateral ventricle trigonum	Total removal	Improved
Sabat, 2010[62]	60/female	IVH, headaches, imbalance, gait disturbances, hydrocephalus	Lateral ventricle	Total removal	Improved
Ohbuchi et al., 2012[6]	67/female	IVH	Lateral ventricle trigonum	Total removal	Improved
Lee et al., 2012[63]	30/female	Headache, short-term memory loss, vomiting	Third ventricle	Total removal	Improved
Bhatia et al., 2013[64]	29/female	Headache, vomiting, hydrocephalus	Third ventricle	Total removal	Improved

Continues

Table 1. Continued

Reference	Age (years)/Sex	Clinical Presentation	Location	Treatment	Outcome
Giannetti, 2013[65]	56/male	Gait disturbances, visual acuity disturbances, confusional state	Lateral ventricle	Total removal, ETV	Improved
Patibandla et al., 2014[2]	35/male	Headache, vomiting	Third ventricle	Total removal	Improved
Faropoulos et al., 2015[66]	56/male	Headache	Septum pellucidum, lateral ventricle	Total removal	Improved
	25/female	Deteriorating conscious state	Lateral ventricle	Total removal	Improved
	61/female	Seizures	Lateral ventricle	Total removal	Improved
	36/female	Headache, neck stiffness, confusion	Lateral ventricle	Total removal	Improved
	31/male	Headache, loss of consciousness	Septum pellucidum, lateral ventricle	Total removal	Improved
Winslow et al., 2015[67]	64/female	Loss of consciousness, hydrocephalus	Lateral ventricle	ETV	Died
Present case	62/female	Headache, gait disturbances, memory loss, urinary incontinence, hydrocephalus	Cerebral aqueduct	ETV	Improved

VC, ventriculocisternal; IVH, intraventricular hemorrhage; ND, no data; VP, ventriculoperitoneal; DI, diabetes insipidus; N/V, nausea/vomiting; CN, cranial nerve; ICH, intracerebral hemorrhage; ETV, endoscopic third ventriculostomy.

unclear if the bleeding risk of aqueduct cavernomas is as high as the risk of cavernomas in the third ventricle. However, as the only symptoms of our patient were symptoms related to intracranial hypertension and as the microsurgical removal of the lesion would have been possible but would have carried significant morbidity risks, we decided to treat hydrocephalus with an ETV and to follow up the cavernoma with MRI. We did not leave an external ventricular drain in place because the patient could be awakened immediately after the procedure, and clinical monitoring is preferable in these cases. The location of the IVC is crucial when assessing intraoperative risks.[3] The role of neuroendoscopy in IVCs has been well established. In all cases, it is possible to directly inspect the lesion, even in deep locations such as the aqueduct or the fourth ventricle, using rigid or flexible endoscopes. This inspection can provide crucial information about the final diagnosis and possible signs of previous bleeding, such as hemosiderin traces. In most cases, MRI provides only nonspecific imaging findings, leading to doubtful diagnosis.[28] Moreover, it is possible to treat the associated hydrocephalus with an ETV. Although endoscopic removal of aqueductal tumors has been shown to be possible, and a complete endoscopic removal of cavernomas has been reported in 2 cases, it should not be the first option.[42,59,71] Bleeding can be copious and difficult to manage through such a small approach, and walls of the cavernoma may collapse resulting in a more challenging resection. These complications have been reported in 3 cases when a purely endoscopic removal was attempted but soon abandoned.[57,59] It has been hypothesized that IVCs can rapidly grow also because there is no brain tissue surrounding them to counteract their expansion.[4] To a certain extent, the cerebral aqueduct is a very peculiar location from this perspective, as its walls can act as a counteracting force when the IVC reaches a volume that balances the aqueduct compliance. This was another consideration leading us to treat hydrocephalus first, avoiding the immediate attempt to microsurgically remove the cavernoma. When symptoms are related only to hydrocephalus, and the site of the cavernoma is difficult to reach with significant risks of morbidity (aqueduct, fourth ventricle), the treatment of hydrocephalus alone must be considered.[51] Some authors claim that the placement of a ventriculoperitoneal shunt can contribute to further growth of the lesion by lowering intraventricular pressure.[72] ETV is the best choice in such cases because it minimizes the risks related to quick, substantial reduction of CSF pressure with consequent increase in differential pressure between the ventricle and the vascular system and higher risk of rupture.[73]

CONCLUSIONS

Aqueductal cavernomas are exceedingly rare and usually lead to obstructive hydrocephalus. Their position makes their removal difficult, with potentially high morbidity. We presented a case of aqueductal cavernoma and showed that flexible neuroendoscopy allows for direct visual inspection of the lesion growing in the aqueduct lumen, for confirmation of the diagnosis, and for ETV to treat

the associated hydrocephalus. ETV can lead to complete resolution of symptoms resulting from CSF blockage. It is the treatment of choice when a cavernoma blocks the aqueduct without any sign of previous bleeding.

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