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Case Report

Atypical spinal endodermal cyst presenting with contralateral C2 neuralgia and aseptic meningitis

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ARTICLE INFO

Article history:

Received 4 January 2019

Revised 14 February 2019

Accepted 14 February 2019

Available online 27 February 2019

Keywords:

Endodermal cyst

Contralateral occipitalgia

Aseptic meningitis

ABSTRACT

A previously healthy 43-year-old man presented with durable occipitalgia for 1 month. Neurological examination revealed severe pain in the right C2 area accompanied by neck stiffness. Magnetic resonance imaging revealed an enhancing, polycystic intradural mass at the C2 level, occupying the left dorsolateral part of the spinal canal. In addition, a rim-like enhancement was found along the surfaces of the spinal cord. Blood tests did not show signs of infection. A lumbar spinal tap revealed albuminocytologic dissociation without cultured organisms. Cranial computed tomography scans taken 20 days later revealed an overt ventriculomegaly. The patient underwent a total tumor resection through hemilaminectomy of the C2. Intraoperatively, the left dorsal C2 roots were found to be extremely redundant due to the tumor and surrounding thickened arachnoids. The roots restored normal morphologies after resection of the arachnoids and tumor. Postoperatively, the patient's symptoms resolved and histological diagnosis was endodermal cyst. Four weeks later, a ventriculoperitoneal shunt was placed to treat progressive ventriculomegaly. A polycystic intradural mass of the upper cervical spine should assume an endodermal cyst that may cause contralateral occipitalgia and aseptic meningitis.

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Introduction

Endodermal cyst (EC) is an infrequent entity comprising 3.4% of cystic lesions in the central nervous system [1] or 0.5% of intraspinal cysts [2]. ECs are thought to originate from the failed

separation of the neurenteric canal in the third week of gestation and are commonly identified in the ventral aspect of the lower brainstem and spinal cord. Headache, motor weakness, and neck pain are common symptoms [3–5], while aseptic meningitis is an infrequent manifestation of ECs [6]. On neuroimages, lobulated or cystic intradural extramedullary mass

Acknowledgment: This work was not supported by a grant.

Conflict of interest: The authors have no conflicts of interest to declare regarding the materials and methods or the findings specified in this paper.

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<https://doi.org/10.1016/j.radcr.2019.02.013>

