

CAS CLINIQUE/CASE REPORT

CAVERNOUS HEMANGIOMA OF THE PAROTID GLAND

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ABSTRACT : In adults, hemangioma is a rare presentation of a parotid mass. In this report, we present a case of a 40-year-old lady complaining of a swelling in the right inferior parotid region. Imaging studies revealed a cystic lesion of the right parotid gland. Fine needle aspiration cytology was not significantly helpful in revealing the diagnosis.

In the operating room, a vascular cystic lesion was found in the deep lobe of the right parotid gland. Pathological analysis confirmed the diagnosis of cavernous hemangioma.

In this case report, we discuss a rare pathology in the adult population, and review different aspects in its diagnosis and treatment.

INTRODUCTION

Facial swelling involving the major salivary glands often poses a difficulty in diagnosis. If the diagnosis is merely based on clinical features and history, the clinical picture and behavior may be quite misleading, and usually suggest a benign epithelial tumor of the salivary gland or sialolithiasis [1]. Further investigation is required in order to provide greater understanding of the nature of the lesion. Still the final diagnosis may not be confirmed until the lesion is excised, and the pathological assessment is completed.

In adults, hemangioma is a rare presentation of a parotid mass with only few cases reported in the literature. Whereas both capillary and cavernous hemangiomas are seen in children, only cavernous hemangiomas have thus far been reported in adults [2].

Cavernous hemangiomas tend to be larger and less well circumscribed than capillary and juvenile hemangiomas. They do not show a tendency to regress. Most cases require surgery. Other treatment methods include vascular ligation, and corticosteroids.

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RÉSUMÉ : L'hémangiome de la glande parotide est une pathologie rare chez l'adulte. On présente une patiente âgée de 40 ans qui s'est présentée au département d'oto-rhino-laryngologie pour une masse au niveau de la région parotidienne inférieure droite. Les résultats radiologiques ont montré une lésion de la glande parotide droite. La cytoponction n'a pas beaucoup contribué au diagnostic de la lésion. Une parotidectomie a été effectuée révélant une lésion kystique avec de multiples branches vasculaires. L'analyse pathologique a montré un hémangiome cavernoux parotidien.

Cet article discutera cette pathologie rare, avec une mise au point sur les techniques diagnostiques et la prise en charge de ces tumeurs.

CASE REPORT

A 40-year-old lady presented to the ENT clinic complaining of intermittent right submandibular swelling. The patient described facial swelling that appeared intermittently at the right inferior parotid region during the past year. The swelling was described as painless, not related to meals, increasing in size as the patient bends forward, and decreasing in size in the supine position. At time the swelling just disappears, according to the patient.

On examination, the patient was found to have a soft and mildly tender mass postero-inferior to the right angle of the mandible that was approximately 3 x 2 cm in size. Facial nerve examination revealed intact function.

Ultrasound and MRI revealed a cystic lesion in the anterior third of the right parotid gland, measuring 34 x 12 mm, with an imprint on the posterior edge of the right submandibular gland. Fine needle aspiration yielded hemorrhagic material that was considered inadequate for pathological assessment.

Intra-operatively, a vascular cystic lesion was found in the deep lobe of the right parotid gland with multiple large feeding vessels. A right total parotidectomy was performed for excision of the superficial and deep lobes of the right parotid gland except for the superior part of the gland. The facial nerve was dissected and preserved.

Pathology showed a well circumscribed mass composed of multiple large dilated vascular channels containing red blood cells, as well as fibrin thrombi in the variable stages of organization. Some thrombi were centrally calcified. The thrombi were centrally calcified.

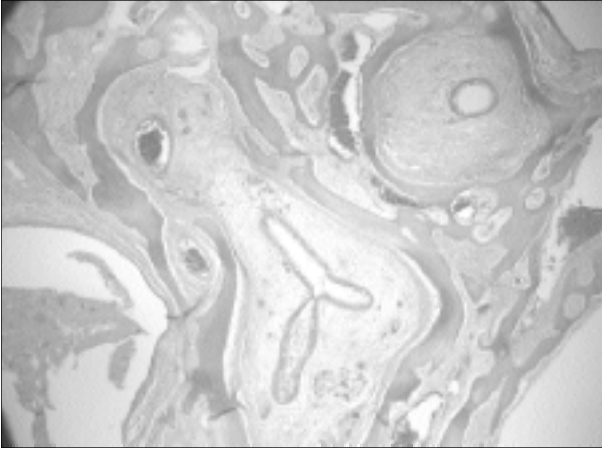


FIGURE 1. Variably sized, sometimes dilated, glands lined by respiratory type columnar ciliated cells admixed with fibro-osseous nodules.

The endothelial cells were cytologically bland with absence of multilayering. No arteries were present.

The lesion was diagnosed as a benign cavernous hemangioma of the deep lobe of the right parotid gland.

