Mexican Journal of Oncology
Gaceta Mexicana de Oncología

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CLINICAL CASE

Thalamic infarct after surgical resection in craniopharyngioma. Case report

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Abstract

Craniopharyngioma (CP) is a rare benign brain tumor. Lipid thrombosis and thalamic infarction are uncommon complications following CP surgery. We report a rare autopsy case of CP that presented with difficult-to-control diabetes insipidus and thalamic infarction after surgery. Macroscopic examination of the brain revealed hemorrhagic infarcts in the thalamus and histologically showed necrosis and lipid droplets in the thalamic vessels and neurons. No tumor was observed. This case report highlights the possible occurrence of intracranial thalamic infarction due to surgery-induced vasculopathy following CP resection.

Keywords: *Craniopharyngioma. Surgical complications. Lipid thrombosis. Thalamic infarct. Case report.*

Infarto talámico después de la resección quirúrgica de un craneofaringioma. Reporte de caso

Resumen

El craneofaringioma es un tumor cerebral benigno poco frecuente. La trombosis lipídica y el infarto talámico son complicaciones raras posteriores a la cirugía de craneofaringioma. Reportamos un raro caso de autopsia de craneofaringioma que presentó diabetes insípida de difícil control e infarto talámico después de la cirugía. El examen macroscópico del cerebro mostró infartos hemorrágicos en el tálamo, histológicamente reveló necrosis y gotas de lípidos en los vasos y neuronas talámicas. No se observó tumor. Este reporte de caso ilustra la posible aparición de infarto talámico intracraneal debido a vasculopatía inducida por la cirugía tras la resección de craneofaringioma.

Palabras clave: *Craneofaringioma. Complicaciones quirúrgicas. Trombosis lipídica. Infarto talámico. Reporte de caso.*

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Date of reception: 24-09-2024
Date of acceptance: 04-12-2024
DOI: 10.24875/j.gamo.24000073

Available online: 13-08-2025
Gac Mex Oncol. 2025;24(Supl):35-40
www.gamo-smeo.com

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Introduction

Craniopharyngioma (CP) is a rare benign brain tumor that arises from remnants of the epithelium of the craniopharyngeal duct or odontogenic tissue[1], located in the sellar and parasellar region due to embryonic malformation[2,3]. The incidence of confirmed cases is 0.16/100,000 people. The age distribution is bimodal, with a peak between 5 and 9 years and another between 55 and 69 years[4]. The typical initial manifestations at the time of diagnosis are symptoms of elevated intracranial pressure, such as nausea and headaches; clinical features are non-specific and include visual impairment and endocrine disorders[5-8]. Diabetes insipidus (DI) is observed in up to 17% of children and up to 30% of adults[5]. Although histologically benign, these tumors are amenable to surgical treatment[9]. Complete or partial resection of CPs presents a challenge for neurosurgeons due to the anatomical location of the tumor, its proximity to optic structures (nerves and chiasm), the circle of Willis, the pituitary gland, and the hypothalamus, representing a potential negative risk associated with high morbidity and mortality[10,11]. Post-surgical quality of life is related to the specific anatomical relationship of each neoplasm and its proximity to the aforementioned structures[12-14]. In addition to the difficulties encountered with tumor resection, post-operative complications, such as endocrine disruption and extensive hypothalamic involvement should suggest a less aggressive approach[13,15,16].

We report a rare case of adamantinomatous CP that was treated surgically and histologically showed inflammation, clef cholesterol, and abundant dirty yellow macrophages secondary to the rupture of CP cystic structures. Subsequently, the patient developed significant metabolic disturbance and a thalamic infarction, and 3 days later, he died.

