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Exophytic bulbar pilocytic astrocytoma and post-operative cerebral salt wasting syndrome

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ABSTRACT

Cerebral salt wasting syndrome (CSWS) is a well-described consequence of several neurological disorders. Although the exact etiology of CSWS is still not completely elucidated, it is believed that the hypothalamus plays a pivotal role in the genesis of this disorder. We report for the first time 3 cases of CSWS occurring during the post-operative course following surgical resection of exophytic bulbar pilocytic astrocytomas in children. Since these 3 cases shared in common a medial implication of the medulla, we suggest that specific interconnectivity between the dorso-medial portion of the medulla oblongata and the hypothalamus might thus represent an anatomical pathway of interest in the pathogenesis of CSWS. Our findings suggest that the resection of medially located exophytic bulbar tumors might constitutes a risk factor in the development of CSWS. Particular care should thus be carried towards the prompt detection and treatment of CSWS in the post-operative courses of exophytic bulbar tumors.

Introduction

Cerebral salt wasting syndrome (CSWS) is characterized by renal sodium loss due to an intracranial disorder that leads to hyponatremia and decreased extracellular fluid volume. Several distinct neurological conditions may lead to CSWS, such as subarachnoid hemorrhage (SAH) and cranio-cerebral trauma [1,2]. Prompt diagnosis and treatment of CSWS in the post-operative setting are of particular importance, as hyponatremia could have deleterious consequences in an already ill neurosurgical patient. Brain swelling due to hyponatremia can indeed precipitate cerebral herniation and lead to dramatic neurological deterioration [3,4].

The exact pathogenesis of CSWS is still not fully understood. Natriuretic factors, such as atrial natriuretic peptide (ANP) and brain natriuretic peptide (BNP), may play an important role in the genesis of this syndrome [5]. Several studies have shown an increase in the levels of these natriuretic peptides in patients with SAH, as well as in patients after traumatic brain injury who developed CSWS [6,7]. Similarly, the levels of ANP and BNP can be increased in patients presenting with CSWS in the postoperative course of craniosynostosis surgery [8].

The underlying mechanisms leading to an increase in natriuretic peptide levels in CSWS remain controversial. An increase in the cardiac production of these factors mediated by a sympathetic discharge has been proposed [9]. BNP may also be released by a dysregulated hypothalamus, particularly in conditions such as SAH and brain trauma [10,11]. In addition, CSWS with elevated levels of BNP has been observed in a patient with a cystic mass in the suprasellar region, as well as in other patients following pituitary surgery [12,13]. Thus, the hypothalamic region seems to be central in the pathogenesis of CSWS through the release of natriuretic factors.

We report for the first time the occurrence of CSWS following the surgical resection of exophytic bulbar pilocytic astrocytomas in three children and suggest an original hypothesis explaining this occurrence.

Hypothesis

Our hypothesis supports that connectivity between the central part of the medulla and the hypothalamus could lead to CSWS following surgical manipulations of this region.

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Case 1

An 18-month-old boy presented with failure to thrive since the age of 6 months. The child also had recurrent vomiting and dysphagia since the age of 12 months. On clinical examination, a right Babinski sign was present. Bloodwork results showed a normal natremia on presentation. A dorsally exophytic bulbar tumor was diagnosed on MRI (Figs. 1 and 2A). Subtotal resection of the tumor by a posterior fossa craniotomy was performed. Histopathological examination was consistent with a pilocytic astrocytoma. The child was re-intubated at day 1 post-operation following a convulsive seizure due to marked hyponatremia (sodium plasma level 124 mmol/L). Clinical and laboratory findings were consistent with the diagnosis of salt wasting syndrome (Table 1). An early post-operative MRI showed the extent of tumor resection without any local complication (Figs. 1 and 2B). The CSWS was treated by hydro-electrolytic replacement with resolution of the syndrome within 12 h. The patient was extubated at post-operative day 2 post and did not present any further seizures or other complications. 2 years follow-up reveals a radiological asymptomatic progression of a tumor residue which is closely followed by imaging.

Case 2

A 7-month-old girl presented with gastroesophageal reflux and failure to thrive. Sodium levels were within normal limits on admission. A dorsally exophytic bulbar tumor, very similar to the tumor observed in case 1, was diagnosed on MRI (Figs. 1 and 2C). Subtotal resection of the tumor by a posterior fossa craniotomy was performed. Histopathological examination revealed a pilocytic astrocytoma. Similarly to our first case, a salt wasting syndrome was diagnosed at day 1 post-operation (sodium plasma level 126 mmol/L) (Table 1). An early post-operative MRI showed the extent of tumor resection without any local complication (Figs. 1 and 2D). Hydro-electrolytic replacement was undertaken and the condition showed resolution within 24 h. The child did not have any other complications. During follow-up 18 months later, she presented a radiological asymptomatic progression of her tumor residue (data not shown) which was treated by chemotherapy, with subsequent tumor regression.

