Cerebrospinal fluid shunt-induced chorea: case report and review of the literature on shunt-related movement disorders

Claudio M de Gusmão,1 Aaron L Berkowitz,2 Albert Y Hung,2 M Brandon Westover2

INTRODUCTION
Cerebrospinal fluid (CSF) diversion by shunting provides effective management of hydrocephalus.1 However, complication rates of CSF shunts range from 5% to 50%.1–3 Most common are shunt infections and mechanical failures; these may lead either to underdrainage (with re-emergence of hydrocephalus) or overdrainage (with intracranial hypotension and its potential complications, eg, subdural hematoma).3 4 Misplacement and migration of shunt catheters may cause seizures, intracerebral haemorrhage, and/or focal neurologic deficits, such as hemiparesis.1 2 We report a case of hemichorea after CSF shunt placement, and review the literature on CSF shunt-related movement disorders.

CASE REPORT
A 20-year-old woman with congenital hydrocephalus treated by CSF shunting presented with a purulent discharge from the site of a recent shunt revision and had a shunt infection. Her previous ventriculoperitoneal shunt was removed and a new ventriculopleural shunt was placed through a left occipital approach. There were no recent medication changes. Five days later, she developed debilitating involuntary choreoathetotic movements of the right upper and lower limbs (see online supplementary video 1), interfering with feeding and walking. Her examination, apart from the involuntary movements, was normal, with no rigidity, tremor, weakness, or ataxia. Her serum electrolytes were normal. CT scan of head showed that the shunt catheter tip penetrated the ventricular wall into the brain parenchyma (figure 1); MR scan of brain showed that its tip abutted the posterior aspect of the putamen (figure 2). There was no worsening of hydrocephalus, stroke, or haemorrhage. The shunt was surgically repositioned. Her choreoathetosis started improving immediately.
Figure 2  MR scan of brain. Axial (T2-FLAIR) and sagittal (T1 postcontrast) views show the cerebrospinal fluid shunt catheter tip terminating in the posterior putamen (arrows).

