—Case Reports—

Sellar Chondroma with Endocrine Dysfunction that Resolved after Surgery: Case Report

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Chondromas originating from the sella turcica are rare, and the most common initial symptoms are headache and visual disturbance. We describe a case of sellar chondroma with endocrine impairment as an initial manifestation that completely resolved after surgery. A 40-year-old Japanese woman with amenorrhea and galactorrhea for the last 2 years was referred to our department of neurosurgery for the evaluation of high prolactin levels and a tumor in the sella turcica. A biochemical assessment indicated endocrine dysfunction. Magnetic resonance imaging and computed tomography indicated a tumor in the sella turcica. The patient's presentation favored the preoperative diagnosis of pituitary adenoma or Rathke's cleft cyst. However, because calcification was detected, other types of tumors, such as craniopharyngioma, meningioma, and chordoma, were also considered. Endoscopic transsphenoidal surgery was performed, and the possibility of a bony tumor was recognized. Finally, the tumor was completely removed, and the histopathological findings confirmed chondroma. The postoperative course was uneventful, and endocrine function improved. Five years after surgery, the patient is doing well without pituitary insufficiency, pituitary hormone medications, or signs of tumor recurrence. In cases of sellar chondroma, endocrine dysfunction sometimes precedes other symptoms, such as headache and visual disturbance. When examining a patient with an intrasellar tumor harboring calcification, clinicians must consider the possibility of sellar chondroma. Furthermore, to the best of our knowledge, this case is the first of sellar chondroma treated with endoscopic surgery to be reported.
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Key words: sellar chondroma, endocrine impairment, endoscopic transsphenoidal surgery

Introduction

Chondromas are benign tumors that develop most often in the small bones of the hands and feet[1]. Intracranial chondromas are rare and usually occur at the base of the skull but can develop in the cavernous sinus[2] and dura mater of the falx[3]. However, chondromas originating in the sella turcica are extremely rare. Since the first case of intrasellar chondroma was described in 1937, only 16 cases have been reported[1,4–15]. In addition, the initial symptoms in most previously described patients with of sellar chondroma were headaches or visual disturbances. In all these reports, disturbance of endocrine function persisted or was not described. Here we describe a case of sellar chondroma in which the initial manifestation was endocrine impairment.

Case Report

A 40-year-old Japanese woman who had her second child at 38 years and had been experiencing amenorrhea and galactorrhea for the previous 2 years was referred to our

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Table 1 Hormone loading test before operation. (A) Responses of pituitary and adrenal hormones to intravenous injection of CRH (100 μg), TRH (500 μg), and LHRH (100 μg). (B) Responses of GH to intravenous injection of GHRP-2 (100 μg).

1A

	0 min	30 min	60 min
ACTH (pg/mL)	6.9	35.7	35.1
Cortisol (μg/dL)	3.3	7.6	8.4
LH (mIU/mL)	1.4	4.8	5.2
FSH (mIU/mL)	6.1	8.1	8.8
TSH (μU/mL)	0.044	0.401	0.291

1B

	0 min	15 min	30 min	45 min	60 min
GH (ng/dL)	0.70	4.66	4.29	2.29	1.12

department of neurosurgery for evaluation of high prolactin levels and a tumor in the sella turcica that had been detected in the department of obstetrics and gynecology. The medical history included bronchial asthma but no pituitary disorder. On arrival, the patient was awake, alert, and oriented. Headache, fatigue, and polyuria were absent. Vital signs were normal, and there were no abnormal physical findings except for galactorrhea. Eyesight and visual fields were normal.