Clinical case

A 50-year-old male patient with a medical history of type 2 diabetes for 20 years and rheumatoid arthritis for 10 years, along with hyperuricemia and alcoholism for the past 4 years, presented with no other significant familial or personal medical history. His clinical condition worsened, displaying signs of intracranial hypertension during physical examination, including nausea, headaches, and altered consciousness with disorientation. In addition, the patient experienced visual hallucinations and reduced visual acuity. The neurological examination revealed no abnormalities. As a result, a magnetic resonance imaging (MRI) scan was performed, which revealed a suprasellar lesion measuring 2.1 cm in diameter (Fig. 1A). The diagnostic evaluation was not hindered and based on the MRI findings, a structural suprasellar lesion or neoplasm, such as CP, was suspected to be causing the clinical symptoms. To address the visual disturbances and intracranial pressure, the patient underwent neurosurgery, aiming for resection until healthy tissue was reached. An extrinsic lesion with a soft/gelatinous consistency was found in the third ventricle, with areas of microcalcification. The tumor resection was reported as an adamantinomatous CP, grade 1, according to the World Health Organization classification. In addition, a strong lymphoplasmacytic inflammatory response with polymorphonuclear cells was identified, along with the formation of cholesterol crystals and multinucleated giant cells (Fig. 1B). Numerous macrophages with a dirty yellowish cytoplasm (Fig. 1C) and several blood vessels, some containing crystallized yellowish material within them (Fig. 1D), were observed, showing vessel rupture with abundant foamy macrophages (Fig. 1E and F).

After surgery, the patient experienced significant hydroelectrolytic and metabolic imbalance, difficult-to-control DI, and fluid balance fluctuations with sodium levels ranging from 120 to 15 mEq, leading to respiratory deterioration and asystole. A follow-up computed tomography scan showed thalamic infarction (Fig. 2A), and he died with a probable diagnosis of pulmonary thrombosis. A partial autopsy was performed, limited to the cranial cavity, where the following findings were observed.

Results

Neuropathology the brain weighed 1500 g and showed herniation of the cerebellar tonsils, bilateral uncal and subfalcine herniation, significant edema, and an area of hemorrhage in the thalamus. Hemorrhage with acute inflammation was identified in the thalamus. Edema with necrosis was observed in the hypothalamus, and the neurons showed abundant hemosiderin pigment, which was noted in thick clumps (Fig. 2B). The presence of lipid droplets in the blood vessels is striking (Fig. 2C), along with the destruction of the neuropil and thickening of the axons (Fig. 2D). Reactive gliosis and perivascular cuffs were also observed (Fig. 2E), along with a diffuse lymphocytic infiltrate. Thalamic neurons showed accumulation of an amorphous material forming yellowish crystals or oil myelin figures (OMF) (Fig. 2F). We observed these same changes in the

