



Case Report

# Cavernous hemangioma in adult parotid gland encroaching on the parapharyngeal space: A case report

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## ABSTRACT

Hemangiomas account for 0.4-0.6% of all tumors of the parotid gland. With about 50 cases reported worldwide in adults, diagnosing a hemangioma in the parotid gland can be difficult. An accurate diagnosis of parotid hemangiomas in adults before excision is challenging. We report a case of a 54-year-old woman with a left parotid mass of five years duration, which was rapidly increasing in size over the last year. Magnetic resonance imaging was suggestive of a left parotid pleomorphic adenoma invading the parapharyngeal space. Fine needle aspiration cytology was inconclusive. However, with a strong clinical suspicion of a pleomorphic adenoma, a left total parotidectomy was performed. The histopathological findings were of cavernous hemangioma. The rarity of such conditions in the head and neck region and misdiagnosis or failure to diagnose make this case more interesting to pathologists and clinicians alike. Nevertheless, hemangioma should be considered in the differential diagnosis of parotid tumors in adults.

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## INTRODUCTION

Hemangiomas are benign tumors of vascular origin characterized by the increased proliferation and turnover of endothelial cells. Hemangiomas are quite common in the body. The head and neck region account for 65% of these benign tumors involving a variety of tissues like the skin, muscles, and salivary glands.<sup>1</sup> Hemangiomas principally affect salivary glands with parotid as the most common site (81-85 %) in the head and neck. However, the frequency is only a meager 0.4% to 0.6% of all tumors in the parotid.<sup>2</sup> Parotid hemangiomas typically occur in children and account for approximately 50% of parotid tumors that develop in the

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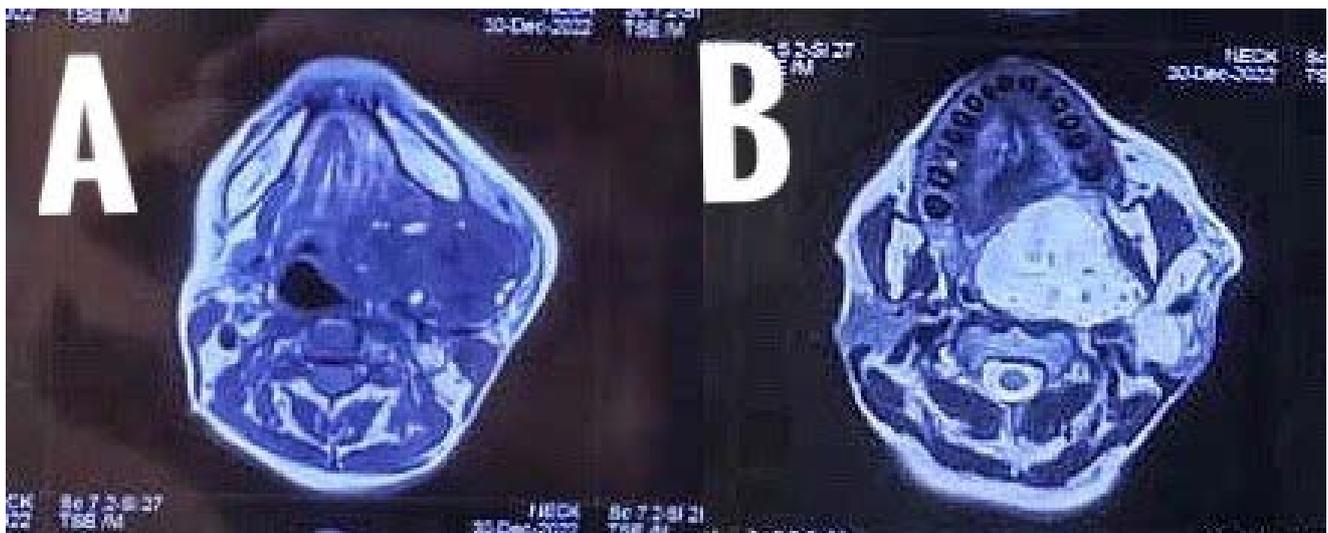
first year of life.<sup>3</sup> Most parotid hemangiomas grow during a child's first 6 to 8 months, followed by involution during the first decade of life.<sup>4</sup> By contrast, adult parotid hemangiomas are quite rare and usually do not regress spontaneously.<sup>3,5</sup> In infants, hemangiomas typically manifest as reddish skin lesions. In adults, asymptomatic swelling of the parotid gland occurs.<sup>6</sup> Due to the rarity of this condition in adults, clinical behavior may be quite misleading and less reported.<sup>7</sup> We hereby report a case of cavernous hemangioma of the parotid gland in an adult female, with an objective to add into consideration cavernous hemangiomas as part of the differential diagnosis of parotid masses in adults.

