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Case Report

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Percheron Artery Implicance in Bi-Thalamic Stroke Following Endoscopic Endonasal Approach for Infundibulo-Neurohypophysitis: A Combination of Two Rare Entities

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ABSTRACT

Bi-thalamic stroke is rarely reported in the literature as a complication of an endonasal endoscopic procedure. It has been associated with presence of a Percheron artery variant, as well as with top of the basilar syndrome, both of which significantly increase patient surgical morbidity. Infundibulo-neurohypophysitis in turn, is an unusual inflammatory disorder affecting the infundibulum, the pituitary stalk, and the neurohypophysis. We present the case of a patient with visual impairment and an abnormal hormone profile compatible with infundibulo-neurohypophysitis, in whom tumor resection was conducted through an endoscopic endonasal approach (EEA). Patient developed postoperative bi-thalamic stroke due to Percheron artery infarct. A review of both conditions is included. This is the fourth case reported in the literature of a Percheron artery infarct, and to the best of our knowledge, the first linking it to endoscopic treatment of neurohypophysitis, itself an infrequent condition.

KEYWORDS: Bi-thalamic stroke, Endoscopic endonasal approach, Percheron artery, Infundibulo-neurohypophysitis, Diagnosis, Outcome

ABBREVIATIONS: AOP: Artery of percheron, **EEA:** Endoscopic endonasal approach, **INHP:** Infundibulo-neurohypophysitis, **MRA:** Magnetic resonance angiography, **MRI:** Magnetic resonance imaging, **PCA:** Posterior cerebral artery

INTRODUCTION

Infundibulo-neurohypophysitis (INHP) is an uncommon inflammatory condition affecting the infundibulum, the pituitary stalk, and the neurohypophysis (3,6,13,14). Patients typically present with central diabetes insipidus and normal anterior pituitary function. The most common pathogenesis involves lymphocytic infiltration, with destruction of normal structures and replacement by fibrotic tissue (6,14). Gadolinium-enhanced pituitary magnetic resonance imaging (MRI) is considered the diagnostic gold standard. Findings suggestive of INHP include pituitary stalk thickening and absence of posterior pituitary bright spot on T1 weighted images (3,4). Surgery is usually indicated in cases not responding to conservative treatment, or presenting significant mass effect and visual deficit (3,4). Endoscopic endonasal surgery may be indicated when diagnosis is unclear and a biopsy is needed for diagnostic confirmation, or for total lesion excision for symptom relief (10,11,14).

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The artery of Percheron (AOP) is a normal anatomical variant of the posterior cerebral circulation. When present, it will impact classification of the small branches at the P1 segment of the posterior cerebral artery (PCA) as well as affect circulation and hemodynamic blood flow characteristics within this part of the brain (14).

Although extremely uncommon, injury to the AOP will usually result in bi-thalamic stroke. AOP is present in 4-12% of the population, and responsible for infarction in 0.5-2% of all strokes (1,8,9,11).

Correlation with use of an endonasal sellar approach for diagnosis or treatment is even more infrequent. To date, only three AOP infarctions have been reported following pituitary surgery. These occurred after endonasal resection of pituitary adenomas (2,10,11). This case we believe is the first to be linked to an inflammatory etiology.

CASE REPORT

We present the case of a 47-year-old female patient with a 7-year history of pituitary incidentaloma, initially diagnosed as a Rathke cleft cyst. Progressive growth observed on control MRI prompted referral to our neurosurgical center. Imaging at this time showed a 24.5 x 14.5 x 15.5mm retro-pituitary cyst (Figure 1). Hormonal profile and ophthalmological examination were abnormal. Patient presented symptoms of ADH deficiency and a bilateral inferior longitudinal visual defect. Differential diagnoses considered included craniopharyngioma, Rathke cleft cyst and hypophysitis.

Resection of the cyst and its dense content was performed through an endoscopic endonasal approach (EEA) (Figure 2) with no intraoperative complications.

Patient was extubated after surgery, but remained obtunded for 24 hours. Signs of somnolence persisted after 48 hours, and non-reactive right mydriasis, consistent with incomplete third cranial nerve (CN III) palsy, as well as mild right hemiparesis were observed. No signs of hematoma or acute hydrocephalus were detected on CT. MRI and magnetic resonance angiography (MRA) showed acute bi-thalamic infarction, and presence of a fetal variant of the right posterior cerebral artery (PCA), with Percheron artery origin from the P1 segment of the right PCA. Vessel caliber reduction and distal signal attenuation suggested artery of Percheron infarct (Figure 3).

