

Intrasellar Symptomatic Salivary Gland Rest

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Ectopic salivary gland tissue in sellar turcica is frequently observed in microscopic examination at autopsy. This tissue is considered clinically silent. Only 2 symptomatic cases have been previously reported. Here we report a 28-year-old woman presenting with galactorrhea and hyperprolactinemia. Magnetic resonance imaging revealed a 6×5-mm nodule in the posterior aspect of the pituitary gland. This nodule showed isointensity on T1- and T2-weighted images and less enhancement on post-contrast T1-weighted images. Transsphenoidal exploration revealed a cystic lesion within the pituitary gland, which consisted of a grayish gelatinous content. The pathologic examination confirmed the diagnosis of salivary gland rest. [*J Chin Med Assoc* 2007;70(5):215–217]

Key Words: ectopic salivary gland, magnetic resonance imaging, pituitary gland

Introduction

Salivary glands resting in human posterior pituitary have been observed at autopsy when serial sections were employed.¹ The tubular glands in the posterior pituitary often communicate with Rathke's cleft or its cystic subdivisions.² However, these tissues are considered clinically silent. Rarely, large lesions can cause hormonal abnormality, and only 2 cases have been reported in the world literature.^{3,4}

Case Report

A 28-year-old woman presented with persistent galactorrhea and menstrual irregularities after her second delivery. Endocrine evaluation revealed mild hyperprolactinemia. The prolactin level was 93 ng/mL. She was prescribed bromocriptine for 2 years. The galactorrhea and menstrual irregularities responded well to bromocriptine treatment. The repeated prolactin level was 12.8 ng/mL, and the other pituitary hormone levels were within normal range.

Magnetic resonance imaging (MRI) demonstrated a 6×5-mm nodule near the posterior pituitary gland. This nodule showed isointensity on T1-weighted images

(T1WI) and T2-weighted images (T2WI), and less contrast enhancement on post-contrast conventional T1WI (Figure 1). Post-contrast dynamic T1WI also revealed less enhancement of the pituitary lesion. Preoperative imaging diagnosis of pituitary adenoma with cystic change or Rathke's cleft cyst was impressed. There was no change in the size of the nodule when compared with MRI studies outside hospital 2 years before. Because of the unacceptable adverse symptoms of headache and sleep disturbances, as well as the unchanged lesion size despite bromocriptine treatment, an operation was decided upon. The operation was performed smoothly via transsphenoidal approach, revealing a lesion with grayish gelatinous content within the posterior pituitary gland.

Microscopic examination showed the tissue consisted of mixed nests of acidophilic, basophilic and chromophobic cells in a delicate fibrovascular network; there was colloid-like material and salivary gland rest with mild chronic inflammation (Figure 2). There was no evidence of neoplasm. These findings pointed to the diagnosis of ectopic salivary gland tissue. Postoperative course was smooth, except that transient diabetes insipidus occurred. The prolactin level and other pituitary hormone levels were all within normal ranges. The patient remained well in the year following the operation.

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Figure 1. A small nodular lesion was noted near the posterior pituitary region. The nodule shows relative isointensity on (A) T1-weighted image and (B) T2-weighted image, and (C) less enhancement on post-contrast T1-weighted image.

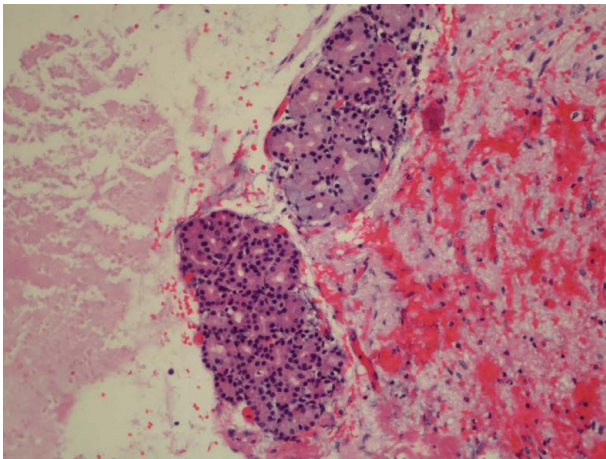


Figure 2. Lobules of salivary glands composed of serous type acini with eosinophilic granular cytoplasm (hematoxylin & eosin stain, 40 \times).

