Int J Clin Exp Med 2016;9(10):20380-20382
www.ijcem.com /ISSN:1940-5901/IJCEM0031473

Case Report

Fatal intratumoral hemorrhage in pineal tumors following cerebrospinal fluid diversion

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Received April 29, 2016; Accepted August 6, 2016; Epub October 15, 2016; Published October 30, 2016

Abstract: We report two patients with massive intratumoral hemorrhage in pineal tumors. A 23-year-old male with pineocytoma, whose (MRI, CT) image demonstrated obstructive hydrocephalus, was subjected to a right ventriculoperitoneal shunt. His consciousness deteriorated, and he experienced massive intratumoral hemorrhage and later died. Another case of pineal tumor was a 47-year-old male who underwent emergency external ventricular drainage for associated obstructive hydrocephalus and deteriorating consciousness. He had a sudden deterioration following the procedure and imaging revealed massive intratumoral hemorrhage. He remained vegetative at discharge and died 2 weeks later. Although Intratumoral hemorrhage in pineocytoma after the placement of a ventriculoperitoneal shunt or external ventricular drainage is rare, it is an important cause of morbidity and mortality and should be kept in mind when cerebrospinal fluid (CSF) diversion was conducted before a craniotomy.

Keywords: Pineocytoma, intratumoral hemorrhage, ventriculoperitoneal shunt, external ventricular drainage

Introduction

Massive intratumoral bleeding following CSF diversion for pineocytoma with obstructive hydrocephalus is rare. We report our experience with two patients and review the literature.

Case reports

Patient 1

A previously healthy 23-year-old male presented with a 3-day history of progressive headache, vomiting and visual blurring. He experienced a worsening headache for 1 day before the hospital presentation. The neurological examination revealed no abnormalities except for diminution of vision and bilateral papilledema. CT showed an isodense lesion in the pineal region with obstructive hydrocephalus (**Figure 1A**). MRI revealed a pineal region tumor measuring 2.6 cm×3.1 cm×2.9 cm, with hypointense on T1-weighted images and slight hyperintense on T2-weighted images (**Figure 1B**). The initial pressure was 39 cmH_2O. A ventriculoperitoneal shunting procedure with a Delta shunt kit (adult, regular, performance level 1.5) was performed (Delta shunt kit manufactured by Medtronic Inc., Minneapolis, U.S.A.) to alleviate the patient's hydrocephalus. 5 hours after the shunt procedure, the patient deteriorated to deeply comatose state with absent brainstem reflexes. Immediate CT showed a massive intratumoral hemorrhage in the third ventricle which had ruptured into the lateral ventricles (**Figure 1C**). As he was in poor neurological state, decompressive surgery was not conducted and he died later.

Patient 2

A 47-year-old male was admitted after presenting with severe headache, vomiting over the course of 3 days on examination, he was drowsy and had bilateral papilledema. CT of chest showed a right lung cancer with mediastinum and right hilus pulmonis lymphatic metastasis. NSE was 42.38 ng/mL↑ (normal range 0-16.3 ng/mL), CEA and SCC were normal. Brain MRI with and without gadolinium administration revealed a pineal tumor with obstructive hydrocephalus. The tumor was hypointense on T1-weighted images and was slight hyperin-

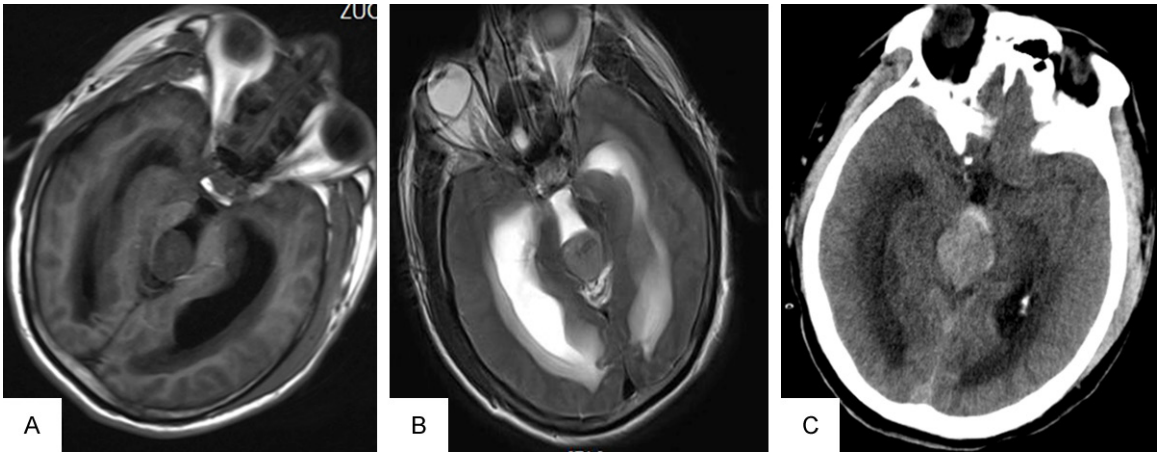

Figure 1. CT and transverse T2-weighted MRI revealed a pineal region tumor with obstructive hydrocephalus (A and B). CT showed a massive intratumoral hemorrhage in the third ventricle which had ruptured into the lateral ventricles (C).