An endocrine assessment at the initial visit obtained the following results: prolactin, 139.7 ng/mL; adrenocorticotropic hormone, 6.9 pg/mL; cortisol, 3.3 μg/dL; thyroid stimulating hormone (TSH), 1.179 mIU/L; growth hormone (GH), 0.68 ng/mL; insulin-like growth factor 1, 118 ng/mL (–1.3 SD at her age); luteinizing hormone (LH), 1.6 mIU/mL; and follicle-stimulating hormone (FSH), 7.3 mIU/mL. Subsequent hormone loading tests revealed that the cortisol response to corticotropin-releasing hormone, the TSH response to TSH-releasing hormone, and the LH and FSH responses to LH-releasing hormone were low (**Table 1A**). A growth hormone-releasing peptide-2 (GHRP-2) infusion test revealed a low GH response (**Table 1B**).

Magnetic resonance imaging (MRI) revealed a 23×13×20-mm tumor occupying the sella turcica which appeared hypointense on T1-weighed images and hyperintense on T2-weighed images (**Fig. 1A, 1B**). The compressed but otherwise normal pituitary gland was located anterosuperior to the tumor, and there was no sign of invasion to the cavernous sinus. However, we were not able to perform a contrast study with a gadolinium-containing contrast agent because of the patient's bronchial asthma. Computed tomography of the head showed a low-density mass with calcification in the sellar region (**Fig. 1C, 1D**).

The patient's presentation favored the preoperative diagnosis of pituitary adenoma or Rathke's cleft cyst. However, because calcification was detected, other tumors, such as craniopharyngioma, meningioma, and chordoma, were included in the differential diagnosis. Endoscopic transsphenoidal surgery was performed. When the sphenoidal sinus was opened, an enlarged sella turcica was observed. Floor bone fenestration and incision of the dura revealed a whitish, well-demarcated tumor. The tumor, partially associated with calcification, was resilient and cartilaginoid and did not extend into the cavernous sinus. At that moment, we first recognized the possibility of a bone tumor. A normal pituitary gland was located above and anterior to the tumor, and the two were separated. Finally, the tumor was completely removed, and the cavity was plugged with an abdominal fat graft (**Fig. 2**).

Pathological examination showed a hyaline cartilage matrix and chondrocytic cells scattered throughout the tumor (**Fig. 3**). In addition, the tumor cells were not immunoreactive for cytokeratin or epithelial membrane antigen but were immunoreactive for S-100; in addition, the Ki-67 index was less than 1%. These features were consistent with chondroma.

The postoperative course was uneventful, and the amenorrhea and galactorrhea resolved completely. The prolactin levels decreased significantly to 1.9 ng/mL, and a GHRP-2 infusion test revealed a normal GH response (**Table 2B**). Other indicators of endocrine function were also improved (**Table 2A**), including the level of insulin-like growth factor 1 (194 ng/mL, +0.9 SD at her age). Five

