Tension Subdural Hygroma following Resection of Posterior Fossa Tumor in a Child – A new Clinical Pathological Radiological entity

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Case Report

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Abstract

Subdural hygroma following posterior fossa tumor resection is very rare, with only three cases reported in the literature. Authors describe a case of subdural hygroma following posterior fossa tumor resection with distinct clinical, pathological and radiological features, through an illustrative case of a child, who underwent gross total resection of a fourth ventricular tumor. The clinical, radiological and pathological sequelae following development of post-operative bilaterally symmetrical subdural cerebrospinal fluid collection, under high pressure, explained. The term “tension subdural hygroma” introduced to describe this unique condition, not described elsewhere, to date. Features that differentiate this condition from previously reported cases of subdural hygroma following posterior fossa tumor resection, explained. Subduro-peritoneal shunt as the choice of treatment, rather than ventriculo-peritoneal shunt, burr hole drainage of the subdural hygroma or aspiration of the pseudomeningocele, suggested.

Introduction:

Hydrocephalus (HC) occur in 70–90% of children with posterior fossa tumors (PFT) [1–3]. HC persist after tumor resection in 30% [1, 2, 4–6]. Earlier studies which categorized these children into low-risk and high risk groups, for the development of HC, based on preoperative variables such as age younger than 2 years, moderate/severe hydrocephalus, preoperative tumor diagnosis, and transependymal edema [6, 7]. One recent study showed that these variables failed to reliably predict which children would require permanent CSF diversion following PFT resection when applied to this cohort and suggested clinical judgment as mainstay of choosing the perioperative treatment of hydrocephalus [8]. The lack of class I evidence, to guide management of HC, lead the Neurosurgeon to choose one of the cerebrospinal fluid (CSF) diversion options, based on clinical status of the child, severity of HC and their experience [1, 9]. The options adopted may be pre-operative or post-operative and temporary or permanent. The procedures include external ventricular drainage (EVD), ventriculo-peritoneal shunt (VPS) and endoscopic third ventriculostomy (ETV). ETV being the most physiological, should be considered as the best option, if a pre-operative CSF diversion was planned, because this can reduce the risk of development of post-operative HC from 27 to 7% [3, 6]. Though postoperative HC involving the ventricles is common, there are no reports of development of tense subdural and interhemispheric CSF collection, without significant ventriculomegaly, after posterior fossa tumor resection, to date. We introduce the term “tension subdural hygroma” (tSH) to describe this rare, but distinct clinical, radiological and pathological phenomenon.

Illustrative Case:

14 month old male child was evaluated by the Pediatric Neurologist, for irritability, truncal ataxia and vomiting of 2–3 weeks duration. Magnetic Resonance Imaging (MRI) of brain showed a large midline posterior fossa tumor, the features of which were suggestive of medulloblastoma. There was associated HC with periventricular lucency (PVL) (Fig. 1a,b). Clinically the child was awake, alert, irritable and had truncal ataxia. Pupils were equal and reacting and fundi showed no papilledema. He underwent modified telo-velar approach and gross total resection of the tumor. An EVD was placed through a right sided
Frazier burr hole, immediately before the surgery [1]. Postoperative Computerized Tomography (CT) scan, after 24 hours, showed slight reduction in the ventriculomegaly. The EVD which was kept clamped post-operatively, was removed after the CT scan which showed clear tumor bed, reduced ventriculomegaly with PVL (Fig. 2a). Postoperative MRI brain and spine performed on the third day, showed no residual mass and the ventriculomegaly was less compared to the post-operative CT brain (Fig. 2b,c). The child developed a pseudomeningocele (PMC) at the surgical and EVD site, which was progressive over the next two weeks. Two attempts of drainage of the CSF via lumbar puncture (LP) on the 5th and 12th post-operative day, were futile. By the 14th day the PMC was tense (Fig. 3a) and CT brain showed very large hypodense collections at the surgical site, bilateral convexity and interhemispheric subdural space with stable ventriculomegaly (Fig. 3b-e). The child remained fully awake and alert, but irritable. He was afebrile and was tolerating breast feeds well. Analysis of CSF, obtained during drainage LP, did not show any evidence of infection. He underwent emergency CSF diversion using a medium pressure shunt system. Upon nicking the dura, to insert the ventricular catheter, the CSF, under high pressure, in the subdural space, jetted out. The catheter had to be advanced for 4 cm, in an attempt to enter the ventricle, to drain out CSF. Postoperatively the PMC completely resolved within a day and the child improved clinically. Postoperative CT scan of brain showed distal end of the catheter with holes, in the interhemispheric subdural space, stable ventriculomegaly with no PVL, complete resolution of the subdural collection and PMC, suggesting a functioning shunt system (Fig. 4). Though a VPS was planned, it turned out to be a subdural-peritoneal shunt (SPS) by default. Histopathology examination confirmed the diagnosis of medulloblastoma. 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), and shunt tube tip in the interhemispheric sudural space (Fig. 5).

