



Case Report

Massive intratumoral hemorrhage following ventriculoperitoneal shunting procedure in an 8-year-old boy: A case report

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ABSTRACT

Background: Pineal region tumors comprise 0.4% of all central nervous system tumors in adults and 2.8% in children aged up to 19 years. Pineal gland tumors exhibit histological variability and necessitate a multidisciplinary approach. The first step in managing hydrocephalus is to treat it with either endoscopic third ventriculostomy (ETV) or a shunt surgery.

Case Description: We present a case of a child with a massive intratumoral hemorrhage located in the pineal region. An 8-year-old boy with a suspected germinoma in the pineal area accompanying noncommunicating hydrocephalus underwent a right ventriculoperitoneal shunt procedure. He experienced a sudden decline of consciousness following the procedure. The subsequent imaging showed an extensive hyperdense lesion within the brain ventricles, indicating intratumoral hemorrhage. Given that intratumoral hemorrhage in pineocytoma following the placement of ventriculoperitoneal shunt surgery is uncommon, it is a significant factor contributing to morbidity and mortality. It should be considered when cerebrospinal fluid (CSF) diversion is performed before a craniotomy.

Conclusion: A hypothesis suggests that changes in CSF flow after diverting CSF can be the main trigger for intratumoral hemorrhage although uncommon. In this study, we present our experience with our patients and provide a comprehensive evaluation of the existing literature.

Keywords: Germinoma, Intratumoral hemorrhage, Neurosurgery, Pediatric, Ventriculoperitoneal shunt

INTRODUCTION

Pineal region tumors comprise 0.4% of all central nervous system tumors in adults and 2.8% in children aged up to 19 years. Germ cell tumors (GCTs) constitute around 50–75% of all pineal tumors. They mostly develop in the pineal region and are more common in males. GCTs can be further classified as germinomas, which account for about 50–75% of cases.^[3]

Pineal gland tumors exhibit histological variability and necessitate a multidisciplinary approach. The intricate anatomical structure of the region poses a significant challenge to surgical procedures, and the efficacy of these procedures varies depending on the specific histological type. The first step in managing hydrocephalus is to treat it with either endoscopic third

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ventriculostomy (ETV) or a shunt surgery. In addition, a tissue diagnosis is obtained to guide further treatment.^[2]

In 2023, Cavalheiro *et al.* conducted a study which revealed that 92 patients (61%) underwent the placement of ventriculoperitoneal (VP) shunts, with a success rate of 72.5% at the 12-month mark. Patients with pineoblastomas showed the highest rate of dependence on these shunts. Out of the overall number of subjects, thirteen individuals experienced surgical complications, with nine instances of intracranial hemorrhage reported. It is important to note that Cavalheiro *et al.* did not specify whether these hemorrhages were located within the ventricles or the tumor. Furthermore, the study indicated that there were no such complications following ETV. This distinction emphasizes the potential differences in complication profiles between VP shunts and ETV, highlighting the need for careful consideration of shunt-related risks.^[1]

The occurrence of extensive bleeding within the tumor after cerebrospinal fluid (CSF) diversion for pineocytoma with obstructive hydrocephalus is uncommon. In this study, we present our experience with our patients and provide a comprehensive evaluation of the existing literature.

CASE DESCRIPTION

A previously healthy 8-year-old boy was admitted with complaints of seizures that began 4 days before admission. The seizures manifested as stiff convulsions throughout his body, lasting approximately 2 min. During the seizures, the patient was unconscious and subsequently became drowsy afterwards. Initially, he experienced one seizure per day; however, in the past 2 days, the frequency increased to 3–4 seizures daily. The patient also reported recurring headaches for 2 weeks before admission without any associated nausea or vomiting. According to his family, the headaches intensified significantly over the past 5 days, and they observed that his eyes had begun to squint.

A neurological examination revealed no significant abnormalities except for decreased light reflexes in both eyes. A computed tomography (CT) scan showed an isodense lesion in the pineal region, accompanied by noncommunicating hydrocephalus [Figure 1], with an initial intracranial pressure (ICP) of 25 cm H₂O. To address the hydrocephalus, a VP shunt procedure was performed using a medium-pressure, burr hole-type shunt.

However, 1 day after the shunt procedure, the patient's neurological status deteriorated significantly, progressing to a deeply comatose state, and the shunt chamber collapsed. Immediate follow-up CT showed a massive hyperdense lesion indicating intratumoral hemorrhage affecting the lateral, third, and fourth ventricles, along with extensive perifocal

edema [Figure 2]. Neurologically, the patient's condition worsened, particularly with a decline in the Glasgow Coma Scale, resulting in a poor prognosis. Unfortunately, the patient was unable to receive chemotherapy or radiotherapy before passing away. Follow-up continued until day 22 postoperation, at which point the patient succumbed to his condition. Given the poor neurological state, decompressive surgery was not conducted, and the patient is currently receiving treatment in the intensive care unit.

DISCUSSION

There is a hypothesis suggesting that changes in CSF flow after diverting CSF can be the main trigger for intratumoral hemorrhage. The suggested pathophysiological mechanisms include that a sudden decrease in CSF pressure in one ventricular compartment leads to an increasing pressure difference throughout the tumor, causing gradual damage to delicate blood capillaries within the tumor. The migration of the tumor can cause additional compression of the venous drainage, resulting in venous hypertension, greater swelling of the tumor, and spontaneous bleeding inside the brain tissue. The continuous increase in ICP may have worsened the venous congestion, resulting in the occurrence of microhemorrhages, edema, venous infarction, and, ultimately, a massive hemorrhage.

