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Tension Subdural Hygroma Following Resection of Posterior Fossa Tumour in a Child

A new clinico-radio-pathological entity

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Abstract

Persistent hydrocephalus is common in child after resection of posterior fossa tumours. Occurrence of subdural hygroma, but is very rare, with only few cases reported. We report the rare case of a child who developed a tense subdural hygroma with stable hydrocephalus, in the early postoperative period, following posterior fossa tumour resection. We describe the distinctive clinical, radiological and pathological features associated with the development of a tense subdural hygroma. We also discuss the management by cerebrospinal fluid diversion, which includes either a ventriculoperitoneal or subduroperitoneal shunt. This unique condition is distinguished from external hydrocephalus by features that are critical to the management strategy.

Keywords: Child, Posterior fossa tumour, Postoperative period, Hydrocephalus, Subdural hygroma, Hygroma, External hydrocephalus.

Introduction

The incidence of non-communicating hydrocephalus (HC) in children with posterior fossa tumour (PFT) is 70-90%.[1] Hydrocephalus (HC) will persist in approximately 30% of them,

after resection of the tumour and this HC may have a communicating component to it.[1-6] No report of development of tense pseudomeningocele (PMC), due to tense subdural and interhemispheric CSF collection associated with regression of HC, after PFT resection, exists to date. We report such a case and introduce the term "tension subdural hygroma" (tSH) to describe this rare, but distinct clinical, radiological and pathological phenomenon.

Case Report

A 14 month old male child presented to Paediatric Neurology clinic with vomiting, irritability and imbalance while sitting/walking of 2-3 weeks duration. Clinically the child was awake and alert, but was irritable and had truncal ataxia. Pupils were equal and reacting to light and fundi showed no papilledema. Magnetic resonance imaging (MRI) of brain showed a large midline PFT, the features of which were suggestive of medulloblastoma. There was associated HC with periventricular lucency (PVL) (Fig A). He underwent a modified telo-velar approach and gross total excision of the tumour. An external ventricular drain (EVD) was placed through a right sided Frazier burr hole, immediately before surgery. The EVD was kept clamped post-operatively. The postoperative Computerized Tomography (CT) scan, after 24 hours showed a clear tumour bed, some blood in the frontal horns of lateral ventricles and the third ventricle. The cerebral aqueduct was patent. The HC was persisting with Evans index remaining the same as in preoperative scan (Fig 1B). EVD was removed since there was no worsening of the HC after clamping the drain for 24 hours. MRI scan of the brain and spine on the third postoperative day, showed a new bilaterally symmetrical subdural hygroma (SH) and reduction in HC (Fig 1C). There was no residual tumour (Fig 1D) or spine metastasis. The child developed a PMC at the surgical and EVD site, on the fifth postoperative day. The PMC progressed despite drainage of CSF via lumbar puncture (LP) on the fifth and twelfth postoperative days. By the fourteenth day the PMC was tense (Fig 2A). The child was afebrile but irritable and oral intake was poor with episodes of vomiting. CT brain showed very large hypodense collections at the surgical site and bilateral convexity and interhemispheric subdural space, with reduction of HC (Fig 2B-D). Analysis of CSF obtained during LP did not show any evidence of infection. He underwent emergency CSF diversion using a medium pressure shunt system. Per-operatively, upon nicking the dura to insert the ventricular catheter, the CSF in the subdural space under high pressure, jetted out. The catheter had to be advanced for 4-5cm, in an attempt to enter the ventricle and obtain continuous drainage of CSF. Postoperatively the PMC resolved completely within a day (Fig 3 A) and the child improved clinically. Follow up CT scan of the brain after one month

showed malposition of distal end of the catheter in the interhemispheric subdural space, a stable HC with no PVL and complete resolution of the subdural collection and PMC, suggestive of a functioning shunt system (Fig 3B-D). Though a ventriculoperitonal shunt (VPS) was planned, it turned out to be a subdural-peritoneal shunt (SPS) by default. Histopathology examination confirmed the diagnosis of medulloblastoma with extensive nodularity. The child was subsequently transferred to Department of Oncology for further management. Follow up MRI brain after 6 months showed complete resolution of the HC and subdural hygroma (SH) (Fig 4A-E). The child is under regular surveillance since then. Follow up imaging of the brain, spine and abdomen did not show evidence of recurrence of tumour, spine metastasis or CSF seeding of tumour in the peritoneal cavity.[7] Intra-thecal chemotherapy was not given because there was no spine metastasis.

