Table 1
Literature reports of intractable singultus that improved after microvascular decompression

Author, year	Sex, age	Singultus	Other deficits	Onset to surgery	Responsible vessel	Responsible neural element	Postoperative course
Johnson, 1993 [1]	Female 16 y.o	Constant	Hoarseness	2 months	A branch of PICA	REZ of vagus nerve	Recurrence after 1 year. No recurrence after 2nd surgery
Farin, 2008 [2]	Male 41 y.o	Almost constant. Less than 1 day break	None	15 years	VA and several loops of PICA	Medulla and REZ of vagus nerve	Improved. One or two 20-minute episodes one or two days per week
Present case	Male 50 y.o	Intermittent. Continued for 8–10 days, 2–3 times per month	None	30 years (Aggravated 6 months before surgery)	VA	Lower medulla	70% improved 10 months after surgery

PICA = posterior inferior cerebellar artery, REZ = root entry zone, VA = vertebral artery.

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Rathke's cleft cyst: A case report of recurrence and spontaneous involution

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ABSTRACT

Rathke's cleft cysts (RCC) are sellar lesions that typically remain asymptomatic throughout life. Symptomatic patients present with headache, visual disturbance and/or pituitary dysfunction and are treated with resection. We present a 61-year-old woman diagnosed with RCC which was resected twice then recurred before undergoing spontaneous resolution. RCC are often managed without surgical intervention. Some of these lesions may spontaneously resolve without surgical intervention while others may become symptomatic. In patients with asymptomatic recurrent RCC conservative management is recommended. Spontaneous involution may occur following initial resection and recurrence of RCC.

1. Introduction

Rathke's cleft cysts (RCC) are benign lesions that account for 1.8% of all tumors in the pituitary and sellar region [1]. The cysts develop between the pars distalis and the pars nervosa when the craniopharyngeal duct develops, with the proximal cleft closing early while the distal cleft remains open [2]. The majority will remain asymptomatic and require no intervention [3]. Spontaneous regression of cysts is uncommon with regression following recurrence being extremely rare. Herein we present a case of a 61-year-old woman diagnosed with a RCC, which recurred twice after two surgical resections and then spontaneously regressed.

2. Case report

A 61-year-old woman with no significant past medical history presented with severe right supraorbital and temporal headaches. She had been treated with antibiotics for sinusitis with minimal relief. She was neurologically intact and denied any visual changes, heat/cold intolerance, polydipsia or polyphagia. She experienced moderate weight loss over the past few months which she attributed to increased exercise. A CT scan demonstrated no sinus pathology. The MRI revealed a mass in the sella suggestive of a pituitary adenoma with minimal suprasellar extension or compression of the optic apparatus (Fig. 1A, D).

She underwent a transsphenoidal resection of the pituitary mass without complications. Histology revealed a RCC with a squamous epithelium lining with associated inflammation and fibrosis of the adjacent adenohypophysis (Fig. 2A, C). On follow up she reported no headaches or visual symptoms.

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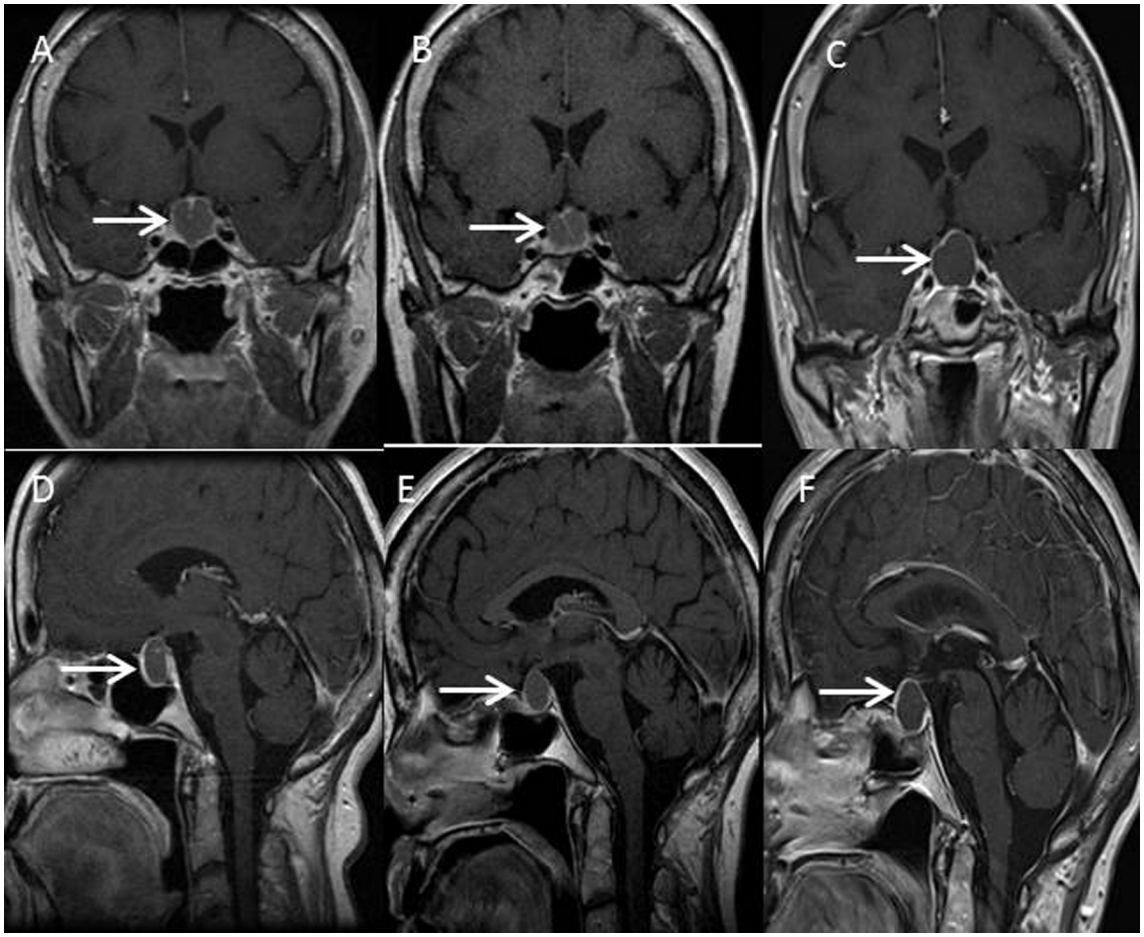

