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Pituitary

Xanthomatous sypophysitis secondary to a ruptured Rathke's cleft cyst

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Introduction: Hypophysitis (HP) is a rare disease which develops secondary to chronic or acute inflammation of the pituitary gland and may cause symptoms related to pituitary dysfunction and mass compression. HP can be classified based on the anatomical location of pituitary involvement (adenohypophysitis, infundibulo-neurohypophysitis, or panhypophysitis), the etiology (primary or secondary), and the histopathology (lymphocytic, granulomatous, xanthomatous, IgG4, necrotizing, or mixed forms). Lymphocytic HP is the most common subtype of primary HP, while xanthomatous HP (XHP) is considered the rarest form, with 35 reported cases, to date. In our report, we describe a case with panhypopituitarism due to XHP secondary to the rupture of an Rathke's cleft cyst (RCC). **Clinical Case:** A 35-year-old woman was initially admitted to a Gynecology clinic with a 2-year history of amenorrhea and headache. She was started on cabergoline 0.5 mg twice a week for macroprolactinoma. Due to persistent amenorrhea with low gonadotropins, she was referred to our Endocrinology clinic. Her pituitary function profile revealed panhypopituitarism (Table 1) and a 13×11×12 mm sized sellar mass with diffuse enhancement which sustained toward the infundibulum and dura was observed on the gadolinium-enhanced pituitary MRI. The patient underwent an endoscopic endonasal transsphenoidal approach for tumor resection and thick yellowish fluid draining from the lesion was observed (Figure 1). The histopathological diagnosis was reported as a rupture of an Rathke's cleft cyst and a xanthomatous hypophysitis. The surgery did not improve the symptoms/pituitary functions, however, headache recovered immediately after the first dose of high dose methylprednisolone treatment. **Conclusion:** The exact mechanism by which XHP occurs still remains unclear. The development of the characteristic histopathological lesion which consists of lipid-laden "foamy" histiocytes and CD68 positive macrophages has been previously attributed to the extension of an autoimmune/lymphocytic spectrum. On the other hand, various recent case presentations have reported to consider the inflammatory process of XHP related to the rupture of a RCC.

XHP is an uncommon inflammatory process of the pituitary gland which should be included in the differential diagnoses, if a patient presents with headache, panhypopituitarism and if yellowish fluid is drained from a cystic lesion intraoperatively. In agreement with the recent reports, our case presentation may also indicate a recommendation to classify XHP as secondary hypophysitis which could be caused by a ruptured cystic lesion. Here, we present a patient with XHP which developed as an inflammatory response to a ruptured RCC and the transsphenoidal surgery did not improve the symptoms/pituitary functions, while headache recovered immediately after the first dose of high dose methylprednisolone treatment.

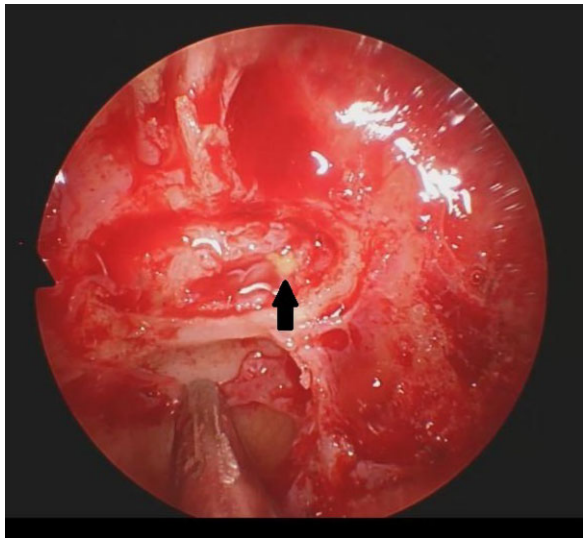


Figure 1. Intraoperative image demonstrating thick yellowish fluid draining from the pituitary lesion.

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Table 1

	Before Surgery		After Surgery		Normal Range
	Before Cabergoline	Under Cabergoline	1 month	8 months	
Prolactin (ng/ml)	125	0.65	0.25	5.18	4.8-23.3
FSH (mIU/ml)		2.14	3.95	7.9	
LH (mIU/ml)		1.17	2.82	9.94	
Estradiol (pg/ml)		<10	<10	<10	
fT4 (ng/dl)		0.45	0.80	1.12	0.6-1.2
TSH (mIU/ml)		2.35			0.4-5.3
Cortisol (µg/dl)		2.5			6.7-22.6
ACTH (pg/ml)		36			0-45
GH (ng/ml)		1.02	0.96	1.95	>8
IGF-1 (µg/ml)		61	69	128	72-233
Headache	+	+	+	-	
Amenorrhea	+	+	+	+	

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