

Quadrigeminal plate arachnoid cyst presenting with eye movement related migraine: a rare case report

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ABSTRACT

Type II arachnoid cyst of the quadrigeminal cistern is the rarest type of arachnoid cyst (10% prevalence) in adults and is generally asymptomatic. We reported an unusual case of chronic right-sided migraine provoked by right eye adduction, right eye adduction soreness, and dry eye symptoms in a 47-year-old woman with quadrigeminal arachnoid cyst confirmed by radiological findings with the compression of the tectal plate, vermis, and superomedial cerebellum's part. She was treated conservatively without improvement for 1 year before surgical intervention was conducted. Microsurgery for cyst excision and fenestration was done, followed by immediate relief from all her complaints after 3 months of follow-up. These findings should help clinicians consider surgical intervention for patients with chronic symptoms related to nerve function that have no improvement with the initial treatment.

KEYWORDS arachnoid cysts, eye movement, microsurgery, migraine disorder

Arachnoid cysts are non-neoplastic cerebrospinal fluid (CSF)-filled cysts lined with the arachnoid layer of the brain. The etiology of arachnoid cysts is yet to be determined. However, according to one theory, these cysts arise from abnormal formation of the arachnoid layer of the brain during embryogenesis and have been associated with a history of head trauma, surgery, intracranial hemorrhage, or infection.¹

Arachnoid cysts are more common in men than women, with a lifetime prevalence of 1.4%. They are generally found in the middle cranial fossa, with a prevalence of 34%, or occur as retrocerebellar cysts, accounting for 33% of all arachnoid cysts.^{2,3} Arachnoid cysts in the quadrigeminal cistern have a prevalence

of 5% in children and 10% in adults and are usually asymptomatic, with only 10% of arachnoid cysts being symptomatic.^{1,4,5}

Quadrigeminal arachnoid cysts are classified into the following three types: (1) type I with supratentorial and infratentorial extensions; (2) type II, when the location of the cyst is only infratentorial, mainly supracerebellar or supraretrocerebellar; and (3) type III, when the cyst has a lateral extension toward the temporal lobe.⁶ Garg et al⁷ reported that only four out of 18 cases of quadrigeminal arachnoid cysts were type II. Similarly, Cinalli et al⁸ found only two cases of type II cysts among 14 patients with quadrigeminal cysts over a 13-year period. This suggests that type II

quadrigeminal arachnoid cysts are one of the rarest arachnoid cysts. To date, no multicenter studies have reported the prevalence of the three types of quadrigeminal arachnoid cysts.^{1,7-9} We report a case of a 47-year-old woman with quadrigeminal cistern arachnoid cyst type II who presented with right-sided migraine accompanied by soreness and dryness of the right eye.

CASE REPORT

A 47-year-old Asian woman complained of a right-sided migraine for the past 1 year that was intermittent, chronically progressive, and almost disabling, affecting her daily routine and working ability. The migraine was often provoked by adduction of the right eye and was accompanied by soreness and dryness

of the eye. She was referred to an ophthalmologist, who diagnosed multiple chalazia in the right eye. Surgical excision of all chalazia was performed to relieve her symptoms. However, 1 month after the chalazion excision, there was still no improvement in the migraine, eye soreness, and dryness. She was initially treated with minor tranquilizers and various painkillers, which slightly alleviated her symptoms.

Non-contrast head computed tomography (CT) showed a hypodense lesion of size 3.66 cm × 2.24 cm × 2.1 cm with attenuation of the CSF at the quadrigeminal cistern, which extends into the superior part of the cerebellum and compresses the tectal plate, superior vermis, and superomedial portion of the cerebellar hemisphere. Due to worsening symptoms, she was referred to a neurosurgeon (MT). Physical examination revealed impaired right eye adduction;