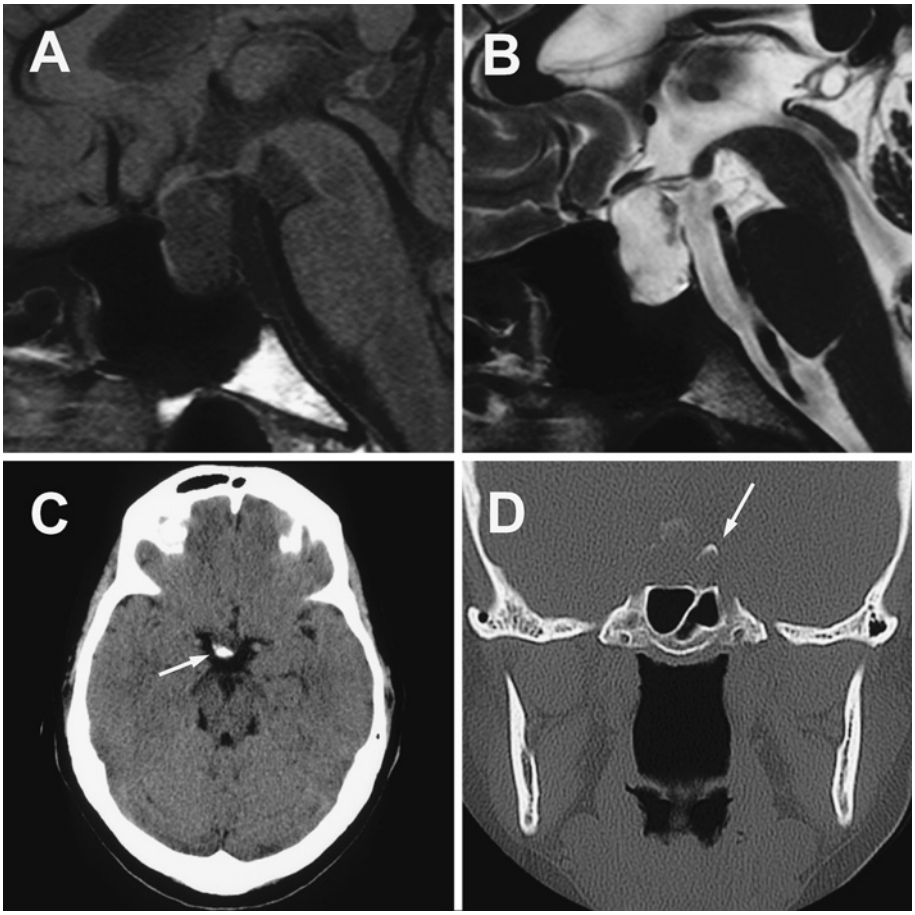

Fig. 1 (A) Plain sagittal T1-weighted magnetic resonance image and (B) sagittal T2-weighted image reveal a mass. (C) Axial and (D) coronal computed tomograms reveal a calcification (**arrow**).

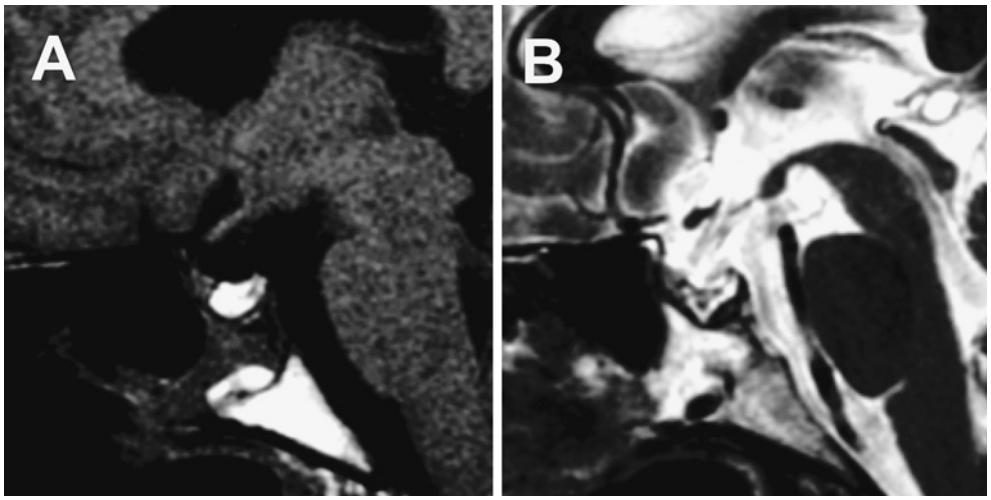

Fig. 2 (A) Plain sagittal T1-weighted magnetic resonance image and (B) sagittal T2-weighted image after operation. The tumor was totally removed.

years after surgery, the patient is doing well without pituitary insufficiency, pituitary hormone medications, or signs of tumor recurrence.

