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Case Report

RECURRENT SWELLING IN THE PAROTID REGION: A CASE REPORT

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A B S T R A C T

Lipoma is the most common neoplasm of mesenchymal origin. Inspite of being common, only 13% of them arise in the head and neck region and most of these occur subcutaneously in the posterior neck. Rarely, they can develop in the anterior neck, infratemporal fossa, and in or around the oral cavity, pharynx, larynx, and parotid gland. Clinically, they can be confused with other benign lesions; however, ultrasonography allows a specific diagnosis to be made in virtually all cases. This case report highlights a recurrent swelling situated on the parotid gland later diagnosed as lipoma with the literature review including etiology, clinical presentation and management.

INTRODUCTION

Lipoma is the most common benign soft tissue mesenchymal neoplasm with a prevalence rate of 2.1 per 1000 people.[1] It occurrence are mostly in the fifth and sixth decades of life.[2] Most placements of areas of lipomas are the arm, shoulder, back, legs, forehead and face.[1] The most common location of lipoma extraorally is the posterior triangle and intraorally is the tongue followed by buccal mucosa and lip. Rarely these tumors reach huge size (greater than 10 cm) to call a giant lipoma.[1] Recurrence in lipomas is considered rare, some authors reported it was from 1% to 5%.[14] Here we describe a case of recurrent lipoma present superficial to the right parotid gland.

CASE REPORT

A 35year old male patient reported to the department with a chief compliant of painless swelling (Fig 1) below right ear since 3 years. The patient reported the swelling to be insidious in onset, and gradual in progression. Past history revealed swelling in the same region 6 years back (with no previous records) which was excised and had again recurred 3 years back. On examination, a diffuse, solitary swelling was present below lobule of right ear, in the region of angle of mandible. It measured 4 x 4 cm in greatest diameter, roughly round in shape, extended anterior to angle of mandible by 1cm and superior-inferiorly from lobule of ear to 2cm inferior to lower border of mandible and had smooth surfaces and normal skin. On palpation, swelling was soft in consistency, fluctuant with slip sign positive.

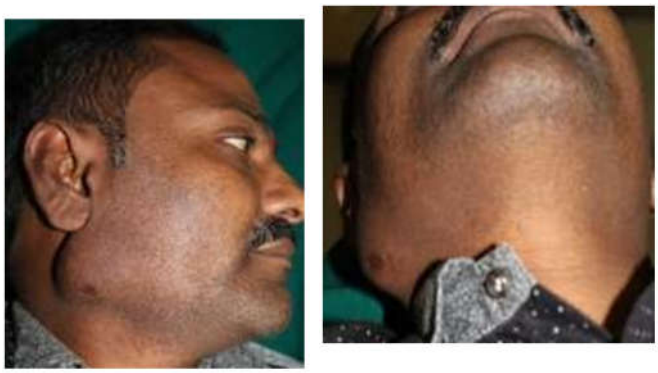

Fig 1 A diffuse, solitary swelling was present below lobule of right ear , in the region of angle of mandible.

Based on the above findings a provisional diagnosis of Recurrent Benign Tumor of Right Parotid gland was given. A differential diagnosis of Lipoma, Pleomorphic adenoma and Parotid Lymphadenopathy was considered.

Lipomas are the most commonly encountered benign mesenchymal tumors, arising in any location where fat is normally present. Concerning the age of onset, lipomas are most common from the fifth to the sixth decades of life. Males are 10 times more affected than females and manifest clinically as a slow growing, painless mass.

Pleomorphic adenoma (PA), is also known as benign mixed tumor, is the most common salivary tumor, constituting up to two-thirds of all salivary gland neoplasms. Mostly, they are located in the parotid glands (85%), minor salivary glands (10%), and the submandibular glands (5%). The age of occurrence is 4th -6th decade of life, and are more common in females than in males, the ratio approximating 6:4. Within the Parotid Gland, they occur most commonly in the superficial lobe. They manifest clinically as a slowly

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progressing asymptomatic, parotid gland swelling without facial nerve involvement. On Palpation, it is firm in consistency without presence of slip sign.

Fine Needle Aspiration Cytology (FNAC) was done and 0.2 ml of hemorrhagic fluid was aspirated. Smears showed presence of mature adipocytes along with few spindle cells.