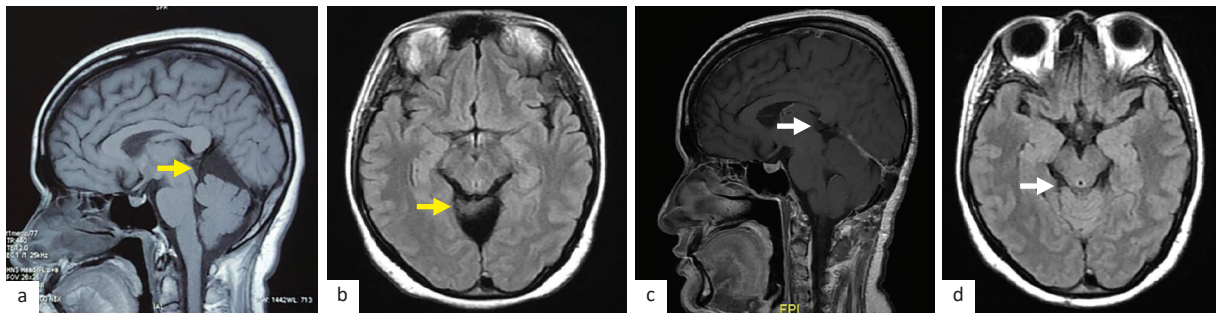


Figure 1. Preoperative non-contrast brain MRI of sagittal (a) and axial (b) views showing a hypointense lesion with a well-defined margin in T1WI that compressed cerebellar roof and tectal plate. An infratentorial extension confirmed the head CT findings of a type II quadrigeminal arachnoid cyst (yellow arrows) compared with the postoperative follow-up of the head MRI without contrast of sagittal (c) and axial (d) views that showed a complete resolution of the arachnoid cyst and a significant cerebellar roof decompression (white arrows). CT=computed tomography; MRI= magnetic resonance imaging; T1WI=T1-weighted images

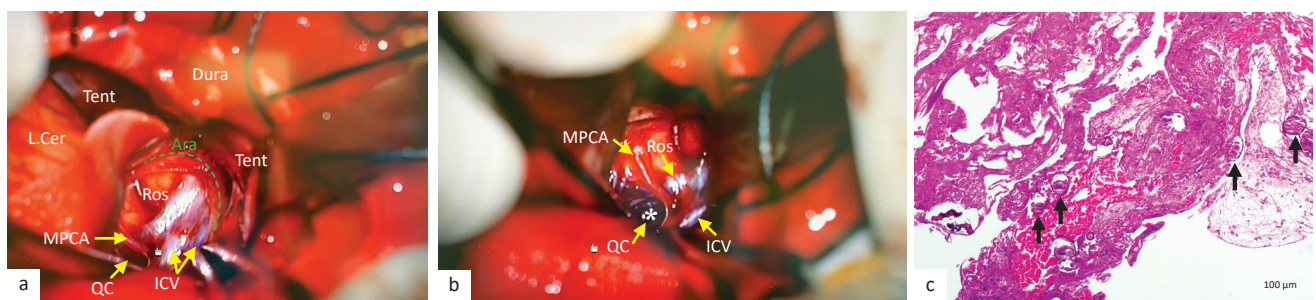


Figure 2. Arachnoid cyst. (a) Intraoperative findings at supracerebellar space showing the quadrigeminal supracerebellar area after some cyst walls removal with a microscope tilting inferolateral toward the brainstem and ambient cistern. The flap of the dura mater was mobilized superiorly to widen the surgical corridor. The remaining thick arachnoid cyst (green dashed line) was still visible at the level of incisura tentorium. In addition, the left side basal vein of Rosenthal and bilateral internal cerebral veins were visible and became the deepest border of the surgical dissection; (b) a more inferolateral tilting microscope aiming to fenestrate to the ambient cistern. The remaining arachnoid cyst wall shown in Figure 2a had already been removed; (c) histopathological examination results of the H&E stained cyst wall (200× magnification) showing a degenerative cyst wall without epithelial lining cells, several foci of calcification were present (black arrows). Ara=arachnoid cyst; Dura=dura mater of the posterior fossa; H&E=hematoxylin and eosin; ICV=internal cerebral veins; L.Cer=left cerebellum; MPCA=medial posterior choroïdal artery; QC=quadrigeminal cistern toward quadrigeminal plate; Ros=basal vein of Rosenthal; Tent=tentorium. *Fenestration to the ambient cistern

eye movement was stalled and painful. The right-sided eye soreness was exacerbated when she performed eye adduction; otherwise, the other examinations were within normal limits. Brain magnetic resonance imaging (MRI) revealed a hypointense lesion in the supracerebellar area, confirming the findings of the head CT scan (Figure 1, a and b).