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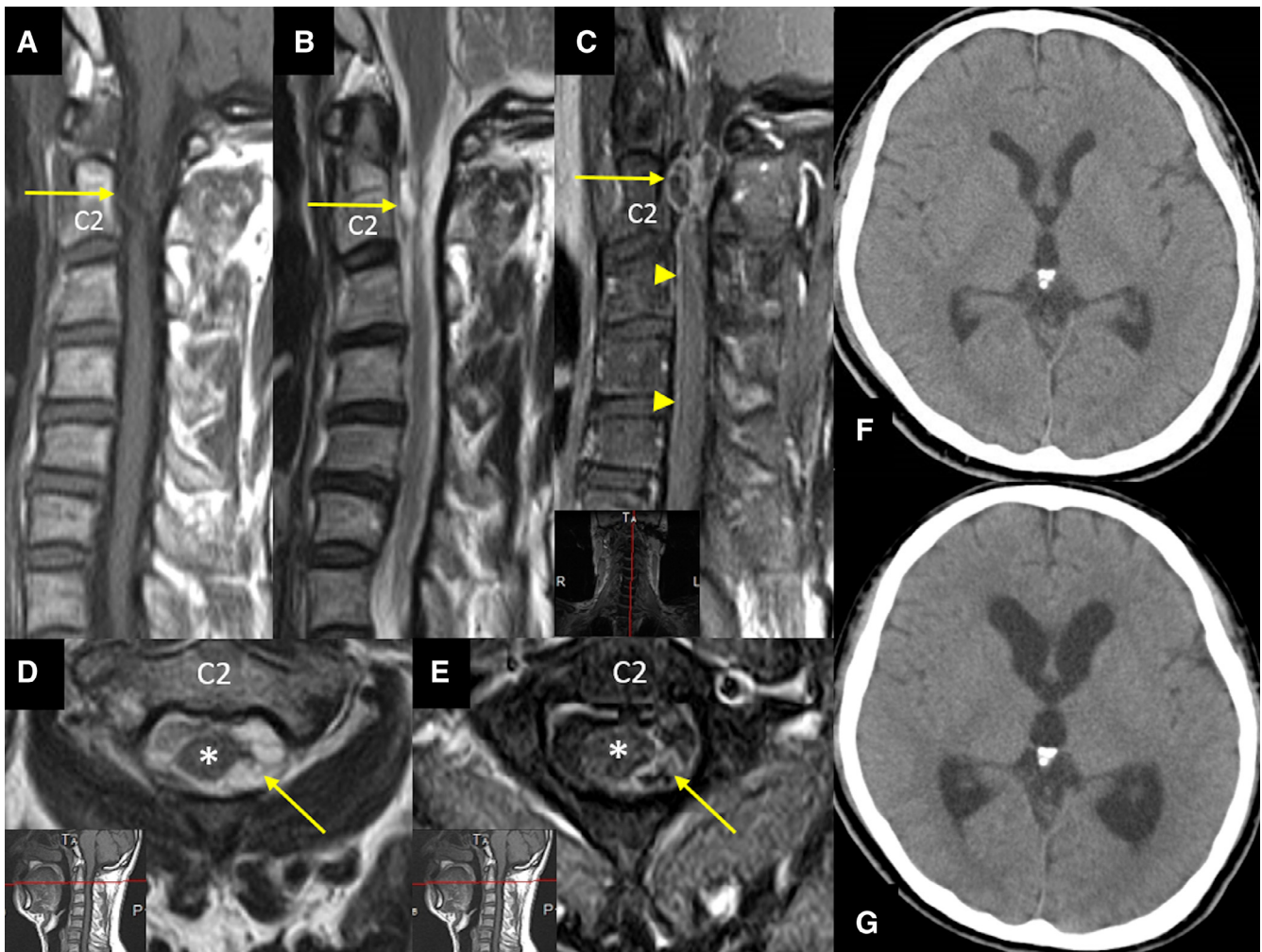


Fig. 1 – Sagittal T1- (A), sagittal and axial T2-weighted (B, D), postcontrast sagittal (C), and axial (E) magnetic resonance images showing a heterogeneously enhancing, polycystic intradural mass at C2, occupying the left dorsolateral part of the spinal canal (A-E, arrow). Rim-like enhancement is found along the surfaces of the spinal cord (C, arrowheads). Asterisk: cord. (F, G) Cranial computed tomography scans of the same level taken at presentation (F) and 20 days later showing the presence of ventriculomegaly (G).

is the characteristic appearance of ECs, especially in the presence of various types of vertebral anomalies [4]. Total resection, not a partial resection or biopsy, is the principle of treatment [3–6]. Here, we present a unique EC case of the upper cervical spine that presented with contralateral C2 neuralgia, aseptic meningitis, and hydrocephalus.

Case presentation

A 43-year-old, previously healthy man, sustained durable occipitalgia for 1 month. At presentation, the patient was afebrile. Neurological examination found severe pain in the right C2 area with marked neck stiffness. Magnetic resonance imaging (MRI) revealed a heterogeneously enhancing, polycystic intradural mass at the C2, occupying the left dorsolateral

part of the spinal canal. Rim-like enhancement was also found along the surfaces of the spinal cord (Fig. 1A-E). On computed tomography (CT) and MRI, vertebral anomalies were not identified, and blood tests showed normal findings without signs of infection. A lumbar spinal tap revealed albuminocytologic dissociation (Cell 22/ μ L, Protein 213 mg/dL) without any cultured organisms. Cranial CT scan taken 20 days later revealed asymptomatic tetraventricular dilation (Fig. 1F, G). The patient underwent tumor resection through hemilaminectomy of the C2. The tumor was located extra-axially in the subarachnoid space and firmly attached to the left dorsal C2 roots. The arachnoids surrounding the tumor showed a marked thickening. The right dorsal C2 roots were extremely tortuous and redundant due to the compression of the tumor and thickened arachnoids. A total resection was achieved for the tumor with surrounding arachnoids (Fig. 2). After resection, the C2 roots restored normal morphologies. The histological diagnosis was

