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CASE STUDY

SOLITARY CERVICAL LYMPHATIC CYSTS IN ADULTS

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ABSTRACT

Solitary lymphatic cysts in neck of adults is a very rare clinical entity, as 90% cases of cystic hygromas are present in children below 2 years. Most of the solitary cystic hygromas in adults are benign and complete surgical excision is the preferred treatment. However, in all patients above 40 years of age, a solitary cystic lesion in neck must be thoroughly investigated to rule out presence of malignancy such as occult metastasis from papillary carcinoma of thyroid or squamous cell carcinoma arising from Waldeyer's ring. We present a case of solitary lymphatic neck cyst in adult, along with a comprehensive review of the literature.

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INTRODUCTION

Neck presents a bewildering museum for a variety of solid and cystic masses in all age groups. A surgeon is occasionally confronted with a solitary cystic lesion in neck of adult patient. Apart from the common benign clinical presentation, the occasional presence of malignancy should be kept at the back of mind especially in adult patients over 40 years of age. A case of adult cervical lymphatic cyst is presented here along with guidelines of management and review of literature.

Case Report

A 32 year old female patient presented with a painless swelling in right upper part of neck for 10 years. There was no history of major illness or any constitutional symptoms. On examination, a nontender, soft, fluctuant, transilluminant oval swelling, of 3"x 2" was noticed in the right submental and submandibular triangle of neck with no evidence of enlarged regional nodes. All hematological and radiological investigations were normal and USG showed a fluid filled cyst encroaching onto carotid sheath posteriorly and submandibular gland superiorly. FNAC revealed clear fluid with few epithelial cell debris. Complete excision of the swelling was done along with its wall, taking care of the surroundings (Fig – 1). Histopathological examination confirmed the nature of lymphatic cyst. Postoperative course was uneventful.

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DISCUSSION

Cystic masses of neck include a wide range of congenital and acquired lesions and can be classified (Mahesh Kumar Mittal, *et al.*, 2012) as – (A) Cystic Masses : Branchial cleft cysts, Thyroglossal cysts, Lymphangiomas, Dermoid and epidermoid cysts, Thymic cysts, Laryngocele, Bronchogenic cysts, Ranulas (B) Cyst-like masses : Cystic metastatic lymph node (including papillary carcinoma of thyroid), Neurogenic lesions (Schwannomas and neurofibromas), Vascular lesions (AV malformations, pseudoaneurysms), Infective lesions (Tuberculous lymphadenitis, abscess), Cystic lesions of salivary glands (silocysts, cystic tumours) and cystic lesions of thyroid gland (cystic degeneration of multinodular goiter, suppurative thyroiditis).

Lymphangiomas are benign cystic malformations of lymphatic vessels, arising from early sequestrations of embryonic lymphatic channels, commonly developing along jugular chain (Mohammad Ahmed Thandar and E Jonas, 2004) and classified into cystic hygroma, cavernous lymphangioma, capillary lymphangioma and vasculolymphatic malformations (Mahesh Kumar Mittal *et al.*, 2013). Cystic lymphangioma or cystic hygroma is a subtype of lymphangioma characterized by large macroscopic cystic spaces histologically believed to arise from a congenital malformation of lymphatic system leading to failure of communication between lymphatic and venous pathways causing accumulation of lymph (Gow *et al.*, 2011). Most of them arise in utero or in infancy and hence 50% of

them are diagnosed at birth while 90% diagnosed before 2 years of age (Qin Zhou *et al.*, 2011).



Fig. 1. Operative photograph showing the lymphatic cyst in neck

They mainly occur in head and neck (tongue, lips, buccal mucosa and neck being the common sites), but can be found on thoracic wall, shoulder, pharynx and mediastinum (Veeraraghavan *et al.*, 2009). 75% of them occur in neck, either in posterior triangle or submandibular region, the cysts forming them varying in sizes from few mms to several cms. They are infiltrative, often extending into facial planes, even axilla or mediastinum (Mahesh Kumar Mittal *et al.*, 2013). Sudden enlargement may occur due to haemorrhage or infection. Usually they are soft, fluctuant and brilliantly translucent lesions. The vesicles may appear red or yellow blisters, filled with blood or purulent fluid when accompanied with haemorrhage or infection. They can be divided into macrocystic or microcystic clinical types, but mixed variety is usually present clinically (Qin Zhou *et al.*, 2011). Histologically, they show multiple dilated locules or sacs, lined by single layer of endothelium supported by connective tissue stroma, interspersed with lymphocytic follicle cells. Macrocystic lesions are localized, composed of large cystic cavity, lined by single or multilayered epithelium. Diffuse lesions are aggressive, extensive and ill defined with finger like projections from wall into the neighbouring tissues (Qin Zhou *et al.*, 2011).