Discussion

We have reported a case of sellar chondroma that initially manifested as endocrine impairment that resolved after surgery. We believe endocrine function improved

because the compression of the otherwise normal pituitary gland was relieved after surgery. In addition, the high prolactin levels at the initial visit were considered to be due to a stalk effect. Previous case reports have made no mention of endocrine function; however, endocrine disorders presumably became obvious after surgery in most cases because the operative field is not clearly visible during microscopic surgery, especially in cases of upper or lateral lesions; therefore, removing the tumor with minimal damage to normal tissue is difficult. We have recently been able to perform precision endoscopic transsphenoidal surgery because of advances in endoscopic and surgical technologies. With an endoscope, a large surgical field, particularly the anterosuperior area, can be clearly observed. As a result, we could break up adhesions between the compressed pituitary gland in the anterosuperior direction and the tumor with visual confirmation. Furthermore, to the best of our knowledge, this case is the first of sellar chondroma treated with endoscopic surgery to be reported.

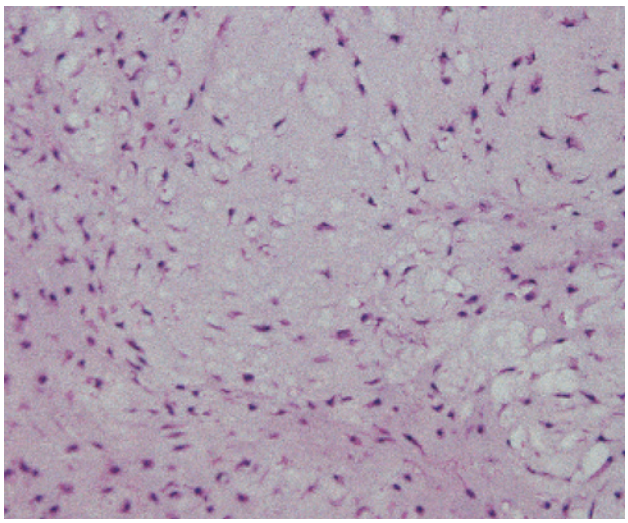

Fig. 3 Hyaline cartilage matrix and chondrocytic cells are scattered throughout the tumor (hematoxylin and eosin staining; 200× magnification).

We were not able to perform contrast-enhanced MRI because of the patient's bronchial asthma. Reportedly, chondroma shows enhanced scalloped margins and curvilinear septa (ring-and-arc pattern) on gadolinium-enhanced T1-weighted images[16]. In addition, a benefit of using a contrast agent is that the location of the normal pituitary gland can be accurately determined. Contrast-enhanced MRI is helpful for differential diagnoses, and an accurate preoperative diagnosis may improve surgical safety.

The most common initial symptoms of sellar chondroma are headache and visual disturbance[4], and the reason our patient presented with endocrine symptoms remains unclear. A likely explanation is that the tumor was detected at an early stage before intracranial pressure had increased or focal neurological deficits had developed. Intracranial chondromas can occur as part of Ollier's disease (multiple enchondromatosis) or Maffucci's disease (multiple enchondromatosis plus subcutaneous hemangiomas)[1,3,5,17]; however, in the present case, only sellar chondroma was observed, and neither recurrence nor de novo appearance of chondroma or hemangioma has been observed in the 5-year follow-up period.

Conclusions

This case report indicates that the endocrine dysfunction of a patient with sellar chondroma sometimes precedes other symptoms, such as headache and visual distur-

Table 2 Hormone loading test after operation. (A) Responses of pituitary and adrenal hormones to intravenous injection of CRH (100 μg), TRH (500 μg), and LHRH (100 μg). (B) Responses of GH to intravenous injection of GHRP-2 (100 μg).

2A

	0 min	30 min	60 min
ACTH (pg/mL)	17.4	51.2	41.3
Cortisol (μg/dL)	12.7	15.5	16.7
LH (mIU/mL)	3.2	7.9	9.9
FSH (mIU/mL)	8.7	11.0	13.8
TSH (μU/mL)	0.525	1.462	1.154

2B

	0 min	15 min	30 min	45 min	60 min
GH (ng/dL)	0.43	24.77	18.09	8.13	3.81

bance. For patients with intrasellar tumors harboring calcification, clinicians must consider the possibility of sellar chondroma.

Conflict of Interest: The authors have no conflicts of interest.

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