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A Rare Case Report of Parotid Cavernous Hemangioma in an Adult

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Authors' contributions

This work was carried out in collaboration between all authors. Author GJ designed the study. Author SR wrote the manuscript. Authors WXY, KKS and NBF helped with the final editing of the manuscript. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Hemangioma is a common soft tissue lesion in oral and maxillofacial region. However, hemangioma in the parotid gland is a rare presentation in adult population with only few reports of cavernous hemangioma within the parotid gland. Hemangiomas are classified as benign vascular tumours which are further categorized into infantile and congenital types. A 40-year old male presented with the mass on the left parotid region to the Oral and Maxillofacial Surgery Unit of second Affiliated Hospital of Jiamusi University. Magnetic Resonance Imaging (MRI) revealed the presence of vascular malformations. The mass was removed with total parotidectomy via standard lazy S incision. Histopathological examination confirmed the diagnosis of cavernous hemangioma. It is often challenging for clinicians to reach a definitive diagnosis based on clinical and/or radiographic findings as it is a rare pathology in adults.

Keywords: Cavernous hemangioma; vascular tumor; parotid gland; total parotidectomy.

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1. INTRODUCTION

Hemangiomas are vascular malformations characterized by increased proliferation and turnover of endothelial cells. Traditionally, they were categorized as capillary, cavernous and mixed [1]. Both capillary and cavernous hemangiomas are mostly reported in children, but only cavernous hemangiomas are so far reported in adults [2]. Cavernous hemangioma is characterized by large blood filled cavities and clinically presents as a slowly growing soft or firm, movable, painless masses [3,4]. According to International Society for the Study of Vascular Anomalies (ISSVA), vascular anomalies are divided into two main groups: proliferating vascular tumours (hemangiomas) and vascular malformations [5]. Recently, Hemangiomas are classified as benign vascular tumours, divided into infantile and congenital types, with further subdivision of congenital hemangiomas into non-involuting congenital hemangiomas (NICHs) and rapidly involuting congenital hemangiomas (RICHs) and partially involuting congenital hemangioma(PICHs) [6].

Among 6% of salivary gland neoplasms arising in head and neck, 80% arise in the parotid gland and out of all parotid gland tumours, 80% are benign [7]. Hemangiomas account only for 0.4-0.6% of all the tumors of the parotid gland and cavernous hemangioma of parotid gland particularly in adult is very uncommon with about 50 cases reported globally [8]. Nevertheless, hemangioma of parotid gland is most common in pediatric population representing approximately 50% of parotid tumor in the first year of life and are usually evident on physical examination. They generally show female predilection [9-11]. Due to rarity of this condition in adults, clinical behavior may be quite misleading and also less reported as they are rarely biopsied. We hereby report a case of cavernous hemangioma of parotid gland in an adult with emphasis on the clinical diagnosis and treatment challenges.

2. CASE REPORT

A 40-year old male reported with a mass on the left parotid region to the Oral and Maxillofacial Surgery Unit of second Affiliated Hospital of Jiamusi University. The patient complained of noticeable swelling below the left ear 6 weeks prior to admission and had been increasing since then without obvious clinical signs and symptoms. The patients mentioned that the mass was about ‘broad-bean’ size, hard and non-tender initially but showed a rapid progressive growth over a period of 15 days. He primarily visited a local hospital where he was treated with anti-inflammatory drugs and then referred to our hospital due to lack of improvement.

On physical examination, a firm, mildly tender, prominent oval mass about 5.0x4.5x5.0 cm size with well-defined margins was palpable in the left parotid region. The mass was hard in texture with smooth surface and reddish-blue in color. The lesion was non-pulsatile and attached to the underlying structures. There were no signs of facial nerve palsy. Intra-oral examination revealed normal parotid gland duct opening. The mouth opening was moderately restricted with Grade-II trismus and poor oral hygiene. Cervical lymph nodes were not palpable. The differential diagnosis were Pleomorphic Adenoma and Adenolymphoma.

Magnetic Resonance Imaging (MRI) showed irregular signal mixed clumps on both T1 and T2 weighted image. A coronal T2 weighted image showed hyper-intense left parotid gland with thick, tortuous vascular flow with void shadow. The radiological diagnosis was left parotid gland vascular malformations.

The treatment plan followed was a left total parotidectomy. The usual S-shaped incision about 10cm was done and the flap was raised to fully expose the parotid gland. Under general anesthesia, retrograde dissection of the facial nerve was carried out and the deep-seated, red, soft tumor of size 4.0x3.0x2.0 cm was completely removed with complete hemostasis.

For microscopic examination, the specimen was fixed in 10% buffered formalin, embedded in paraffin. The findings consisted of large, irregular shaped dilated endothelial lined channels that contained large erythrocyte aggregates.