Table 1  Movement disorders complicating shunts

<table>
<thead>
<tr>
<th>Age</th>
<th>Type of movement</th>
<th>Cause of hydrocephalus</th>
<th>Time after shunting</th>
<th>Recovery and time</th>
<th>Mechanism</th>
<th>Reference</th>
</tr>
</thead>
<tbody>
<tr>
<td>17</td>
<td>Parkinsonism</td>
<td>Pineal gland germinoma</td>
<td>Not reported (but 3 days after roller coaster ride)</td>
<td>Full, 1 month</td>
<td>‘Shunt displacement with hydrocephalus’</td>
<td>Lau et al11</td>
</tr>
<tr>
<td>28</td>
<td>Parkinsonism</td>
<td>Ommaya reservoir for methotrexate injection</td>
<td>‘Following treatment’</td>
<td>‘Immediate after shunt insertion’</td>
<td>Not reported</td>
<td>Cheshire et al16</td>
</tr>
<tr>
<td>32</td>
<td>Parkinsonism</td>
<td>‘Obstructive hydrocephalus’</td>
<td>3 months</td>
<td>Improved with levodopa</td>
<td>‘Nigrostriatal pathway congestion’</td>
<td>Ochiai et al13</td>
</tr>
<tr>
<td>59</td>
<td>Parkinsonism</td>
<td>‘Obstructive hydrocephalus’</td>
<td>3 months</td>
<td>Improved with levodopa</td>
<td>‘Nigrostriatal pathway congestion’</td>
<td>Ochiai et al13</td>
</tr>
<tr>
<td>38</td>
<td>Parkinsonism</td>
<td>Neurocysticercosis</td>
<td>3 days</td>
<td>Partial, 3 months</td>
<td>‘Changes in intraventricular pressure or malpositioning of the shunt’</td>
<td>Prashantha et al6</td>
</tr>
<tr>
<td>46</td>
<td>Parkinsonism</td>
<td>Aqueductal stenosis</td>
<td>1 year</td>
<td>Partial with levodopa</td>
<td>‘Parkinsonism associated with V-P shunting’</td>
<td>Sakurai et al17</td>
</tr>
<tr>
<td>64</td>
<td>Parkinsonism</td>
<td>Aqueductal stenosis</td>
<td>4 months</td>
<td>None</td>
<td>‘Slit-ventricle syndrome’</td>
<td>Yomo et al18</td>
</tr>
<tr>
<td>26</td>
<td>Parkinsonism</td>
<td>Encephalitis</td>
<td>7 months</td>
<td>Full, 2 months</td>
<td>‘Shunt malfunction’</td>
<td>Tokunaga et al12</td>
</tr>
<tr>
<td>11</td>
<td>Akinetia/ parkinsonism</td>
<td>Tectal glioma</td>
<td>‘Days’</td>
<td>2 weeks, Partial improvement with bromocriptine and levodopa</td>
<td>‘Recurrent hydrocephalus and tectal glioma’</td>
<td>Rebai et al19</td>
</tr>
<tr>
<td>32</td>
<td>Tremor/ parinaud’s syndrome</td>
<td>Neurocysticercosis</td>
<td>Not reported</td>
<td>‘Several days after shunt revision’</td>
<td>Hydrocephalus or direct trauma to basal ganglia</td>
<td>Keane7</td>
</tr>
<tr>
<td>68</td>
<td>Ballism</td>
<td>Normal pressure hydrocephalus</td>
<td>‘Upon awakening from surgery’</td>
<td>Not reported</td>
<td>Not reported</td>
<td>Walker et al20</td>
</tr>
<tr>
<td>14</td>
<td>Torticollis</td>
<td>Aqueductal stenosis</td>
<td>12 years</td>
<td>Full, 1 year</td>
<td>‘Local in the neck’</td>
<td>Singh et al21</td>
</tr>
<tr>
<td>14</td>
<td>Ataxia cerebrospinal fluid deficits</td>
<td>Congenital Chiari I</td>
<td>13 years</td>
<td>‘Near complete’, 1 week</td>
<td>‘Positive rostral pressure gradient through stenotic foramen magnum’</td>
<td>Elliot et al22</td>
</tr>
<tr>
<td>24</td>
<td>Hemichorea</td>
<td>Meningitis</td>
<td>2 weeks</td>
<td>Complete, ‘few days’</td>
<td>Not reported</td>
<td>Alakandy et al5</td>
</tr>
</tbody>
</table>
postoperatively, and had largely resolved spontaneously at 2 weeks when she was discharged to rehabilitation (see online supplementary video 2)

DISCUSSION
Movement disorders related to cerebrospinal fluid (CSF) shunting are rare, though parkinsonism, ataxia, ballism and torticollis may occasionally occur (table 1). We could find only one previous report of shunt-related hemichorea, which, as here, resolved after shunt repositioning.5

The mechanism of ventriculoperitoneal (VP) shunt-related movement disorders has been attributed to ‘altered CSF dynamics’,6 whereby shunt obstruction causes acute hydrocephalus, with shearing, torsion, or ischaemia of striatal projections.7–14 Although there is a report of chorea accompanying acute hydrocephalus and resolving after shunting, in that case the movements were generalised and presumed secondary to caudate head compression by the dilated lateral ventricles.15 In our case, the movement disorder probably resulted from direct striatal irritation by the shunt catheter itself, as the movement disorder emerged in the setting of normal shunt function with no radiological evidence of worsening hydrocephalus, and promptly resolved after shunt repositioning.

CONCLUSION
Hemichorea is an unusual complication of CSF shunt placement. Clinicians should be attentive to focal deficits including unilateral movement disorders in patients with CSF shunts, as prompt surgical intervention may alleviate debilitating neurological symptoms and minimise the risk of long-term complications.

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REFERENCES
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