#### # CASE REPORT

A 54-year-old female presented to the otolaryngology department of Patan Hospital with a 5-year history of

swelling over the left side of the face which was rapidly increasing in size for one year. The swelling was not accompanied by pain. On physical examination, a dumbbell-shaped mass was palpated over the left parotid region. The mass was non-tender, nonfluctuating, and non-pulsatile, and the overlying skin was normal in color, texture, and temperature. No palpable mass over the right parotid area or any associated cervical adenopathy was detected.

Fine needle aspiration (FNA) and magnetic resonance imaging (MRI) were done to guide the diagnosis. The result of FNA revealed blood and its elements only. On MRI, a well-defined homogeneously enhancing dumbbell-shaped mass of 8.6 x 8 x 3.5 cm size in the left parotid space was identified involving both superficial and deep parotid gland with lobulated margin invading the parapharyngeal space, hyperintense on T2, isointense on T1. (fig.1)



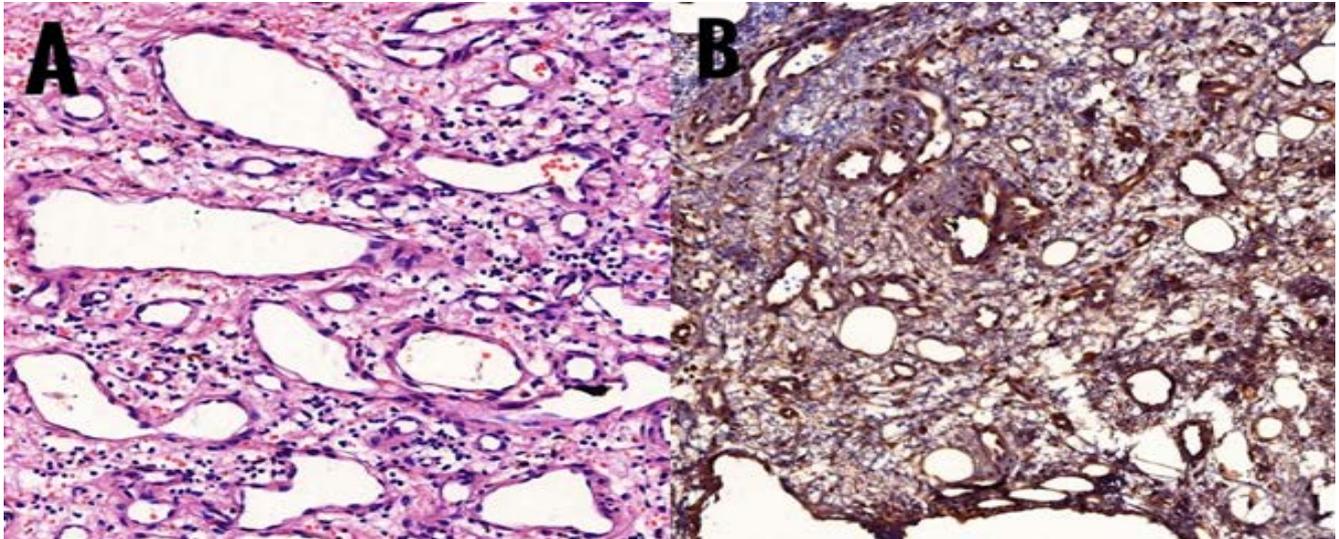
**Figure 1:** A) MRI T1 reveals a well-defined, hypointense intraparotid tumor of approximately 8 cm in the left parotid gland. B) MRI T2, shows the same image but the tumor is hyperintense.

