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Case Report

The treatment and follow-up for pituitary adenoma and concomitant rathke cleft cyst: a cases report and review of the literature

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Abstract: Pituitary adenoma (PA) concomitant with Rathke cleft cyst (RCC) is extremely rare. The risk of recurrence of both PA and RCC necessitates post-operative follow-up. Here we describe a 22-year-old woman was admitted for headache and hypopsia. Magnetic resonance imaging (MRI) revealed a sellar mass. During surgery, a small RCC was incidentally found inside the PA. Both tumors were completely resected. Follow-up MRI 6 years later demonstrated a sellar lesion and a following surgery were performed, during which an RCC was validated without evidence of PA recurrence. This is the first report of the recurrence of PA concomitant with RCC 6 years after surgery, suggesting that PA and concomitant RCC requires periodical post-operative follow-up. For recurrent tumors, especially for symptomatic ones, a second surgery is needed. During the surgery, lesion should be removed as completely as possible, without damage of the normal pituitary.

Keywords: Pituitary adenoma, rathke cleft cyst

Introduction

Pituitary adenoma (PA) and Rathke cleft cyst (RCC) are both common sellar diseases. Statistics showed the incidence of PA was 16.7% [1]. Famini et al. [2] retrospectively analyzed MRI information of 2598 patients and found RCC the second most common sellar lesion after PA, accounting for 19% of all sellar lesions. These two diseases often separately occur, with concomitance rarely seen. Sumida et al. [3] investigated 374 cases of sellar lesions, and only found 8 PA concomitant with RCC (2.1%). Currently, merely 44 PA concomitant with RCC cases were reported in English [4-28], with fewer studies having post-operational follow-up information.

Recently, we received a case of PA concomitant with RCC. It was a small-sized incidental RCC inside a PA discovered by the first surgery. In regular follow-up 6 years after the surgery, recurrence of a sellar lesion was noticed, which was validated during the second surgery as relapse of RCC rather than PA. Since there is no similar report before, we studied the features of PA concomitant with RCC combined with literature search and review.

Case report

A 22-year female was admitted on July 21th, 2008 for 2 years of headache and 6 months of hypopsia. Ophthalmoscopic examination revealed binocular vision 0.6, normal fundus oculi, no defect of visual field or other positive signs were identified. Pituitary MRI revealed a sellar mass with suprasellar extension, measured 11 mm×20 mm×23 mm, isointense on T1-weighted image (T1WI), slightly hyperintense on T2-weighted image (T2WI) and mild to moderate enhancing, which was considered a PA (**Figure 1A-C**). The serum prolactin level was 55.6 ng/ml, with all other pituitary hormones within normal range. Then the patient underwent transsphenoidal tumor resection under general anesthesia on July 26, 2008. During surgery, a soft gray tumor was seen. When the deeper part of the tumor was resected, a capsule of 8 mm in diameter was noticed in the