Discussion

In the course of normal human embryogenesis, the pituitary gland is classically thought to come from 2 anlagen.⁵ The first one arises as an up-growth from the lining of the roof of the oral cavity, rostrally located to the oropharyngeal membrane. The second one develops as a down-growth from the floor of the diencephalon. The up-growth arising from the roof of the mouth gives off a funnel-shaped extension called the adenohypophyseal pouch, or Rathke's pouch. The tubular gland in the posterior pituitary often communicates with Rathke's cleft, and is considered to be the origin of the salivary gland.¹ Kusakabe et al⁶ reported that during the organogenesis of the mouse's anterior pituitary, there exists a developmental stage (Day 8.5–11 *in utero*) when prospective pituitary epithelium can respond to the heterotopic submandibular gland mesenchyma, leading to subsequent development of a submandibular gland-like tissue. This experiment

supported that the origin of salivary cells in the pituitary was from the epithelium of Rathke's pouch.

Salivary gland ectopia has been found widely in the head and neck regions, including external ear canal, mastoid process, tonsils, tongue, body of mandible and thyroglossal duct.^{7,8} It is normally thought to result from the misplacement of salivary gland rests along the embryonic pathway of migration of the major salivary gland analog, during development, or, more rarely, by differentiation from vesicular remnants of primitive embryologic structures.⁸ Ectopic salivary tissue located in regions as far away as the submucosa of the rectum have also been reported.⁹

Intracranial ectopic salivary glands are very rare. There are 2 cases reported in the cerebellopontine angle region.^{8,10} Cases of salivary gland rests in the posterior pituitary were reported in as many as 78 out of 2,300 consecutive autopsies.¹ However, only 2 symptomatic cases were reported in the literature.^{3,4} Kato et al³ reported an ectopic salivary gland within the pituitary gland of an 11-year-old boy who presented at age 9 with growth hormone deficiency. Tatter et al⁴ reported a symptomatic salivary-rest-cyst of the sella turcica of a 22-year-old woman who presented with progressive menstrual irregularities and mild hyperprolactinemia. MRI found the lesion in the posterior aspect of the pituitary gland, and the lesion was isointense on T1WI, hypointense on T2WI, and showed no enhancement after intravenous gadolinium administration. A similar picture was also observed in our case. Post-contrast dynamic and conventional T1WI in our case revealed less enhancement of the lesion.¹¹ The imaging differential diagnosis of the intrasellar salivary gland cyst might include cystic pituitary adenoma, Rathke's cleft cyst, craniopharyngioma, arachnoid cyst, and pituitary abscess.^{11–14}

Other than non-neoplasm mass, very few salivary gland-like tumors within the sellar regions have been

reported. Hampton et al¹⁵ described 5 salivary gland-like tumors of the sellar region. These tumors included cellular pleomorphic adenoma, monomorphic adenoma (2 cases), oncocytoma and low-grade adenocarcinoma of the salivary gland. They exhibited morphologic characteristic of various salivary neoplasms. Chimelli et al² reported a pleomorphic adenoma of the sellar region. The appearance indicated that it was a salivary gland pleomorphic adenoma arising within the wall of a Rathke's cleft.

Moderate hyperprolactinoma in the range of 25–150 ng/mL is a common feature in patients with nonprolactin-secreting pituitary adenomas or other intrasellar neoplasms, such as craniopharyngiomas, non-functioning macroadenomas, hypothalamic tumors and lymphocytic hypophysitis.^{16,17} Elevated prolactin is probably caused by compression of the pituitary stalk and interruption of the dopaminergic inhibition of lactotrophs.^{17,18} The above viewpoint may explain the elevated prolactin level (93 ng/mL) and clinical presentations in our case.