Parental consent was obtained for the publication of case report with photograph and radiological images.

Discussion

In children with PFT the factors leading to persistence of HC, after tumour resection includes an age of less than 3 years, duration of illness of less than 3 months, midline location of the tumour, subtotal resection, pre-operative EVD placement, prolonged EVD requirement, early PMC formation, post-operative CSF leak, medulloblastoma/ependymoma histology and greater ventricular index on presentation.[4,5,8-12] In this case, the child underwent right sided EVD, immediately before resection of the tumour. Child developed PMC at the surgical and EVD site, in five days, which was progressing to a large and tense PMC, despite two attempts of drainage of CSF. The MRI and CT scans (on the 3rd & 14th postoperative days) showed progression of the SH and PMC, as well as regression of the HC. The mechanism of development of postoperative SH is still a topic of debate. The possible explanation based on our case is as follows. The CSF from the ventricles tracked into the subdural space via the iatrogenic communication created by the EVD and modified telo-velar approach. The progressive egress of CSF, compounded by the communicating nature of HC, led to increase in SH, in terms of both volume and pressure. This in turn led to the development of PMC. The CSF in the subdural space was under high pressure. This was unlike in a SH caused by loss of cerebral volume and SH due to other causes, where there was no direct communication subdural space and the ventricles. We hence consider this as a separate entity and term it “tension SH” (tSH). One possible explanation of missing the ventricle, while

performing VPS, was the transient change in the configuration of the right cerebral hemisphere and lateral shift of the right lateral ventricle, due to sudden egress of CSF from the subdural space, upon opening the dura. Use of intra-operative image guidance could have avoided the malposition of ventricular catheter. The complete resolution of PMC, SH and HC as seen in the post shunt imaging of the brain, confirms the dynamic communication between all the three CSF compartments, in this child (ventricles, PMC, subdural space). Other management options were a VPS, burr hole and drainage of SH or endoscopic third ventriculostomy (ETV). VPS would have resulted in resolution of the SH, PMC and HC. Burr hole drainage would have certainly resulted in re-accumulation of the SH as evidenced by recurrence of PMC after drainage via LP. ETV would have been a failure due to the communicating nature of the HC in this case.[13] Included with the manuscript is a supplement with flow chart of management algorithm, covering all the possibilities. A literature review showed four articles reporting development of SH following tumour resection, one being supratentorial and the other three being infratentorial tumors.[14-17] A case report by Behera et al, closely resembles our case. They termed the SH as "periencephalic subdural panhygroma". This case, but did not have a PMC, possibly due to lack of enough tension in the SH to produce a PMC. Other possibility was water tight closure of the dura and replacement of the bone flap. The HC also did not regress with the development of SH. Moreover the SH did not resolve completely after VPS, as evidenced by the postoperative CT scan.[17] Eguchi et al reported three paediatric cases, with tumour in the supratentorial region.[14] They called these as post-operative extra axial CSF collections, irrespective of whether the collection was in the subarachnoid or subdural space. Anokha et al reported cases of two adults with posterior fossa tumour, who developed post-operative SH (one being a intra axial metastatic cerebellar lesion and the other an intra-fourth ventricular lesion).[15] These collections were asymmetrical and did not resolve completely after VPS, unlike tSH. Stavrinos et al reported another case in an adult who developed SH following excision of intra axial cerebellar mass.[16] The SH was symmetrical and was managed by burr hole drainage of the supratentorial SH and aspiration of the PMC. The other differential diagnosis was external hydrocephalus (EH), where the CSF accumulates in the subarachnoid space. The visualization of subdural bridging veins over the convexity and absence of widening of cortical sulci in the CT brain, excluded the possibility of EH.

Conclusion

Ours is the second reported case of SH following posterior fossa tumour resection in a child. The hallmark features which makes it distinct include, a tense PMC, progression of SH with regression of the HC, CSF in the subdural space under high pressure and CSF diversion resulting in complete resolution of the PMC, SH and HC. We introduce the term "tSH" to name this distinct condition. A SPS may be the procedure of choice compared to VPS, because of easy access of shunt tube to convexity subdural space, compared to ventricles. Further studies and similar case reports are warranted to establish this entity.

Authors' Contribution

MKP contributed to the concept and design, data acquisition, drafting, critical review and literature review. RK and RC were involved in the data analysis, critical review and literature search. VG contributed to the literature search and data collection. KKK was involved in the data collection. All authors approved the final version of the manuscript.

