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SURGICAL INTERVENTION OF SYMPTOMATIC PINEAL CYST: CASE REPORT

HIROYUKI HASHIMOTO, TAIJI YONEZAWA, JUNICHI IIDA,
KATSUYA MASUI and TOSHISUKE SAKAKI*
Department of Neurosurgery, Okanami General Hospital, and
*Department of Neurosurgery, Nara Medical University**

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Abstract: We report a rare case of symptomatic pineal cyst detected by magnetic resonance imaging. This patient presented with intermittent headache and gaze paresis. Subtotal resection of the pineal cyst was performed by the occipital transtentorial approach. It was proved histologically that the surgical specimen was composed of reactive glial tissue and normal tissue of the pineal gland with calcification. The procedure completely relieved the patient of refractory symptoms. Surgical management should be considered as an option to treat this rare lesion.

Index Terms

Pineal Cyst, Parinauds' syndrome, Surgery

Benign cystic lesions of the pineal gland are relatively common incidental findings in as high as 40 % of autopsy series[5,7,27]. Pineal cysts were once seldom detected by neuroradiological examinations. Since the advent of magnetic renonance (MR) imaging asymptomatic pineal cysts have been demonstrated with increasing frequency.

However, symptomatic pineal cysts are rare. This rare lesion has been highlighted in recent reports[2,4,8–11,24,19–22,25,26,28,29]. We describe a case of symptomatic pineal cyst. The patient has made a full recovery by subtotal resection of the cyst. Surgical management, and etiological, radiological aspects of this rare lesion are discussed.

CASE REPORT

A 38-year old woman complained of intermittent progressive headache and developed mild emotional disorder 4 months later. She had a syncopal episode with Parinauds' sign, and was referred to our unit. On admission she had moderate vertical gaze paresis, and was depressive.

A computed tomographic (CT) scan on admission showed a nonenhancing, round cystic lesion in the pineal region without hydrocephalus. MR images disclosed a cyst (2.5×1.5 cm) with slight enhancement of its wall on T 1-weighted images. On T 2-weighted images the cystic component was revealed to be of relatively higher intensity than cerebrospinal fluid (CSF) intensity. The lesion apparently compressed the tectal plate (Fig. 1). Then the cystic mass lesion was diagnosed as a nonneoplasmic, symptomatic pineal cyst.

The patient underwent open surgery with right occipital transtentorial approach to the pineal region. The yellowish green cyst was visualized beneath the arachnoid membrane in the quadrigeminal cistern. The cyst wall was glistening, and partially calcified. Slightly viscous, yellowish fluid was obtained after cyst opening. Subtotal resection of the cyst was performed

as the ventral part of the cyst showed a refractory adhesion to the tectal plate. The cyst wall was composed of two different parts. : a thin, glistening, calcified part, and a relatively thick elastic one.

The patient was uneventful after the operation (Fig. 2). The surgical specimens were examined histologically by hematoxylin and eosin staining, and immunohistochemical stain for glial fibrillary acidic protein (GFAP). It was microscopically found that the thin calcified part,

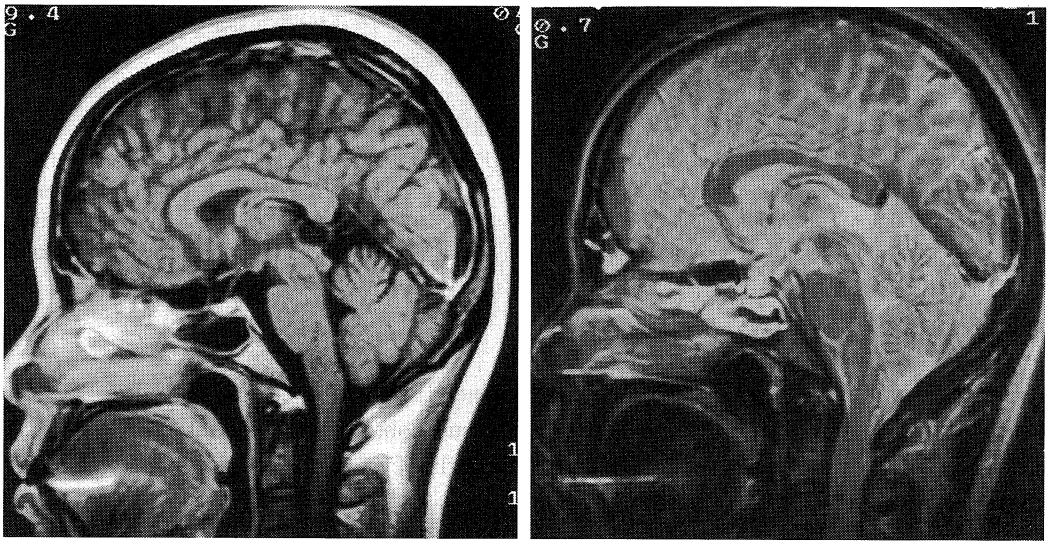

Fig. 1. Sagittal T1-weighted image (left) and T2-weighted image (right) showing a round, high signal (T2-weighted image) mass in the pineal region, compressing the superior colliculus.