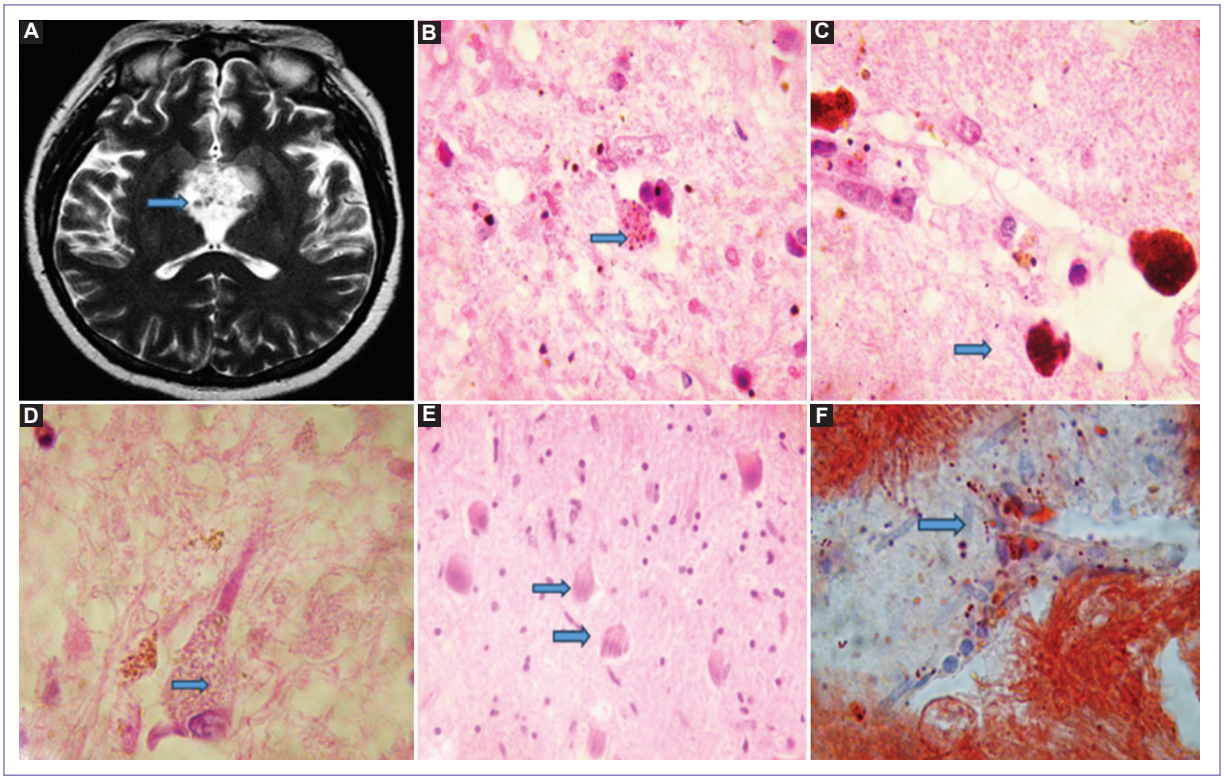

Figure 1. **A:** cerebral magnetic resonance imaging showed a suprasellar tumor. **B:** lymphoplasmacytic inflammatory response of polymorphonuclear leukocytes and multinucleated giant cells. **C:** abundant macrophages with yellowish cytoplasm. **D:** numerous blood vessels with yellowish crystallized material inside. **E** and **F:** rupture of blood vessels with abundant foamy macrophages.

neurons of the thalamic nuclei (supraoptic and paraventricular) as well as in the dentate nuclei of the cerebellum. Immunohistochemical staining (Fig. 3) for glial fibrillary acidic protein revealed reactive gliosis, astrocytes with intracytoplasmic vacuoles, diffuse axonal damage with myelin, and neurons and vessels exhibiting lipid vacuoles, which were more evident with oil red O staining in fresh tissue. Lipid vacuoles were also observed in the brain. GLUT1 was diffusely positive, and GLUT3 was faintly positive, as was the insulin-like growth factor (IGF) in some cells. Therefore, the diagnosis was lipid thrombosis with thalamic infarction and neuronal damage in the hypothalamus with lipid accumulation.

Discussion

The treatment of CP is not specific; present therapy almost always involves surgical intervention, either complete or partial, followed by radiotherapy[10,17]. The prognosis and quality of life improve with reduced invasion of the hypothalamic and pituitary structures[5,7,18]. Given the present conditions of our patient and CP in general, more demanding treatments and more controversial neurosurgical procedures are often recommended compared to conventional neoplasms. Our patient exemplifies the post-surgical complications that can arise from these procedures. Pre-operative central DI has been reported in 8-35% of CP patients, and in 70-90% after surgery[19,20]. The classic three-phase pattern of endogenous vasopressin secretion includes an initial phase of symptomatic DI occurring within 24 h post-surgery; a second phase of inappropriate vasopressin secretion that may lead to hyponatremia; and a third phase with a return to DI that can persist up to 2 weeks later, often complicated by cerebral salt wasting and disturbances in thirst regulation[21].

Furthermore, the mechanism of CP cyst rupture may involve cyst wall weakness induced by cyst expansion leading to degeneration[15]. In this case, a biopsy revealed an intense inflammatory response characterized by abundant yellowish foamy macrophages and crystallized proteinaceous material within the lumen of blood vessels, along with cholesterol crystals and multinucleated