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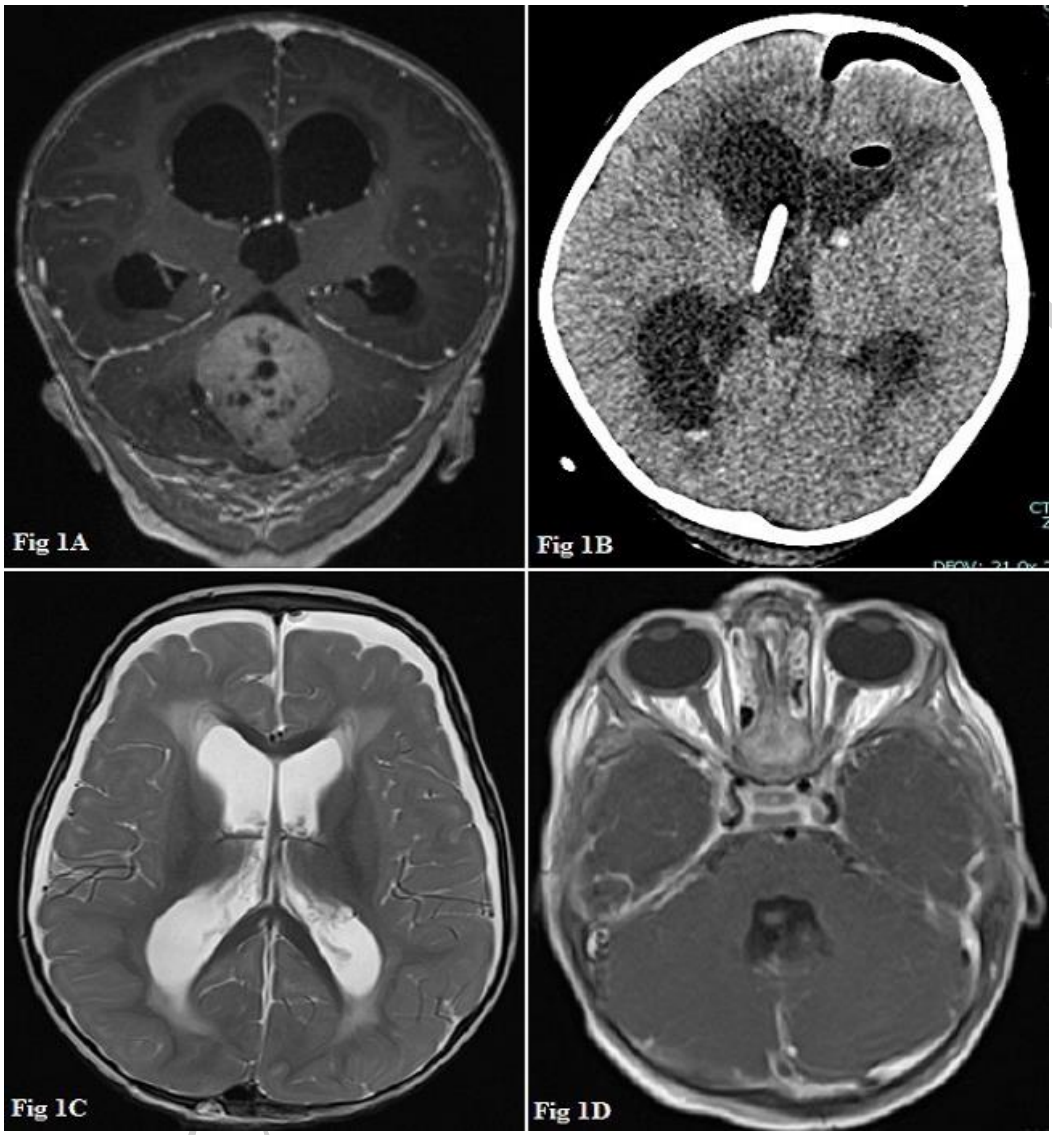

Figure 1: **A** - MRI brain, T1W, gadolinium enhanced, coronal image, showing midline posterior fossa tumour with hydrocephalus. **B** - Postoperative CT scan axial section, 24 hours after clamping the external ventricular drain, showing persistent hydrocephalus and ventricular catheter in the right lateral ventricle (arrow); **C** - Postoperative MRI brain, axial T2W image, 72 hours after removal of external ventricular drain, showing thin subdural hygroma and regression of hydrocephalus; **D** - T1W, gadolinium enhanced, axial image, showing no residual tumour.