Following a provisional clinical and radiological diagnosis of a benign parotid tumor: pleomorphic adenoma, a left parotidectomy was performed. During surgery, the macroscopic appearance was of an encapsulated and hemorrhagic, well-defined tumor suggestive of a vascular lesion. (fig.2)



**Figure 2:** A gross image of serial sections of the parotid mass, exhibiting a vascular tumor.

On microscopic evaluation, there was a well-circumscribed tumor composed of variably calibered vascular channels, mostly widely dilated and congested that showed a lobular pattern of distribution. The vessels were lined by flattened endothelium without atypia and with a thin wall supported by a dense collagenous layer. Overall findings were compatible with a Cavernous hemangioma of the parotid gland. The peripheral parotid tissue was unremarkable, with no evidence of malignancy. Further immunohistochemistry (IHC) revealed CD34 positivity in endothelial cells, supporting the histomorphological diagnosis. (fig.3) The patient is doing well on a three-month follow-up.



**Figure 3:** *A) Microscopic image showing vascular proliferation of different caliber, lined by a single layer of endothelium without atypia and with a thin wall supported by a collagen layer. (H&E 400X); B) CD34 positivity in endothelial cells (IHC, X400)*

## DISCUSSION

Traditionally, hemangiomas are categorized as capillary, cavernous and mixed, and cavernous type is the only type described in adult parotid glands so far. There is still not enough data regarding recent prevalence of parotid gland cavernous hemangioma in adults. It is considered extremely rare, with the 2005 WHO classification of tumors stated about 50 cases reported globally mostly as individual case reports.<sup>3</sup> Since then only a few cases have been described in the literature.<sup>8</sup>

The classic clinical presentation of a parotid hemangioma is the presence of a mass at the parotid region associated with or without skin lesions which can be red, red-bluish, or blue macules and/or papules, as well as with accompanying features such as vibration or pulsation when palpating the parotid region. The presence of radiological phleboliths is suggestive of hemangioma or vascular malformation. However, these only occur in 2-3% of cases and it should be differentiated from sialoliths by a sialography. If these signs are absent, similar to this case report, the diagnosis could be challenging, particularly in an adult patient in whom this diagnosis is not suspected as the main possibility.<sup>9</sup>

Nagao et al. in a series of 20 cases of cavernous hemangioma of the parotid gland, showed that there was a female predilection with a 2:1 female-to-male ratio.<sup>10</sup> It usually fluctuates in size with pregnancy and menarche. These phenomena suggested that the endothelial cells may be quite responsive to circulating hormones.<sup>11</sup> While juvenile hemangioma was predominant in males.<sup>9</sup> An observed left-sided preference has been noted in both parotid and submandibular gland cavernous hemangioma.<sup>10</sup> Hemangioma of the major salivary gland especially parotid is quite common in the pediatric population. Few published

studies have provided quantitative evidence of adult presentations of parotid cavernous hemangioma.<sup>12</sup>

Cavernous hemangiomas in adults do not regress, and they tend to have a chronic course and a slowly progressive growth.<sup>9</sup> Due to the low prevalence of hemangiomas in adults, they are not usually taken into consideration in the differential diagnosis of parotid masses. In adults, the pleomorphic adenoma and the Warthin tumor are the most common benign tumors of salivary glands.<sup>13</sup>

