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Case Report

Adult cystic Lymphangioma - A Rare Clinical Entity with Review of Literature

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Abstract:

Cystic lymphangioma is a congenital lesion and usually occurs in children below 2 years of age. Cystic lymphangioma in submental and submandibular tringle in adult without involving the submandibular gland is clinically very rare with scarce reports in literature. We report one such case here with extensive review of the literature because of its rarity in adults and also to emphasize the need to consider cystic lymphangioma in differential diagnosis of submental and submandibular swelling. We have treated one such case of cystic lymphangioma in a 65 years old patient, who presented to us as a right sided upper neck swelling of 1 year duration. The awareness of occurrence of cystic lymphangioma in adults is important for its proper management.

Key words: ovarian endometrioma; in vitro fertilization embryo transfer (ivf-et); single-port laparoscopy; ethanol sclerotherapy; infertility

1.Introduction

Cystic lymphangioma is a malformation of lymphatic channels that usually develop when lymphatic channel fails to connect with the normal jugular vein. Cystic lymphangioma can be found more commonly in head and neck region and 80% of them are in the children below the age of 2 years. Adult lymphangioma are extremely rare and reported with only little over 100 cases in literature till 2009 (1) and because of its rarity the clinician may often miss the diagnosis while doing the clinical evaluation. In children a big sized cervical lymphangioma can cause dysphagia and airway obstruction, however adult lymphangioma are mostly asymptomatic excepting the neck swelling. One such case of right sided neck swelling involving the right submental and submandibular triangle in an adult is presented here because of its rarity and also documented its stepwise complete resection of the cyst to prevent itch recurrences.

Case Report

A 65 years old male presented to our ENT OPD with the history of right sided upper neck swelling of one year duration, along with minimal irritation and itching over the swelling. There were no other associated symptoms of pain, discomfort, dysphagia or any other complaint. The swelllig was insidious on onset and gradually progressed to the presenting size. He has no known comorbidities. A thorough ENT examination was done followed by a laryngeal endoscopy and CEMRI neck. On clinical examination, a swelling approximately measuring 1.5 to 2 centimeters in size, involving the right submental submandibular triangle was found. On palpation, a firm to soft in consistency, non tender and mobile on bimanual palpation was present. The skin over the swelling appears normal.

The imaging reported a cystic lesion with thin T2 hypointense non enhancing capsule in the right submandibular region measure approximately 35mm*15mm.The lesion was seen beneath right platysma, anterior to right submandibular gland and lateral to right mylohyoid muscle.

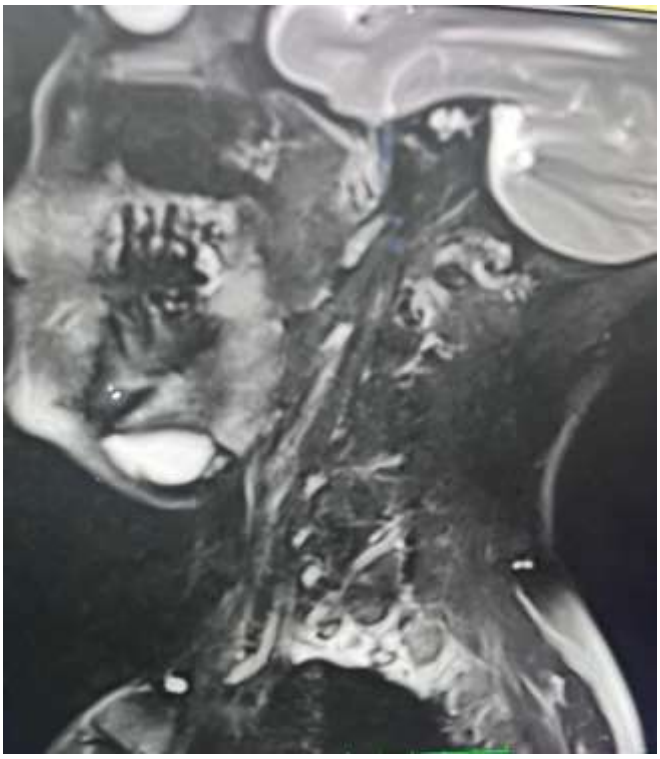

Figure 1- MRI showing saggital section of a cystic lesion with thin T2 hypointense, non-enhancing capsule seen in right sub mandibular region, measuring approximately 35*15 mm in cross section.

