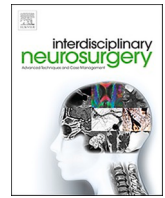




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Unexpected presentation of diabetes insipidus following pineal tumor resection: A case report

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ABSTRACT

Background: Neuroendocrine dysfunction is a common complication of several neurosurgical conditions. In particular, central diabetes insipidus (CDI) can occur subsequent to traumatic brain injury, subarachnoid hemorrhage, cerebral tumors or as a result of a complication following pituitary neurosurgery. In contrast, surgical resection of non-sellar tumors does not commonly result in CDI, with only a few cases reported in the literature. **Case description:** We report the case of a 40-year-old man who presented a transient CDI following surgical resection of a pineal papillary tumor via an occipital interhemispheric trans-tentorial approach. The underlying pathogenesis of CDI occurring post resection of tumors arising at a distance from the sella is not yet clearly understood, especially since there is no evidence of direct compression of the pituitary stalk. With regards to our case, we hypothesize that restauration of the initial obstructive hydrocephalus might induce a rapid intracranial pressure variation leading to hemodynamics changes of the portal hypophyseal vascular system. Postoperative air entrapment in the sellar region might also lead to irritation of the pituitary stalk. **Conclusion:** This case highlights that CDI may happen as a consequence of pineal tumor resection and advocates for a careful postoperative monitoring.

1. Introduction

Several neurosurgical conditions are complicated by neuroendocrine dysfunction [1]. In particular, central diabetes insipidus (CDI) can occur after traumatic brain injury and subarachnoid hemorrhage [2,3]. It may be present, before surgery, at the time of diagnosis of cerebral tumors such as craniopharyngioma or germinoma, or as complication following pituitary neurosurgery [2,3]. In contrast, surgical resection of non-sellar tumors does not commonly result in CDI, with only a few cases reported in the literature [2,4].

2. Case description

2.1. Clinical presentation

A 40-year-old man with no prior medical history was referred to our

neurosurgical outpatient clinic following diagnosis of a pineal tumor on magnetic resonance imaging (MRI) performed in the context of head and neck pain.

2.2. Diagnostic assessment

Neurological and ophthalmic examinations were normal, with no evidence of papillary edema on fundoscopy. Blood tests were normal, including alpha-foetoprotein and beta-hCG.

Brain MRI revealed a unifocal pineal mass, with solid and cystic components and strong enhancement after gadolinium injection (Fig. 1 A, B and C). The lesion was responsible for obstructive hydrocephalus with transependymal edema (Fig. 1 D). A complementary MRI of the whole spine was normal.

Abbreviations: CDI, Central Diabetes Insipidus; MRI, Magnetic Resonance Imaging.

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2.3. Management

He underwent an elective subtotal surgical resection via an occipital interhemispheric trans-tentorial approach. There was no complication during the procedure.

2.4. Outcomes and follow-up

Histopathological analysis revealed a papillary tumor. Postoperatively, the patient only complained of a slight visual blurring without any visual field deficit.

Postoperative brain CT performed at day 1 showed slight diffuse pneumocephaly, with an intrasellar air bubble against the posterior aspect of the pituitary stalk (Fig. 2 A). Brain MRI performed on day 3 showed a subtotal resection of the tumor and attenuation of hydrocephalus, with resolution of the transependymal edema (Fig. 2 B and C).

On the second and third day after tumor resection, the patient unexpectedly presented with hypotonic polyuria and hypernatremia, features leading to the diagnosis of CDI (Table 1).

He was successfully treated with desmopressin for two consecutive days. On the fourth postoperative day, CDI resolved. Anterior hypopituitary function was preserved.

One month after surgery, the patient presented signs of intracranial hypertension, revealing an acute relapse of his hydrocephalus. A ventriculostomy was therefore performed. Thereafter, the patient remained stable, with no recurrence of CDI during the second hospitalization and subsequent 12 months ambulatory follow-up period.

3. Discussion

CDI is a well-known complication of the surgical management of intra and parasellar tumors. In contrast, surgical resection of non-sellar tumors does not commonly result in CDI.

Some authors have described pituitary dysfunction following surgical resection of benign or malignant tumors that do not originate from the pituitary [2,4,5]. In their retrospective study, Schneider et al. [2] examined the endocrine function in 68 patients with non-pituitary intracranial tumors, some of them located in the posterior fossa. Persistent diabetes insipidus occurred in three cases. Not surprisingly diabetes insipidus was only present in patients harboring tumors located close to the sellar region including a dysgerminoma, a tuberculoma sellae transitional meningioma and a pilocytic astrocytoma of the third ventricle [2]. All of these patients also developed a panhypopituitarism which could suggest that they underwent more radical surgery responsible for multiple endocrine deficits [2]. In their prospective study on 51 patients with neurosurgical intracranial procedures excluding pituitary tumors, Fleck [4] reported two cases of CDI early after surgery, both of which resolved at long-term follow-up. However, no data on the histological nature of the tumor or its localization were available.

We report a case of a patient with a pineal tumor who developed CDI in the early postoperative course, which resolved rapidly. Tumors in the pineal region are rare, representing less than 1% of all intracranial tumors in adults [6]. If some cases of pre-operative CDI concomitant to pineal tumor have been described, they mainly concern bifocal germinomas invading the hypothalamic-hypophyseal tract on MRI or the floor of the third ventricle on endoscopic inspection [7].