Case 3

A 4-year-old boy had the incidental finding of a right laterally exophytic bulbar tumor on an MRI performed for epileptic seizures due to peri-natal ischemic frontal lesions (Fig. 3A). Peri-tumoral edema extending beyond the midline of the medulla oblongata was observed on T_2-weighted MRI sequences (Fig. 3A). Subtotal resection of the tumor was performed via a right subtonsillar approach and histopathological examination revealed a pilocytic astrocytoma. Similarly to our first two cases, a profound hyponatremia was diagnosed at day 1 post-operation (sodium plasma level 121 mmol/L). Clinical and laboratory findings were consistent with the diagnosis of salt wasting syndrome (Table 1), although a combined syndrome of inappropriate antidiuretic hormone secretion (SIADH) could not be excluded, as the condition showed resolution in 24 h with electrolytic replacement and liquid restriction. The child did not have any further complications. 4 years follow-up revealed a small tumor residue on the right posterior part of the medulla (Fig. 3B), which has shown a slight regression over time.

Discussion

We report three cases of exophytic bulbar pilocytic astrocytomas in young children who underwent surgery for tumor

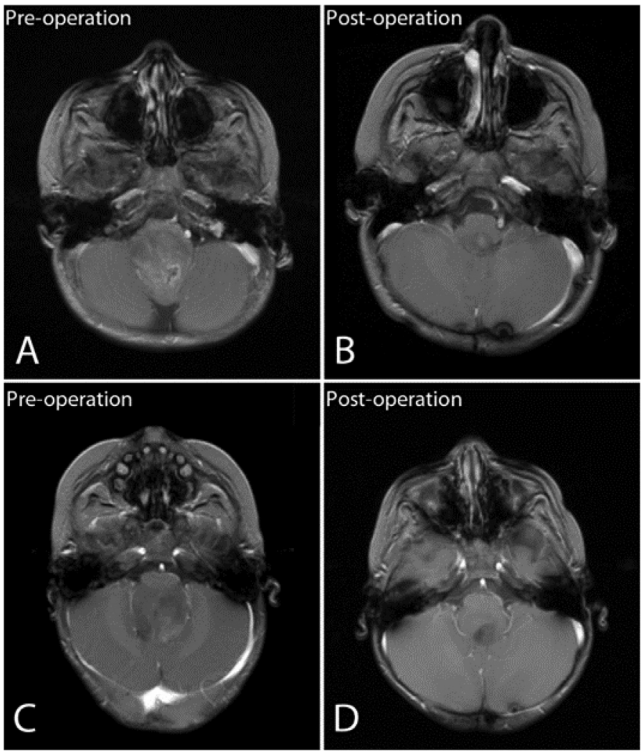

Fig. 1. Pre- and post-operative head axial injected T_1-weighted MRI sequences of cases 1 and 2 with dorsally exophytic bulbar tumors. (A) A sequence in case 1 showing a dorsally exophytic bulbar tumor with partial enhancement. (B) An early post-operative sequence in case 1 showing a small tumor residue and no local complication. (C) A sequence in case 2 showing a dorsally exophytic bulbar tumor with partial enhancement. (D) An early post-operative sequence in case 2 showing a small hypo-intense non-enhancing tumor residue on the right dorsal part of the bulba without any local complication.

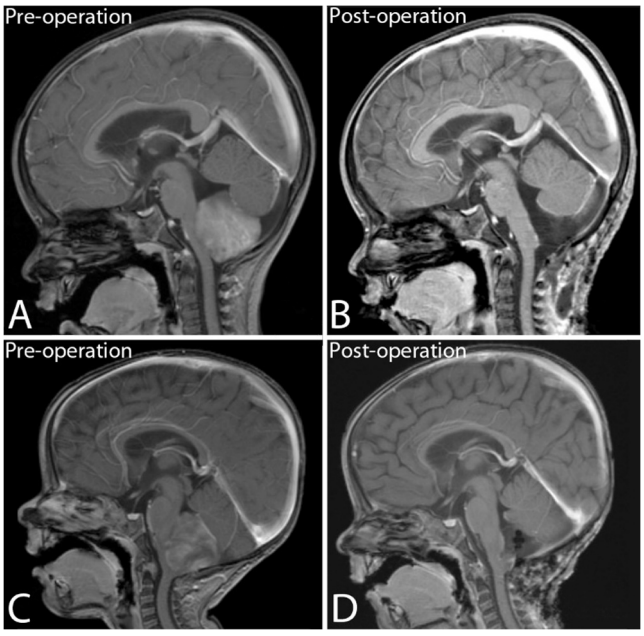

Fig. 2. Pre- and post-operative head sagittal injected T_1-weighted MRI sequences in cases 1 and 2 with dorsally exophytic bulbar tumors. (A) A sequence in case 1 showing the caudo-rostral extent of the tumor. (B) An early post-operative sequence in case 1 showing a very small enhancing tumor residue. (C) A sequence in case 2 showing a very similar lesion as compared to (A). (D) An early post-operative sequence in case 2 showing a small tumor residue on the dorsal part of the bulba.