Figure 2 showing axial section of MRI

(Figure 1 and 2). FNAC was done outside in a local hospital but was found to be inconclusive. The case was posted for surgical excision under general anesthesia. A collar incision was given, two fingers breadth below the margin of mandible (Figure 3).

Figure 3 a collar incision two fingers below the lower border of the right mandible the mass was excised in toto following a collar incisio

Figure 4 showing the exposure of a cystic mass in the right submandibular region after flap elevationn

Figure 5 showing the dissection of the cystic mass along with the subcutaneous tissue from the right sub mental and sub mandibular area

Figure 6 showing progressive resection with intact submandibular gland

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Figure 7 showing the separation of the mass from the underlying structure

Figure 8 showing the underlying structures after complete excision of the mass

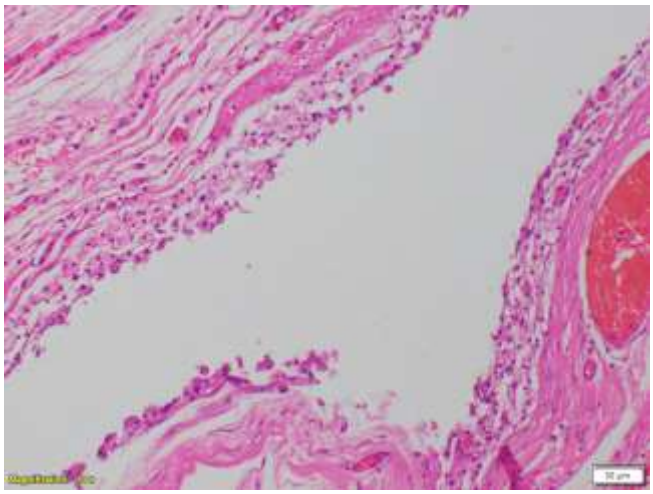

Figure 9 showing the histopathological picture with dilated lymph spaces separated by fibrocollagenous tissue

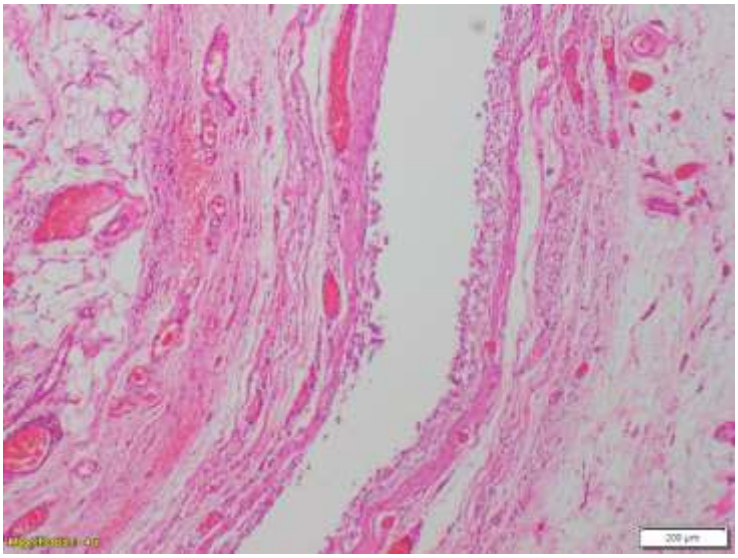

Figure 10 showing the histopathological picture with foamy histiocytes and sparse inflammation.