Length of hospital stay was 10 days (6 in ICU). Patient was discharged to a rehabilitation center, in a stupor awareness state, with no significant changes on neurological examination.

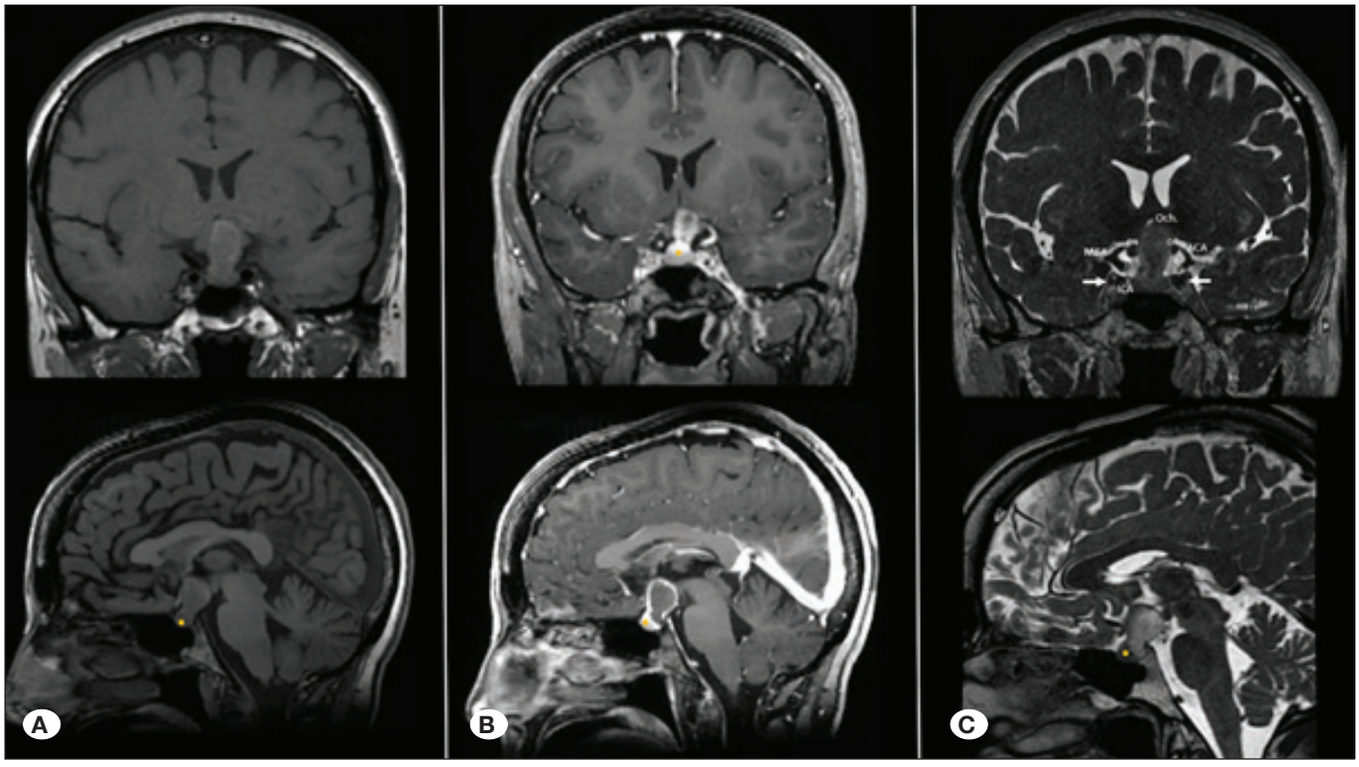

Figure 1: Preoperative T1 **(A)**, T1 post-gadolinium **(B)**, and T2-weighted **(C)** images showing a homogeneous, mild T1 spontaneously hyperintense, and T2 hypointense retro pituitary and infundibular lesion, with suprasellar extension and upward displacement (C) of the optic chiasm (Och.). Notice pituitary gland enhancement (yellow dot) and stalk thickening due to the lesion (B). Evidence of loss of posterior pituitary bright spot is observed on T1(A), sellar floor remains intact. In combination with clinical findings these images suggest presence of infundibulo-neurohypophysitis. **ON:** Optic nerve; **ACA:** Anterior cerebral artery; **MCA:** Middle cerebral artery; **ICA:** Internal carotid artery; **White arrows:** Oculomotor cistern and third cranial nerve (CN III).

