ARTERIOVENOUS HEMANGIOMA INVOLVING SUBMANDIBULAR SALIVARY GLAND

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ABSTRACT : *We present a case of an arteriovenous hemangioma involving the submandibular salivary gland in a 20 year old girl . Hemangiomas in this region are rare. Out of the reported cases most have been cavernous hemangiomas. The rarity of an arteriovenous malformation in the submandibular salivary gland prompted us to report this case.*

Key words : *Hemangioma, Submandibular salivary gland*

INTRODUCTION

Majority of the swellings in the submandibular region are either lymph node swellings or cases of chronic sialadenitis involving the submandibular salivary gland. The clinical picture and the history gives a clue as to the etiology of the lesion. Hemangiomas involving the submandibular salivary gland are a rare cause of swelling in this region. We present a case of arteriovenous hemangioma involving the submandibular gland.

CASE REPORT

A 20 year old girl presented with a swelling in the left submandibular region since childhood, which had gradually increased to its present size. There was no change reported in the size of the swelling with meals or on bending or straining. There was no history of sudden increase in the size of the swelling.

Clinical examination revealed a solitary, firm swelling, approximately 5x3 cm in size, mobile from side to side with the overlying skin normal in colour, texture, and a temperature similar to the surrounding skin.

Fine needle aspiration cytology done twice revealed only blood. A sialogram revealed a normal duct system with no evidence of any duct calculi (Fig.I). In view of the inconclusive fine needle aspiration cytology, a doppler ultrasound was done. The ultrasound showed multiple, anechoic, poorly encapsulated masses almost totally replacing the submandibular salivary gland parenchyma. The anechoic masses showed scanty blood flow at rest but filled up with blood on performing the valsalva manoeuvre. No stone was detected on ultrasound. The

Fig. I : Sialogram showing normal duct structure with no calculi.

Fig. II : Doppler ultrasound picture showing, multiple, anechoic poorly encapsulated vascular masses with some feeder vessels

radiologic picture was suggestive of an arteriovenous malformation (Fig.II).

At surgery an ill-defined, poorly encapsulated vascular mass was found lying around and within the submandibular gland. A hard stone like structure approximately 0.5 cm

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Fig. III : Photomicrograph showing medium size vessels in close association with one another. (Haematoxylin and Eosin x 40)

Fig. IV : Vessel wall showing Calcification (Haematoxylin and Eosin x 40)

in diameter was found lying within i e vascular lesion. The mass was excised in toto and submitted for histopathological examination. Microscopic examination revealed medium and large sized arteries and veins lying in close association with one another (Fig.III). However no communication between the vessels was demonstrable in any of the sections.There was a focus of calcification in a vessel wall seen protruding into the lumen (Fig.IV). The surrounding submandibular salivary gland parenchyma showed marked dilatation of vessels and congestion. No features suggestive of sialadenitis were seen. Based on these features, a diagnosis of arteriovenous hemangioma was arrived at.

DISCUSSION

Hemangiomas are the most common lesions of the major salivary glands reported during infancy and early childhood (Batsakis JG, 1986). Salivary gland hemangiomas in the adult have been rarely reported (Nussbaum et al 1976). In a reference series from the Armed Forces Institute of Pathology (AFIP), only 1.4% of all salivary gland tumors were found to be benign mesenchymal tumors. Out of these, 30% were hemangiomas. As many as 87.5% of these mesenchymal tumors occurred in the parotid gland and only 12% in the submandibular gland (Me Menamin M et al 1997). A probable explanation of this difference is the lack of a well-defined capsule and the presence of neurovascular structures in the parotid gland.

Hemangiomas are found to occur more frequently in females. It has been observed that the left side is more frequently involved (Batsakis JG, 1986), a finding supported by the case in study.

Arteriovenous hemangiomas can be divided into two types-one occurring in deep locations and associated with varying degrees of arteriovenous shunting, the other type superficial, occurring in the dermis with no significant shunting.

The deep form occurs mostly in young persons and is regarded by some as an arteriovenous malformation (Enzinger FM and Weiss SW 1995). It is thought to be, due to partial persistence of fetal capillary bed, causing abnormal connections between the arteries and veins. These lesions are commonly seen in the head, neck and the lower extremities. Lesions, which have large shunts, may e a bruit over the mass and raised temperature of the ng skin. In some lesions there may be an increase in if the extremity with enlarged veins.

H iy, these lesions are difficult to diagnose. There ies and veins seen lying in close association tther. Serial sections may document presence tween them. At times these lesions may ipillary or cavernous hemangioma (Enzinger SW, 1995).

ave been mostly seen in soft tissue cavernous s and should be distinguished from salivary i (Hopkins R, 1969). Plain radiographs may multiple calcified phleboliths in upto 2% to (Enzinger FM & Weiss SW, 1995). In the diagnosis, ectopic oral calcification including lymph nodes, myositis ossificans and arasitic lesions must be included. A single pulebol vas found in our case from the lesion. It was missed plain radiographs as well as on ultrasound.

Ultras* d shows hemangiomas as heterogeneous hypoe ic lesions (Hopkins, R 1969). CT dynamic scanni ind magnetic resonance imaging are useful investi 'e techniques for hemangiomas.

Another differential diagnosis in such a case is an intramuscular hemangioma, first reported in the anterior belly of the digastric muscle (Slack et al, 1989).

Submandibular salivary gland hemangiomas are exceedingly rare, with a total of 13 cases reported so far (McMenamin M et al, 1997).

Most of these hemangiomas have been of the cavernous type. The clinical features, as seen in our case, can lead to an erroneous diagnosis of other benign neoplasms or chronic sialadenitis involving the submandibular gland. Hemangiomas of the salivary glands should be considered in the differential diagnosis because there is a risk of profuse bleeding per-operatively. The rarity of such a lesion in this area prompted us to report this case.

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