over the swelling and raising superior and inferior subplatysmal flaps. (Figure 4,5,6,7). The mass was sent for HPE examination, wound was sutured with vicryl 3-0 suture material. Patient was followed up after a week from the surgery, sutures removed and the wound was found to be healing well. The latest follow up was two months post operative day, with no clinical evidence of recurrence. The HPE reported features showed dilated lymphatic space separated by fibrocollagenous tissue with collections of foamy histiocytes and sparse inflammation,suggestive of cystic lymphangioma. (Figure 9,10)

Discussion

Cystic lymphangioma is a benign lesion commonly seen in the children under 2 years of age (80-90%) with 1.2-2.8 per 100 000incidence rate [2], whereas cervical cystic lymphangioma in adult is very rare.[3] Precise etiology of cervical cystic lymphangiomais controversial but have postulated three theories, one is blockage or arrest of normal growth of the primitive lymphatic channels during embryogenesis, secondly primitive lymphatic sac fail to reach the venous system in jugular vein and thirdly during embryogenesis, lymphatic tissue lays in wrong area. It is supposed to be congenital but also can be a result of obstruction and retention of lymph fluid of developing lymphatic vessels caused by infection, trauma and neoplasm. There is no notable preference for sex, both male and female are equally affected by the disease.

It can develop in any part of the body but commonly seen in head and neck area followed by clavicular and axillary area. Cystic lymphangioma in submandibular and submental area will mostly be arising from lymphatic tissue in submandibular and submental triangle and most of the reported cases showed involvement of the submandibular salivary gland also by the lymphangioma and may have to excise the salivary gland for complete excision of the lymphangioma. But present case, lymphangioma was arising separately from lymphatic tissue from submental space with no involvement the submandibular salivary gland but only causing minimal displacement. Histopathologically it is classified as four types [1] cavernous lymphangioma [2] cystic hygroma or cystic lymphangioma [3] lymphangioma circumscriptum and [4] acquired progressive lymphangioma or benign lymphangioendothelioma .[4,5]

Adult lymphangioma are mostly asymptomatic but may have a complaint of presence of a swelling with minimal occasional irritation or pain at the site of the cyst but patients have a fear of increase in size and transformation to malignancy whereas cervical lymphangioma in children may complain of difficulty in swallowing and respiratory obstruction. Clinically it may be a well defined, non tender, cystic and mobile swelling but mobility may be restricted in case of deeper extension of the cyst. Commonly differentiated from conditions like thyroglossal cyst, bronchogenic cyst, cystic dermoid and malignant lymph node. Investigations like ultrasound neck, FNAC, contrast CT or MRI neck

recommended, CT and MRI may also be used to assess the intrathorasic extension of the cyst. [6]

Complete surgical excision of the cyst is the best line of management [7,10]. Partial or piecemeal removal of the cyst is associated with high rate of recurrence, as high as 88% (Kennede TL). We could excise the cyst in our patient in toto and same has been documented and showed no evidence of recurrence so far. Other recommended treatments are sclerotherapy, radiotherapy, laser therapy, electrocoagulation and cryotherapy. OK-432 is considered to be a sclerosing agent of choice [11]. Usually, these additional therapies are used in excision of large, expensive and complicated cyst along with the primary surgery or also may be used separately to excise the lymphangioma in high-risk patients.

The definitive diagnosis of cervical lymphangioma comes after histopathological examination of the excised cyst. Microscopically, cervical lymphangioma are characterize by ectatic and dilated lymphatic spaces lined by an attenuated endothelium. Some of the lumina may contain lymph fluid. The spaces are surrounded by fibrocollagenous septa harboring varying degrees of lymphoid aggregates. Histopathology of our excised mass showed dilated lymphatic space separated by fibrocollagenous tissue with collections of foamy histiocytes and spares inflammation, suggesting cystic lymphangioma.

Conclusion

We have reported a case of upper neck spontaneous swelling of 5 years duration with the main complaints of minimal irritation, itching and constant fear in mind of transformation of the swelling into malignant growth. MRI with contrast was done as main investigation procedure. Complete surgical excision of the cyst done.

Appendix

CEMRI – contrast enhanced magnetic resonance imaging

FNAC – fine needle aspiration cytology

MRI - magnetic resonance imaging

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