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Intralesional abscess in choroid plexus carcinoma. A case report

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ABSTRACT

Background: Choroid plexus carcinoma (CPC) is a rare, aggressive intraventricular tumour that predominantly affects young children. While CPC typically presents with hydrocephalus and mass effect, intratumoral abscess formation has not been previously documented.

Case Presentation: We report the case of a previously healthy 3-year-old female presenting with central facial hemiparesis and anisocoria. Imaging revealed a solid-cystic, contrast-enhancing lesion in the left ventricular atrium with spinal dissemination. Endoscopic-guided total resection via a parietal approach was performed, revealing a friable, haemorrhagic mass. Histopathology confirmed CPC with high proliferative index (Ki-67: 70%). Postoperative treatment followed the HEAD START III chemotherapy protocol without hematopoietic stem cell transplantation, followed by craniospinal radiotherapy. The patient remains recurrence-free after 12 months of follow-up.

Conclusion: This case highlights the successful management of a CPC complicated by an intratumoral abscess, an unprecedented presentation. Total surgical resection followed by multiagent chemotherapy and radiotherapy resulted in favourable early outcomes. Given CPC's rarity and variable presentation, individualised, multidisciplinary approaches are essential.

Keywords
intralesional abscess, choroid plexus carcinoma

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INTRODUCTION

Choroid plexus carcinoma (CPC) is a rare malignant neoplasm of neuroepithelial and primary of the central nervous system (1,2). The average age of diagnosis of CPC is 3 years and the annual incidence rate is 0.3 per million individuals (3,4). CPC has profound clinical and molecular differences compared to its lower-grade counterpart, choroid plexus papillomas (1). Tomography imaging typically reveals a large, hyperdense, contrast-enhancing intraventricular mass. Associated findings include hydrocephalus, calcifications, and/or hemorrhages. CPC is characterized histologically by friable papillary or cauliflower-like appearance, increased mitotic figures, pleomorphic nuclei, and necrosis. Intraoperatively, CPC may loosely or densely

adhere to the ventricular wall and minimally or extensively invade adjacent brain parenchyma (2). No abscessed lesions have been described so far, so in this text we report a pediatric case of CPC complicated with intratumoral abscess successfully treated with resection + systemic chemotherapy.

Case presentation

A previously healthy 3-year-old female patient presents with two months of central facial hemiparesis that progressed with anisocoria due to left mydriasis. A computed tomography (CT) scan of the skull showed a space-occupying lesion of a solid and cystic nature in the left ventricular atrium with extraventricular extension and discrete perilesional vasogenic edema. A brain Magnetic Resonance Imaging (MRI) contrast-enhanced with spectroscopy revealed a neoplastic-like lesion located within the left atrium, hinted at the occipital and temporal horn (Figure 1A). Spectroscopy showed a choline/NAA ratio of 1.31.

Extension workup through MRI of the cervical, thoracic and lumbosacral spine showed secondary lesions with contrast enhancement at the C5/C6, T1/T2 and L1 level, on the right side, that compromised the subarachnoid space and the medullary contour (Figure 2).

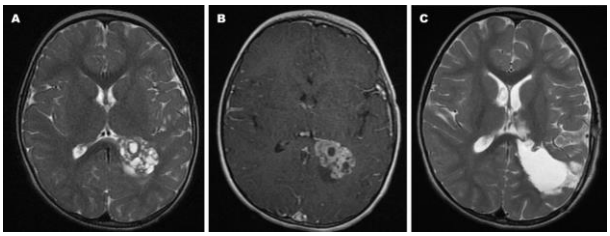

Figure 1. Baseline magnetic resonance of brain in axial T2-weighted (A) and contrast-enhanced (B), and postoperative T2-weighted imaging.

During the hospital stay, she presented vomiting and headaches and received symptomatic management. Endoscopic resection was performed through a parietal approach and guided by neuronavigation. During surgery, the tumor lesion had malignant appearance with a grayish, friable, profusely bleeding and easy to aspirate. A biopsy sample was taken and total macroscopic resection was achieved. There were no complications, with persistence of central facial hemiparesis and left mydriasis in the postoperative period (Figure 1B). Pathology and immunohistochemistry report evidenced a malignant tumor made up of small, ovoid and spindle-shaped cells, marked cytonuclear pleomorphism and mitotic activity. Positive tissue for Cytokeratin, S100, E-Cadherin, Vimentin and P53, with Ki-67 showing 70%, configuring the diagnosis of choroid plexus carcinoma.