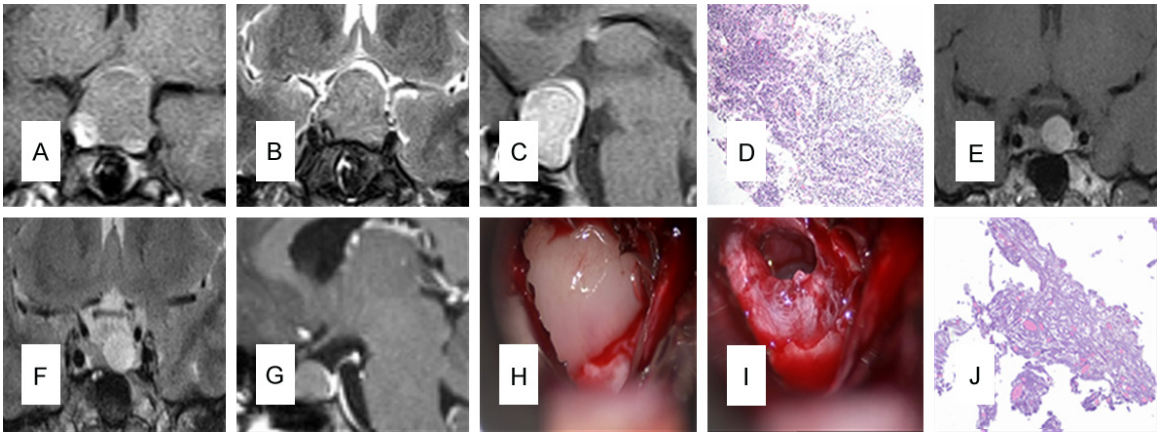

Figure 1. MRI and histopathological manifestations of the patient. A. Coronal T1WI; B. Coronal T1WI; C. Sagittal T1+ MRI before surgery. D. Hematoxylin and eosin staining of tumor tissues (×10); E. Coronal T1WI; F. Coronal T1WI; G. Sagittal T1+ MRI before secondary surgery; H. Greyish white viscous cyst content was seen after opening the dura; I. No abnormal pituitary tissue was recognized after removal of the cyst during the second surgery; J. Hematoxylin and eosin staining of tumor tissues of the second surgery (×100). There was myxoid substance among the tumor tissues.

tumor. The capsule was greyish white without blood supply. Incision of the capsule revealed a greyish white soft mass which was totally aspirated. After aspiration of the cystic content, tumor was totally curetted. The sample of the capsule was not available for pathological examination due to its limited volume and viscosity. The result of pathological examination was consistent with prolactin-producing adenomas (PRL-omas) (**Figure 1D**). The serum prolactin reduced to 25.81 ng/ml on the first day after the surgery. And symptoms of headache and hypopsia were improved. Follow-up pituitary MRI on March 2, 2015 revealed a well-defined roundish abnormal signal intensity, measured 11 mm×9 mm×11 mm, with slightly hyperintense on T1WI, hyperintense on T2WI and homogenous enhancing of normal pituitary, which was considered post-operative relapse of PA (**Figure 1E-G**). The serum prolactin level was 30.64 ng/ml, with other pituitary hormones within normal ranges. Another transsphenoidal tumor resection was carried out under general anesthesia on March 5, 2015. During the surgery, greyish white viscous fluid was released after puncture. The dura was opened followed by discharge of greyish white viscous mass (**Figure 1H**, **1I**). The capsule was well-defined and tightly adhered to adjacent tissues with poor blood supply, and was completely aspirated. The hormone assays were all normal on the first post-operative day. Pathological examination showed there was myxoid substance among the tumor tissues (**Figure 1J**).

Discussion

Kepes [4] first reported PA concomitant with RCC in 1978. In the previously reported 44 cases of PA concomitant with RCC, PRL-omas is the most common type of PA, followed by growth hormone-producing adenomas (GH-omas), clinlcally nonfunctioning adenomas (NF-omas) and adrenocorticotrophic hormone-producing adenomas (ACTH-omas). To the best of our knowledge, the current study is the first to report the recurrence of PA concomitant with RCC 6 years after surgery.

PA concomitant with RCC has no specific clinical symptoms or pre-operative imaging findings, thus it is often hard to confirm the diagnosis before surgery. In the reported studies, pre-operative diagnosis is often PA or PA with cystic change or with hemorrhage, or craniopharyngioma. Confirmation of the diagnosis is all via surgery or pathological examination. The typical pathological finding of RCC refers to cyst wall composed of simple ciliated columnar epithelium or cuboidal epithelium and some RCCs have squamous metaplasia and inflammatory components. The cyst content has multiple forms such as myxoid or watery fluid, with components like proteins, mucopolysaccharides, cholesterol crystals, debris of necrotic exfoliat-

ed cells and hemosiderin. In the first surgery of the patient, the sample of the cyst was not obtained due to its small size. During the second surgery, viscous fluid was clearly noticed, despite that the typical simple ciliated columnar epithelium or cuboidal epithelium was not observed. Therefore, we believe the patient can be confirmed PA concomitant with RCC.