**Discussion:**

In children with PFT, the factors which can lead to persistence of HC, after tumor resection, necessitating CSF diversion, include age less than 3 years, duration of illness than 3 months, midline location of tumor, 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, 10–14]. In our case, the child underwent right sided EVD, through a Frazier burr hole, immediately before resection of the tumor. Child developed PMC at the surgical and EVD site, in 14 days post-operatively, which was progressive, despite two attempts of drainage of CSF via LP. The MRI scan on third and CT scan on fourteenth post-operative days, immediately before the SPS, showed progression of PMC at the surgical site, bilaterally symmetrical hemispheric convexity and interhemispheric CSF collection, though the ventriculomegaly was significantly less, compared to the pre-operative images. The mechanism of development of post-operative SH is still debated. The possible explanation based on our case is as follows. The development of HC after the surgery and removal of EVD resulted initially in PMC formation. The egress of CSF from ventricle into the PMC stopped once the PMC attained maximum stretch capacity and pressure equalized in both the CSF compartments. The next possible way for the CSF to flow out of the expanding ventricles, was into the subdural space, through the iatrogenic
communication between the IV ventricle and subdural space in the posterior fossa and from there, via tentorial incisura, into the subdural space in the supratentorial region. Another possible way was the iatrogenic port, created by the EVD tube, in the right lateral ventricle, reopened by the high pressure in the expanding ventricles. Thus, there existed a dynamic communication between the ventricle, PMC and subdural space. The expanding PMC and subdural space accommodated the increasing volume of CSF, and contained the progression of ventriculomegaly. The CSF in the subdural space was under high pressure unlike in subdural hygroma (SH). 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 intraoperative image guidance could have avoided the malpositioning of ventricular catheter. The complete resolution of PMC and SH 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). Literature review showed three articles reporting development of SH following tumor resection, one being supratentorial and the other two being infratentorial tumors [15–17]. Eguchi et al reported three cases, all with tumor in the suprasellar, hypothalamic and third ventricular regions [15]. They called these as post-operative extra axial CSF collections. Anokha et al reported two cases with posterior fossa tumors, who developed post-operative SH (one being a intra axial metastatic cerebellar lesion and the other an intra fourth ventricular lesion) [16]. These collections were asymmetrical and did not resolve completely after VPS, unlike tSDH. Stavrinos et al reported another case who developed SH following excision of intra axial cerebellar mass [17]. The SH was asymmetrical and was successfully 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 the cortical sulci in the CT brain, excluded the possibility of EH.

**Conclusion:**

Ours is the third reported case of SH following posterior fossa tumor resection and stands distinct from the other two reported cases. The hallmark features which makes it distinct include, a tense PMC, images showing bilaterally symmetrical supratentorial, infratentorial, and interhemispheric SH, stable ventriculomegaly with no PVL, CSF in the subdural space under high pressure and complete resolution of the SH and PMC, after CSF diversion. 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.

**Statements And Declarations:**

*Authors contributions:*
Mahesh Krishna Pillai - Primary author; involved in surgical management of the child; explain the pathophysiology and coin the term Tension subdural hygroma

Rajeev Kariyattil - Secondary author; involved in surgical management of the child; edited the manuscript; explain the pathophysiology

Rajesh Chhabra - Reviewed and edited the manuscript

Vankatesh Govindaraju - Edited the manuscript

Koshy Kochummen - Prepared the figures

Ethical Declaration Statement: Ethical approval was waived by the ethics Committee of Sultan Qaboos University, in view of this being a retrospective case report, all the procedures performed were part of the international standard care and parent has signed the consent. The study was conducted in accordance with the declaration of Helsinki.

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References:


Figures
Figure 1

MRI brain, T1W, Gadolinium enhanced, axial (a), coronal (b) and sagittal (c) images, showing midline posterior fossa tumor with hydrocephalus

Figure 2
Post-operative CT scan axial section (a), 24 hours after clamping the external ventricular drain, showing dilated ventricles and ventricular catheter in situ (arrow); Post-operative MRI brain, axial T2W images (b-e), 72 hours after removal of external ventricular drain, showing no residual tumor (b), reduction in the ventriculomegaly and bilateral supra and infratentorial thin subdural hygroma (c).

Figure 3

Tense pseudomeningocele (a) at the external ventricular drain site (arrow down) and surgical site (arrow up), immediately before shunt surgery; shunt incision mark (arrow left). CT scan brain axial section (b) showing the pseudomeningocele (arrow) & c-e showing subdural hygroma and stable ventriculomegaly. Note the symmetrical distribution of hygroma all around the hemispheres.
Figure 4

One month post shunt CT scan of brain axial (a-d), coronal (e) and sagittal (f) sections showing complete resolution of the subdural hygroma in the supratentorial and near complete resolution in the infratentorial compartments; note shunt tube tip in the interhemispheric subdural space with stable ventriculomegaly
Figure 5

Six months post shunt MRI scan of the brain showing complete resolution of the subdural and interhemispheric collection and stable ventriculomegaly; axial sections (T2W images a-e; SWI image f) showing the shunt tube traversing medial to the right lateral ventricle & terminal end of shunt tube in the interhemispheric space (arrows)