In conclusion, intrasellar salivary gland rest, although rarely symptomatic, should be taken into consideration in the differential diagnosis when a lesion shows posterior pituitary location and less enhancement after intravenous contrast media administration. From the good result of this case, transsphenoidal surgery could be a proper treatment for those who are intolerant of bromocriptine treatment.

References

- Schochet SS, McCormick WF, Halmi NS. Salivary gland rests in the human pituitary: light and electron microscopical study. *Arch Pathol* 1974;98:193–200.
- Chimelli L, Gadelha MR, Une K, Carlos S, Pereira PJ, Santos JL, Filho PN, et al. Intra-sellar salivary gland-like pleomorphic adenoma arising within the wall of a Rathke's cleft cyst. *Pituitary* 2000;3:257–61.
- Kato T, Aida T, Abe H, Miyamachi K, Hida K, Taneda M, Ogata A. Ectopic salivary gland in the pituitary gland: case report. *Neurol Med Chir* 1988;28:930–3.
- Tatter SB, Edgar MA, Klibanski A, Swearingen B. Symptomatic salivary-rest cyst of the sella turcica. *Acta Neurochir (Wien)* 1995; 135:150–3.
- Dubois PM, el Amraoui A, Heritier AG. Development and differentiation of pituitary cells. *Microsc Res Tech* 1997;39:98–113.
- Kusakabe M, Sakakura T, Sano M, Nishizuka Y. A pituitary-salivary mixed gland induced by tissue recombination of embryonic pituitary epithelium and embryonic submandibular gland mesenchyme in mice. *Dev Biol* 1985;110:382–91.
- Gudbrandsson FK, Liston SL, Maisel RA. Heterotopic salivary tissue in the neck. *Otolaryngol Head Neck Surg* 1982;90:279–82.
- Rodriguez F, Scheithauer BW, Ockner DM, Giannini C. Solitary fibrous tumor of the cerebellopontine angle with salivary gland heterotopia: a unique presentation. *Am J Surg Pathol* 2004;28: 139–42.
- Weitzner S. Ectopic salivary gland tissue in submucosa of rectum. *Dis Colon Rectum* 1983;26:814–7.
- Curry B, Taylor CW, Fisher AW. Salivary gland heterotopia: a unique cerebellopontine angle tumor. *Arch Pathol Lab Med* 1982;106:35–8.
- Rand T, Lippitz P, Kink E, Huber H, Schneider B, Imhof H, Trattig S. Evaluation of pituitary microadenomas with dynamic MR imaging. *Eur J Radiol* 2002;41:131–5.
- Iqbal J, Kanaan I, Al Homsy M. Non-neoplastic cystic lesions of the sellar region presentation, diagnosis and management of eight cases and review of the literature. *Acta Neurochir (Wien)* 1999;141:389–97.
- Shin JL, Asa SL, Woodhouse LJ, Smyth HS, Ezzat S. Cystic lesions of the pituitary: clinicopathological features distinguishing craniopharyngioma, Rathke's cleft cyst, and arachnoid cyst. *J Clin Endocrinol Metab* 1999;84:3972–82.
- Eun Kim J, Hee-Won J. Diagnosis and treatment of symptomatic Rathke's cleft cyst. *Neurosurgery Quarterly* 2003;13: 64–76.
- Hampton TA, Scheithauer BW, Rojiani AM, Kovacs K, Horvath E, Vogt P. Salivary gland-like tumors of the sellar region. *Am J Surg Pathol* 1997;21:424–34.
- Albuquerque FC, Hinton DR, Weiss MH. Excessively high prolactin level in a patient with a nonprolactin-secreting adenoma: case report. *J Neurosurg* 1998;89:1043–6.
- Liu JK, Couldwell WT. Contemporary management of prolactinomas. *Neurosurg Focus* 2004;16:E2.
- Arafah BM, Nekl KE, Gold RS, Selman WR. Dynamics of prolactin secretion in patients with hypopituitarism and pituitary macroadenomas. *J Clin Endocrinol Metab* 1995;80:3507–12.