Surgery is the major treatment for most PA and symptomatic RCC. For the risk of recurrence, regular post-operative follow-up is of significance. At present, only a small number of studies reported post-operative follow-up of PA concomitant with RCC. Noh et al. [18] and Wang et al. [24] reported the follow-up information 6 months after surgery of a GH-omas concomitant with RCC and a case of NF-omas concomitant with RCC respectively, without recognition of any recurrence. Karavitaki et al. [21] followed a case of ACTH-omas concomitant with RCC for 5 years after surgery and didn't find recurrence, whereas the RCC of the patient in this article reappeared 6 years after surgery without relapse of the PA.

The texture of PA is the key factor to affect the extent of resection during transsphenoidal surgery. For a soft tumor, even if it has suprasellar or juxtasellar invasion, the suprasellar or juxtasellar part can gradually fall into the sella with the pulse of brain arteries and satisfying removal can be achieved. Diri et al. [29] pointed out that the resection extent of PA is associated with the risk of recurrence. In the paper, the texture of tumor was soft and beneficial for total resection. We believe that tumor texture is the key factor for its recurrence-free 6 years after surgery.

At present, the study of RCC with the largest surgical cases was published in 2011 by Ogawa et al. [30]. Most researchers adopt microscopic transsphenoidal technique, and endoscopic transsphenoidal technique is also used [31]. The management of cyst wall of RCC varies from biopsy, partial resection to complete resection. Partial resection and decompression is believed by some researchers to achieve satisfying surgical effect and reduced cerebrospinal fluid rhinorrhea, diabetes insipidus and apituitarism at the same time [32, 33], while others hold that the resection extent of the cyst is associated with the recurrence risk, and therefore, promote total resection [34-37].

Surgery approach of the patient in this study was transsphenoidal. Given that the smaller RCC was easy to achieve total resection in the patient and risk of post-operative complications like hypopituitarism is little, we performed microscopically complete resection. However, the RCC recurred 6 years later. Currently, the following factors are believed to associate with post-operative recurrence of RCC. 1) Suprasellar expansion: RCC completely or partially located on suprasellar area is easier to recur than simple sellar cyst [32]. 2) Pathological change of cyst wall: squamous metaplasia or inflammation enhances recurrence risk [33-36]. 3) Infection: infection of cyst is a high risk factor for relapse [34]. 4) Filling materials for cyst cavity: fat tissue or fascia can generate recurrence [34]. 5) Cyst fluid properties: cerebrospinal-fluid-like fluid is more likely to recur than myxoid fluid [9]. 6) Resection extent: Kim et al. [35] hold that resection extent of cyst is associated with recurrence risk. We believe that the relapse of RCC of the patient may be linked to residual tissue of cyst wall due to factors like suprasellar location, limited microscopic field, insufficient illumination during the surgery. Frank et al. [31] adopted neuroendoscopic surgery and obtained significantly decreased recurrence rate than microscopic operation.

In conclusion, PA concomitant with RCC rarely occurs, among which post-operative recurrence of RCC were not reported previously. PA concomitant with RCC necessitates regular post-operative follow-up. For recurrence, especially symptomatic ones, a second surgery is needed, with transsphenoidal approach the first choice. Without damaging of normal pituitary during the surgery, the lesion should be removed as completely as possible.

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Disclosure of conflict of interest

None.

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References

[1] Ezzat S, Asa SL, Couldwell WT, Barr CE, Dodge WE, Vance ML, McCutcheon IE. The prevalence of pituitary adenomas: a systematic review. Cancer 2004; 101: 613-619.

[2] Famini P, Maya MM, Melmed S. Pituitary magnetic resonance imaging for sellar and parasellar masses: ten-year experience in 2598 patients. J Clin Endocrinol Metab 2011; 96: 1633-1641.