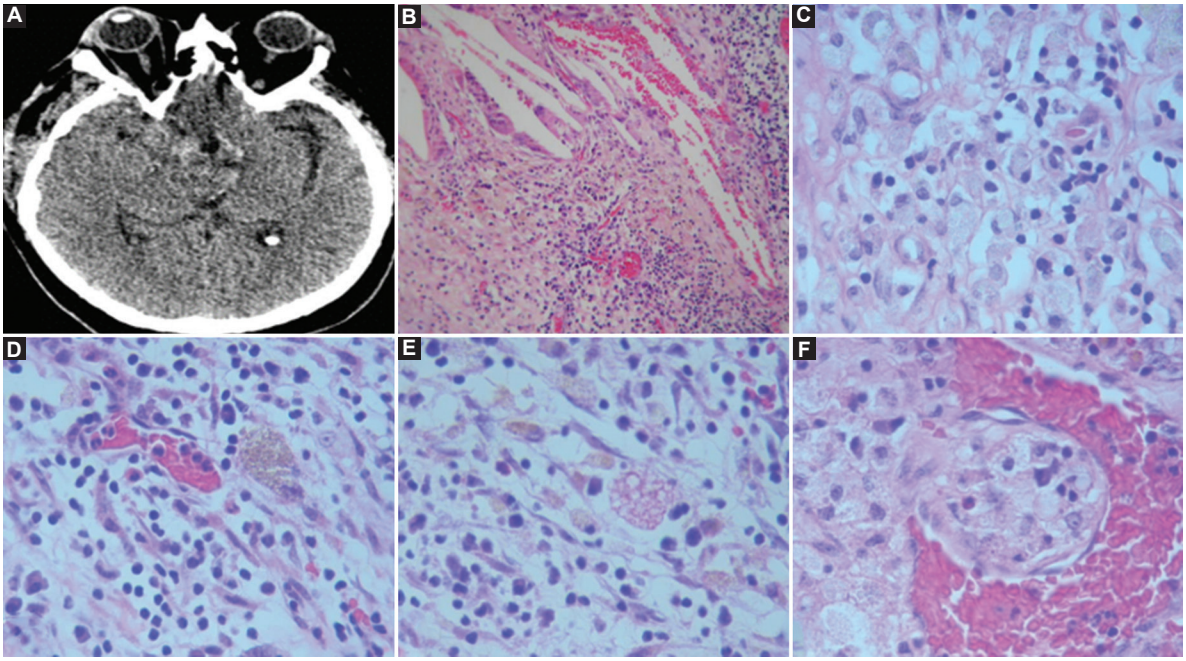

Figure 2. **A:** computed tomography showing thalamic infarction. **B:** histologically, a hemorrhagic thalamic infarction is shown, with soft, necrotic white matter and significant edema. **C:** necrosis with the presence of yellowish material forming variable crystals. **D:** Oil Myelin Figures (OMF) crystals in the neuropil, forming dark spots. **E** and **F:** OMF crystals in astrocytes, macrophages, and in the cytoplasm of some neurons (H and E × 400). The oil red O staining in fresh tissue reveals diffuse speckled oily material in the neuropil as well as in the vascular wall (×400).

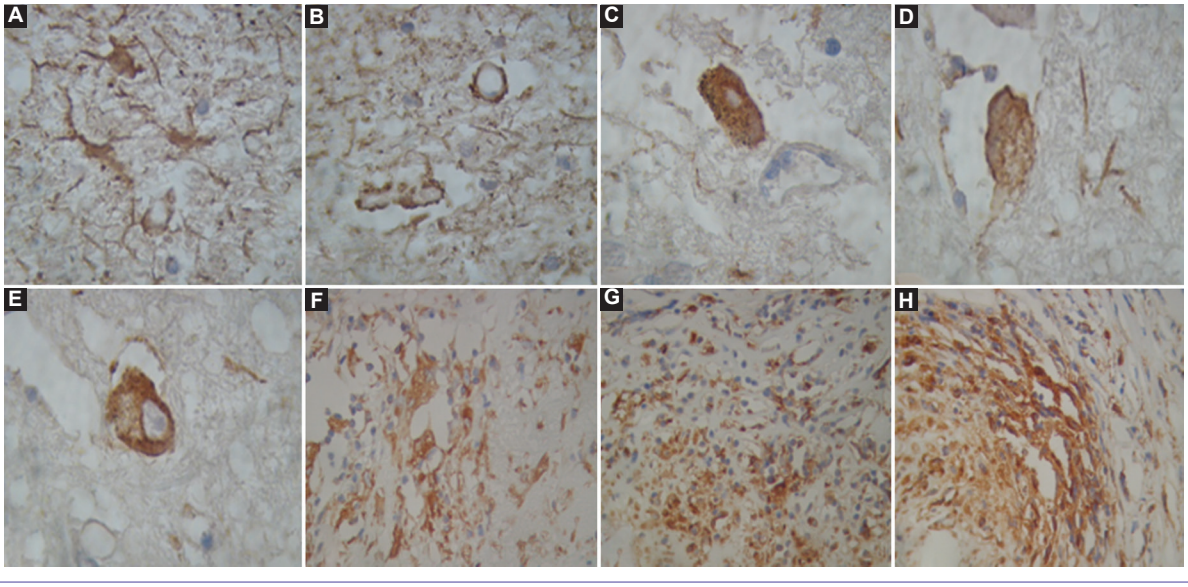

Figure 3. Immunohistochemistry. **A:** we observed glial fibrillary acidic protein (GFAP)-positive astrocytes and in **B:** we observed fragmented GFAP-positive fibrils and empty-looking astrocytes. **C** and **D:** neu-N our positivity in the neurons that show dense granules of variable sizes, **E:** observe that these neurons are Glut3 positive where intracytoplasmic granules are also appreciated. **F:** Glut3 showed a dirty background in a diffuse form and in **G:** Glut1 that is positive in a diffuse form as in the wall of blood vessels and in **H:** the neuropil shows deposits of a diffuse granular material that was positive also for IGF (original magnifications, Immunohistochemistry stain ×400).

