

Case Report

A Rare Case of Submandibular Lymphangioma in an Adult

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ABSTRACT

Lymphatic malformations are uncommon in adults. They are common in the head and neck region because of the abundance of lymphatics. In this report, we have a male patient, 54 years old, who presented with a painless swelling in the submandibular area. He was clinically diagnosed with a submandibular gland mass. Ultrasonography showed a multiloculated cyst arising from the submandibular gland. Complete surgical resection of the cyst was performed along with the submandibular gland. A histopathological study of the specimen confirmed the diagnosis of Lymphatic malformation.

Keywords: Adult, Excision, Lymphatic malformation, Neck, Submandibular gland

INTRODUCTION

Lymphatic abnormalities result from an abnormal embryonic development of the lymphatic system. It is a rare vascular malformation that can occur in any part of the body. Only a few cases are documented in adulthood; about 90 percent of occurrences happen in children under 2 years old. About 75 percent of these are in the head and neck region.^[1] Lymphatic abnormalities can be inborn or acquired. Acquired ones develop when there is a blockage in the lymphatic system due to trauma or infection.^[2] Lymphatic malformations are classified into capillary lymphangioma, cavernous lymphangioma, and cystic hygroma. The head and neck (HN) are the regions most affected, and symptoms are based on the site and the lesion size.^[3]

The two most common routine studies are computed tomography (CT) and magnetic resonance imaging (MRI), which are more advanced than ultrasound for defining extension and demonstrating relationships with other anatomical structures. Surgery is the definitive treatment. Other treatment modalities are LASER, sclerotherapy, and embolisation.

In this report, we describe a 54-year-old male patient with swelling in his submandibular area, which was treated surgically and diagnosed as a lymphangioma using histopathology.

CASE REPORT

A 54-year-old male came to the Ear, Nose, Throat (ENT) OPD with a gradually increasing swelling in the right submandibular region for 2 months with no other contributory history.

Examination revealed a single, well-defined swelling measuring 6 cm × 4 cm in the right submandibular region. On palpation, the swelling was firm in consistency and nontender. Ultrasonography of the neck revealed a well-defined, multiloculated cystic lesion arising from the right submandibular gland, with possible differential diagnoses such as lymphoepithelial cyst. Fine needle aspiration suggested a nonneoplastic lesion favouring lymphoepithelial cyst (category II)

The patient was managed surgically. He underwent excision of the cyst with the right submandibular gland. A 6 cm × 6 cm cyst was noted in the right submandibular gland [Figures 1 and 2]. The specimen was sent for histopathological examination. Microscopic examination showed fatty tissues with numerous lymphatic channels lined with flattened epithelium [Figure 3] and few lymphatic channels with lymph fluid, which suggested lymphatic malformation.

The postoperative period was uneventful. The patient has shown no signs of recurrence in the last 10 months.

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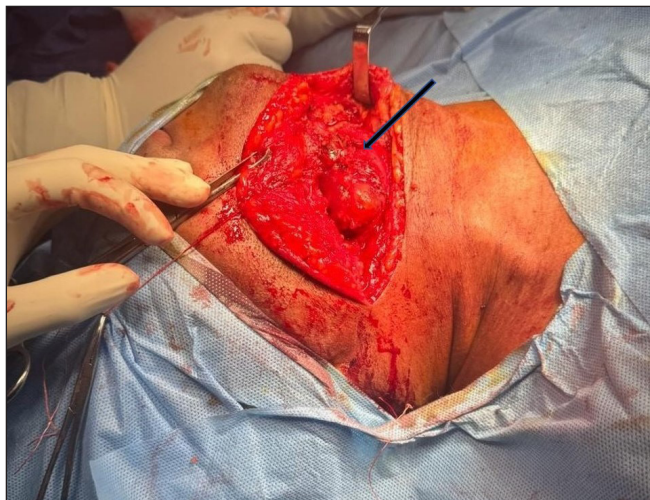


Figure 1: Intraoperative image showing the cyst in the right submandibular region (dark blue arrow).



Figure 2: Specimen of lymphangioma with the submandibular gland.