Cyst excision and fenestration to the surrounding basal cistern were performed by MT. The patient underwent microsurgery using the supracerebellar infratentorial (SCIT) technique while in the prone position. Meticulous arachnoid dissection was performed to access the superior cerebellar area and the cyst located in the quadrangular cistern under high magnification using a surgical microscope. All cerebellar bridging veins were preserved. Once the cyst wall was found, micro scissors were used to open and remove the cyst, followed by fenestration of the surrounding ambient cistern to ensure normal CSF flow. A hemostatic agent was used to manage minor post-dissection bleeding (Figure 2).

The surgery was successful and without any complications, and the patient experienced immediate relief from her chief complaints of migraine, right eye adduction soreness, and dry eye. She was hospitalized for 4 days and discharged with a modified Rankin Scale (mRS) score of 1 due to slight dizziness. Histological examination revealed a degenerative cyst wall with calcified foci (Figure 2).

Her condition had improved during the outpatient follow-up visit 2 weeks after surgery, with an mRS score of 0. Compared with the preoperative brain MRI, brain MRI performed after 3 months with gadolinium-diethylenetriamine pentaacetic acid contrast showed complete resolution of the quadrigeminal cistern arachnoid cyst and no further compression of the superior vermis and superomedial cerebellar hemisphere (Figure 1, c and d).

Written informed consent for the medical procedures and publication of this case report and accompanying images had been obtained from the patient. All ethical principles for medical research established by the hospital were followed.

DISCUSSION

During the last 30 years, only 38 cases of symptomatic quadrigeminal cistern arachnoid cysts have been reported (Table 1).^{7,10-27} Of these, only 15

had visual disturbances as one of the main complaints. These complaints included decreased visual acuity, diplopia, nystagmus, worsening of eye vision, Parinaud's syndrome in the form of supraduction eye movement paralysis, and spasms of the orbicularis oculi muscle.^{8,11-13} To the best of our knowledge, type II quadrigeminal cyst presenting with migraine related to eye adduction, eye adduction soreness, and dry eye has not been reported previously.

In early life, quadrigeminal cysts often cause hydrocephalus by compressing the aqueduct of Sylvius. Compared to cysts in other locations, cysts in the quadrigeminal cistern tend to cause symptoms because of obstructive hydrocephalus.² However, quadrigeminal cyst is commonly an incidental finding because it is usually asymptomatic. If symptomatic, the symptoms are usually nonspecific, such as headache, lethargy, nausea, vomiting, gait ataxia, papilledema, and visual disturbance.^{1,5,25,28} Rarely, typical clinical signs may develop, such as impaired posture, nystagmus, diplopia, Parinaud's syndrome, hearing loss, hemiparesis, body spasticity, and weakness of the lateral rectus muscle of the eye.^{5,25}

As many as 50% of patients with quadrigeminal arachnoid cysts are under 18 years of age and show signs of hydrocephalus in the form of developmental delays. In the present case, the clinical signs of eye adduction soreness were probably due to pressure on the superior vermis and tectal plate. The underlying mechanism may involve: (1) compression of the cerebellar vermis cortex, especially lobes VI and VII (declive, folium, and tuber), also referred to as the oculomotor vermis, which is responsible for initiating saccadic movement, projecting inhibitory signals to stop the gaze accurately at the target during saccadic movement, and calibrating saccadic amplitude, direction, and horizontal alignment of the eye;²⁹⁻³¹ (2) compression of the tectal plate, the deepest layer of the superior colliculus, which plays an essential role in the eye's motor function. The superior colliculus projects saccadic motion stimuli to the tegmentum and controls the oculocephalic reflex and gaze shift.³² Hence, compression of these two anatomical structures may cause limited eye movement, eventually generating soreness and headaches.

