SHORT REPORT

Pneumoencephaly following lumbar puncture in association with an ethmoidal osteoma and porencephalic cyst

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A 50-year-old woman developed pneumoencephaly following a CSF examination for evaluation of dysequilibrium. Previous investigations had demonstrated a number of high signal T2 lesions on MRI of the brain. In addition, there was what was thought to be an asymptomatic cystic lesion in the left frontal lobe communicating with the lateral ventricle. After the lumbar puncture she developed extensive pneumoencephaly with pressure dilatation of the ventricular system. There was CSF rhinorrhoea. Further CT scans showed an osteoma in the ethmoidal air sinus with protrusion into the cystic area. This was the site of both the CSF leak and air entry. Caution must be taken when considering a CSF examination in the presence of either a presumed asymptomatic porencephalic cyst or ethmoid osteoma.

CASE REPORT

A 56-year-old female presented with a 2 year history of disequilibrium. This was intermittent with episodes lasting up to 2 weeks' duration. She described subjective swaying "as if I just came off a boat". There was no associated vertigo. She remained asymptomatic between these attacks. Apart from saccadic intrusion on pursuit eye movements, her examination, including Dix Hallpike's manoeuvre, was normal. She underwent MRI of the brain.

MRI demonstrated multiple subcortical lesions which gave a high signal on both T2 weighted and fluid attenuated inversion recovery (FLAIR) sequences (fig 1). In addition, there was a cystic lesion in the left frontal lobe with signal characteristics of CSF (fig 2). It was felt this communicated with the non-dilated horn of the left lateral ventricle.

In order to evaluate the high signal changes, she was further investigated with a series of blood tests and CSF examination. The CSF examination was performed as a day case in the left lateral position. This was uncomplicated with an opening pressure of 8.5 cm H₂O. Having been discharged on the same day after a 4 h period of observation in the ward, she developed sudden onset of severe generalised headache associated with nausea. There was no postural variation in the headache complex. She was admitted and rested on the assumption that this was probably low pressure headache. Over the next 24 h she began to deteriorate with confusion, intermittent dysphasia and mild left-sided weakness. An urgent CT scan of the brain was performed.

This demonstrated the presence of extensive pneumoencephaly with pressure dilatation of the ventricular system (fig 3). She was treated with bed rest and advised not to blow her nose. A nasal discharge was confirmed to be CSF.

She was observed, given adequate fluid and bed rest. Over 2 weeks her condition stabilised and she gradually improved. A repeat scan showed near complete resolution of the air in the subarachnoid space, although there was still air in both lateral

ventricles and the left frontal cyst. It was therefore felt that she had a CSF fistula and further investigations were undertaken.

T2 weighted MRI showed high signal change within the left anterior ethmoidal sinus air cell compatible with a CSF leak (fig 4). Fine cut CT imaging with bony window settings demonstrated an osteoma in the ethmoidal air cells, previously unsuspected on MRI (fig 5). This appeared to protrude up into the cystic area.

Four weeks after the onset of pneumoencephaly, she underwent repair of her CSF leak and removal of the osteoma. A left frontal craniotomy was performed. The floor of the left frontal fossa was explored where a intradural osteoma arising from the ethmoid area was seen. There was no dura above it and there was an associated dural fistula and brain herniation in the region. Once this was dissected the osteoma was drilled and removed.

The hole was then identified and repaired using a combination of Tisseal, glue and dural patch. Haemostasis was then secured and the dura was repaired in the usual fashion.

Further imaging confirmed successful repair and gradual improvement with absorption of intracranial air and resolution of her symptoms. Her CSF constituents were normal with no evidence of an inflammatory process. It was felt that the high signal changes on T2 weighted imaging were probably vascular in origin and that her dysequilibrium was vestibular.

Figure 1 Initial fluid attenuated inversion recovery (FLAIR) imaging demonstrating subcortical high signal change.

Figure 2 MRI demonstrating left frontal lobe cyst.

DISCUSSION

It is well established that spontaneous pneumoencephaly can occur as a result of osteomas.¹⁻⁸ Our patient underwent a straightforward, atraumatic, first pass lumbar puncture, with constant CSF flow throughout. At no point, either during or after the procedure, was it apparent that air had entered via the lumbar puncture site.

The site of the CSF leak correlated anatomically with that of the ethmoidal osteoma.

The precise method of formation of CSF fistula and pneumocephaly is unclear. It may be in the presence of CSF leakage, the

Figure 4 CSF in ethmoidal air cells.

escaping fluid through the defect, be it dural or arachnoidal, draws air by a mechanism comparable with that of a siphon into the relatively low pressure intracranial space. Alternatively, the breached dura acts as a simple flap valve, and air will enter the cranium whenever ambient pressure exceeds that of intracranial pressure. In our case we postulate that the lumbar puncture resulted in the creation of a negative pressure system and that this perpetuated the inflow of air into the cranium.

We do not have an adequate explanation for the presumed porencephalic cyst but postulate that a previous leak may have occurred at some time. The porencephalic cyst was of CSF intensity and had smooth walls. No abnormal signal was seen in the adjacent brain parenchyma. No mass effect or volume loss

Figure 3 CT demonstrating extensive pneumocephaly.

Figure 5 Osteoma in ethmoidal air cells.

was appreciated. In view of these findings, the porencephalic cyst is likely to have occurred in early life, probably in utero (congenital). Post traumatic sequelae would be associated with adjacent gliosis and haemorrhagic residua. Such cysts would have irregular margins, unlike what was seen in this case.

In conclusion, a CSF examination may be contraindicated in the presence of an ethmoidal osteoma, as pressure effects may result in an iatrogenic pneumocephaly. Also, incidental frontal porencephalic cysts should be further investigated for the presence of an osteoma which may be missed on standard MRI and even CT scans.⁹ A high index of suspicion is necessary as consequences could be grave if not treated.

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ECHO

Heart defects generally increase migraines

Please visit the *Journal of Neurology, Neurosurgery, and Psychiatry* website [www.jnnp.com] for a link to the full text of this article.

A small study has emphasised that migraines may have more than one source, after finding a link with random congenital heart disease defects in adults, not just right to left shunt, as previously accepted.

Prevalence of migraines, particularly those with aura, was significantly higher for patient groups with obligate right to left shunt or random congenital heart defect compared with a general practice population but not between groups with congenital defects with or without obligate right to left shunt. A link was also found between raised haemoglobin concentration and migraine, leading researchers to suggest that migraine caused by raised haemoglobin concentration, as seen elsewhere, might occur through activation of blood compounds and endothelium by shear stress.

The three groups comprised 40 patients, each age and sex matched, all aged over 16 years without Down syndrome or mental retardation. Patients with obligate right to left shunt were selected first from a database of patients with congenital heart disease and matched with patients from the entire database and with others from general practice. Migraine with or without aura was diagnosed by a neurologist from patients' self completed questionnaires, according to internationally recognised criteria.

One other report has suggested a link between migraine and congenital heart defects generally, but right to left shunt has been the focus of attention previously.

▲ Hermans H, *et al.* *Heart* 2007;**93**:361–362.