Histopathological examination confirmed the diagnosis of cavernous hemangioma with chronic inflammatory cell infiltration inside parotid tissue.

Until a year of follow up, there was no recurrence of tumor.

3. DISCUSSION

Hemangiomas arising in parotid gland are rare in adults. The typical characteristic is asymptomatic, soft swelling below and/or in front of the ear lobule [8]. The tumor expands slowly

but sometimes sudden increase in size can lead to compression of vital structures especially at later stage [12]. Invasion is commonly by continuity with complete or partial replacement of parotid tissue[12]. In presence of red, reddish-blue, bluish or purplish discoloration of skin, vibration or pulsation while palpating the region and particularly the presence of radio-opacities representing phleboliths on radiographs, suspicion of vascular malformations(cavernous hemangioma) can be made [8,13,14]. Phleboliths should be differentiated from sialoliths and other cause of head and neck calcifications [15]. However, in the absence of such signs as in our case, diagnosis might be difficult particularly in adults and should be differentiated from major differential diagnosis of parotid gland tumor like pleomorphic adenoma and Warthin's tumour [8, 14]. Due to the low prevalence of hemangioma in adult, it is usually not considered as differential diagnosis for parotid masses [8].

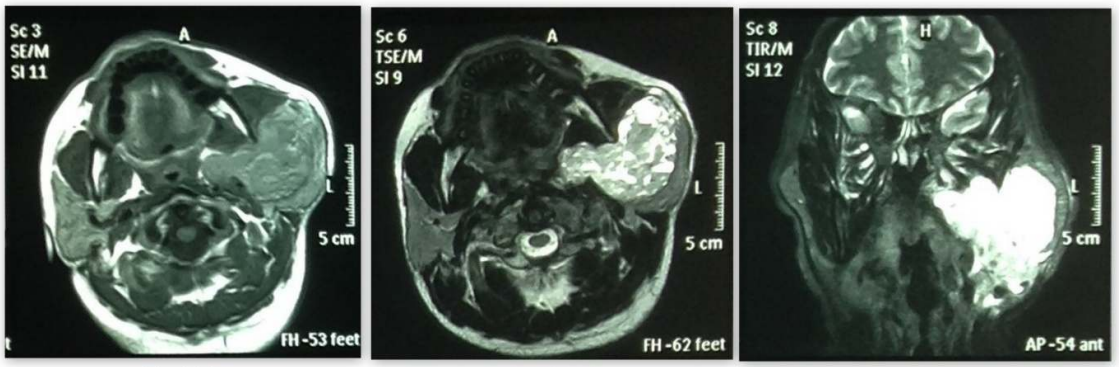

Fig:1(a) Fig:1(b) Fig:1(c)

Fig. 1. MRI image showing hypointense axial T1-weighted lesion (a) hyperintense axial T2-weighted lesion (b) hyperintense coronal T2-image (c)