Various processes have been suggested, such as the rapid reduction of ICP after the insertion of a ventriculoperitoneal VP shunt or CSF drainage. These methods result in an elevated pressure difference between the blood arteries within the tumor, leading to bleeding within the tumor itself. Another reason could be attributed to the abrupt reduction in CSF volume and pressure following a ventriculoperitoneal VP shunt procedure, which disrupts the balance between different compartments inside the skull. This disruption leads to an elevation in cerebral blood flow and vascular congestion. Due to the constant intracranial capacity, any sudden fall in CSF volume is counterbalanced by a reciprocal rise in cerebral blood volume, resulting in vascular congestion and intratumoral hemorrhage.^[5]

Previous reports have documented similar findings. Matsumoto *et al.* reported a case of pineocytoma with massive intratumoral hemorrhage following a VP shunt procedure, highlighting that the sudden decrease in CSF pressure may have contributed to hemorrhage within the tumor due to altered pressure dynamics.^[6] Harada *et al.* also described a similar case, where intratumoral hemorrhage occurred after a VP shunt, suggesting that the changes in ICP led to vascular congestion and increased risk of bleeding.^[4] These cases support the hypothesis that changes in CSF flow and pressure can disrupt the ICP equilibrium, leading to damage to fragile tumor vasculature and subsequent hemorrhage. Including

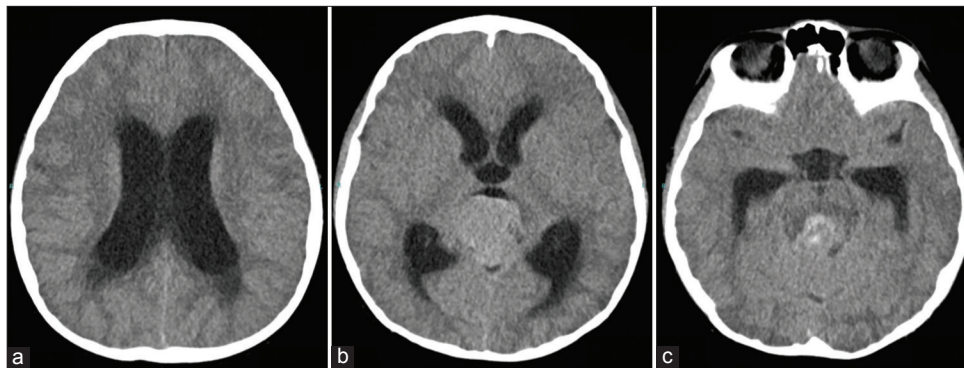


Figure 1: Preshunt computed tomography showed an isodense lesion in the pineal region with noncommunicating hydrocephalus, indicating a pineal region tumor. (a) View of lateral ventricle, (b) View of third ventricle, (c) view of temporal horn.

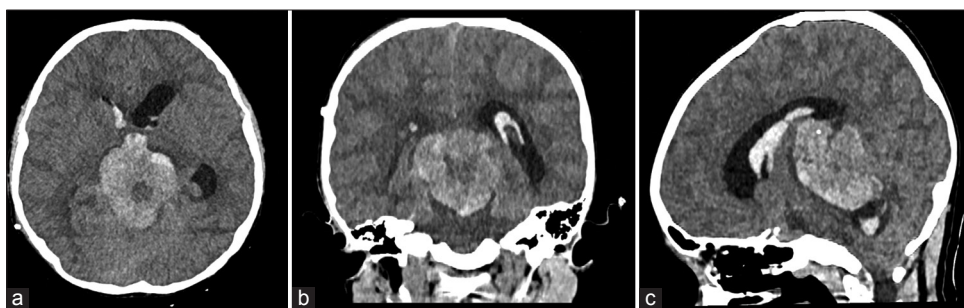


Figure 2: Postshunt computed tomography showed a massive hyperdense lesion indicating intratumoral hemorrhage in the lateral, third, and fourth ventricles with extensive perifocal edema. (a) Axial view, (b) Coronal view, (c) Sagittal view.

these studies enriches the discussion on the pathophysiology of intratumoral bleeding linked to CSF diversion procedures.

A consensus is growing that simultaneous ETV and endoscopic biopsy are becoming the treatment of choice for pineal region tumors. Although, as highlighted in the Cavalheiro series, this approach is not yet fully supported by strong evidence, and a large proportion of patients still receive VP shunts. The technique of simultaneous ETV and biopsy offers significant advantages, particularly in minimizing the risks associated with massive volume changes due to overdrainage, which may help prevent complications such as those observed in our cases. Cavalheiro *et al.* also emphasized that no such hemorrhagic complications occurred following ETV, suggesting that this approach could offer a safer alternative to VP shunt placement.^[1]

Recent literature further supports this approach. A systematic review and meta-analysis by Porto Sousa *et al.* provide a detailed overview of the outcomes associated with simultaneous ETV and biopsy in patients with pineal region tumors. The study reports favorable clinical outcomes and highlights the reduced necessity for CSF diversion procedures when ETV is performed alongside a biopsy.^[7] Including this option in our treatment discussion emphasizes the evolving

strategies and the importance of selecting the safest and most effective approach for managing these complex cases.

CONCLUSION

Based on this study, we conclude that the potential occurrence of intratumoral hemorrhage should be considered while planning the preoperative approach for obstructive hydrocephalus caused by pineal tumors. The occurrence of this significant complication, which is associated with morbidity, should be taken into mind and taken into account by the surgeon who is managing patients after CSF diversion for obstructive hydrocephalus in the setting of a pineal germinoma.

Author's contribution

RNW, WS, and MAP contributed to the design and implementation of the research, to the analysis of the results and the writing of the manuscript equally. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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