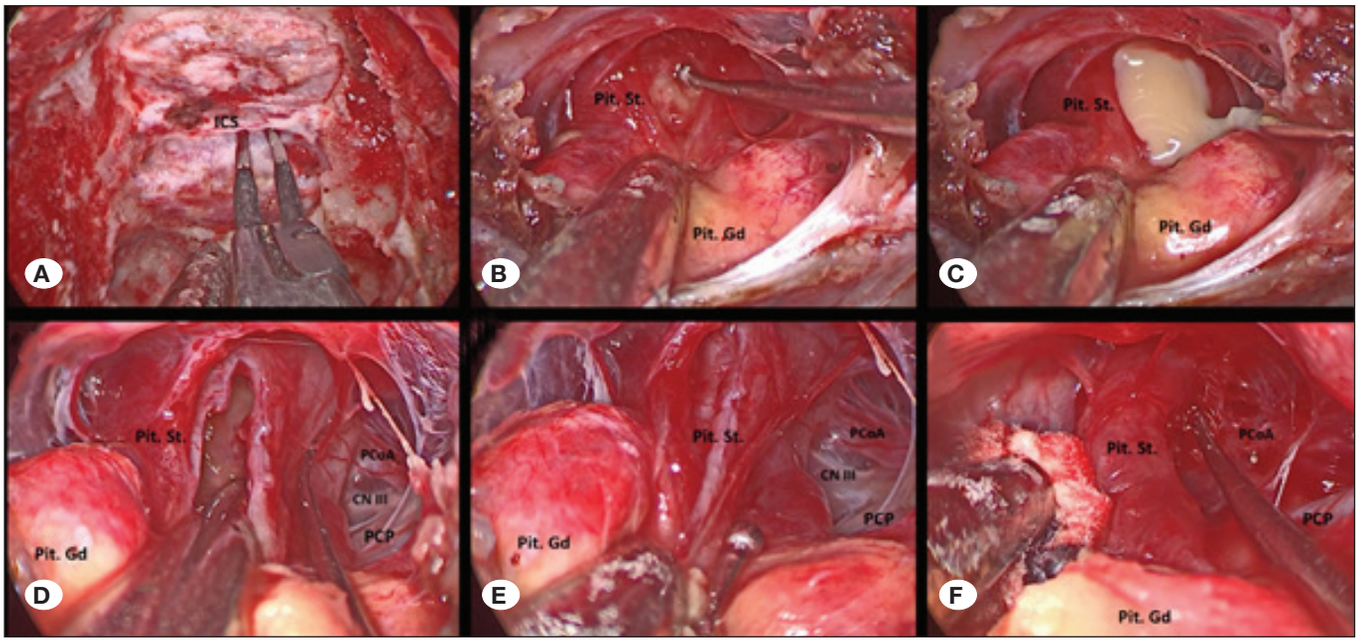

Figure 2: Intraoperative endoscopic images illustrating extended EEA to the sella **(A)**. Notice how the lesion is widening and displacing the pituitary gland (Pit. Gd.) and stalk (Pit. St.), with no clear limit between them **(B)**. Careful dissection is performed across the fibers of the stalk to access the cystic lesion from behind. Careful capsulotomy allows purulent inflammatory content evacuation **(B and D)**. Once lesion is emptied, a thinned pituitary stalk is repositioned. The posterior arachnoid is preserved **(E and F)**.

