Int J Clin Exp Med 2017;10(7):11097-11100
www.ijcem.com /ISSN:1940-5901/IJCEM0050895

Case Report

Cavernous hemangioma with multiple phleboliths of the parotid gland in adult masquerading assialolithiasis

Bo-Nien Chen[1,2]

[1]*Department of Otolaryngology-Head and Neck Surgery, Hsinchu MacKay Memorial Hospital, Hsinchu City, Hsinchu 30071, Taiwan;* [2]*Mackay Junior College of Medicine, Nursing, and Management, New Taipei City, Taiwan*

Received January 18, 2017; Accepted May 26, 2017; Epub July 15, 2017; Published July 30, 2017

Abstract: Objectives: Hemangiomas account for 0.4%-0.6% of all tumors of the parotid gland and are extremely rare in adults. Changes in blood flow dynamics within hemangiomas result in thrombus formation and phleboliths. We present a case of cavernous hemangioma with multiple phleboliths of the parotid gland masquerading assialolithiasis in an adult patient. Methods: A 43-year-old woman visited our hospital, presenting with a 3-year history of a slowly progressive painless mass at the left infraauricular area associated with intermittent swelling episodes related to meals. Computed tomography revealed multiple calcified nodules located within the enlarged, contrast-enhanced left parotid gland. The patient subsequently underwent total parotidectomy. Results: Cavernous hemangioma with multiple phleboliths of the parotid gland was diagnosed. The patient recovered well from the surgery and was disease free at the most recent follow-up, 10 years later. Conclusions: No previous case reports have described a cavernous hemangioma with multiple phleboliths of the parotid gland. This condition requires further study because its rarity, clinical presentation, and imaging features often lead physicians to a misdiagnosis of sialolithiasis. It emphasizes that the possibility of a cavernous hemangioma should be considered in the differential diagnosis of parotid tumors when multiple intraglandular calcification nodules are observed in imaging studies.

Keywords: Cavernous hemangioma, phlebolith, parotid gland, sialolithiasis, mealtime syndrome

Introduction

Hemangiomas account for 0.4%-0.6% of all tumors of the parotid gland and are extremely rare in adults [1]. Changes in blood flow dynamics within hemangiomas result in thrombus formation and phleboliths. The changes may originate from injury to a vessel wall or result from stagnation of the flow of blood [2-4]. To date, a few reports of hemangioma with phlebolith of the parotid gland have been reported (**Table 1**) [4-6]. Herein, we present a case of cavernous hemangioma with multiple phleboliths of the parotid gland masquerading assialolithiasis in an adult patient. Based on our research, this is the first case report of cavernous hemangioma with multiple phleboliths of the parotid gland in the English-language literature.

Case report

A 43-year-old woman visited our hospital, presenting with a 3-year history of a slowly progressive painless mass at the left infraauricular area associated with intermittent 10-minute period of swelling episodes related to meals (mealtime syndrome). On physical examination, an elastic, nonfluctuating, nonpulsatile mass with a maximum diameter of 5 cm × 5 cm was palpated in the left parotid gland without trophic skin changes. Her white blood cell count and serum amylase were within normal ranges. Computed tomography (CT) revealed multiple calcified nodules located within the enlarged, contrast-enhanced left parotid gland (**Figure 1**). The tentative diagnosis was hemangioma with phleboliths of the left parotid. The patient subsequently underwent total parotidectomy. During surgery, the macroscopic appearance of the lesion was compatible with that of a vascular lesion, and there was unusual bleeding from the hemangiomatous areas during dissection. The histopathological sections of the lesion revealed dilated, thin-walled vascular proliferation with formation of numerous conglobate fibrocalcified nodules (**Figure 2**). Thus, cavernous hemangioma with multiple phleboliths of the parotid gland was diagnosed.