DISCUSSION

There are two interesting points in this case. First, it presents a rare pathology of the major salivary glands, namely cavernous hemangioma of the parotid gland. Second, the clinical and radiologic features which often mislead physicians into making an erroneous diagnosis of sialolithiasis are reviewed, in order to outline the findings that should promote high index of suspicion of a cavernous hemangioma of the salivary gland.

Parotid hemangiomas are rare in adults. Adult salivary gland hemangiomas are of the cavernous type, while infantile hemangiomas are usually capillary [2]. Hemangiomas may arise from the gland proper, or by invasion of subcutaneous blood vessels into the gland structure. It is currently believed that hemangiomas are benign and congenital neoplasms, which are usually undetected for long periods of time until sudden growth induces pain or cosmetic deformity.

Clinically, cavernous hemangiomas occur twice as often in females as in males. More than 90% of cases present before the fourth decade of life. They usually present as slowly growing, soft or firm, movable, painless parotid mass. Severe pain and swelling can occur, however, depending on the size of the hemangioma or in particular in the presence of acute hemorrhage or thrombosis. Cavernous hemangiomas may typically fluctuate in size with pregnancy and menarche. These phenomena suggest that the endothelial cells may be quite responsive to

circulating hormones [1].

Plain radiography demonstrates multiple calcified phleboliths in 2-3% of cases [1]. On ultrasound, hemangiomas are heterogeneous hypoechoic lesions in which calcified phleboliths are identifiable [3]. To differentiate parotid from extraparotid lesions, ultrasound has been largely supplanted by CT with contrast enhancement or in combination with sialography [4]. CT dynamic scanning may show a tumor with enhancing quality similar to that of blood vessels, depending on the rate of blood flow through the hemangioma. With magnetic resonance imaging, lesions with isointensity to muscle on T1-weighted images and characteristic hyperintensity on T2-weighted images have been reported [5]. Enlarged vessels may be seen as signal voids within and around the lesions. Nuclear medicine imaging with ^{99m}Tc red blood cell scintigraphy has also been reported to be useful in differentiating sialadenitis from hemangiomas [3].

Prior to surgery, magnetic resonance angiography or intra-arterial digital subtraction angiography should be performed to investigate the vascular supply of the tumor [6]. Surgical excision is the treatment of choice for small lesions. Large cavernous hemangiomas usually require superficial or total parotidectomy. Especially in the case of extended lesions, the facial nerve may be difficult to identify and should be monitored intraoperatively.

Hemangiomas of the cavernous variant, as seen in this case, are characterized by dilated, thin-walled vascular spaces filled with blood and lined with flattened, cytologically bland endothelial cells.

In conclusion, cavernous hemangioma with phleboliths should be included in the differential diagnosis of a swelling in the parotid and submandibular regions.

REFERENCES

1. Enzinger FM, Weiss SW : Soft Tissue Tumors, 3rd edition, St Louis : Mosby, 1995 : 579-626.
2. Mussbaum M, Tan S, Som ML. Hemangiomas of the salivary glands. Laryngoscope 1976 ; 86 : 1015-19.
3. Bradley M, Stewart I, King W, Metreweli C. The role of ultrasound and ^{99m}Tc RBC scintigraphy in the diagnosis of salivary gland hemangioma. Br J Oral Maxillofac Surg 1991 ; 29 : 164-5.
4. Som P, Biller F. The combined computerized tomographysialogram : a technique to differentiate deep lobe parotid tumors from extraparotid pharyngomaxillary space tumors. Ann Otol Rhinol Laryngol 1979 ; 88 : 590-5.
5. Levine E, Wetzel LH, Neff JR. MR imaging and CT of extrahepatic cavernous hemangiomas. Am J Roentgenol 1986 ; 47 : 1299.
6. Hamdan AL, Hadi U, Moukarbel RV, Younes A, Tawil A. Arteriovenous malformation of the parotid gland : Case report and review of the literature. Oto-Rhino-Laryngologia Nova 2001 ; 11 : 214-17.

ورم وعائي دموي كهفي في الغدة النكفية

موجز : الورم الوعائي الدموي في الغدة النكفية هو اعتلال نادر الحدوث عند البالغين. نروي سيرة امرأة عمرها ٤٠ عاماً راجعت قسم الاذن والانف والحنجرة لكتلة في الناحية السفلية اليمنى للنكفية. تبين بالصورة الشعاعية افة في الغدة النكفية اليمنى. البزل الخلوي لم يساعد على التشخيص. استؤصلت الغدة وتبين فيها افة كيسية مع عدة عروق دموية وتبين بالفحص النسجي وجود ورم وعائي دموي كهفي. المقالة تناقش هذا الاعتلال النادر وتبحث تقنيات التشخيص والاهتمام بهذه الاورام.