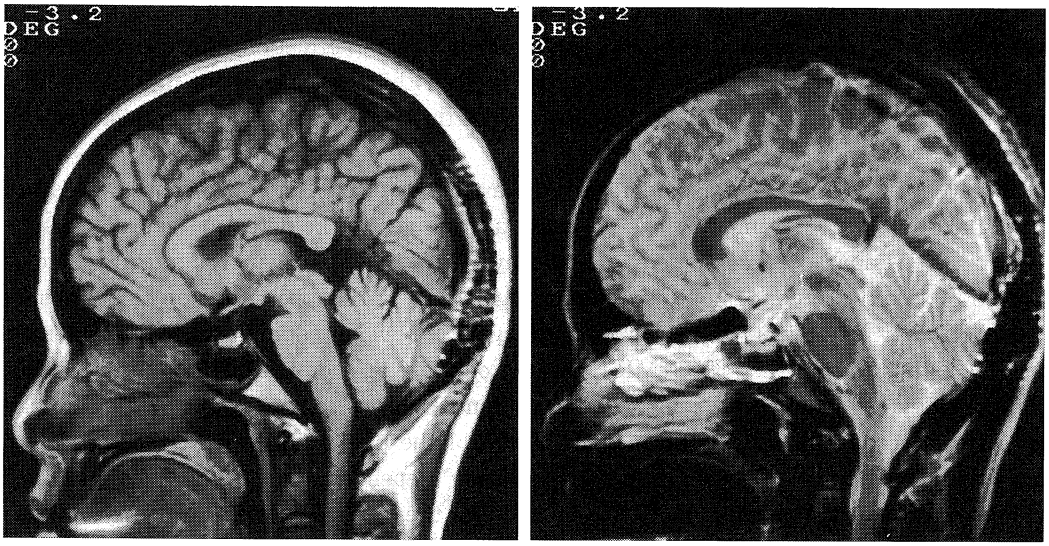

Fig. 2. Sagittal, postoperative T1-weighted image (left) and T2-weighted image (right) showing subtotal resection of the cyst and relief of compression to the superior colliculus.

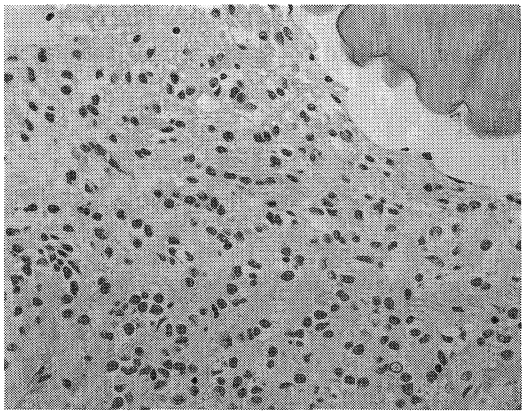

Fig. 3. Photomicrograph of the thin calcified tissue resected from the cyst wall, showing normal pineal cells, collagenous fibers, and calcification (hematoxylin and eosin, × 200).

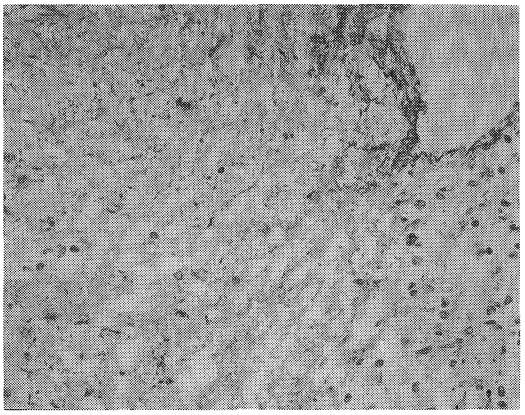

Fig. 4. Photomicrograph of the elastic tissue resected from the cyst wall, showing neuroglial tissue with strong immunoreactivity for GFAP (immunohistochemical staining for GFAP ×200).

noted above, was normal pineal parenchyma distended without a typical cell (Fig. 3). The elastic part was composed of dense neuroglial tissue containing abundant GFAP (Fig. 4). Consequently the cystic lesion was diagnosed to be a glial cyst of the pineal gland. The patient remains asymptomatic at 18-month follow-up.