Another interesting clinical presentation in our patient was chronic dry eye, which was relieved after cyst resection. The mechanism was unclear, but we believed that removing the cyst eventually released

Table 1. Adult symptomatic quadrigeminal cistern arachnoid cysts case report in the last 30 years

| First author, year | Age/sex | Clinical presentations | Radiologic findings | 1st surgical procedure | 2nd surgical procedure | Postoperative complications | Follow-up (months) | Outcomes |
|-----------------------------|---------|--|---|--|----------------------------------|---|--------------------|----------|
| Garg, ⁷ 2015 | 34/F | Headache, gait ataxia, and vision diminution | Supratentorial extension of the cyst | ETV | Shunt followed by shunt revision | Pseudomeningocele | 24 | ~ |
| | 22/F | Headache and vomiting | Supratentorial and infratentorial extension of the cyst | ETV | NA | NA | 44 | ↑ |
| | 24/M | Parinaud's syndrome and ataxia | Supratentorial and infratentorial extension of the cyst and compression of the dorsal midbrain | Endoscopic ventriculostomy | Shunt | Intraoperative bleeding during endoscopic ventriculostomy | 16 | ~ |
| | 22/F | Headache and vision diminution | Supratentorial and infratentorial extension of the cyst | Endoscopic ventriculostomy and ETV | NA | NA | 27 | ↑ |
| | 50/F | Headache, gait ataxia, vision diminution, and Parinaud's syndrome | Supratentorial and infratentorial extension of the cyst and compression of the dorsal midbrain | Endoscopic ventriculostomy and ETV | NA | NA | 12 | ↑ |
| | 28/M | Vision diminution, gait ataxia, and headache | Supratentorial and infratentorial extension of the cyst | Endoscopic ventriculostomy and ETV | NA | NA | 6 | ↑ |
| | 19/M | Headache and vision diminution | Supratentorial and infratentorial extension of the cyst | Endoscopic ventriculostomy and cysto-cisternostomy | Shunt | NA | 36 | ~ |
| Silva, ¹⁰ 2022 | 33/F | Headache | Hydrocephalus | ETV | NA | Superficial skin infection | 24 | ↑ |
| Takaki, ¹¹ 2021 | 19/M | Headache and gait disturbance | Hydrocephalus | Endoscopic fenestration | NA | NA | 60 | ↑ |
| | 68/F | Intermittent spasms in the left orbicularis oculi and orbicularis oris muscles spasm as well as left upper and lower extremities cerebellar ataxia | Obstructed cerebral aqueduct and narrowing of the prepontine cistern with moderate ventriculomegaly and elongated left vertebral artery | Endoscopic ventriculostomy | NA | NA | NA | ↑ |
| Hayashi, ¹² 1999 | 71/M | Gait instability, memory impairment, and disorientation | Supracollicular cyst, 3rd and lateral ventricle's anterior horn enlargement, and cerebellar vermis compression | ETV and ventricle-cystostomy | NA | NA | 5 | ↑ |
| Inamasu, ¹³ 2003 | 35/F | Headache | Hydrocephalus and infratentorial extension of the cyst | ETV and ventricle-cystostomy | NA | NA | 6 | ↑ |
| Gangemi, ¹⁴ 2005 | 45/F | Intracranial hypertension symptoms | NA | ETV and ventricle-cystostomy | NA | NA | NA | ↑ |