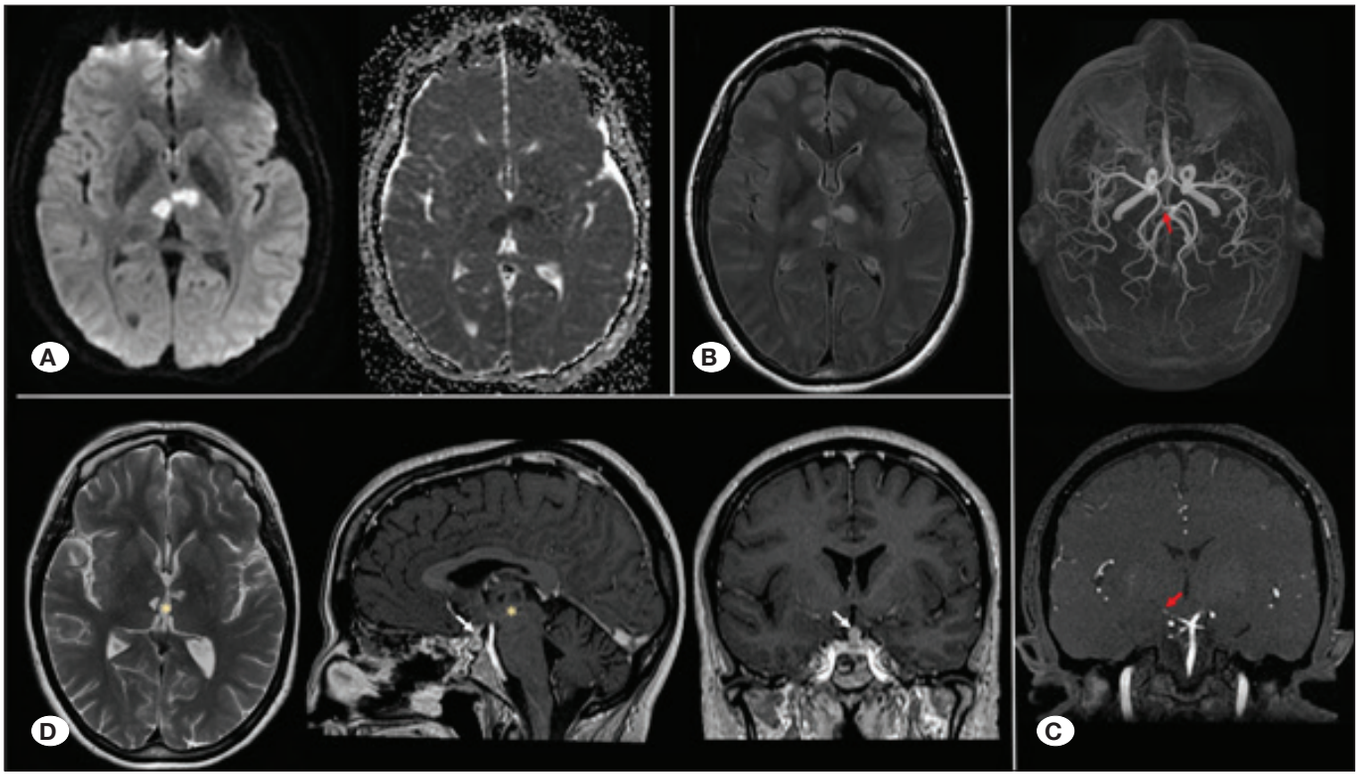

Figure 3: Early postoperative MRI shows bi-thalamic restrictive signal **(A)** on DWI sequence (diffusion-weighted), ADC (apparent diffusion coefficient), and hyperintensity on T2/FLAIR correlating with bi-thalamic ischemia **(B)**. MRA shows fetal variant of right PCA and Percheron artery origin at P1 segment of the right PCA, with evidence of caliber reduction and distal signal attenuation (red arrow) **(C)**. Postop MRI at three months **(D)** showing bi-thalamic increased T2 and decreased T1 signal (yellow asterisk), as well as persistence of pituitary stalk thickening (white arrow).

Pathology report confirmed inflammatory disease compatible with hypophysitis. Tissue cultures were negative.

Considerable neurological improvement was observed after rehabilitation. Patient was discharged after 4 months, and continued treatment as an outpatient. Both neurological and visual field symptoms improved after the surgery. However, panhypopituitarism and diabetes insipidus persisted, requiring hormone replacement therapy and desmopressin.

Despite the presence of minor visual deficit and mild cognitive impairment 1-year after surgery, patient was able to conduct daily life activities independently.

DISCUSSION

The artery of Percheron (AOP) is a normal anatomical variation of the posterior cerebral circulation, originally described by Gerard Percheron in 1973 (1). When present, not only does it impact classification of the small branches at the P1 segment of the posterior cerebral artery (PCA), it also affects circulation and hemodynamic blood flow characteristics within this region of the brain (14).

