CASE REPORT

Trapped ipsilateral lateral ventricle: a delayed complication of hemispherotomy for Rasmussen's encephalitis

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SUMMARY

Hemispherotomy is the currently preferred surgical treatment option for refractory unihemispheric epilepsies. The incidence of hydrocephalus is greatly reduced in this disconnective procedure when compared with the resective procedure of anatomical hemispherectomy. We describe the occurrence of ipsilateral trapped lateral ventricle months after hemispherotomy for Rasmussen's encephalitis. There is enough evidence to suggest that this rare and interesting complication is due to the local inflammatory changes associated with the surgical trauma.

BACKGROUND

Hydrocephalus is more commonly seen after the resective procedure of hemispherectomy than after the disconnective procedure of hemispherotomy. Entrapment of the lateral ventricle after hemispherotomy is not described so far. We describe an 18-year-old woman who presented with new onset neurological symptoms in association with entrapment of the ipsilateral lateral ventricle months after hemispherotomy for Rasmussen's encephalitis.

CASE PRESENTATION

An 18-year-old woman presented with recurrent seizures from the age of 12. Her birth and childhood were uneventful. Initially, she suffered from occasional unprovoked seizures involving the right

side of the body which gradually became more frequent. From the age of 16, she had epilepsy partialis continua (EPC) of the right face and upper and lower limbs. She required frequent hospitalisation for secondary generalised seizures and status epilepticus. Seizures did not respond to multiple anti-epileptic drugs tried in varying combinations. She never developed persisting motor or speech deficits.

INVESTIGATIONS

Serial brain MRI scans revealed progressive atrophy of the left hemisphere predominantly involving the caudate nucleus and perisylvian area and subtle white matter hyperintensities (figure 1). Video-scalp electroencephalographic monitoring revealed persistent slow waves and background attenuation over the left hemisphere. Multiple complex partial seizures of left hemispheric origin were recorded. Serum was negative for autoimmune antibodies including N-methyl-D-aspartate receptor antibodies. A diagnosis of probable Rasmussen's encephalitis was made based on the above clinical, radiological and electrographic features. A trial of intravenous methyl prednisolone (1g daily for 5 days) did not give any benefit.

TREATMENT

The patient underwent a vertical parasagittal hemispherotomy (described by Delalande *et al*¹) of the left hemisphere. Histopathology of tissue

Figure 1 Brain MRIT2-weighted (T2W) axial image (A) and fluid attenuation and inversion recovery (FLAIR) axial image (B) showing atrophy of left caudate nucleus (arrow); T2W axial image (C) shows subtle left hemispheric atrophy especially in the orbitofrontal region (arrow).

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Figure 2 Brain MRI T2-weighted (T2W) axial images (A,B) showing trapped left lateral ventricle with dilated frontal, temporal and occipital horns. Fine septation is noted within the trapped ventricle (arrow). T2W coronal (C) image shows subfalcine herniation of the left cerebral hemisphere (arrow). There is midline shift to the right and compression of the right foramen of Monro with resultant dilatation of the right lateral ventricle. Fluid attenuation and inversion recovery (FLAIR) axial image (D) shows medial caudal herniation of the left lateral ventricle into the left midbrain with subjacent oedema (arrow).

obtained from the frontal lobe was consistent with Rasmussen's encephalitis. Postoperatively, she developed dense right hemiplegia. However, her speech remained preserved. She became seizure-free after surgery. In the immediate postoperative period, she developed fever which lasted for around 12 days. Cerebrospinal fluid (CSF) study done on the 10th postoperative day showed glucose of 62 mg%, protein of 130 mg% and 41 cells per cubic mm of which 5 were polymorphs, 17 were lymphocytes and 19 were red blood cells. CSF culture was negative for organisms. She recovered with supportive measures and antipyretics. Nine months later, she presented with excessive somnolence. Brain MRI revealed trapped left lateral ventricle with prominent dilation of its frontal, occipital and temporal horns ([figure 2](#)). Fine septations were also noted within the trapped left lateral ventricle. There was subfalcine herniation of the left cerebral hemisphere which compressed the right interventricular foramen resulting in dilation of the right lateral ventricle. The left lateral ventricle was seen herniating caudally into the midbrain, compressing the left cerebral peduncle. A CT ventriculography demonstrated the trapped left lateral ventricle with the contrast not entering the third and fourth

ventricles or the contralateral lateral ventricle ([figure 3](#)). A ventriculo-peritoneal shunt was done on the left side with improvement in the midline shift and resolution of the hydrocephalus ([figure 4](#)). There was very good clinical improvement and she is doing well at 3 years follow-up.