Adult cystic hygromas are very rare and solitary, translucent lymphatic cysts in cervicofacial region is the usual presentation in adults (Gupta and Dayal, 1972). They may occur in late adult life between 40 – 60 years presenting as painless slowly enlarging lump in neck in an otherwise asymptomatic patient. The reason for them to remain dormant for such a long time may be because of local infection precipitating the growth of a previously undiagnosed lesion or may be attributed to tumour or trauma (Veeraraghavan *et al.*, 2009). Rapid enlargement may occur over a short period of time and major structures like larynx, trachea, oesophagus, brachial plexus or great vessels may be compressed. Thus the presentation may be because of pain, hoarseness, dysphagia or breathlessness (Gow *et al.*, 2011).

A solitary cystic mass in lateral aspect of neck in young adult patients (16 – 40 years) is often a benign lymphatic cyst, but a more sinister disorder like metastasis from a papillary carcinoma or occult oropharyngeal squamous cell carcinoma arising from Waldeyer's ring may also have be considered. Isolated cervical cystic metastasis may mimic a branchial cyst initially. The incidence of carcinoma in cervical cysts initially diagnosed as branchial cysts is reported to between 4% - 24%, but this proportion rises upto 80% in patients above 40 years of age (James Sira *et al.*, 2011). The incidence of thyroid malignancy is reported to be 11% in patients with primary diagnosis of lateral cervical cysts, which may be a metastasis from a primary tumour of thyroid gland or carcinoma arising from an ectopic thyroid tissue lying within the cyst (Raul Gonzalez-Garcia *et al.*, 2008). The incidence of cystic lymph node metastasis arising from squamous cell carcinomas of Waldeyer's ring is reported to be as high as 62% in nodal metastasis and in 10% of them, no primary tumour is initially apparent (James Sira *et al.*, 2011).

Fnac is the standard investigation to be done in such cases and has got a very low false positive and false negative rates (1-3%) in the diagnosis of solid masses in neck, but its role in cystic lesions is less convincing, as in malignant cervical cysts its sensitivity ranges from 33%-73% (James Sira *et al.*, 2011). However it is imperative as the first investigation to be done with complete examination of the aspirated fluid to rule out any malignant potential. Ultrasonography reveals the nature of the cyst – whether unilocular or multilocular – and fluid levels may be present with septations if complicated by haemorrhage. CT Scan shows them as poorly circumscribed, uni/multilocular hypodense cystic lesions with fluid attenuation (Mahesh Kumar Mittal *et al.*, 2012). MRI is also very important to identify the location, size and extent of the lesion. All patients above 40 years must be subjected to a complete work up including panendoscopy and biopsy to rule out any evidence of focus of primary malignancy, in all suspicious lesions (James Sira *et al.*, 2011).

Many Treatment modalities are advocated for the cystic lesions in neck, including surgery, sclerotherapy or radiotherapy. Localised microcystic lesions can be excised completely, but excision is very difficult in diffuse and extensive lesions because of poor demarcation and potential complications like damage to surrounding vessels, facial nerve palsy (incidence 3.3 – 5.9 %), Horner's syndrome and postoperative lymphatic leakage (Qin Zhou *et al.*, 2011). Hence nonsurgical conservative treatment like radiotherapy, cryotherapy, electrocoagulation, sclerotherapy and laser therapy have been recommended, as a primary or adjuvant therapy especially in children. Sclerotherapy (with OK 432 or bleomycin) is now well established in neonatal and paediatric cystic hygromas, but its role in adult cases has not been satisfactory because of its less acceptance rate (<67 % reduction in size) and complications associated in treatment of lesions in close association with important structures in neck. OK 432 (Picibanil – lyophilised extract of streptococcus pyocenes treated with benzyl penicillin) also induces a local immune response often resulting in rapid temporary increase in size of cystic hygroma (Gow *et al.*, 2011).

Complete surgical excision is the preferred modality of treatment in solitary cystic hygromas in adults and should be done carefully, avoiding leaving any islands of tissue which may act as foci of recurrence (as high as 20%), and also to avoid damage to the surrounding entangled structures like hypoglossal, lingual or vagus nerves (Veeraraghavan *et al.*, 2009). Care is also taken to avoid injury to brachial plexus, sympathetic chain, carotid sheath contents and lower branches of facial nerve. Complications may include sepsis, infection, fistula formation, facial palsy, vocal cord palsy or shoulder weakness. Laser therapy has been used to treat superficial localized lymphatic malformations, the advantages being its ease of use, less bleeding, minimum pain, reliable effect and repeatable treatment, especially in paediatric age group (Qin Zhou *et al.*, 2011). For recurrent cystic hygromas, IV Cyclophosphamide and intracystic injection of OK 432 has been advocated. However, in recurrent extensive cystic hygromas, a multidisciplinary surgical approach is preferable for optimum cosmetic and functional results (Veeraraghavan *et al.*, 2009).

Conclusion

Solitary cystic lesions in neck are very rare in adults, majority being benign lymphatic cysts, for which complete surgical excision is the preferred treatment of choice. Patients above 40 years of age must be thoroughly investigated to rule out any evidence of malignancy, before embarking on the treatment.

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