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Case Report

Angioleiomyoma of parotid gland: a rare case report

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ABSTRACT

Angioleiomyoma or vascular leiomyoma first described by Virchow is a very uncommon benign soft tissue tumor. Angioleiomyoma has been defined as frequently painful, benign subcutaneous or deep dermal tumor composed of mature smooth muscle bundles which are surrounded and interlaced by vascular channels. Only 8.5% cases arise in the head and neck. A 39 years old male presented with swelling right side face and upper neck past five years. The swelling was non tender, immobile and non-compressible. There was lateral pharyngeal wall bulge which was pushing the tonsil medially. The patient underwent total conservative parotidectomy and histopathology (HPE) came out to be angioleiomyoma. Angioleiomyoma is difficult to diagnose correctly from clinical manifestations and imaging alone, and a biopsy is mandatory to make the diagnosis.

Keywords: Angioleiomyoma, Parotid, Smooth muscle actin, SMA

INTRODUCTION

Angioleiomyoma or vascular leiomyoma first described by Virchow is a very uncommon benign soft tissue tumor.[1-3] Angioleiomyoma has been defined as frequently painful, benign subcutaneous or deep dermal tumor composed of mature smooth muscle bundles which are surrounded and interlaced by vascular channels. Oral leiomyoma accounts for only 0.06% cases because the only primary source of smooth muscle here is the tunica media of blood vessels.[4] Only 8.5% cases arise in the head and neck.[5,6] World Health Organization has defined angioleiomyoma (ALM) as a histological type of leiomyoma that occurs very rarely in the oral mucosa. ALM comprise vascular endothelial and smooth muscle cells. The oral ALM is mainly found on the labial mucosa followed by tongue, buccal mucosa, palate and the retromolar trigone. Rarely, gingivae, floor of the mouth, salivary glands, larynx, uvula & tonsil are involved. Here we are presenting a rare case of ALM of parotid in a young male.

CASE REPORT

39-year-old male presented with, gradually progressive, swelling of right side face and upper neck for last 5 years. On examination, there was a 5×3 cm, firm swelling in right parotid and retromandibular region. The swelling was non-tender with restricted mobility. Swelling was not compressible and transillumination test was negative. Overlying skin was normal and there were no visible pulsations. Right lateral pharyngeal wall was displacing the tonsil and soft palate medially. The larynx and hypopharynx examination was normal. There were no neurological deficits and the cranial nerve functions were normal.

Ultrasonography (USG) neck was done which depicted heterogenous and hypoechoic lesion in right parotid measuring 3.6×2.8 cm suggestive of pleomorphic adenoma.

Contrast enhanced tomographic scan (CECT) was done from base of skull to clavicle which revealed a well-defined heterogeneously enhancing mass showing soft tissue attenuation in the right parotid space. The mass was involving both lobes of parotid gland. The mass was displacing the parapharyngeal fat and was abutting the lateral border of external carotid artery. The mass was causing extension of stylomandibular tunnel. No evidence of any invasion of surrounding structures was seen. Both coronal and axial cuts are shown below.

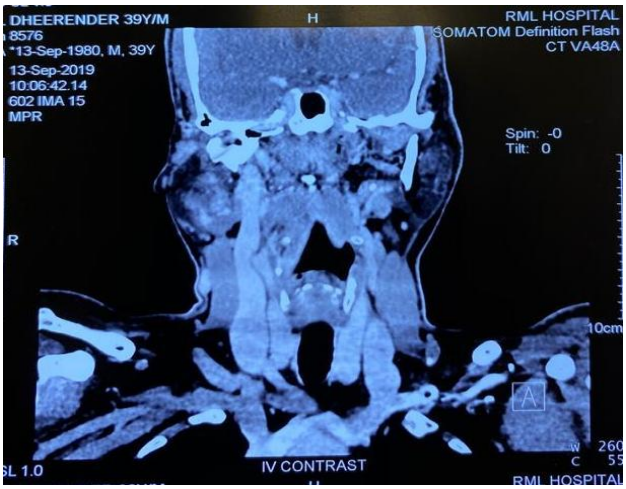

Figure 1: Coronal CT of patient showing lesion in parotid gland involving both the lobes.