Patient was subjected to ultrasonography of right parotid region, which revealed an ill defined non encapsulated lesion with echogenicity and echopattern similar to that of fat, continuous with the adjacent subcutaneous plane and superficial parotid gland.(Fig:2)

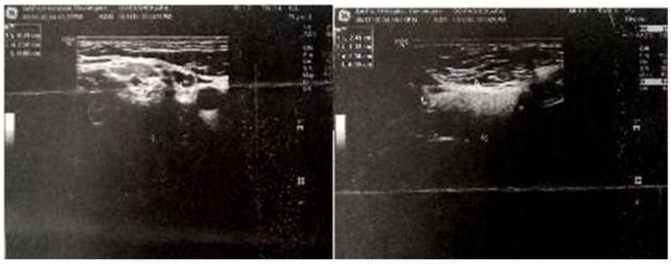

Fig 2 Ultrosonography of right Parotid gland shows an ill defined non encapsulated lesion with echogenicity and echopatternsimilar to that of fat, continuous with the adjacent subcutaneous plane and superficial parotid gland.

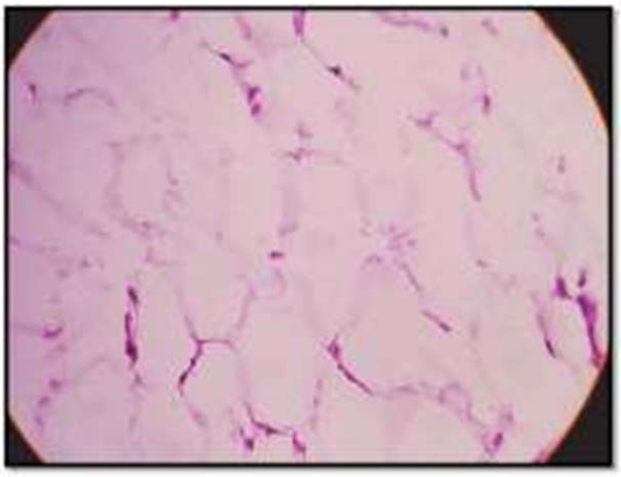

Fig 3 Histopathologic findings revealpresence of adult adipocytes were seen interspersed with connective tissue stroma and thinned epithelium.

The lesion was excised in to. On histopathological examination,(Fig:3) it was reported as lipoma where adult adipocytes were seen interspersed with connective tissue stroma and thinned epithelium.

Based on the above investigations, the **final diagnosis of Recurrent Lipoma of the Right**

Parotid Gland.

DISCUSSION

Lipomas are the most common neoplasms of mesenchymal origin, arising in any location where fat is normally present.[1] Lipoma is a benign tumor arises from fat cells of adult type or mature fat cells. The typical lipoma is soft, rubbery lump located just beneath the skin. They are slow growing and usually painless, being subcutaneous swelling it is freely mobile. Lipoma could be single, multiple and rarely uncapsulated and sometimes painful (neuro lipomas –eg- Dercum's disease). [3]

The etiology of lipomas in general is unknown. Possible theories implicate obesity, trauma, and genetics as contributing to their formation. Lipomas are more common in obese individuals. Although tumor size may increase with weight gain, tumor size does not decrease during periods of weight loss. Two explanations linking trauma to lipoma formation exist. One is that inflammation following trauma may induce lipoma formation through the release of growth factors and cytokines. The other is that necrosis of fatty tissue may cause pre- adipocytes to differentiate and form lipomas. Finally, there may be a genetic component to lipoma formation. Individuals that present with multiple lipomas often report a positive family history. Familial cases of angiolipomas, a variant of lipomas, have reported an autosomal dominant inheritance. Studies in cytogenetics have linked translocations and inversions involving regions 12q13–15 and 6p13q to lipoma formation anywhere on the body.[4] A genetic link has been demonstrated that cites that about two-thirds of lipomas exhibit genetic abnormalities. In a subgroup of lipomas, the HMGA2 gene (located on 12q14.3) was involved in tumor pathogenesis.[8]

The following structural rearrangements of chromosomes have been associated with lipoma occurrence:[8]

12q13-15 region (65%)
13q portion loss(10%)
6p21-23 region (5%)

Other loci anomalies or normal karyotype.

