Spontaneous concomitant cranial and spinal subdural haematomas with spontaneous resolution

Jain V, Singh J, Sharma R

ABSTRACT
We report a rare case of concomitant cranial and spinal subdural haematoma (SDH) in a 12-year-old boy with severe thrombocytopenia due to aplastic anaemia, and review the available literature. Magnetic resonance (MR) imaging at presentation revealed a cranial SDH confined to the posterior fossa, and spinal SDH extending from the C1 to T3 segments. The child was managed conservatively due to his poor general condition and lack of any neurological deficit. Repeat MR imaging done at six weeks showed complete resolution of the spinal SDH and partial resolution of the cranial SDH. Although rare, a spontaneous spinal SDH can occur simultaneously with a cranial SDH. Urgent surgical decompression is considered the treatment of choice for spinal SDH; however, a conservative approach may succeed in patients with poor general condition, and/or mild/no neurological deficit.

Keywords: child, cranial subdural haematoma, spinal subdural haematoma, spontaneous resolution

INTRODUCTION
Spinal subdural haematoma (SDH) is a rare condition, usually associated with apparent trauma, iatrogenic manoeuvres, such as lumbar puncture, and haemostatic disorders including anticoagulation.1-3 Many cases of spontaneous onset spinal SDH have also been reported,4-6 albeit a few of them without associated coagulation anomalies.4-6 Although urgent surgical management is considered the treatment of choice,4-7 spontaneous resolution of spinal SDH is also well documented.4-6,8-9 Concomitant cranial SDH and spinal SDH are extremely rare, with only 11 cases reported previously.8-18 We describe a case of concomitant cranial and spinal SDH involving almost the entire spinal canal in a child with severe thrombocytopenia due to aplastic anaemia, which showed spontaneous resolution at six weeks of conservative management, and review the available literature.

CASE REPORT
A 12-year-old boy, a known case of aplastic anaemia, presented to the emergency room of our institute with a severe headache localised to the occipital region for three days. There was no history of trauma or any invasive procedure, like lumbar puncture. A non-contrast computed tomography (CT) performed at that time showed hyperdensity along the tentorium cerebelli suggestive of SDH (Figs. 1a–b). His platelet count at admission was 20,000/μL (normal range 1.5–4 × 10^5 per μL). He was put on an infusion of platelet-rich plasma. Initially his symptoms improved; however, after two weeks, his headache worsened. The platelet count at that time was 70,000/μL. A repeat non-contrast CT again demonstrated SDH along the tentorium cerebelli (Figs. 1c–d), which appeared to have increased in comparison with the previous scan.

During the second episode of headache, he also complained of severe back pain, which increased in sitting
Fig. 2 (a,b) Sagittal and (c) axial T1-weighted MR images show multiple biconvex subdural collections in the posterior fossa compressing the cerebellar parenchyma. These collections are hyperintense on T1-weighted images, and (d) show blooming on GRE-weighted image, suggestive of a subacute haematoma. The sagittal images clearly show the continuity between the posterior fossa and spinal SDH.

Fig. 3 Sagittal MR images of the spinal canal show the subdural haematoma extending from the posterior fossa cranially to the third sacral segment caudally. The haematoma is most prominent anteriorly in the cervical and upper dorsal regions, and posteriorly in the lower dorsal and lumbar regions. The signal intensity of the haematoma is hyperintense on (a,b) fat-saturated T1-weighted images, as well as the (c) T2-weighted image.
Fig. 4 (a) Axial T1-weighted MR image at the level of lumbar spine shows circumferential hyperintense subdural collections compressing the cauda equina resulting in a “three-branch star” appearance. A hypointense layer of dura (arrow) is seen dorsal to the collection with presence of hyperintense fat external to it (asterisk), that shows suppression on the (b) fat-saturated MR image (solid white arrow).

Fig. 5 Repeat T1-weighted sagittal MR images of the spine, performed after symptomatic resolution at six weeks, show complete resolution of the spinal SDH. Minimal residual SDH in the posterior fossa is still seen (arrow).
or standing positions, and was least painful when he lay supine. On examination, there was no local tenderness, no neurological deficit, and all the reflexes were normal. The child experienced neck rigidity. Magnetic resonance (MR) imaging of the brain and spine were performed. MR imaging showed multiple extra-axial collections confined to the posterior fossa. The collections were biconvex in shape and had a mass effect on the cerebellar parenchyma. These subdural collections were hyperintense on T1-weighted as well as T2-weighted images, with “blooming” on gradient echo images, suggesting a diagnosis of subacute haematoma (Fig. 2). There was no other intracranial bleed.

