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Case Report

"Turkey Wattle Sign" a Very Uncommon Sign for a Parotid Hemangioma – A Case Report

Priyajeet Panigrahi*

ENT and Head and Neck Surgery Consultant, Tata Steel Medica Hospital, India

***Corresponding Author:** Priyajeet Panigrahi, ENT and Head and Neck Surgery Consultant, Tata Steel Medica Hospital, India.

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Abstract

We have reported a case of a vascular malformation within the parotid gland of an 55 YEARS elderly woman. There was no associated pain or any other related symptoms. Patient was having swollen cervical region below right parotid gland while bending head down and disappear while turning head up (turkey wattle sign). Although the hemangioma of the parotid is not uncommon in young children, it is rare in the adult. Several clues to the diagnosis have been reviewed, all absent in the case reported. The potential confusion with primary salivary pathology is emphasized by this report.

Hemangiomas of the parotid region are relatively uncommon, although they account for the majority of parotid gland tumors in infants. They are very uncommon in adults. In the presence of changes of the overlying skin (red, reddish-blue or blue), an appreciable thrill, bruit or pulsation, or multiple radio-opacities representing phleboliths on plain radiographs, the diagnosis can be suspected and arteriography carried out to delineate the anatomy of the vascular malformation. However, in the absence of such signs, the diagnosis may not be obvious, particularly in the adult patient. This report presents a case of a cavernous hemangioma of the parotid gland of an elderly adult whose diagnosis was doubtful after ultrasound but was confirmed only after looking intraoperative findings and biopsy report.

Keywords: Hemangioma; Vascular Malformation; Arteriography

Case Report

A 55-year-old while female presented at the ENT consulting room of the TATA STEEL MEDICA HOSPITAL complaining of a soft fluctuating mass lesion coming over the right cervical region while bending her head down but it disappear while elevating her head up since 2-3 years noticed by her husband. No history of associated pain.

Physical examination revealed there was no evidence of trauma. Parotid region looks normal while head in normal position but while bending head down there was a non tender, was not warm, fluctuant but non pulsatile mass appears below parotid in the cervical region. Clear saliva was expressible from the right Stensen's duct both side. There was no purulence. No mass or lymphadenopathy was palpable in the neck. There were no bruits in the neck or overlying the mass. The remainder of the examination was within normal limits. Routine admission laboratory tests were unremarkable, including a normal WBC. A screening USG showed an ill defined hypoechoic mass of heterogenous echo texture with multiple cystic spaces with in present with in the parotid tissue between the superficial and deep parotid lobe. Because the diagnosis was unclear she was admitted to the hospital planned for superficial parotidectomy and excision of the mass lesion and biopsy. The following day, the patient was taken to the operating room where, through a standard incision, the main trunk of the facial nerve was located. The cystic area within the superficial portion of the parotid was evident. As the individual branches of the nerve were being dissected distally, troublesome bleeding began, arising from deep to the plane of the nerve branches. Several large clots were noted in the field. They were partially organized rather than fresh clots. No discrete foci of bleeding could be discerned. Instead, there was a diffuse, brisk oozing from the deep portion of the parotid. It was apparent at this point that we were dealing with a vascular lesion and that bleeding was occurring through multiple rents in the vessel walls. Systematic pressure upon the parotid with over sewing of the bleeding points was carried out with difficulty. Surgical and electrocautery served as adjuncts to achieve satisfactory hemostasis. The superficial parotidectomy was completed with preservation of the facial nerve. The patient's postoperative course was benign. The drain was removed on the 3rd postoperative day and no further bleeding was noted. Pathologic diagnosis was vascular malformation of the parotid gland, consistent with arteriovenous malformation or cavernous hemangioma. The patient refused arteriography to delineate the residual lesion and continues in follow-up with no symptomatology 3 months postoperatively.

Figure 1

Figure 2

Figure 3

Discussion and Conclusion

Facial hemangiomas are relatively common in infants but relatively uncommon in the parotid gland. However, of parotid tumors in infants the hemangioma is most common. There have been multiple reports in the literature presenting series of hemangiomas of the parotid gland in children, with the findings that these lesions tend to regress spontaneously after the first 5 or 6 months of life, often completely over a few years or more gradually over a longer period. The amount of involution is generally complete by 4 to 5 years of age. When the mucosa of the lips is involved, involution is often incomplete. Cavernous hemangiomas, unlike the capillary type tend not to regress, however, the cavernous type is found in older children and adults. It is unclear whether this lesion represents a developmental process, perhaps secondary to trauma. In the adult patient, a vascular lesion in the preauricular region is more likely to be within the masseter muscle than within parotid parenchyma. Intramuscular hemangiomas are most common in the lower extremities, but in the face the masseter is most commonly involved, accounting for 4.9% of all intramuscular hemangiomas. Differentiation between a hemangioma in the masseter from one involving the parotid must be made. Arteriography and sialography are important diagnostic tools [1-24].

The hemangioma in the parotid region in children is a widely recognized entity. Because it is uncommon in adults, this tumors rarely entertained in the differential diagnosis of parotid masses. The initial diagnosis may be incorrect in many instances, as emphasized by several authors and there may be confusion with recurrent parotitis, neoplastic or cystic lesions of salivary gland origin, or masseteric hypertrophy. Our search of the literature revealed 20 cases of parotid hemangiomas in adults, aged 16 and older. Reports of facial hemangiomas in the parotid region but not clearly within the substance of the gland were excluded. Distribution between the sexes was essentially equal and the mean age was 37.5 years. Diagnostic aids include the finding of phleboliths in the parotid region, especially if multiple. Their presence is highly suggestive of a vascular malformation although they occur in perhaps 2-3% of angiomas in general. Distinction from parotid sialoliths is aided by sialography which will show, in the instance of parotid stones, faint opaque lesions in a background of sialectasis. In addition, the parotid stone tends to be lamellated. Many studies suggest that a mass or swelling that becomes more prominent with clenching of the jaw should lead one to consider the presence of a vascular lesion of the parotid. However, distinction from masseteric hypertrophy or hemangiomas of the masseter muscle would be difficult based on this sign. Sialography may be helpful in that the mass can be demonstrated as being separate from the ductal tree9, although a mass within the parotid surrounded on all sides by parenchyma could represent a cystic lesion. The "turkey wattle sign" refers to the increased swelling of a facial hemangioma when the head is placed in a dependent position, resulting in further vascular pooling. This finding seems to be relatively uncommon. The case presented in this report represents an error in diagnosis. Our preoperative diagnosis was a benign salivary neoplasm or cyst. There were no signs

suggestive of a vascular lesion of the parotid. In retrospect, aspiration of the mass may have been helpful. However, all of the findings were consistent with a cyst or tumor of the parotid gland. Several authors have emphasized potential confusion of parotid vascular lesions with salivary gland pathology, as was the situation in our case. Despite the apparent cystic nature of the lesion in the present report, malignant processes may present as apparently benign lesions, a point emphasized by RICHARDSON who advocate that the proper approach to any salivary gland lesion, whether solid or cystic, is to remove it with a "generous margin" of apparently normal parenchyma. We followed this general principle in our decision to resect the mass. Other than the clinical and radiographic aids to diagnosis, Doppler studies may be of value in that arterial sounds may be distinguished from venous sounds and each in turn from arteriovenous fistula sounds. Perhaps more than as a diagnostic aid, the Doppler can be useful to follow the progression of a given lesion as may be appropriate in this case in which the entire lesion was not excised. CT angio/MRI angio definitely have more diagnostic value but cost factors restrict these investigations further often.

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