DISCUSSION

The surgical treatment of refractory epilepsy associated with uni-hemispheric pathology has evolved over the past century. Anatomic hemispherectomy, where the affected hemisphere is removed either 'en bloc' or in fragments, was first performed in 1938.^{2,3} Hemosiderosis and hydrocephalus were the two dreaded complications of this procedure which discouraged surgeons from embracing this surgical technique. Anatomic hemispherectomy was hence modified by Rasmussen into a resection-disconnection procedure called functional hemispherectomy.² Here, the central and temporal regions were resected out while the rest of the hemisphere was disconnected. In the 1990s, attempts to refine this technique with minimal resection and more disconnection resulted in the development of hemispherotomy. Two approaches namely

Figure 3 CT ventriculography shows the trapped left lateral ventricle with fine septations inside (A,B). No contrast is seen in the right lateral ventricle or in the third or fourth ventricles (C).

Figure 4 CT Brain after the ventriculo-peritoneal shunt (A, B, C, D) showing the shunt tube in the posterior body of left lateral ventricle (arrow). The trapped left lateral ventricle is reduced in size with improvement in the midline shift and resolution of hydrocephalus.

the vertical parasagittal approach and the lateral perisylvian approach are currently available.^{1,4} Hemispherectomy with its lesser complication rates and good seizure outcome has virtually replaced hemispherectomy in the surgical management of unihemispheric epilepsies. This is one of the very successful forms of epilepsy surgery with seizure freedom rates of around 90%. Functional outcome is also generally good after the surgery with the cognitive function remaining stable. There is an expected contralateral hemianopia and distal motor deficit, but the patients remain ambulatory.

The incidence of hydrocephalus after anatomic hemispherectomy varies greatly between studies and ranges from 9% to 81%.^{5–9} The exact pathophysiology is uncertain. The inflammatory response induced by the blood products and the brain proteins along with the surgical trauma is believed to interfere with the absorption of the CSF. The disconnective procedures could also deleteriously affect the CSF flow dynamics. In the case of functional hemispherectomy, aetiology seems to be the decisive factor than the inflammation induced by the surgery. One study found higher rates of hydrocephalus and shunting procedures in association with hemimegalencephaly and central nervous system infection and less in association with cortical dysplasia and Rasmussen's encephalitis.¹⁰ This was explained by the varying CSF flow dynamics in association with the different aetiologies.

The risk of postoperative hydrocephalus has reduced dramatically after the development of the different hemispherotomy techniques. A shunt rate of 15.7% was reported by Delalande in his series of 83 children after vertical parasagittal hemispherotomy.¹¹ Only 1 out of 40 paediatric patients required CSF shunt after the parasagittal procedure in a centre from Vienna.¹² Similarly, various lateral hemispherotomy techniques have reported varying frequencies of hydrocephalus ranging from 2% to 20%.^{4,13–15}

We present this case of entrapment of the ipsilateral lateral ventricle as a hitherto unreported late complication of hemispherotomy. Entrapment of the temporal horn of the lateral ventricle is a well-described entity.¹⁶ This is especially seen as a complication of tuberculous meningitis. The CT ventriculography test done in our case demonstrated the trapping of the lateral ventricle. The mild dilation of the contralateral lateral ventricle was only its consequence. It also clearly showed that despite the septations, the lateral ventricle was not loculated. Endoscopic fenestration of the trapped ventricle or a CSF diversion procedure were the two options available to restore the CSF outflow from the trapped ventricle.

The left-sided surgical procedure in our case would explain the inflammatory response in the left lateral ventricle. The

opposing walls of the lateral ventricle would adhere and the interventricular foramen of Monro would get obstructed by the inflammatory changes in the CSF induced by blood and brain proteins. The use of hemostatic agents would also contribute to the local inflammatory response. The prolonged fever in the perioperative period and the presence of septations within the ventricle are indirect markers of local inflammation. However, we are unsure why the hydrocephalus developed late, months after hemispherotomy. Such late development of hydrocephalus years after hemispherectomy has been described.¹⁵ This may be due to the slow changes in cerebral compliance associated with cerebral redistribution in relation to the CSF spaces happening over a period of time after hemispherotomy.

Learning points

- ▶ Entrapment of the lateral ventricle can occur as a late complication of hemispherotomy.
- ▶ New-onset neurological symptoms after hemispherotomy should raise the suspicion of this complication.
- ▶ Prompt intervention with diversion of cerebrospinal fluid from the trapped ventricle or endoscopic fenestration is required to control the symptoms.

Contributors RSI was involved in conception, draft and the content. RMR contributed towards the interpretation of the surgical data. Both KM and PK were involved in interpretation and draft of the radiological data.

Competing interests None declared.

Patient consent Obtained.

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