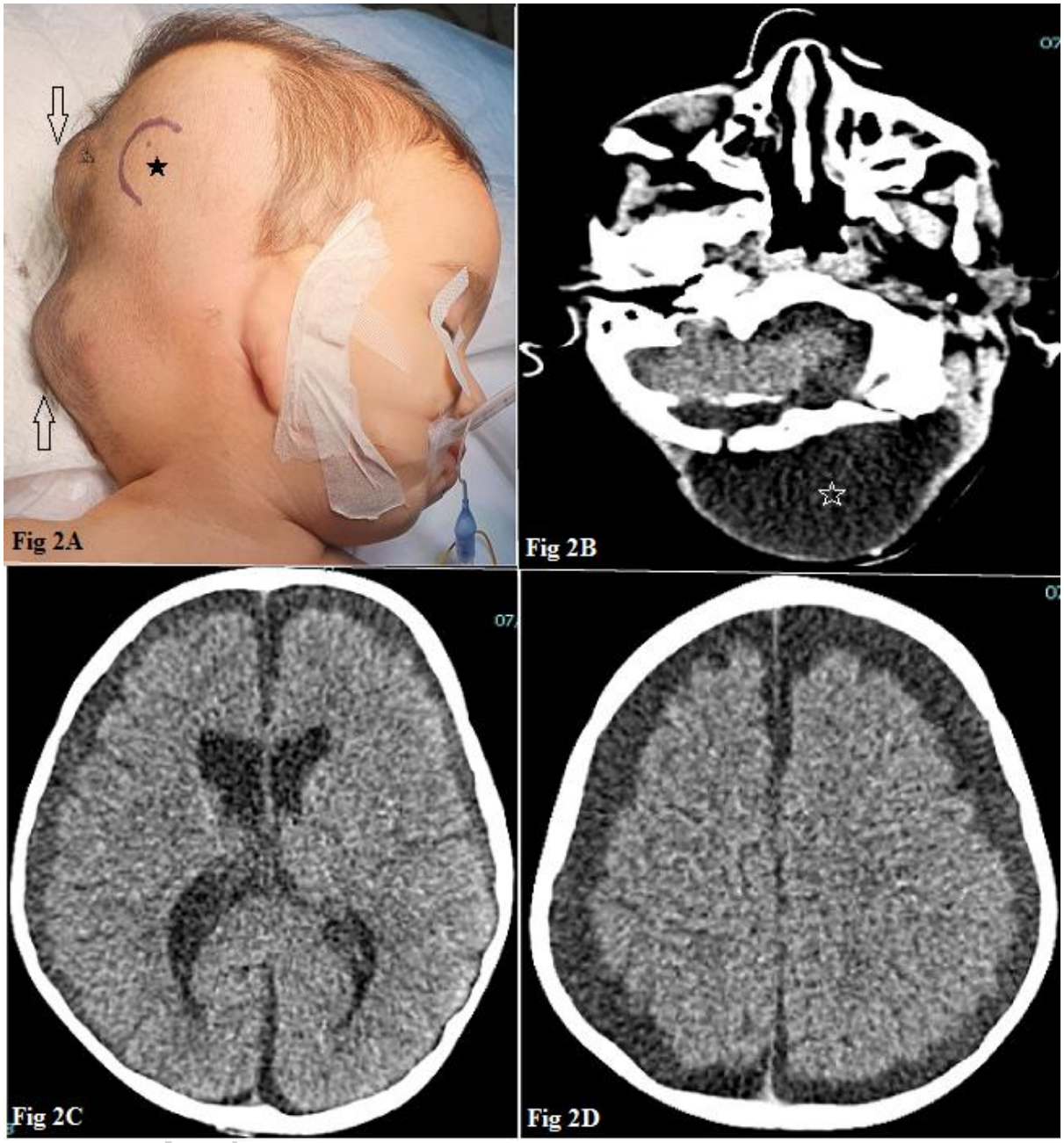

Figure 2: **A** - Tense pseudomeningocele at the external ventricular drain site (arrow down) and surgical site (arrow up), immediately before shunt surgery; shunt incision mark (asterix). **B** - CT scan brain axial sections showing the pseudomeningocele (asterix); **C & D** – progression of subdural hygroma and further regression of hydrocephalus.