Fig. 1. T1-weighted MRI with contrast (A) coronal and (D) sagittal, at initial presentation showing a ring enhancing cystic Rathke's cleft cyst (RCC) (white arrow). T1-weighted MRI with contrast (B) coronal and (E) sagittal, 3 years after first resection showing the first recurrence of the RCC. T1-weighted MRI with contrast (C) coronal and (F) sagittal, 7 years after first resection showing increased size of the RCC with elevation of the optic chiasm.

Three years after her initial surgery, the patient's headaches returned. Her visual exam found a mild superior temporal visual field depression and MRI demonstrated a cystic lesion in the sella with suprasellar extension suggesting a recurrent RCC (Fig. 1B, E). Her neurological exams were unremarkable and she underwent close monitoring with regular ophthalmologic assessments. Four years later she returned with severe right otalgia that was relieved with antibiotics. Her headaches had also returned but she denied any other problems. MRI revealed a mild increase in the size of the cyst (Fig. 1C, F). Visual field exam revealed a subtle superior bitemporal visual field loss. She underwent an endoscopic transsphenoidal resection without complications and the post-operation MRI showed no residual mass or optic chiasm compression (Fig. 3A, D). Pathology showed chronically inflamed fibrous connective tissue with clusters of respiratory epithelial cells supporting diagnosis of a recurrent RCC (Fig. 2B, D). On follow up she had resolution of the headaches and otalgia as well as improvement of the super temporal visual field deficit.

Two years after the second resection, she was asymptomatic with no vision changes. However, MRI showed recurrence of the cyst (Fig. 3B, E). Continued serial imaging was recommended since she was asymptomatic without compression of the optic chiasm. Repeat MRI at 2-year follow up demonstrated cyst regression (Fig. 3C, F).

3. Discussion

RCC are remnants of an ectodermal pouch that failed to regress and then enlarged later in life with an unpredictable natural history [4]. RCC occurs in females and males in a 2:1 ratio most commonly occurring in the sixth decade [4,5]. These are found incidentally in 2%–26% of patients at autopsy, while symptomatic RCC account for only 1.8% of pituitary and sellar region lesions [1,4]. The most common presenting symptoms are headaches, visual disturbance, and pituitary dysfunction [2,4,6]. Additional symptoms may include pituitary apoplexy, aseptic meningitis with visual loss, sphenoid sinusitis, syncope, seizures, ataxia, mood disturbance, and precocious puberty [5].

On MRI, the cysts are in the sella or suprasellar region and are small, well circumscribed, ovoid in shape with a variable density depending on the intracystic contents [4,5,7,8]. However, the diagnosis cannot be made solely on imaging because their appearance is similar to many other pituitary lesions [2,4].

Histopathological analysis is the gold standard for the diagnosis of RCC [8]. Cyst contents can range from white or clear, to gray or green with a consistency that varies from gelatinous to serous. Microscopically, the wall is composed of cuboidal or columnar, ciliated epithelium with, occasionally, goblet cells [3,4]. Inflammation can lead to stratified or pseudostratified squamous metaplasia [5].