Table 1. Previous reports of hemangioma with phlebolith of the parotid gland

Authors	Cases	
Mandel et al. (2010)	1	56 y/o, female
Choi et al. (2013)	1	44 y/o, male, left side, tumor size: 6 × 4 × 3 cm, total parotidectomy
Gao et al. (2014)	5	4 total parotidectomy, 1 partial parotidectomy, no recurrence after 1-8 years of follow up

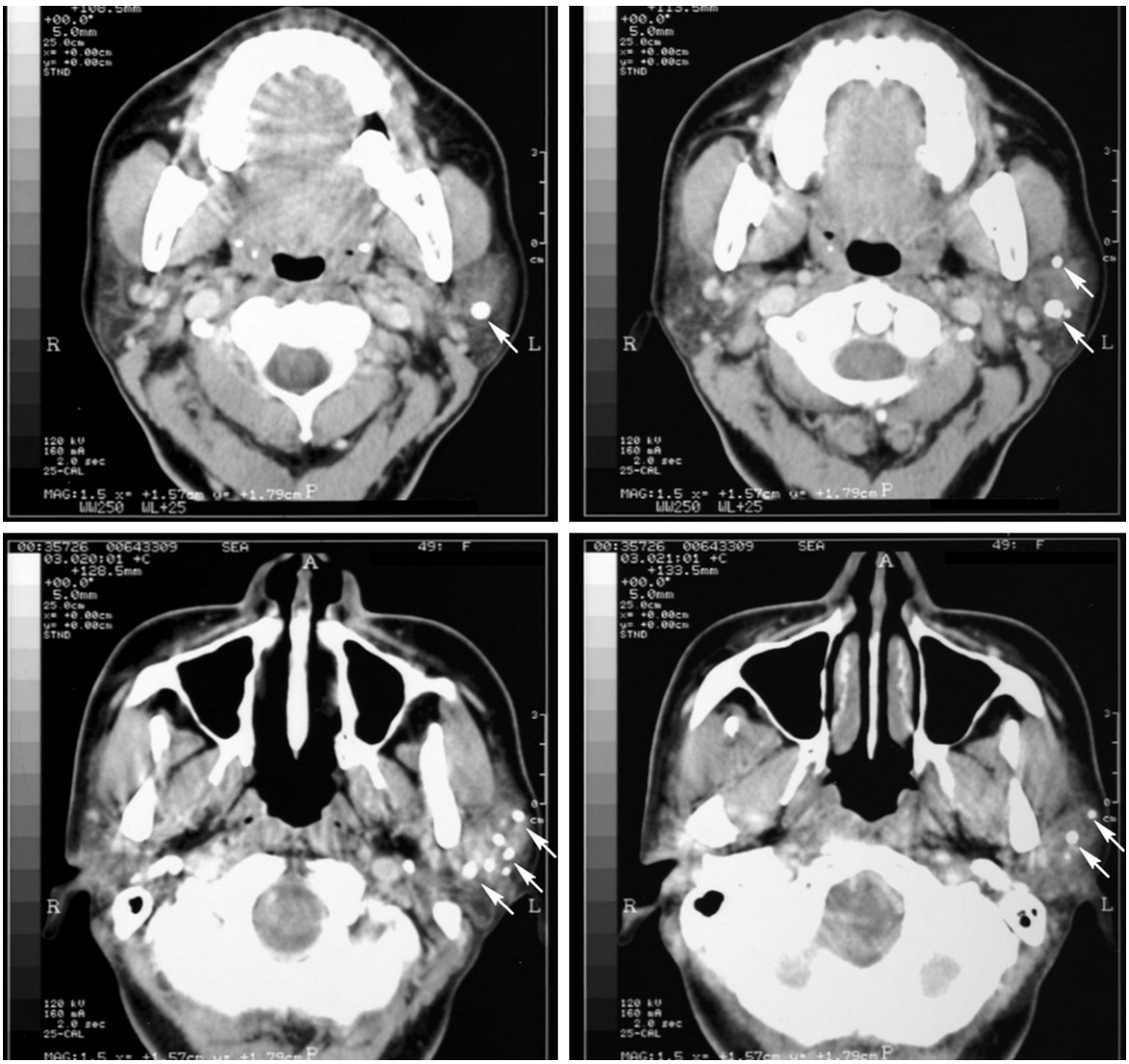

Figure 1. CT revealed multiple calcification nodules (arrows) located within the enlarged, contrast-enhanced left parotid gland.

The patient recovered well from the surgery and was disease free at the most recent follow-up, 10 years later.

Discussion

In the head and neck, hemangiomas principally affect the salivary glands, with the parotid as the most common site. Hemangiomas mostly occur in children [1-3]. Hemangiomas are classified as cavernous, capillary, and mixed hemangiomas [1]. Adult salivary gland hemangiomas are of the cavernous type, whereas infantile hemangiomas are usually capillary [2].