DISCUSSION

Lymphangioma of the neck is a rare malformation of the lymphatic system. Abnormalities of the lymphatic system differ in size from small lumps to large deformities. The majority of the lymphatic abnormalities are in the HN region.^[2]

Several hypotheses have been put forward to explain the origin:

- 1) Blockage or arrest of normal growth of primary lymph channels occurs during embryogenesis, 2) The primitive lymphatic sac does not reach the venous system, and 3) During embryogenesis, lymphatic tissues lie in the wrong area.^[4]

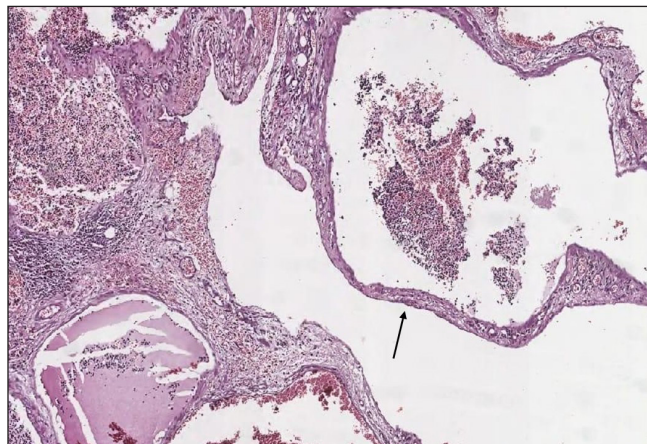


Figure 3: Histopathology showing cystic lymphatic spaces lined by endothelial cells (black arrow). Haematoxylin and eosin, 100x.

Although common in paediatric practice, they are rarely seen in adults.^[5] Nearly 90 percent of cases occur in paediatric patients under the age of two because most symptoms appear by then, and routine prenatal and postnatal imaging tests increase the likelihood of diagnosis. The majority of paediatric patients receive sclerotherapy or surgical excision.^[6]

The most typical initial symptom is a painless, gradually growing mass of the HN.^[7] The late appearance may be explained by the lack of initial pressure symptoms and the typical gradual progressive increase of the oedema. The mass effect occurs when lesions expand and can cause obstructive symptoms.^[8] Pre-operative imaging is a helpful tool for selecting the ideal approach for treatment. Imaging options include ultrasonography, CT, and MRI. The initial method for identifying this kind of lesion and defining its structural cystic-type features is ultrasonography. It is also helpful for post-surgical evaluation, assessing the effectiveness of sclerotherapy, checking for recurrences, and tracking the non-operated lesions during therapeutic abstention. Nevertheless, the ability to identify the deep extension of the lesion is insufficient.

CT and MRI are crucial supplementary diagnostic tools for determining the precise position, extension, and depth of the lesions. Low-flow vascular anomalies known as lymphatic abnormalities occasionally manifest in multiple cysts. The signal intensity is hyperintense on the T2-weighted image and intermediate on the T1-weighted image with no enhancement post-contrast injection. Treatment options are laser therapy, sclerotherapy, surgery, or an amalgam of these.

Differential diagnosis of lymphangioma is often tricky, which may influence the treatment strategy.

Despite being benign lesions, lymphatic abnormalities hardly ever get cured without treatment.^[1] Many

conservative nonsurgical therapies, consisting of radiation, electrocoagulation, cryotherapy, ligation, and embolisation, have been explored for these lesions, in addition to surgical excision.^[2] Sclerotherapy, laser therapy, or surgery are all possibilities for managing localised lymphatic abnormalities. For cosmetic reasons, excision may be considered.^[2]

For lymphatic anomalies, various treatments, including non-invasive surgery to specialised surgery combined with adjuvant therapies, are available. Due to its weak delineation and relationship with nearby critical structures, HN lymphangiomas are difficult to manage. Complete surgical excision becomes more challenging due to its invasive nature and difficulty identifying the damaged essential structures from nearby normal tissues.^[1]

Histopathology validates the diagnosis of lymphangioma. Histopathology can be nonspecific and exhibit clusters of lymphocytes and inappropriately walled vascular spaces with fluid rich in eosinophilic proteins.^[9]

Our subject exhibited a painless swelling in the submandibular region and was diagnosed as a submandibular gland mass. We achieved complete clearance with excision of the gland and the cyst, as the cyst was adherent to the gland.

CONCLUSION

Lymphatic abnormalities are benign lesions of the lymphatic system, and spontaneous regression is rarely seen. These are typically found in the HN area. A thorough clinical, radiological, and histopathological examination is essential in evaluating and treating this condition. Although uncommon in adults, it can often mimic other neck swellings and should be included in the differentials of any neck swelling.

Ethical approval: Institutional Review Board approval is not required.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given consent for their images and other clinical information to be reported in the journal. The patient understand that the patient's names and initials will not be published and due

efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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