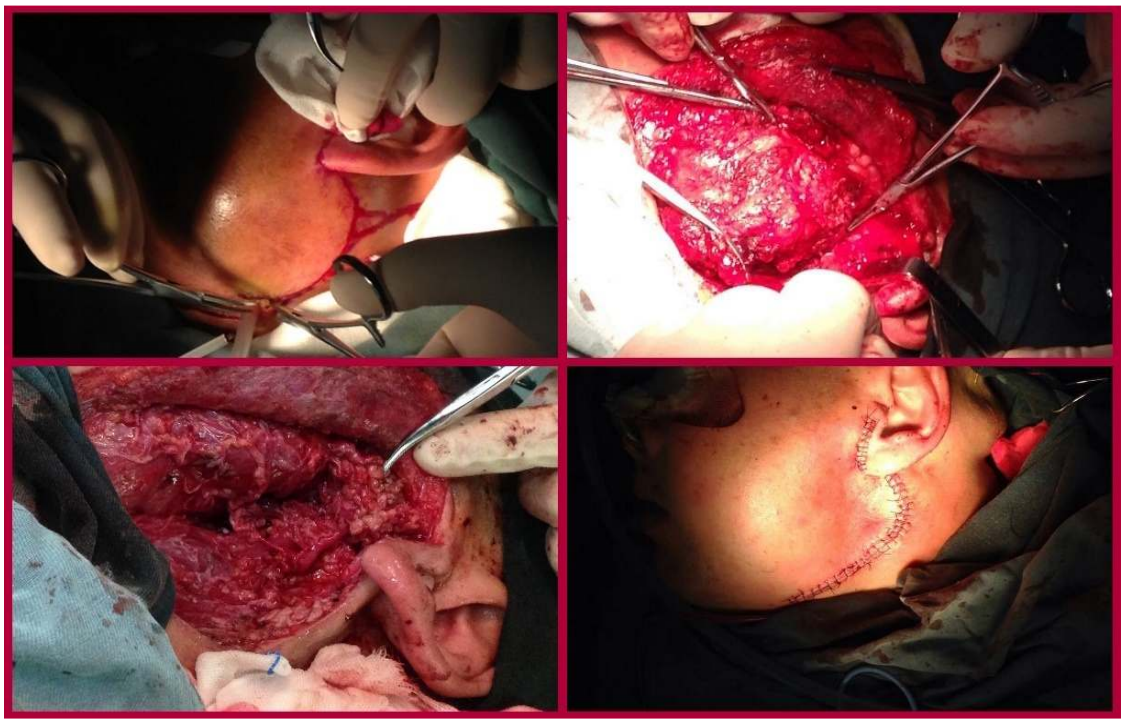

Fig. 2. Intraoperative photographs showing surgical removal of parotid gland

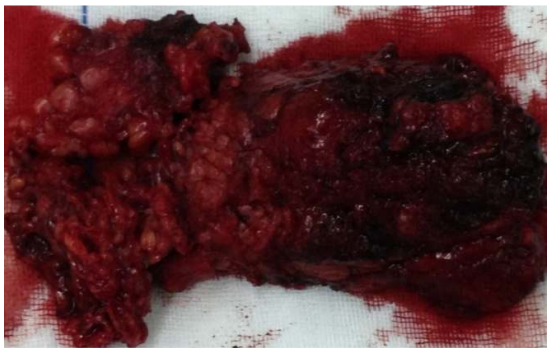

Fig. 3. Excised surgical specimen

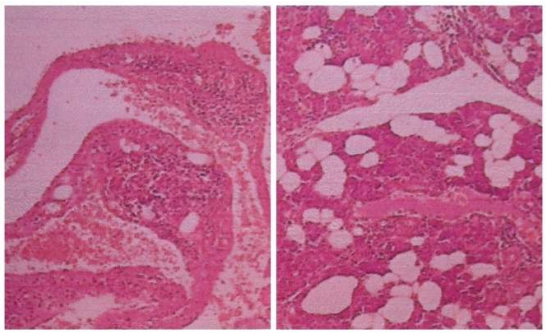

Fig. 4. Histopathological section exhibiting cohesive groups of bland spindle cells with bloody background

The clinical examination to diagnose parotid hemangioma is turkey-wattle sign (lump may get engorged when the head is bent forward or the patient lies flat). If the hemangioma lies outside the gland, there will not be any increase in the size [13,15]. The imaging techniques commonly used for investigation includes Ultrasonography (USG), Computed Tomography (CT) scan, MRI and Fine Needle Aspiration Cytology (FNAC). Ultrasonography is the first investigation of choice in salivary gland imaging [16]. MRI is regarded as the best imaging method for evaluation of parotid hemangioma and also preferred over CT scan for the evaluation of soft tissue lesions in head and neck region[4,8,9, 15,17]. FNAC is the final procedure for a definitive histologic diagnosis but it is regarded unnecessary preoperatively in case of hemangioma because of risk of generating a hematoma [8,16]. Large thin walled tortuous and dilated blood vessel, lined by flattened endothelial cells is a typical sign of cavernous hemangioma [15,18].

Surgical excision of vascular malformation in head and neck region is one of the established clinical practice [16,18]. However, recently preoperative super selective embolization is preferred to minimize blood loss in cavernous hemangioma [8,19]. However, hemangioma in children and in patients unfit for surgery are often treated conservatively using steroids, interferon, sclerotherapy, intra-lesional alcohol injection and compression therapy [8-11,20].

The etiology of hemangioma is still elusive. Controversies surround all explanation from gene mutation to developmental factors for etiopathogenesis of hemangioma. Few evidences demonstrate that dendritic cells play an important role in the formation of hemangioma through some cytokine, such as vascular endothelial growth factor while some experts concluded that hemangioma originates from embryo of angioblasts [4]. Recently, the expression of cyclooxygenase 2 (COX2) protein on endothelial cells of several vascular spaces of cavernous hemangioma have been found but has little proof with the relationship of vascular tumors [8].

4. CONCLUSION

Cavernous hemangioma is rare in adult population thereby posing difficulty for clinicians to reach a diagnosis based on clinical and/or radiographic findings. Hence, feasibility of preoperative diagnosis on clinical and radiological grounds is emphasized so that cavernous hemangioma of the parotid gland in an adult can be distinguished without complications.

CONSENT

All authors declare that written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images.

ETHICAL APPROVAL

All authors hereby declare that all experiments have been examined and approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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