These benign lesions are rare in the first two decades of life, usually developing in the fifth and sixth decades when fat begins to accumulate in inactive, under-exercised individuals. Males are 10 times more affected than females. Prevalence is 1%. 15% of lipomas are found in head and neck. Annual incidence of 1 in 1000 individuals has been reported. They comprise about 0.6% of benign neoplasms of larynx and hypopharynx.[8] Of those lipomas that occur in head and neck region, the most common location is posterior neck. Most of it is present in posterior cervical subcutaneous tissue. Below the clavicles, lipomas are more common in obese female patients over 40 years of age; however, in the head and neck region, men in their seventh decade are most often affected.[1] In a series of 25 cases of head and neck lipomas reported by Ahuja *et al*.[5], 17 cases were in men (68 per cent). Som *et al*.[6] had 11 men out of 21 cases (52 per cent) in their series. Lipomas account for approximately 16% of soft-tissue mesenchymal tumors.[8].Their occurrence in the head and neck is rare, even more at the level of the parotid region where they can be found nearby the parotid capsule, inside the capsule, or within the gland. In addition, lipomas involving the deep parotid lobe are extremely unusual.[9]They grow slowly, usually without symptoms. Lipomas appear as a slow-growing, painless, mobile and well-differentiated mass in the parotid region.[10] In most cases, patients claim surgery for cosmetic concerns. Deep lobe parotid lipomas may extend between the sternocleidomastoid and digastric muscles, causing an asymptomatic parotid region soft swelling. In other cases, they may extend to the parapharyngeal space, causing medial displacement of the lateral pharyngeal wall; facial nerve involvement and pain are uncommon and have been rarely described.[9] Lipomas are nonpainful, usually round, mobile masses with a characteristic soft, doughy feel on palpation, with the skin over them often feeling cool because

of the insulating quality of fat. Although most superficial subcutaneous lipomas can be suspected with a high degree of accuracy by clinical examination alone, very large, deep-seated or infiltrating lipomas, as well as lipomas arising from unusual regions within the head and neck, require imaging for further assessment and diagnosis.[1] The incidence of lipoma among parotid tumours ranges from 0.6 to 4.4 per cent, with most series reporting an incidence of approximately 1.0 per cent. Lipomas of the parotid region can be classified into periparotid lipomas (those tumours that are found to be compressing the lateral surface of the parotid gland) and intraparotid lipomas (tumours that are totally surrounded by salivary tissue).[1] Most parotid lipomas are found in relation to the superficial lobe, with deep lobe lipomas being exceedingly rare. Kimura *et al*.,[11] in a recent review, found only five cases of deep lobe parotid lipomas reported in the literature. In the above case lipoma was present in the right Parotid region superficial to the gland. Intraoral lipomas represent 0.6% to 2.2% of all lipomas, and they account for 1% to 4.4% of all benign intraoral neoplasms. They tend to be solitary, soft, mobile, and superficially located in the buccal mucosa (cheek), tongue, floor of mouth, and buccal sulcus; they are less commonly seen in the gingiva, lip, and palate. Intraoral lipomas are most frequently diagnosed in the 4th and 6th decades, but they have been recognized at all ages.[12]

To assist with clinical diagnosis, ultrasound, computed tomography (CT), or MRI may be utilized. Generally, ultrasound may provide the most rapid assessment and can be used if a patient cannot tolerate an MRI. However, CT and MRI provide a more detailed three- dimensional picture of the tumor in relation to local neurovascular structures.[4] Sonography has been used as an initial imaging study in cases suspected to have head and neck lipomas. The characteristic sonographic appearance is that of an elliptical mass parallel to the skin surface, which is usually hyperechoic to adjacent muscle and contains linear, echogenic lines at right angles to the ultrasound beam. However, lipomas may be sometimes isoechoic or even hypoechoic relative to adjacent muscle and therefore sonography features are less pathognomonic than other, more sophisticated imaging modalities.[1]

A CT scan provides a definitive diagnosis of lipoma in virtually all cases by calculating the actual density of the suspected mass (via the CT attenuation number). The CT attenuation number is related to the radiodensity of a lesion. The attenuation number of water is set arbitrarily at zero. Bone, being radiodense, has a high attenuation number (þ1000) whereas air has a very low number (21000). The CT attenuation number of most soft-tissue tumours would be between 0 and 100. Fat, being the only soft tissue with a density less than water, has a negative CT attenuation number. Thus, lipomas have the typical CT characteristics of a homogeneous mass with few septations, a low CT attenuation number (usually measuring between –50 and –150 Hounsfield Units (HU)) and no contrast enhancement.3,14 Magnetic resonance imaging can also accurately diagnose lipomas pre-operatively, with typical signal intensity patterns simulating that of subcutaneous fat (i.e. high signal intensity on T1-weighted images and intermediate intensity on T2-weighted images, with a weak signal on fat-suppressed images). Moreover, the margin of a lipoma is clearly defined by MRI as a 'black-rim', enabling one to distinguish lipomas from surrounding adipose tissue, a distinction that cannot be made from CT images.[1]