The haematoma was seen extending into the spinal subdural space, involving almost the entire spinal column, extending from the C1 to S3 segments (Fig. 3). Continuity of the SDH between the posterior fossa and spinal component was well defined on the sagittal images (Figs. 2a–b). The spinal SDH was present circumferentially around the cord and cauda equina (Fig. 4), being most prominent anteriorly in the cervical and upper dorsal regions, and posteriorly in the lower dorsal and lumbar regions (Fig. 3). It showed similar signal intensity to cranial SDH, being hyperintense on the T1-weighted and T2-weighted images. In the lumbar spinal canal, the cauda equina was compressed from the anterior and posterolateral aspects, giving a somewhat “three-branch star” appearance (Fig. 4a). The haematoma was seen surrounded posteriorly by a hypointense layer of dura (Fig. 4a) with preserved epidural fat seen dorsal to it (Fig. 4a), establishing its subdural location. Due to the poor general condition of the child, and lack of any neurological deficit, he was put on conservative management with platelet-rich plasma, mannitol and steroids. He showed gradual improvement in symptoms, and was free of head and back pain after six weeks. Repeat MR imaging after

<table>
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<tr>
<th>Author (year)</th>
<th>Possible cause(s)</th>
<th>Age (years) / gender</th>
<th>Level of SDH</th>
<th>MR imaging – SI (T1-W/T2-W)</th>
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<tbody>
<tr>
<td>Bortolotti et al</td>
<td>Trauma</td>
<td>23/F</td>
<td>L4-S2</td>
<td>high/iso</td>
<td>Surgery</td>
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<td>Hung et al (2002)</td>
<td>Trauma</td>
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<tr>
<td>Leber et al (1997)</td>
<td>Trauma</td>
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<td>L1-S2</td>
<td>high/-</td>
<td>Surgery</td>
<td>Good</td>
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<td>Lecouvet et al (2003)</td>
<td>Haemorrhagic brain metastases</td>
<td>31/M</td>
<td>L1-S2</td>
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<td>Conservative</td>
<td>Good</td>
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<td>Ohta et al (2001)</td>
<td>VP shunt</td>
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<td>Surgery</td>
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<td>Shimizu et al (1999)</td>
<td>Cranietomy</td>
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<td>high/iso</td>
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<td>Silver et al (1991)</td>
<td>VP shunt</td>
<td>14/F</td>
<td>D12-L2</td>
<td>not done</td>
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<td>Good</td>
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<td>Tillich et al (1999)</td>
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<td>54/M</td>
<td>L1-S2</td>
<td>high/high</td>
<td>Surgery</td>
<td>Good</td>
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<td>Wurm et al (1996)</td>
<td>VA shunt</td>
<td>16/M</td>
<td>L3-S2</td>
<td>high/-</td>
<td>Conservative</td>
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<td>Yamaguchi et al (2003)</td>
<td>Ruptured i/c aneurysm, CSF drainage, intrathecal thrombolytics</td>
<td>52/F</td>
<td>L1-S2</td>
<td>high/iso</td>
<td>Conservative</td>
<td>Good</td>
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<tr>
<td>Yamaguchi et al (2005)</td>
<td>Antiplatelet drugs</td>
<td>59/M</td>
<td>D11-S1</td>
<td>iso/iso</td>
<td>Conservative</td>
<td>Good</td>
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<td>Present case</td>
<td>Severe thrombocytopenia</td>
<td>12/M</td>
<td>C1-S3</td>
<td>high/high</td>
<td>Conservative</td>
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CSF: cerebrospinal fluid; i/c: intracranial; T1-W: T1-weighted; T2-W: T2-weighted; VA: ventriculoatrial; VP: ventriculoperitoneal
In the present case, MR imaging revealed a large anomaly. Most spinal SDH cases manifest with back pain the patient had a normal platelet count and no coagulation by Yamaguchi et al, was a history of antiplatelet drugs case. The only predisposing factor in the case illustrated anomaly as the sole predisposing factor, as seen in this reported cases, there is no such report with coagulation intervention including a lumbar puncture. Among the due to aplastic anaemia, with no history of trauma, or any case, the only predisposing factor was the altered SDH either had previous cranial surgery (five cases), or out of 11) of the cases of concomitant cranial and spinal SDH is also obscure. The spinal SDH can be caused by rupture of spinal vessels. However, unlike in the brain, the spinal subdural space lacks the bridging veins that may be the source of SDH. Spinal haematoma may migrate from the cranial lesion, a view that has been supported by many authors, Lecouvet et al explained that the propagation of blood from cranial to spinal subdural space is possible due to the presence of anatomic continuity between them. In the present case, as the continuity of the haematoma between the posterior fossa and spine was very well documented by MR imaging (Fig. 2), we also support the migration theory.

The predisposing factors for spinal SDH include trauma, iatrogenic manoeuvres such as lumbar puncture, and disorders of coagulation. Although many reports of spontaneous onset of spinal SDH exist, iatrogenic trauma like lumbar puncture is considered an important superimposed factor in many such cases. In the largest series of spinal SDH, Domenicucci et al found coagulation anomalies in 53% and a prior history of a lumbar puncture in 33% of the 106 cases reviewed. The majority (nine out of 11) of the cases of concomitant cranial and spinal SDH either had previous cranial surgery (five cases), or a history of trauma (four cases) (Table I). In the present case, the only predisposing factor was the altered coagulation profile caused by a severe thrombocytopenia due to aplastic anaemia, with no history of trauma, or any intervention including a lumbar puncture. Among the reported cases, there is no such report with coagulation anomaly as the sole predisposing factor, as was seen in this case. The only predisposing factor in the case illustrated by Yamaguchi et al, was a history of antplatelet drugs for a previous episode of stroke. However, even then, the patient had a normal platelet count and no coagulation anomaly. Most spinal SDH cases manifest with back pain or cauda equina compression. All cases of concomitant intracranial and spinal SDH also presented with back or leg pain, with or without neurological deficit. In the present case, our patient had severe back pain that increased on movement and when sitting. However, he had no neurological deficit.