Prevalence ranges between 4 and 12% in the general population, and AOP infarction accounts for 0.5-2% of all isquemic strokes (1,8,9,11). Because it supplies blood flow to bilateral paramedian thalamic regions, AOP infarction will generate clinical symptoms reflecting midbrain involvement of areas such as the corticospinal tract, subthalamic nucleus, third cranial nerve nucleus, and the dentato-rubro-thalamic tract. Neurological syndromes such as those described by Weber, Benedikt and Claude have also been observed (1,7,9).

The most common underlying causes of an AOP occlusion are small artery disease and cardiac embolism (1,5), but it can also occur secondary to diabetes, smoking, tumors, inflammation, coagulation disorders and hypotension, among others (10).

Not only is the presence of an AOP anatomically significant, as it is a single trunk supplying both the right and left paramedian thalamic nucleus, it also has hemodynamic consequences. This territory is a “watershed zone” presenting increased risk of infarction due to occlusion, inflammation, or hypotension (1). In the case we describe, no direct involvement of the artery or other perforating branches was observed during the surgical procedure, and the arachnoid plane was preserved (see Figure 2). However, the hypoplastic right P1 origin of the AOP with a fetal variation of the CPA (Figure 3), as well as the inflammatory nature of the underlying disease (aseptic purulent hypophysitis) may have played a role in triggering vessel occlusion/stenosis.

Infundibulo-neurohypophysitis (INHP) is an unusual inflammatory disorder affecting the infundibulum, pituitary stalk, and neurohypophysis (3,6,13,14). Overall, hypophysitis is classified according to extension of inflammation and etiology (3,4). In cases of INHP, the usual MRI findings include loss of the posterior pituitary “bright spot’ (which reflects normal vasopressin and oxytocin storage) and thickening of the pituitary stalk. Bright spot absence in INHP indicates a vasopressin storage defect, though it is not considered a pathognomonic sign, as it can be absent in up to 20 % of normal individuals (6).

Other radiological signs that help differentiate INHP from pituitary adenomas or craniopharyngiomas are a homogeneous pre-contrast appearance, full contrast enhancement, an intact sellar floor and in some cases, a dural tail reaction (3,4). (Figure 1).

Hypophysitis is defined as primary, if the inflammation involves the pituitary gland. Autoimmune etiology is usually assumed. Whereas hypophysitis is considered secondary, when focal pituitary lesions are present. Possible causes include craniopharyngioma, adenoma, germinoma, and Rathke cleft cyst (6,3,13). Patients typically present with central diabetes insipidus and normal anterior pituitary function.

Clinical signs and symptoms, biochemical parameters, and MRI findings all contribute to a correct diagnosis (6,14). The endoscopic endonasal approach can be used to obtain a biopsy and confirm histology, or for complete surgical excision in patients not responding to conservative treatment, or presenting significant mass effect and loss of vision (4,6,10,12).

We report a case of Percheron artery infarction, as postoperative complication after endoscopic endonasal INHP resection.

CONCLUSION

Both, artery of Percheron infarction as well as infundibulo-neurohypophysitis are two extremely rare conditions. To the best of our knowledge, their combined occurrence has not been previously reported in the literature. We present the case of a patient who, after endonasal endoscopic resection of a pituitary lesion diagnosed as INHP, developed AOP infarction. No further postoperative complications were observed.

The etiology of the resulting bi-thalamic stroke remains unclear. Nevertheless, the fact that the AOP in this particular case originated from a hypoplastic right P1, with a fetal variation of the CPA, in addition to the inflammatory nature of the underlying disease (purulent aseptic hypophysitis), may have played an important role in causing vessel occlusion/stenosis.

Declarations

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All authors have seen and agree with the contents of the manuscript and have no financial interest to report.

Availability of data and materials: The datasets generated and/or analyzed during the current study are available from the corresponding author by reasonable request.

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AUTHORSHIP CONTRIBUTION

Study conception and design: LDL MR

Data collection: LDL MR PV

Analysis and interpretation of results: LDL MR PV DAK MFB AC

Draft manuscript preparation: LDL MR PV

Critical revision of the article: DAK MFB AC

All authors (LDL, MR, PV, DAK, MFB, AC) reviewed the results and approved the final version of the manuscript.

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