Table continued on next page

Table 1. (continued)

| First author, year | Age/sex | Clinical presentations | Radiologic findings | 1st surgical procedure | 2nd surgical procedure | Postoperative complications | Follow-up (months) | Outcomes |
|-----------------------------|---------|---|---|--|------------------------|-----------------------------|--------------------|-------------------------|
| Ohnishi, ¹⁵ 2007 | 62/F | Pain in 2nd division of right N V and papilledema | Infratentorial extension of the cyst and hydrocephalus | Ventriculo-cysto-cisternostomy | NA | NA | 12 | ↑ |
| Roka, ¹⁶ 2010 | 26/M | Migraine | Supratentorial and prepontine cistern extension of the cyst | Conservative treatment (carbamazepine) | NA | NA | 12 | Symptoms ↑, cyst size ~ |
| Zanini, ¹⁷ 2013 | 66/F | Unsteady gait, headache, and cognitive impairment | Infratentorial extension of the cyst | Endoscopic fenestration | NA | Parinaud's syndrome | 6 | ↑ |
| Arakawa, ¹⁸ 2013 | 28/F | Headache and papilledema | Hydrocephalus and infratentorial extension of the cyst | ETV | NA | NA | 12 | ↑ |
| | 26/F | Headache and nausea | NA | LVC and ETV | NA | NA | 81 | ↑ |
| | 38/F | Headache and nausea | NA | LVC, third ventricle-cystostomy, and ETV | NA | NA | 55 | ↑ |
| Gui, ¹⁹ 2016 | 42/M | Headache, diplopia, and visual impairment | NA | LVC, third ventricle-cystostomy, and ETV | NA | NA | 52 | ↑ |
| | 36/F | Headache and nausea | NA | LVC, third ventricle-cystostomy, and ETV | NA | NA | 37 | ↑ |
| | 23/M | Nystagmus | NA | LVC and ETV | NA | NA | 19 | ↑ |
| | 30/M | Headache, drowsiness, and ataxic gait | Infratentorial extension of the cyst | Third ventricle-cystostomy | NA | NA | 70 | ↑ |
| Yu, ²⁰ 2016 | 19/M | Generalized convulsion and headache | Supratentorial and infratentorial extension of the cyst | Third ventricle-cystostomy | NA | NA | 51 | ↑ |
| | 50/F | Headache and lassitude | Supratentorial and infratentorial extension of the cyst | ETV | NA | NA | 64 | ↑ |
| | 35/M | Ataxic gait and visual complaints | Infratentorial extension of the cyst | Cysto-cisternostomy | NA | NA | 50 | ↑ |
| Yu, ²⁰ 2016 | 26/F | Unsteady gait and visual complaints | Supratentorial and infratentorial extension of the cyst | ETV and third ventricle-cystostomy | NA | NA | 65 | ↑ |
| | 20/F | Increased intracranial pressure | Supratentorial and infratentorial extension of the cyst | ETV and LVC | NA | NA | 11 | ↑ |
| Isaka, ²¹ 1995 | 37/M | Headache and visual impairment | Hydrocephalus | Ventriculoperitoneal shunt | NA | NA | NA | NA |

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Table 1. (continued)

| First author, year | Age/sex | Clinical presentations | Radiologic findings | 1st surgical procedure | 2nd surgical procedure | Postoperative complications | Follow-up (months) | Outcomes |
|------------------------------|--------------|--|--|---|--|------------------------------------|--------------------|----------|
| Laviv, ²² 2017 | 31/F | Migraine, gait ataxia, cognitive impairment, and bilateral horizontal nystagmus | Supratentorial and infratentorial extension of the cyst and bilateral tonsil herniation | EVD and cystoperitoneal shunt | NA | NA | 18 | ↑ |
| Ohtsuka, ²³ 1998 | 54/M | Trochlear nerve palsy (torsional diplopia) and mild truncal ataxia | 3rd, 4th, and lateral ventricle enlargement; compression of the dorsal part of the brainstem, vermis cerebellar anterior, cisterna interpenduncularis, prepontine, and ambient | NA | NA | NA | NA | NA |
| Hayashi, ²⁴ 2005 | 71/M 54/F | Ataxia Headache | NA NA | NA NA | NA NA | NA NA | NA NA | NA NA |
| Garg, ⁷ 2015 | 42/M 45/F | Headache and gait ataxia Headache and vision diminution | Infratentorial extension of the cyst Infratentorial extension of the cyst | Microsurgery cyst removal, ventricle-cystostomy, and cysto-cisternostomy Midline suboccipital craniotomy with ventricle-cystostomy and cysto-cisternostomy | ETV NA | Air embolism hyponatremia NA | 24 36 | ↑ ↑ |
| Topsakal, ²⁵ 2002 | 67/F | Lower cranial nerve paresis, pyramidal sign, respiratory disturbances, disorientation, and memory disturbances | Hydrocephalus, lateral extension, brainstem, tectal plate, cerebellum, and aqueduct compression, and lateral and 3rd ventricle dilation | Microsurgery cyst removal | NA | NA | 18 days | ↑ |
| Wong, ²⁶ 1996 | 42 /F | Right hemiparesis, dysarthria, dizziness, and hypesthesia | Supratentorial and infratentorial extension of the cyst | Microsurgery cyst removal and cysto-cisternostomy | NA | NA | 18 | ↑ |
| Sharifi, ²⁷ 2013 | 52/F | Headache increased intracranial pressure signs. | Hydrocephalus and infratentorial extension of the cyst | Ventriculoperitoneal shunt, microsurgery cyst removal, and fenestration | Third ventriculostomy and third ventricle-cystostomy | Recurrence | 28 days | ↑ |

