Pituitary Symptomatic Salivary Gland Rest Cyst: Case Report

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Tel: +82-53-420-5651 Fax: +82-53-423-0504 E-mail: jhwang@knu.ac.kr Although salivary gland rest in posterior pituitary is frequently observed during microscopic examination at autopsy, this rest tissue rarely causes clinical symptoms. So far, only three symptomatic salivary gland rest cyst in pituitary was reported. The author reports the 4th case of symptomatic pituitary salivary gland rest cyst in 26-year-old woman presenting with severe headache. The lesion showed hyperdense shadow on computed tomography, high signal on T1-weighted image and low signal on T2weighted image magnetic resonance imaging. There was no enhancement of the mass after gadolinium injection. Trans-sphenoidal approach revealed that the cyst wall consisted of normal appearing salivary gland and gelatinous cyst content. Although the pituitary salivary gland rest cyst is rarely symptomatic, it should be take into consideration in the differential diagnosis of pituitary lesions.

Key Words Pituitary gland; Ectopic salivary gland; Sella turcica; Magnetic resonance imaging.

INTRODUCTION

Microscopic salivary gland rests in human pituitary have long been recognized [1]. Theses glandular structures in the posterior pituitary seem to drive from and communicate with Rathke's pouch and its remnants [2]. Despite the well-known knowledge of this salivary gland rest in pituitary, these lesions rarely cause clinical symptoms, and only three cases have been reported in the literature [3-5]. Here, we report the 4th case of symptomatic salivary gland rest cyst in pituitary gland.

CASE REPORT

A 26-year-old woman presented with persistent moderate headache after sudden onset of severe headache and nausea 5 days ago. She had regular menstrual periods since menarche at the age 14. Endocrine evaluation revealed no pituitary hormonal abnormality. The patient showed neither recent vision impairment nor focal neurological symptoms or signs except headache.

Computed tomography (CT) revealed hyperdense shadow on sella turcica in which might suggest hemorrhage within pituitary gland. Magnetic resonance imaging (MRI) demonstrated 19×5×9 mm mass in the postero-inferior sella turcica. This lesion showed high signal on T1-weighted image and low signal on T2-weighted image (T2WI). There was no enhancement of the mass after intravenous gadolinium injection (Fig. 1).

Because of the persistent headache for several days and the suspicion of pituitary hemorrhage, trans-sphenoidal exploration was performed. Cystic mass was identified after opening the dura at the inferior aspect of the sella. Dissection around the cyst revealed a lesion with yellow-gray colored mucoid gelatinous content which looked like a Rathke's cleft cyst within the pituitary gland. There was no macroscopic hemorrhage within the cyst. Normal pituitary gland was well preserved during operation. Microscopic examination showed the cyst wall to consist of well-formed salivary acini with a low-columnar epithelium in a fibrovascular network. The eosinophilic secretions are consistent with the mucinous material found at surgery. There was no evidence of neoplasm. These findings consisted with ectopic salivary gland tissue within pituitary gland (Fig. 2).

Postoperative course was uneventful except transient mild diabetes insipidus for a few weeks. No hormonal deficiency was observed.

DISCUSSION

Salivary gland rest cyst is well-described lesion in posterior pituitary gland [1]. During normal human embryogenesis, the pituitary gland comes from both an up-growth of the lining in the oral cavity roof and a down-growth of the floor of the diencephalon [6]. The up-growth arising from the roof of the

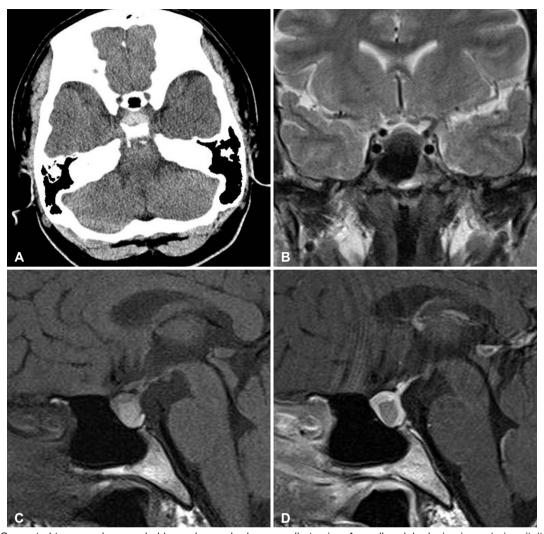


Fig. 1. A: Computed tomography revealed hyperdense shadow on sella turcica. A small nodular lesion in posterior pituitary shows low signal on T2-weighted image (B) and high signal on T1-weighted image (C). D: There was no enhancement of the mass after gadolinium injection.