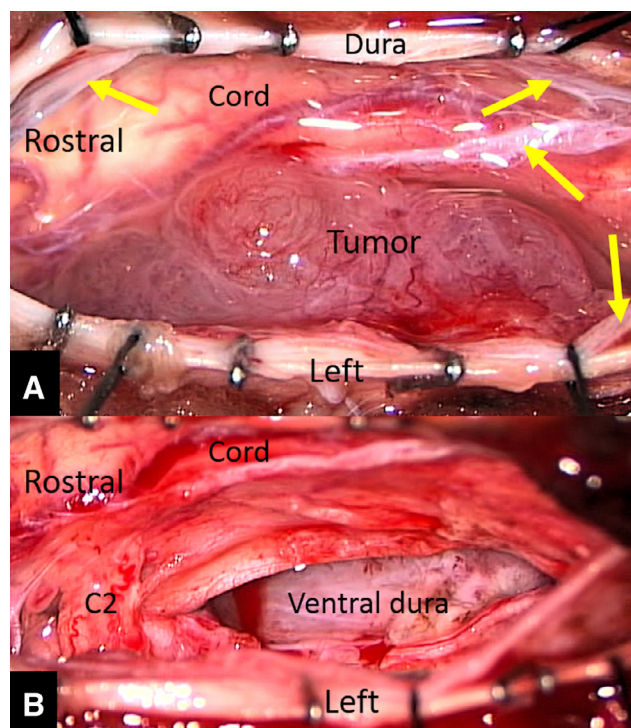


Fig. 2 – Intraoperative photos showing tumor after removal of the surrounding, thickened arachnoids (A) and after resection of the tumor (B). C2: left dorsal C2 roots; Arrows in (A): thickened arachnoids.

consistent with EC (Fig. 3). Postoperatively, the patient's occipitalgia resolved. However, the patient presented with progressive ventriculomegaly. He underwent a ventriculoperitoneal shunting 4 weeks after the initial surgery and was eventually discharged without neurological deficits.

Discussion

Aseptic, or chemical meningitis is an infrequent manifestation that can be caused by various cystic tumors such as colloid cyst, epidermoid or dermoid cyst, craniopharyngioma, and EC, spontaneously leaking cyst contents into the CNS [6–10].

Appearances on serial CT and MRI, intraoperative findings, and postoperative course of the present patient suggested that the aseptic meningitis and irreversible hydrocephalus were caused by the EC. Also, the patient's occipitalgia presenting on the contralateral side of the EC might be caused by the extremely distorted right C2 roots because the pain resolved after restoration of the normal morphologies that was yielded by resection of the tumor and surrounding arachnoids. A recent study suggested that trigeminal neuralgia with no neurovascular compression can be caused by thickened arachnoids that give rise to angulation or torsion of the trigeminal nerve root [11]. The clinical findings and

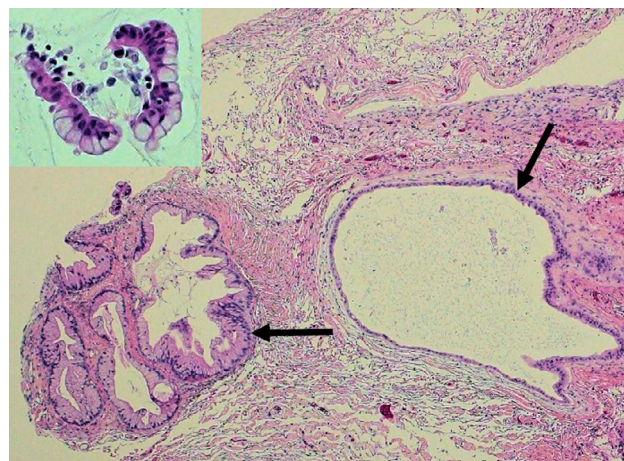


Fig. 3 – Photomicrograph of the resected tumor. Cystic components are lined by cuboidal and columnar cells (Arrows) and intervening fibrous connective tissues. The inset shows the magnified view of these cells with plump cytoplasm. Hematoxylin and eosin stain, $\times 40$.

neuroimaging appearance of the present case were indicative of spontaneous leakage of the cyst contents.

Conclusion

A polycystic intradural mass of the upper cervical spine should assume EC that can be a cause of contralateral occipitalgia and aseptic meningitis.

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