[3] Sumida M, Migita K, Tominaga A, Iida K, Kurisu K. Concomitant pituitary adenoma and Rathke's cleft cyst. Neuroradiology 2001; 43: 755-759.

[4] Kepes JJ. Transitional cell tumor of the pituitary gland developing from a Rathke's cleft cyst. Cancer 1978; 41: 337-343.

[5] Bader LJ, Carter KD, Latchaw RE, Ellis WG, Wexler JA, Watson JC. Simultaneous symptomatic Rathke's cleft cyst and GH secreting pituitary adenoma: a case report. Pituitary 2004; 7: 39-44.

[6] Hiyama H, Kubo O, Yato S, Kagawa M, Kitamura K. A case of pituitary adenoma combined with Rathke's cleft cyst. No Shinkei Geka 1986; 14: 435-440.

[7] Ikeda H, Niizuma H, Fujiwara S, Suzuki J, Sasano N. A case of prolactinoma in close association with Rathke's cleft cyst. No Shinkei Geka 1987; 15: 999-1003.

[8] Ikeda H, Yoshimoto T, Katakura R. A case of Rathke's cleft cyst within a pituitary adenoma presenting with acromegaly--do "transitional cell tumors of the pituitary gland" really exist? Acta Neuropathol 1992; 83: 211-215.

[9] Kaku S, Tanaka T, Sawauchi S, Dobashi H, Ohtsuka T, Numoto RT, Murakami S, Komine K, Abe T. A case of non-functional pituitary adenoma associated with Rathke's cleft cyst. No Shinkei Geka 2005; 33: 797-803.

[10] Matsumori K, Okuda T, Nakayama K, Miyasaka Y, Beppu T, Kubo O. Case of calcified prolactinoma combined with Rathke's cleft cysts. No Shinkei Geka 1984; 12: 833-838.

[11] Miyagi A, Iwasaki M, Shibuya T, ido G, Kushi H, Miyagami M, Tsubokawa T. Pituitary adenoma combined with Rathke's cleft cyst--case report. Neurol Med Chir 1993; 33: 643-650.

[12] Nakasu S, Nakasu Y, Kyoshima K, Watanabe K, Handa J, Okabe H. Pituitary adenoma with multiple ciliated cysts: transitional cell tumor? Surg Neurol 1989; 31: 41-48.

[13] Nishio S, Fujiwara S, Morioka T, Fukui M. Rathke's cleft cysts within a growth hormone producing pituitary adenoma. Br J Neurosurg 1995; 9: 51-55.

[14] Nishio S, Mizuno J, Barrow DL, Takei Y, Tindall GT. Pituitary tumors composed of adenohypophysial adenoma and Rathke's cleft cyst elements: a clinicopathological study. Neurosurgery 1987; 21: 371-377.

[15] Swanson SE, Chandler WF, Latack J, Zis K. Symptomatic Rathke's cleft cyst with pituitary adenoma: case report. Neurosurgery 1985; 17: 657-659.

[16] Trokoudes KM, Walfish PG, Holgate RC, Pritzker KP, Schwartz ML, Kovacs K. Sellar enlargement with hyperprolactinemia and a Rathke's pouch cyst. JAMA 1978; 240: 471-473.

[17] Vancura RW, Jacob KM, Damjanov I. A 70-year-old man with diplopia, nausea, and vomiting. Rathke cleft cyst concomitant with pituitary adenoma. Arch Pathol Lab Med 2006; 130: 403-404.

[18] Noh SJ, Ahn JY, Lee KS, Kim SH. Pituitary adenoma and concomitant Rathke's cleft cyst. Acta Neurochir (Wien) 2007;149: 1223-1228.

[19] Radhakrishnan N, Menon G, Hingwala DR, Radhakrishnan VV. Non-functioning pituitary adenoma and concomitant Rathke's cleft cyst. Indian J Pathol Microbiol 2011; 5: 649-651.