Histologically, simple lipomas consist of mature adipocytes with uniform nuclei and scanty connective tissue; separated by thin fibrous septa and blood vessels may be present within the fibrous septa. Typically, a lipoma is surrounded by a fibrous capsule. Other variants of lipomas include neural fibrolipomas, intramuscular and intermuscular lipomas, angiolipoma, and spindle cell lipoma. Neural fibrolipomas are composed of fibrofatty tissue that surrounds and/or infiltrates local nerves. Lipomas that have a mixture of adipose tissue and skeletal muscle are considered intramuscular and/or intermuscular lipomas. Angiolipomas show an increased vascular component that can range from 5 to 50% or more of the tumor. Unlike most lipomas, angiolipomas typically present as multiple, painful masses. Spindle cell lipomas consist of spindle cells and adipose tissue. Typically, they arise in the subcutis and may extend into the dermis. [4] Fibrolipomas, however, consist of fat cells interspersed in broad bands of dense connective tissue. Simple lipomas have no site, age, or sex predilection unlike fibrolipomas which are more frequent in the cheek mucosa and show a slight female predominance. Angiolipoma is a rare histological subtype seen due to overgrowth of vascular tissue and usually affects adolescent males and subjects in their early 20s. Myxoid lipomas of the oral cavity are rare. Microscopically, these lipomas were well-circumscribed and contained adipocytes of variable size and myxoid areas.[8]

The accepted treatment protocols for subcutaneous head and neck lipomas include surgical excision through incisions made in the skin overlying the lipoma, as well as liposuction-assisted removal. Using endoscopes for better vision within the cavity has also been described to further enhance liposuction removal in some anatomic locations. Liposuction techniques are attractive because of the potential for improved cosmetic results. However, this technique is not without associated risks, including skin irregularities such as dimpling paraesthesias and numbness, pigmentation changes, oedema, and a higher risk of recurrence. All our subcutaneous lipoma cases were surgically resected through appropriate skin incisions, with no complications. Skin incisions were properly placed and meticulously closed, achieving an acceptable cosmetic result.[1]

A 5% rate of recurrence has been reported among patients with simple lipomas, and these recurrences are generally attributed to inadequate excision.[12] .The recurrence rate is highest with infiltrating type of lipomas and is estimated to be between 3% and 62.5%. The propensity for recurrence is probably due to the infiltrative nature of this type of tumor and the great difficulty in achieving a complete surgical excision.[13]

Lipomas usually occur sporadically, but rarely they can be associated with several inherited disorders, including hereditary multiple lipomatosis, Gardner's syndrome and Madelung's disease. While solitary lipomas are generally more common in women, multiple tumours (referred to as lipomatosis) are more common in men. Hereditary multiple lipomatosis, an autosomal dominant condition, is characterized by widespread, symmetric lipomas appearing most commonly over the extremities and trunk. Lipomatosis may also be associated with Gardner's syndrome, an autosomal dominant

condition involving intestinal polyposis, cysts and osteomas. The term Madelung's disease, or benign symmetric lipomatosis, refers to a peculiar distribution of fatty tissue around the cervical region, shoulders and proximal upper extremities. Patients with Madelung's disease, often men who consume alcohol, may present with the characteristic 'horsecollar' cervical appearance. These subcutaneous deposits of fat are non-encapsulated and poorly circumscribed, with tongue-like extensions of fat between muscle groups. Occasionally, these patients experience swallowing difficulties, respiratory obstruction and even sudden death.[1]

CONCLUSION

Lipoma is an extremely rare entity in the region of the Parotid Gland. With a maximum prevalence rate of 4.4%, they can be successfully treated by complete surgical excision. However a 5% recurrence rate has been reported. This case report highlights the importance of identifying lipomas from other benign tumors that occur in the region of Parotid Gland.

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