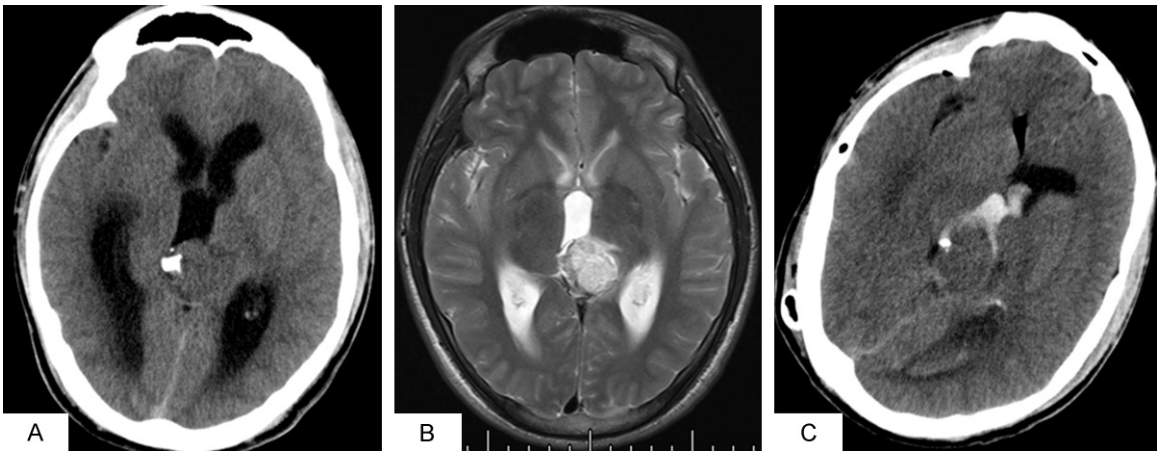

Figure 2. Transverse T1-weighted images and T2-weighted images showed a pineal region tumor with obstructive hydrocephalus (A and B). CT scan revealed a massive intratumoral hemorrhage (C).

tense on T2-weighted images and enhanced heterogeneously (**Figure 2A** and **2B**). He underwent medical treatment with mannitol and was admitted for tumor resection. Due to progressive decline in consciousness level, an emergency CSF diversion was planned and he underwent right external ventricular drainage. Intraoperatively, the CSF was under high pressure. Minimal CSF was allowed to drain intraoperatively. However, two hours after external ventricular drainage, he collapsed into deep coma with pupils inequality. Urgent brain CT scan revealed a massive intratumoral hemorrhage in the third ventricle which had ruptured into the lateral ventricles (**Figure 2C**). Families chose to give up any treatment and discharge from hospital.

Discussion

Pineal tumors are rare primary brain tumor (incidence 0.4% to 3.4%) [1] that commonly present with local mass effect or obstructive hydrocephalus [2]. The treatment of hydrocephalus includes external ventricular drainage and ventriculoperitoneal shunt. Intratumoral hemorrhage in pineal tumors is a rarely reported complications following treatment of obstructive hydrocephalus by ventriculoperitoneal shunt or external ventricular drainage. To date only four cases of pineal tumors are reported to have bled massively following CSF diversion [3-5]. Bleeding into other tumors following CSF diversion includes medulloblastoma, ependymoma, cerebellar astrocytoma, thalamic [3, 6, 7].

Different mechanisms are postulated to cause intratumoral hemorrhages after CSF diversion. At the high ICP values, the compliance of the brain decreased, hence, ICP decreased markedly even we released small amounts of CSF intra-operatively. This may cause minimal but rapid motion that distorts intracranial structures and creates a sudden imbalance between intracranial and intratumoral pressures [5]. This imbalance can lead to vascular insufficiency, venous congestion, and subsequent hemorrhage within the tumor [3]. As we know, the intracranial volume is constant. Any rapid decrease in CSF volume is compensated with increase in the cerebral blood volume reciprocally. Vascular congestion leads to intratumoral hemorrhage [4].

The ventricles in our patients were considerably smaller after CSF diversion was conducted. We believe that the tumor bleeding in our case was the result of the sudden decrease of the elevated ICP during CSF diversion procedure. After CSF diversion and the ventricles were reduced in size, the tumors shifted away from the surrounding structures. This shift may have altered the venous circulation in the tumors, causing the intratumoral hematoma [8].

An extremely high mortality of tumor hemorrhage has been shown in earlier reports [3-5, 9, 10]. In our patients the hemorrhage also resulted in subsequent death despite intensive care. Although releasing CSF gradually during the operative procedure the bleeding could not be prevented. In certain cases presenting with high ICP due to tumor-induced hydrocephalus it might be better to restore patency of the obstructed CSF pathways by subsequent tumor removal.

On the basis of this experience, we conclude that the possibility of intratumoral hemorrhage should be taken into consideration when planning the preoperative management of obstructive hydrocephalus caused by pineal tumors. CSF diversion is potentially hazardous and should be restricted to that selected group of patients who are acutely ill from increased intracranial pressure that is refractory to temporizing pharmacological management.

Disclosure of conflict of interest

None.

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