DISCUSSION

Cysts in the pineal region are usually classified according to pathological and clinical views as follows: 1) small single or multiple cavities within pineal parenchyma without gland enlargement; 2) cysts associated with tumors, particularly teratomas and pinealomas[4,11]; 3) large single cysts with distention of the pineal gland and a mass effect on adjacent structures[29].

Benign asymptomatic pineal cysts are detected with a frequency of up to 40% of autopsies[5,7,27]. Symptomatic nonneoplasmic cysts of the pineal gland that were histologically confirmed, are rare.

The advent of MR imaging has demonstrated a number of detection of benign cysts in the pineal gland[10,11,13,20]. MR imaging has apparent advantages in the detection and diagnosis of pineal cysts, compared to computed tomography (CT)[11–13,16,20]. Symptomatic pineal cysts reported of late, were confirmed by MR imaging [11–13,29]. MR appearance of pineal cysts is characterized by isointensity or nearly isointensity on T 1-weighted images and hyperintensity on T 2-weighted images[11–13,16,20,24]. MR images of our case are compatible with those findings.

The etiology of pineal cysts is not still clear. Some theories regarding cyst formation have been postulated as follows: the normal involution of the pineal gland[3,10,16]; remnant of the embryonic cavum pineale; ischemic glial degeneration[5]; ependymal invasion of glial lacunae[4]; and invagination of the pineal gland[10]. The reason for the cyst enlargement remains unknown. Hemorrhagic events have been closely related to the enlargement of cysts in several cases[1,8,10,20,25]. However, most cases did not have any macroscopic or microscopic confirmation of hemorrhage in the cysts. Carr[4] suggestively ascribed cysts to the coalescence of smaller

cysts or the embryonic inclusion of ependymal cells. Klein and Rubinstein[10] threw doubt on these hypotheses by reason of the frequency occurrence of pineal cysts in young adults[10,11,14], and the lack of ependymal cells in nearly all cases[2,10]. Wisoff and Epstein[29] summed up clinical presentation of symptomatic pineal cysts with three syndromes : 1) paroxysmal headache with gaze paresis ; 2) chronic headache, gaze paresis, papilledema, and hydrocephalus ; or 3) pineal apoplexy with acute hydrocephalus. The headache will often be paroxysmal in onset. This probably results from intermittent obstruction of the aqueduct of Sylvius[29]. The symptoms of our case are consistent with paroxysmal headache with gaze paresis as noted above.

Maurer et al.[14] presented a case with multiple episodes of loss of consciousness as our case also had. These interesting episodes most likely result from acute obstruction of the aqueduct by shifting of cysts with bending of the head. Several patients including our case exhibited abnormal affect, hallucination, or suicidal behavior[4,10,17]. Surgical resection of cysts improved those psychiatric symptoms although the etiological mechanism has been unclear.

A surgical intervention should be considered when a patient of symptomatic pineal cyst is encountered. There are three options as surgical treatment : 1) open resection of the cyst, 2) cerebrospinal fluid shunt to bypass the obstruction, and 3) stereotactic aspiration of the cyst. Patients of symptomatic pineal cysts have been usually treated by open surgery, by the supracerebellar or the occipital transtentorial approach[10,14,15,28]. Fetell et al.[6] reported several patients that required shunting following open resection of the cyst. Vaquero et al.[28] treated a patient of simple glial cyst with shunting and supracerebellar resection. There seem to be patients that need shunting after resection of the cyst if accompanied with hydrocephalus. Stern et al.[26] described two patients of benign pineal region cyst that were treatable by stereotactic aspiration of the cyst. One patient required reaspiration of the cyst 71 months after the first procedure. Simple stereotactic aspiration may be a favorable means to attenuate symptoms of the cyst. However, we can not always put our whole reliance on such a procedure as this treatment has the possibility of cyst recurrence and the difficulty of obtaining an adequate sample of cyst wall. Wisoff and Epstein[29] reported six cases of symptomatic pineal cysts that underwent open surgical resection. Two of these patients underwent subtotal resection of the cyst ; total removal was limited due to dense adhesions to the tectal plate. There was no case with recurrence of the cyst in their series.

Miyatake et al.[15] documented a case of glial cyst of the pineal gland in which a disturbance of upward gaze postoperatively occurred due to total resection of the cyst. It is still obscure whether total resection of the cyst is most desirable to cure a nonneoplasmic cyst in the pinal gland. We obtained a satisfactory outcome with subtotal resection of the cyst. We think that subtotal resection of the cyst would be enough to obtain persistent relief in a patient with the symptomatic pineal cyst. Further retorted cases are desirable to resolve what is the most effective way to treat this rare lesion.

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