Magnetic resonance imaging (MRI) is useful in demonstrating lesions of the parotid region and its extension. Hemangiomas usually appear as lobulated lesions with an intermediate signal on T1 and hyperintense on T2. MRI also helps determine the surgical approach for the tumor and to reveal the relationship with adjacent structures.<sup>14</sup>

Fine needle aspiration is useful in the preoperative diagnosis of tumors of the head and neck. It is considered unnecessary in a hemangioma because of the probability of generating a hematoma and when MRI is highly suggestive of the diagnosis. Therefore, a typical clinical presentation and characteristic radiologic findings are sufficient for the diagnosis.<sup>14</sup>

## CONCLUSIONS

Cavernous hemangioma of the parotid gland in adults is rare. Nevertheless, it should be included in the differential diagnosis of salivary gland swellings. Definitive diagnosis, although difficult, can be accomplished through clinical and radiographic findings

## REFERENCES

1. Eltohami YI, Alrofaey AH, Suleiman AM. Cavernous hemangioma of the parotid gland in adults: a review of the literature & case report. *Adv Dent Oral Health*. 2018;10(2):555782. [Website](#)
2. Gampper TJ, Morgan RF. Vascular anomalies: hemangiomas. *Plast Reconstr Surg*. 2002;110(2):572-85. 10.1097/00006534-200208000-00032
3. Lara-Sanchez H, Peral-Cagigal B, Madrigal-Rubiales B, Verrier-Hernandez A. Cavernous hemangioma of the parotid gland in adults. *J Clin Exp Dent*. 2014;6(5):e592-e594. 10.4317/jced.51750
4. Childers EL, Furlong MA, Fanburg-Smith JC. Hemangioma of the salivary gland: a study of 10 cases of a rarely biopsied/ excised lesion. *Ann Diagn Pathol*. 2002;6(6):339-44. [Crossref](#)
5. To VS, Chan JY, Tsang RK, Wei WI. Review of salivary gland neoplasms. *ISRN Otolaryngol*. 2012:872982. [Crossref](#)
6. Brown NA, Elenitoba-Johnson KS. Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Hematolymphoid Tumours. *Head Neck Pathol*. 2017;11(1):96-109. [Crossref](#)
7. Suwal R, Guan J, Wang XY, Khatri KS, Nyimi BF. A Rare Case Report of Parotid Cavernous Hemangioma in an Adult. *BJMMR*. 2017;19(5):1-5. [Crossref](#)
8. Tisch M, Kraft K, Danz B, Maier H. Cavernous hemangioma of the parotid gland in adults. *HNO*. 2005;53(10):879-83. German. [Crossref](#)
9. Chaubal R, Tavri O, Sawant A, Singh C. Infantile Hemangioendothelioma of the Parotid Gland. *J Clin Imaging Sci*. 2017 Jan 27;7:5. [Crossref](#)
10. Nagao K, Matsuzaki O, Shigematsu H, Kaneko T, Katoh T, et al. Histopathologic studies of benign infantile hemangioendothelioma of the parotid gland. *Cancer*. 1980;46(10): 2250-6. [Crossref](#)
11. Halwani C, Zgolli C, Tbini M, Ben M hamed R, Akkari K. Cavernous intra parotid hemangioma: a rare etiology of parotid tumors. *J TUN ORL*. 2018;p:1-3. [Crossref](#)
12. Elsayed SA, Alahmady HH. Hemangioma in parotid gland encroaching on the temporomandibular joint: A case report. *Tanta Dental Journal*. 2014;11(2): 109-13. [Crossref](#)
13. Choi HJ, Lee JC, Kim JH, Lee YM, Lee HJ. Cavernous Hemangioma with large phlebolith of the Parotid Gland. *J Craniofac Surg*. 2013;24:621-2. [Crossref](#)
14. Kaneko K, Kanai R. Cavernous Hemangioma of the Accessory Parotid Gland. *J Craniofac Surg*. 2011;22:28-9. [Crossref](#)