Management was started with the HEAD START III protocol without consolidation therapy with autologous transplantation of hematopoietic precursors, there were no major complications during the chemotherapy cycles. Craniospinal radiotherapy and boost in the tumor bed were used as consolidation therapy. MRI of the brain and axis did not reveal neoplastic lesions with the respective post-surgical parenchymal defect. Patient continues in multidisciplinary follow-up 12 months after diagnosis, with no evidence of recurrence.

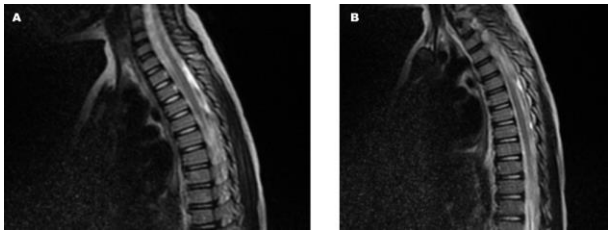

Figure 2. Baseline magnetic resonance of spinal cord showing diffuse lesions in upper (A) and lower (B) thoracic segments.

Discusion

CPC is a rare cause of a hemispheric brain tumor arising from the lateral ventricles in children. Possible differential diagnoses for a hemispheric brain tumor of this type include choroid plexus papilloma, ependymoma, atypical teratoid rhabdoid tumor, glioma, astrocytoma, and primitive neuroectodermal tumor (PNET). Radiopathological correlation with tissue immunohistochemistry is essential to differentiate and establish a confirmatory diagnosis. (5).

Due to the extremely low incidence of CPC, it is not possible to standardize medical conduct, so individualization based on case studies and clinical expertise determine the therapeutic vector in these patients. However, higher success rates have been reported in patients undergoing total resection with subsequent multiagent chemotherapy. (6). Radical surgery is the most important prognostic factor, although survival remains relatively low. (7). Despite the merits of surgical resection, due to the

macroscopic characteristics of the tumor (large size, high vascularity, diffuse infiltrative nature, and excessive friability), it is difficult to achieve complete resection, resulting in the necessity of a surgical resection. neoadjuvant therapy later. In the pediatric population, the risk of bleeding from surgical resection is considerable, as all circulating blood volume may be lost during resection of these highly vascularized tumors.

The planned surgical approach should allow good visual access to tumor blood flow and maximum exposure of the mass. An effective intraoperative surgical strategy is to identify and ligate the choroidal feeding vessel, which facilitates block-removal of the tumor mass. (5). McEvoy et al. reported a mortality rate attributable to intraoperative bleeding between 5 and 12%. In this report, four of nine surgeries for CPC in their series were suspended due to excessive bleeding, achieving only 33% of CPCs grossly resected completely. (8).

Due to this, it is recommended that if for some reason complete resection is not achieved, reoperation is a reliable option, which can be preceded by the administration of chemotherapy to reduce intraoperative bleeding and tumor size. (1), which will allow for a complete posterior resection rather than an incomplete resection (5). In order to reduce bleeding, for certain anatomical dispositions of the tumor it is feasible to submit the patient to preoperative embolization and thus achieve total gross resection. (9). Embolization also reduces the production of cerebrospinal fluid, the reduction of which is a goal of comprehensive CPC treatment. (10).

Like surgical resection, there is a lack of global consensus on chemotherapy and neoadjuvant regimens and it has not yet been standardized. The following drugs are used in treatment: carboplatin, etoposide, cyclophosphamide, high-dose methotrexate, and vinca alkaloids (1). The best chemotherapy regimen has yet to be determined, but a combination using platinum and etoposide as a base is preferred (11). Its combination with radiotherapy is also controversial due to evidence that it presents a greater benefit in chemotherapy alone (12). Radiotherapy is an important aspect of management, but its implementation is limited in young children (<2 years). Among a database of 524 patients, 5-year survival was better in irradiated CPC patients (13).

The effects of surgery and radiation therapy were evaluated in patients with or without TP53 germline mutations to provide vital information to help define neuro-oncology response assessment specific to pediatric patients with CPC. Li et al. showed an improvement in patients who received total tumor resection and avoided radiation therapy. CPC have been linked to germline TP53 mutations. In particular, patients with the TP53 germline mutation showed a significant survival advantage, demonstrating that radiation therapy should not be considered for patients who have the TP53 germline mutation (4). Despite these findings in the literature, it was decided to use radiotherapy in this patient due to the high risk of consolidating high doses of myeloablative chemotherapy and rescue with autologous transplantation of hematopoietic precursors.

Conclusion

CPC is a rare malignant neoplasm of neuroepithelial origin and primary central nervous system, there is still no consensus on the neoadjuvant alternative for the management of these neoplasms, on the other hand, robust evidence suggests that total resection achieves greater survival, although it continues being relatively low.

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