In our case, a pineal papillary tumor was diagnosed. Headache and signs of intracranial hypertension due to obstructive hydrocephalus are the most common manifestations of these tumors [6]. To our knowledge, occurrence of CDI during the preoperative evaluation of pineal papillary tumor or after its surgical resection has not been previously described.

Several mechanisms leading to hypothalamic and pituitary dysfunction after traumatic brain injury or subarachnoid hemorrhage have been proposed. The pituitary gland or the hypothalamic-hypophyseal tract can be affected by different mechanisms following direct mechanical injury or hypoperfusion leading to ischemia and/or infarction [1]. The hypothesis of vascular dysfunction due to intracranial hypertension has also been considered in the context of hormonal insufficiency following non-pituitary tumor resection [5]. However, the underlying pathogenesis of CDI occurring post resection of tumors arising at a distance from the sella is not clearly identified, especially since there is no evidence of direct compression of the pituitary stalk, nor by the tumor, nor by the surgeon.

In our case, a possible explanation could be linked to the irritation of the pituitary stalk due to air entrapment in the sellar region, favored by the prolonged supine positioning of the patient during surgery. In fact, a pneumocephalus localized in the sella was identified on postoperative CT imaging.

Nevertheless, CDI is an extremely rare complication of papillary pineal tumor resection. Systematically reporting these cases can help us to better understand the underlying mechanisms responsible for the onset of central diabetes insipidus.

4. Conclusion

Central diabetes insipidus is a common complication following pituitary surgery, brain trauma or subarachnoid hemorrhage. Our case report shows that CDI may also rarely happen as a consequence of other brain surgery, such as pineal tumor resection. Careful clinical and biological postoperative monitoring remains essential to correctly diagnose and manage CDI following neurosurgery.

Disclosure

This study obtained written consent from the patient and approval from the local ethics committee (reference: P2020/619).

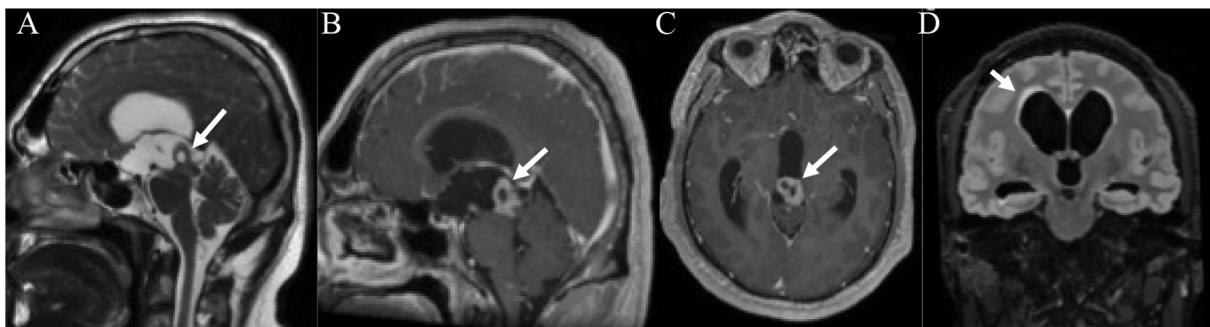


Fig. 1. Preoperative MRI: sagittal T2-weighted (A), sagittal (B) and axial (C) and post-gadolinium T1-weighted images showing a strongly enhancing pineal mass composed of solid (white arrow) and cystic components. Coronal (D) FLAIR weighted-image demonstrating an obstructive hydrocephalus with transependymal edema (white arrow).

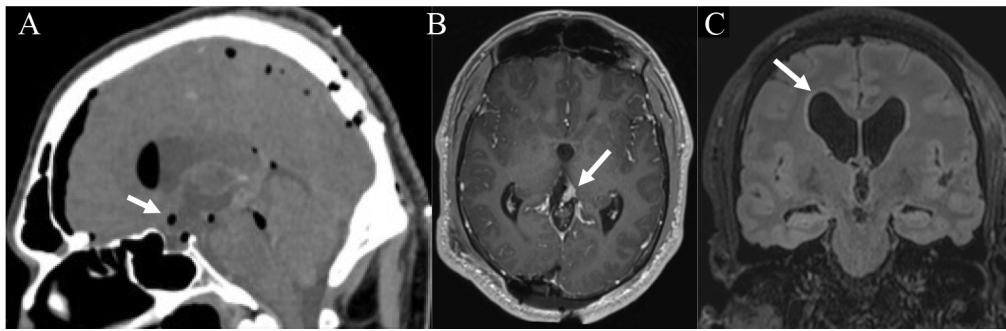


Fig. 2. (A) Postoperative CT showing diffuse pneumocephaly with air entrapment around the posterior aspect of the pituitary stalk (*white arrow*). Axial (B) post-gadolinium T1-weighted postoperative MRI demonstrating a subtotal resection (*white arrow*). Coronal FLAIR weighted-image (C) illustrating regression of the hydrocephalus and resolution of the transependymal edema (*white arrow*).

Table 1

Postoperative biological data.

| Time after surgery | Day 2 | Day 3 | Day 4 | 1 Month |
|---|-----------|-----------|-----------|---------|
| Plasmatic sodium in mmol/L (range 136–145) | a.m.: 148 | a.m.: 140 | a.m.: 143 | 143 |
| | p.m.: 145 | p.m.: 142 | p.m.: 139 | |
| Urinary osmolality in mOsmol/kg | | 118 | | 538 |

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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