MR imaging is considered the modality of choice for identifying the presence and extent of spinal SDH. In the present case, MR imaging revealed a large spinal SDH that spanned almost the entire spinal canal, extending from the first cervical segment, in continuity with the posterior fossa, to the S3 level. In all previously-reported cases of concomitant cranial and spinal SDH, the spinal SDH was confined below the lower thoracic spine, and cervical involvement has not been reported. The authors are also unaware of any report of isolated spinal SDH without associated intracranial SDH, with such an extensive involvement; the largest reported extent comprised 18 spinal segments. Therefore, the unusual features of our case include a very long segment haematoma with involvement of the cervical segment, as well as a circumferential nature of the haematoma. In a review of 35 cases of spinal SDH, Boukobza et al found only two cases with a circumferential involvement, and involvement of the cervical segment was very rare.

The signal intensity of spinal SDH was hyperintense on T1-weighted as well as T2-weighted images. Since the evolution of signal intensity changes in spinal haematoma parallels that of cranial haematoma, the present case corresponded to the subacute stage of haematoma. All cases of concomitant intracranial and spinal SDH, except that reported by Yamaguchi et al, were subacute, showing hyperintensity on T1-weighted images. Also, the signal intensity of the intracranial SDH and spinal SDH were similar, suggesting same origin of these two haematomas, and further supporting the migration theory for the origin of spinal SDH.

The localisation of spinal haematoma is important, and there are several points to differentiate extradural haematoma (EDH) from SDH. EDH has a less marked craniocaudal extension (two to four vertebrae), biconvex or lentiform shape, tapering superior and inferior margins on sagittal images, and regular anterior border on axial images. By contrast, spinal SDH appears concave on sagittal images and irregular on axial images. Post et al established that the presence of dura and a layer of fat outside the haematoma and/or dura matter confirmed the subdural localisation of the haematoma. Therefore, the most definitive point of distinction between SDH and EDH is the presence of a well-defined layer of dura limiting the SDH posteriorly, and a well-preserved epidural fat external to it seen on the axial images. Both these features were very well seen on the axial T1-weighted images (Fig. 4) in

DISCUSSION

Spinal SDH is a rare entity, and its concomitant occurrence with cranial SDH is even rarer, with only 12 such cases (including the present case) being reported in the literature (Table I). The origin of bleeding in spinal SDH is controversial. Spinal SDH may originate from subarachnoid haemorrhage that dissects through the arachnoid membrane into the subdural space; or a sudden increase in abdominal and thoracic pressures may lead to rupture of the intraspinal vessels and consequent haematoma. The aetiology of occurrence of concomitant intracranial and spinal SDH is also obscure. The spinal SDH can be caused by rupture of spinal vessels. However, unlike in the brain, the spinal subdural space lacks the bridging veins that may be the source of SDH. Spinal haematoma may migrate from the cranial lesion, a view that has been supported by many authors. Lecouvet et al explained that the propagation of blood from cranial to spinal subdural space is possible due to the presence of anatomic continuity between them. In the present case, as the continuity of the haematoma between the posterior fossa and spine was very well documented by MR imaging (Fig. 2), we also support the migration theory.

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the present case. Axial images also showed a similarity to the “three-branch star” appearance described by Lecouvet et al in lumbar subdural haematomas,(8) which could result from the accumulation of nerve roots of the cauda equina within the dural sac, the posterior “branch” representing stretched filum terminale and anterolateral “branches” representing the emerging nerve roots.

Management of spinal EDH and SDH is considered a neurosurgical emergency, with extended laminectomy and complete evacuation of haematoma being the treatment of choice, specially if the neurological status of the patient is deteriorating.(2-4,7) However, spontaneous resolution has also been well documented.(5,6) The majority of the cases (seven out of 11) of concomitant cranial and spinal SDH also showed good recovery on conservative management.(8,9,11,14,15,17,18) This case substantiates the fact that spinal SDH may resolve with conservative management. Therefore conservative management can be recommended for patients with no or minimal neurological deficit and/or poor general condition. To conclude, the illustrated case of spinal SDH was unusual. Not only was the association of spinal SDH with intracranial SDH a rare entity, a purely spontaneous origin, as well as such a large extent of the spinal component, involving almost the entire spine, have not been reported before. However, in spite of such a large extent, the spinal SDH showed complete resolution after conservative management for six weeks.

REFERENCES