mouth gives off a funnel-shaped extension called the adenohypophyseal pouch, or Rathke's pouch. The lumen of Rathke's pouch develops into Rathke's cleft. The tubular glands in the posterior pituitary which are identical to salivary glands often communicates with Rathke's cleft, and is considered to be the origin of the salivary gland within pituitary [1]. However, intracranial ectopic salivary glands other than pituitary are very rare, and few cases were reported in the cerebellopontine angle region [7].

The cases of salivary gland rests in posterior pituitary were occasionally reported in as many as 78 in 2,300 consecutive autopsies [1]. However, symptomatic salivary tissue in pituitary is exceptionally rare, only three cases were reported in the literature [3-5]. Kato et al. [3] reported an ectopic solid salivary gland tissue in pituitary in an 11-year-old boy with growth hormone deficiency. Tatter et al. [4] also reported a pituitary

symptomatic salivary rest cyst in a 22-year-old woman who present with menstrual irregularities and mild hyperprolactinemia. Chen et al. [5] reported a cystic lesion in a 28-year-old woman presenting with galactorrhea and hyperprolactinemia.

With regard to usual small size of pituitary salivary rest cyst and the rarity of symptoms, it is postulated that the lack of parasympathetic and sympathetic innervation of the posterior pituitary may prevent the secretion of mucinous material [8]. It is also suggested that local reabsorption mechanisms through lymphatics and veins remove any secretions from salivary rests in pituitary. In case of symptomatic cyst, a developmental anomaly leading to an increased mass of benign salivary tissue or clonal propagation of an abnormal cells with an autoregulatory defect favoring increased secretion is likely cause of the mucinous cyst formation [4]. Meanwhile, mucous-secreting glands have been reported in association with an intra-

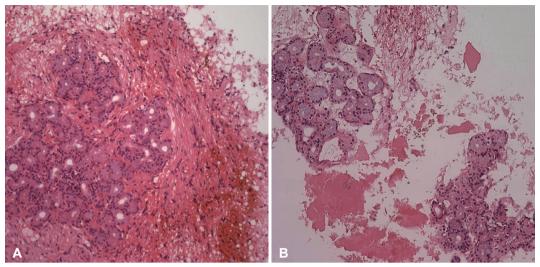


Fig. 2. A: Microscopic examination showed well-formed salivary acini with a low-columnar epithelium in a fibrovascular stroma in cyst wall. B: The eosinophilic secretions are consistent with the gelatinous material found at surgery (hematoxylin and eosin, x40).

sellar epithelial cyst [9]. Considering embryonic salivary gland structures often communicate with Rathke's cleft [2], it may represent coincidental existence of a salivary gland rest adjacent to Rathke's cleft cyst. However, in the present case, there was no evidence of Rathke's cleft cyst in histopathological specimen.

In practice, the incidence of salivary gland rest cyst may be higher than we expected. During trans-sphenoidal surgery for symptomatic Rathke's cleft cyst, surgeons usually performed simple drainage of cystic content without cyst wall biopsy. Therefore, it is possible that some salivary rest cyst may be misdiagnosed with Rathke's cleft cyst.

Radiographically, it is practically indistinguishable to identify salivary gland rest cyst from other sellar lesions, particulary mucoid Rathke's cleft cyst [10]. High density on CT and low signal intensity on T2WI MRI may be helpful to differentiate salivary gland cyst from Rathke's cleft cyst or other non-mucoid pituitary cyst in which have a low CT density and various MR intensities [10]. The absence of contrast enhancement as well as their posterior location should help to distinguish salivary gland rest cyst from most solid or cystic macroadenoma [4].

In conclusion, the incidence of salivary gland rest cyst in sella turcica may be higher than expected. Although the pituitary salivary gland rest cyst is rarely symptomatic, it should be take into consideration in the differential diagnosis of posterior pituitary lesions. Trans-sphenoidal surgical resection could be an adequate treatment for those who have symptoms.

Conflicts of Interest

The author has no financial conflicts of interest.

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