ETV=endoscopic third ventriculostomy; EVD=external ventricular drainage; F=female; LVC=lateral ventricle cystostomy; M=male; NA=not available; N V=trigeminal nerve. ~: unchanged; ↑: improved

the stretching and angulation of the ophthalmic branch (V₁) of the trigeminal nerve (cranial nerve [CN] V). Previously, Hayashi et al³³ have also reported a relationship between quadrigeminal arachnoid cysts and CN V involvement. They suggested that trigeminal neuralgia was caused by stretching, angulation, and demyelination at the root entry zone by a quadrigeminal arachnoid cyst.

Microsurgical fenestration and excision of the quadrigeminal cyst using the SCIT technique were preferred, considering the clear evidence of cerebellar compression, cyst size, predominant infratentorial extension, and progressive worsening of the symptoms without hydrocephalus findings.^{7,10,34} Microsurgery is preferred in type II quadrigeminal cysts without hydrocephalus, as sudden cyst decompression may invoke bleeding in the surrounding fragile neurovascular structures.^{6,7,34} Bleeding can be more efficiently controlled with microsurgery than an endoscopic approach. The reported success rate of microsurgery (85%) is comparable to that of endoscopic fenestration (88.5%).^{10,34}

In the present case, open surgery was preferred because of the degree of cyst wall removal required. A wide fenestration, with a minimum size of 10 to 15 mm, was made to reduce the possibility of cyst recurrence in the future.^{1,34} Post-intervention cerebellar complications were minimized by preserving all the cerebellar bridging veins while avoiding coagulation to handle minor bleeding and using a hemostatic agent instead. The larger bridging veins, mainly located at the midline, must be preserved; however, some smaller bridging veins can be removed if they cover a limited surgical corridor.³⁴

Intraoperative findings in the supracerebellar space showed the quadrigeminal supracerebellar area after removing some cyst walls with a microscope tilted inferolaterally toward the brainstem and ambient cistern. The remaining thick arachnoid cyst (green dashed line) was still visible at the level of the tentorium of the incisura. In addition, the left-side basal vein of Rosenthal and bilateral internal cerebral veins were visible and became the deepest border of the surgical dissection. Figure 2b shows an inferolateral tilting microscope aimed at fenestrating an ambient cistern. The remaining arachnoid cyst wall, shown in Figure 2a, had already been removed.

In conclusion, this was the first report of type II quadrigeminal arachnoid cyst presenting with migraine

associated with eye adduction, eye adduction-induced eye soreness, and dry eye symptoms, which were immediately relieved after microsurgical cyst resection and fenestration. These findings may help clinicians consider surgical intervention in patients with chronic symptoms related to nerve function that do not improve with initial medical treatment.

Conflict of Interest

The authors affirm no conflict of interest in this study.

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