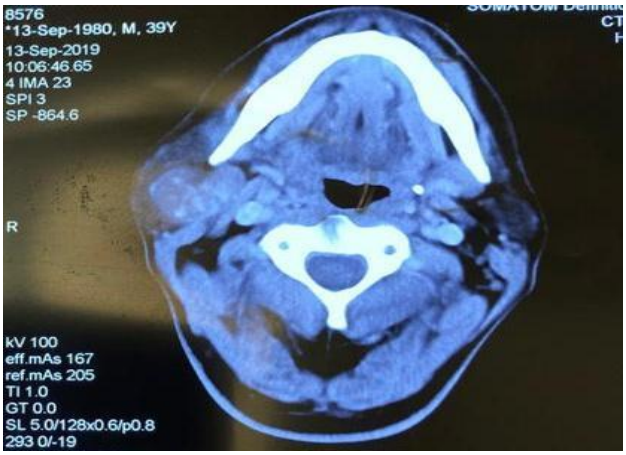

Figure 2: Axial CT scan of patient.

Fine needle aspiration (FNA) smears were moderately cellular, showing sheets of epithelial cells along with few stromal fragments, suggestive of pleomorphic adenoma.

Patient underwent total conservative parotidectomy under general anesthesia. All the branches of facial nerve along with main trunk are preserved while removal of tumor.

Histopathology (HPE) was suggestive of circumscribed tumor with many dilated vascular channels, with thick n thin walls in between fibromuscular septae. The vessel walls showed focally attenuated smooth muscle and surrounding the tumor was unremarkable serous salivary gland tissue. Smooth muscle actin (SMA) stain was positive in the septae wall as well as focally in vessel wall. The features were suggestive of cavernous type angioleiomyoma.

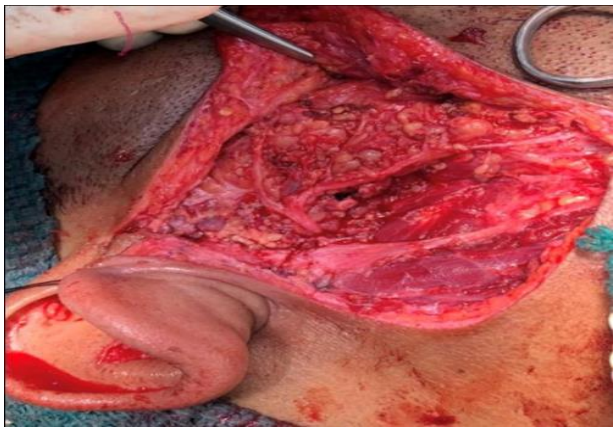

Figure 3: Intraoperative picture after removal of specimen with preservation of all the branches and main trunk of facial nerve.

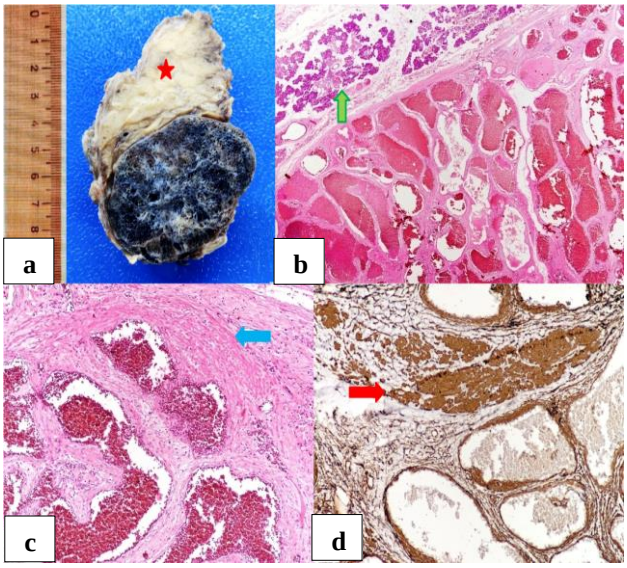

Figure 4: (a) Gross image showing normal salivary gland tissue (red star) along with a well encapsulated tumor with areas of hemorrhage and comprising of numerous cystic spaces (vascular channels); (b) microscopic examination at low magnification showing normal salivary gland parenchyma (green arrow) along with a capsulated tumor comprises of many dilated vascular channels; (c) higher magnification image showing thick and thin walls vascular spaces in between fibro-muscular stroma (blue arrow); and (d) immunohistochemistry for smooth muscle actin (SMA) demonstrating smooth muscle bundles (red arrow) and vessel walls.