giant cells in a hypervascularized stroma. This suggests that the cyst ruptured, subsequently releasing the oily material. An autopsy revealed extensive necrosis with lipid vacuoles in the neuropil. In astrocytic neurons, yellowish proteinaceous material was observed, both in crystalline form and as free material. We propose that tumor rupture or the release of oily contents from CP cystic structures may contribute to thrombosis and lipid dissemination throughout the brain[22,23]. The secretion of vasoactive peptides, combined with water-electrolyte imbalances, forms the basis of cerebral circulation disturbances[19]. Arginine vasopressin (AVP) acts by binding to cell membrane receptors and activating adenylate cyclase, leading to intracellular accumulation of cyclic adenosine monophosphate. DI is associated with either a deficiency or resistance to AVP. Central DI results from diminished AVP due to damage to neurosecretory nuclei or the pituitary stalk[24]. Our patient exhibited signs of respiratory failure, likely due to pulmonary thrombosis; however, this finding could not be confirmed as the autopsy was partial. Thrombosis of the vessels can lead to extensive basal ganglia and vascular lesions, resulting in adverse outcomes[22,25].

The genesis of thrombotic complications has been clarified through specialized hemostasiological studies confirming the presence of acquired protein S deficiency[26]. Although the association between coagulopathy and neoplastic disease is well-documented, there are few reports of primary central nervous system involvement[11,19]. Recent studies have revealed an unexpectedly high incidence of deep vein thrombosis and pulmonary embolism in neurosurgical patients, associated with elevated blood coagulability[27]. Brain tumors, especially suprasellar masses with hypothalamic dysfunction, have been suggested to frequently cause thromboembolic disorders and are seen in systemic tumors exhibiting hypercoagulable states[28]. Experimental studies have examined the effect of external contact on the femoral vessels in rats to assess its potential role in cerebral vascular disease[23,29], concluding that preventing the spillage of this fluid and the routine use of cerebral vasodilators to prevent ischemic complications after CP surgery warrants further evaluation[30]. We propose that the dense oily material (OMF) may pass into blood vessels, occlude them, and subsequently leak or rupture, causing severe secondary damage to brain tissue.

Conclusion

We present a case of CP that histologically showed rupture of the cystic structures with an intense inflammatory response and abundant foamy, dirty macrophages containing microthrombi of yellowish crystalline material (OMF), which led to poor clinical progression, resulting in a significant electrolyte imbalance and thalamic infarction, representing one of the rare cases of CP evolution. Post-operative management of complications, such as DI and electrolyte imbalances, is critical. Establishing protocols to monitor and manage these post-operative complications, particularly endocrine and metabolic disorders that may arise after surgery, is essential. Finally, further research is needed to explore strategies for preventing ischemic and thrombotic complications in neurosurgical patients, managing DI, and evaluating coagulability, which may be a key to preventing post-operative thrombotic events and improving clinical outcomes.

Acknowledgments

We thank the Armstrong Foundation for its unconditional support for the benefit of neuroscience and the student Alejandro López-Landa.

Authors' contributions

ALL: Methodology and writing. SVM: Methodology and writing. CR: Original draft preparation, conceptualization, review, and editing. MTS: Original draft preparation, conceptualization, methodology, project administration, review, and editing.

Funding

This research has not received any specific grant from agencies in the public, commercial, or for-profit sectors.

Conflicts of interests

The authors declare no conflicts of interest.

Ethical considerations

Protection of humans and animals. The authors declare that no experiments involving humans or animals were conducted for this research.

Confidentiality, informed consent, and ethical approval. The authors have followed their institution's confidentiality protocols, obtained informed consent from patients, and received approval from the Ethics Committee. The SAGER guidelines were followed according to the nature of the study.

Declaration on the use of artificial intelligence. The authors declare that no generative artificial intelligence was used in the writing of this manuscript.

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