[20] Koutourousiou M, Kontogeorgos G, Wesseling P, Grotenhuis AJ, Seretis A. Collision sellar lesions: experience with eight cases and review of the literature. Pituitary 2010; 13: 8-17.

[21] Karavitaki N, Scheithauer BW, Watt J, Ansorge O, Moschopoulos M, Llaguno AV, Wass JA. Collision lesions of the sella: co-existence of craniopharyngioma with gonadotroph adenoma and of Rathke's cleft cyst with corticotroph adenoma. Pituitary 2008; 11: 317-323.

[22] Babu R, Back AG, Komisarow JM, Owens TR, Cummings TJ, Britz GW. Symptomatic Rathke's cleft cyst with a co-existing pituitary tumor; Brief review of the literature. Asian J Neurosurg 2013; 8: 183-187.

[23] Zhou P, Cai B, Ma W, Jiang S. Combined pituitary adenoma and Rathke's cleft cysts: two multicystic cases and literature review. Neurol India 2012; 60: 665-667.

[24] Wang K, Ma L, You C. Pituitary adenoma and concomitant Rathke's cleft cyst: a case report and review of the literature. Neurol India 2012; 60: 309-310.

[25] You C, Qiao F, Jiang S, Xiao A. Growth hormone secreting pituitary adenoma associated with Rathke's cleft cyst. Neurol India 2012; 60: 310-311.

[26] Inder WJ, Macfarlane MR. Hyperprolactinaemia associated with a complex cystic pituitary mass: medical versus surgical therapy. Intern Med J 2004; 34: 573-576.

[27] Gessler F, Coon VC, Chin SS, Couldwell WT. Coexisting rathke cleft cyst and pituitary adenoma presenting with pituitary apoplexy: report of two cases. Skull Base Rep 2011; 1: 99-104.

[28] Kamoshima Y, Sawamura Y, Iwasaki YK, Fujieda K, Takahashi H. Case of Carney complex complicated with pituitary adenoma and Rathke cleft cyst. No Shinkei Geka 2008; 36: 535-539.
[29] Diri H, Ozaslan E, Kurtsoy A, Tucer B, Simsek Y, Ozturk F, Durak AC, Bayram F. Prognostic factors obtained from long-term follow-up of pituitary adenomas and other sellar tumors. Turk Neurosurg 2014; 24: 679-687.
[30] Ogawa Y, Watanabe M, Tominaga T. Prognostic factors of operated Rathke's cleft cysts with special reference to re-accumulation and recommended surgical strategy. Acta Neurochir (Wien) 2011; 153: 2427-2433; discussion 2433.
[31] Frank G, Sciarretta V, Mazzatenta D, Farneti G, Modugno GC, Pasquini E. Transsphenoidal endoscopic approach in the treatment of Rathke's cleft cyst. Neurosurgery 2005; 56: 124-128; discussion 129.
[32] Benveniste RJ, King WA, Walsh J, Lee JS, Naidich TP, Post KD. Surgery for Rathke cleft cysts: technical considerations and outcomes. J Neurosurg 2004; 101: 577-584.
[33] Potts MB, Jahangiri A, Lamborn KR, Blevins LS, Kunwar S, Aghi MK. Suprasellar Rathke cleft cysts: clinical presentation and treatment outcomes. Neurosurgery 2011; 69: 1058-1068; discussion 1068-1057.
[34] Tate MC, Jahangiri A, Blevins L, Kunwar S, Aghi MK. Infected Rathke cleft cysts: distinguishing factors and factors predicting recurrence. Neurosurgery 2010; 67: 762-769; discussion 769.
[35] Kim JE, Kim JH, Kim OL, Paek SH, Kim DG, Chi JG, Jung HW. Surgical treatment of symptomatic Rathke cleft cysts: clinical features and results with special attention to recurrence. J Neurosurg 2004; 100: 33-40.