The patient is under our regular follow up and he is asymptomatic.

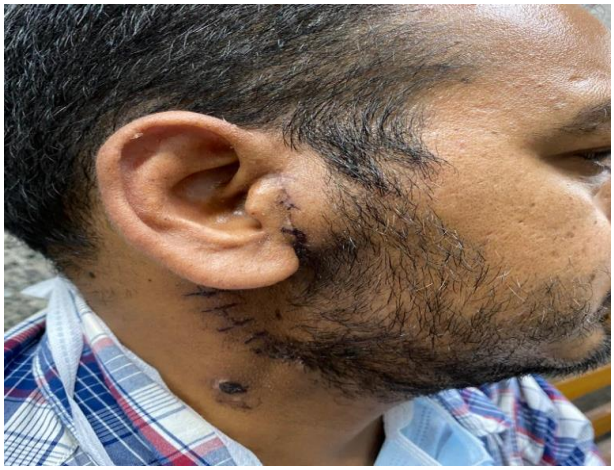

Figure 5: Post-operative picture of the patient with healthy suture line and healthy facial muscle functions.

DISCUSSION

Angioleiomyomas are extremely rare tumors, particularly in the head and neck. They are benign, solitary slow-growing tumor that originates from the vascular smooth muscles. Histologically, these can be subdivided into three types, depending on the vascular and smooth muscle components present, solid or capillary, venous and cavernous.

Morimoto classified the tumors: solid - tumor comprises of closely compacted smooth muscle and abundant blood vessels, which are small and slit-like, the smooth muscle bundles surround the vessels and interdigitate with them; venous - tumor lacks compacted smooth muscle bundles but the blood vessels have thick muscular walls of varying size; and cavernous - tumor consists of numerous dilated vascular channels and smaller quantities of smooth muscle bundles, which are difficult to distinguish from the muscular walls of the vascular channels.[7]

Morimoto further classified angioleiomyomas into two groups- larger group of extremity tumors that are frequently painful and predominantly solid and a smaller group of painless tumors on the head that are predominantly venous.[7] Lip, palate and tongue are the most common sites for angioleiomyomas in the oral cavity however salivary gland involvement is very rare.[8,9] Pain provoked by angioleiomyomas is described as paroxysmal, stinging or radiating, which are essentially the symptoms of neuropathic pain.[10,11] Only a small proportion (7.8%) of oral angioleiomyomas are painful.

Our patient had no complaint of pain. FNA was not beneficial in our case as it supported pleomorphic adenoma. Even imaging was also not helpful in this regard but it yielded information that helped in surgical planning.

Treatment option for angioleiomyoma is surgical excision only with no recurrence reported so far. Our patient underwent surgical excision that is total conserative parotidectomy with tumour removal in toto.

The differential diagnosis of angioleiomyoma includes vascular tumors (hemangiomas and lymphangiomas), benign mesenchymal tumors (lipomas, schwannomas and neurofibroma), pleomorphic adenomas and cyst. HPE remains the most reliable method for diagnosis. It is important to differentiate angioleiomyoma from other types of spindle cell tumor.[12]

Table 1: Types of spindle cell tumour and IHC marker correlation.

Spindle cell tumour	Markers		
	Desmin	CD34	S-100
Leiomyoma	-	-	-
Myofibroma	-	-	-/+
Myopericytoma	-	-	-

It is particularly important to distinguish angioleiomyomas from malignant mesenchymal tumors, including leiomyosarcoma.[13] Angioleiomyomas are predominantly composed of mature smooth muscle cells while leiomyosarcomas consist mainly of undifferentiated mesenchymal cells or fibroblast and myofibroblast like cells.[14] Immunohistochemical (IHC) and molecular markers like SMA, proliferating cell nuclear antigen, B-cell lymphoma 2, cyclin-dependent kinase 4, p53 and mouse double minute-2 homolog also allow accurate discrimination between benign and malignant smooth muscle tumors.[15] The positive expression of desmin along with SMA demonstrates the presence of smooth muscle cells. IHC vastly contributes to diagnosis of angioleiomyoma.

CONCLUSION

Angioleiomyoma is difficult to diagnose correctly from clinical manifestations and imaging alone, and a biopsy is mandatory to make the diagnosis. Immunohistochemistry and molecular markers are also very helpful in diagnosis of angioleiomyoma as we have done in our case.

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