The classic clinical presentation of a parotid hemangioma is an intraglandular mass that may be associated with skin lesions characterized by reddish macules, and a vibration or pulsation when palpating the parotid region

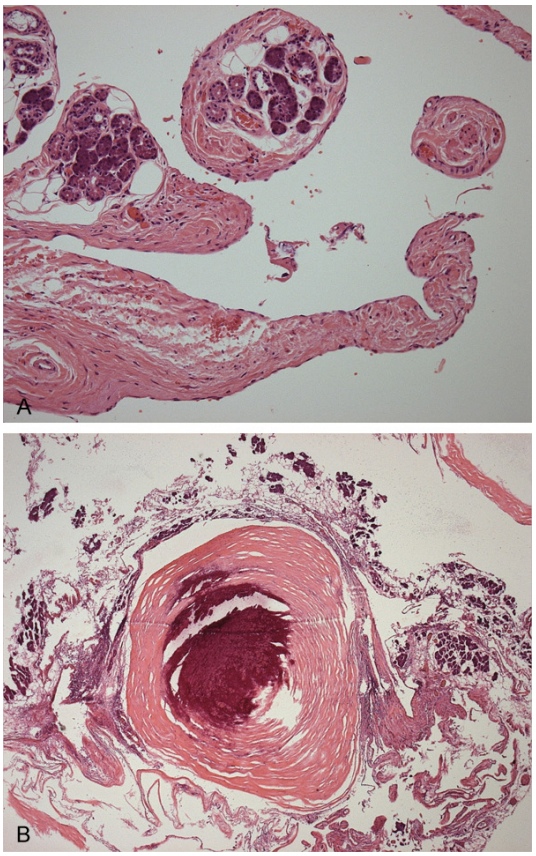

Figure 2. Histopathological sections revealed (A) vascular proliferation (H&E 100 ×) (B) with formation of conglobate fibrocalcified nodule (H&E 20 ×).

[1]. Hemangiomas occur twice as often in females as in males, and may fluctuate in size with pregnancy and menarche [2]. If these signs are absent as in the present case, diagnosis can be challenging; this is especially true in adult patients, in whom this disease is extremely rare and not usually considered in a differential diagnosis.

The findings of ultrasonographic, CT, and magnetic resonance imaging are useful in diagnosing hemangiomas preoperatively [2]. Changes in blood flow dynamics within hemangiomas result in thrombus formation and phleboliths. Imaging studies enable the observation of phleboliths that are highly suggestive of a hemangioma or vascular malformation; however, these occur in only 2%-3% of cases [1].

Moreover, hemangioma with phlebolith within the salivary gland is often mistaken for sialolithiasis in preoperative imaging studies, because sialolithiasis has a higher incidence [2-5, 7]. The presence of mealtime syndrome is also prone to leading physicians to a misdiagnosis of sialolithiasis. Sialography may be a useful method for the evaluation of radiopaque lesions localized intraglandularly in the parotid area to rule out sialoliths [5].

Fine needle aspiration cytology is not particularly helpful in diagnosing hemangioma. It carries the risk of generating a hematoma and is considered unnecessary when a typical clinical presentation and characteristic radiologic findings are highly suggestive of the diagnosis [1]. Thus, preoperative fine needle aspiration was not performed for this case because a tentative diagnosis of hemangioma was made through CT before surgery.

Because cavernous hemangiomas tend not to regress, the treatment of choice forintraparotid cavernous hemangioma is surgery, taking into consideration a presurgical embolization [1, 2]. However, other treatment options exist for infantile hemangiomas, such as endovascular sclerotherapy, intralesional or systemic corticosteroids, vincristine, and propranolol [1].

Our case offers three pertinent contributions. First, no previous case reports have described a cavernous hemangioma with multiple phleboliths of the parotid gland. Second, this condition requires further study because its rarity, clinical presentation, and imaging features often lead physicians to a misdiagnosis of sialolithiasis. Third, it emphasizes that the possibility of a cavernous hemangioma should be considered in the differential diagnosis of parotid tumors when multiple intraglandular calcification nodules are observed in imaging studies.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Bo-Nien Chen, Department of Otolaryngology-Head and Neck Surgery, Hsinchu MacKay Memorial Hospital, No. 690, Sec. 2, Guangfu Rd., East Dist., Hsinchu City, Hsinchu 30071, Taiwan. Tel: +88636119595; Fax: +88636110900; E-mail: chenbonien@yahoo.com.tw

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