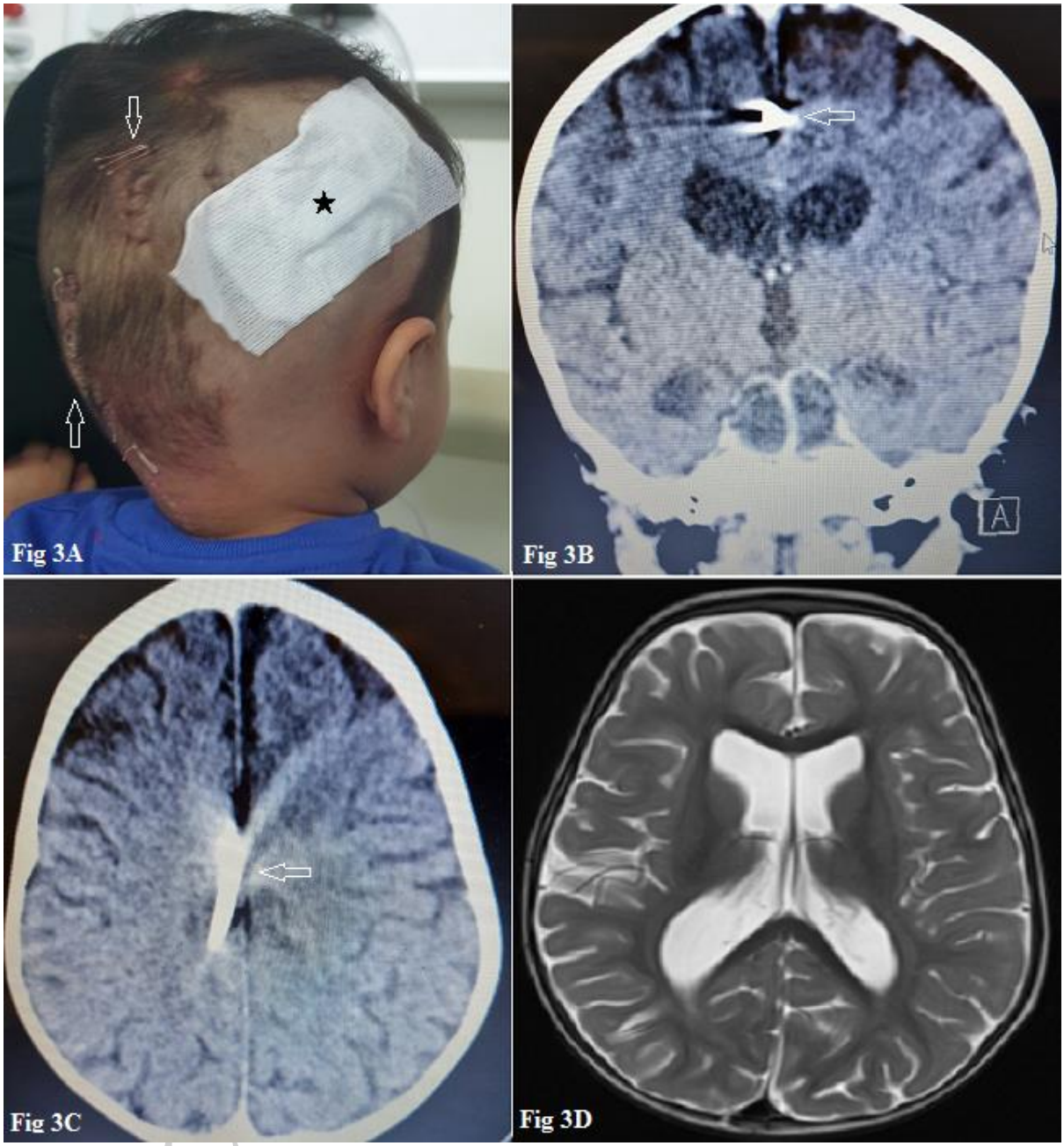

Figure 3: **A** – Complete resolution of pseudomeningocele 24 hours after shunt (arrows up & down; asterix - dressing over shunt insertion site). **B & C** - CT scan of the brain coronal and axial sections, after one month, showing complete resolution of the subdural hygroma, regression of the hydrocephalus and shunt tube tip in the interhemispheric subdural space (arrows). **D** – MRI brain T2W axial section, after 5 months, showing complete resolution of the hydrocephalus and subdural hygroma.