Case Report

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Unraveling of Radiological and Therapeutic Approach to Cervical Cystic Lymphangioma in Elderly Patient: A Rare Case Report

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Abstract

Background: Cervical cystic lymphangioma is rarely reported in elderly patients. This case report aimed to establish a diagnosis and determine the proper treatment by considering the probability of cervical cystic lymphangioma in the elderly based on radiological examination.

Objective: To report an uncommon case of lymphatic malformation in the neck region was reported in an elderly woman.

Case Report: A 58-year-old woman presented with a right neck mass that had progressively enlarged over the year. Ultrasound (US) and contrast-enhanced computed tomography (CECT) scans of the neck revealed clustered cystic lesions in the right submandibular, upper anterior and posterior jugular, mid and lower jugular, upper and lower posterior triangle, and medial and lateral supraclavicular regions, consistent with cervical cystic lymphangioma. Fine-Needle Aspiration Biopsy (FNAB) revealed an acellular cystic lesion, which was later confirmed to be a lymphangioma. The tumor was reduced with sclerotherapy and bleomycin injection.

Conclusion: Lymphangioma is a rare benign lession characterized by fluid-filled masses of lymphatic tissue that typically occur in the head and neck region. Although it can be congenital in origin, these lesions usually present in children under the age of two. Adult lymphangioma is a rare condition, with fewer than 80 documented cases in the literature. Cervical cystic lymphangioma can be diagnosed with US and CECT examinations, as well as histological examinations. The preferred treatment is sclerotherapy and surgical resection.

Keywords: Lymphangioma, cervical, cystic, elderly, radiology

Introduction

Lymphangiomas are rare, benign abnormalities of the lymphatic system that can develop anywhere on the skin and mucous membranes due to aberrant growth of lymph channel. These are thought to be the effects of lymphatic tissue that has been sequestered, yet still retains the capacity to expand.[1] Lymphangioma is typically diagnosed at birth or within the first few years of life.[2] Most often, it affects the head and neck area and also axillary region.[3] However, it can also be found in the abdomen (hepatic, splenic, pancreatic, and renal), as well as in retroperitoneal and mediastinal locations.[4]

Adult cervical cystic lymphangioma is an uncommon lymphatic system abnormality caused by the separation of lymph sacs in venous drainage channels. A growing benign cystic tumor characterizes it and typically manifests in the posterior triangle of the neck.[5] Their location determines the clinical

Fig. 1 A 58-year-old Woman Presented with An Enlarged, Soft, Undulating Mass on Her Right Neck

presentation. When lesions expand and press on the surrounding important structures, obstructive symptoms can arise.[6] The literature reveals very few reports of adult occurrences. To date, fewer than 150 cases of adult lymphangioma have been documented in the literature.[7]

Lymphangiomas account for about 25% of all benign pediatric vascular tumors and 4% of all vascular tumors, with no gender or race preference.[2] Adult lymphangioma development is idiopathic, with infection, trauma, or iatrogenic damage being the risk factors.[8] Nevertheless, the clinical features and underlying pathophysiology of lymphangiomas are still unknown.[7]

The modalities of choice in diagnosing lymphangioma are US, CECT, and magnetic resonance imaging (MRI).[6,4] Histopathology then confirms a definitive diagnosis. Treatment of choice is total surgical excision, but there are also other alternative treatments, one of which is sclerotherapy, the goal being to diminish the size of the tumor and harm to adjacent organs.[8]

Case

A 58-year-old woman came with clinical symptoms of a tumor in her right neck that had been growing in size for a year. The patient has complained of mild shortness of breath, trouble swallowing, and sometimes palpitations for the last three months.. During a localized status assessment in the right neck region, a soft mass measuring ± 9 x 10 cm was palpated. It moved when swallowing (Fig. 1). No tenderness was present.

Chest x-ray revealed opacity with indistinct borders in the soft tissue of the right neck (Fig. 2). US examination of the neck revealed multiple complex cystic lesions in the

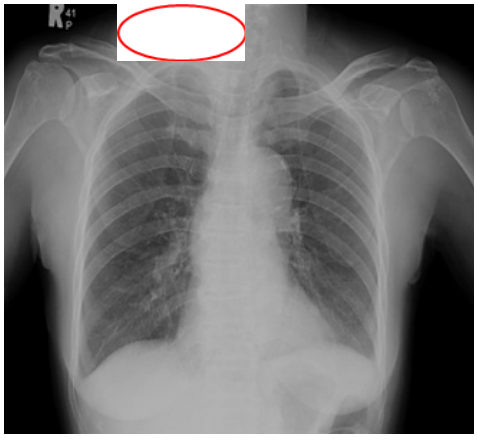

Fig. 2 Chest X-Ray Showed Opacity with Indistinct Borders on The Right Neck Soft Tissue (Red Circle)

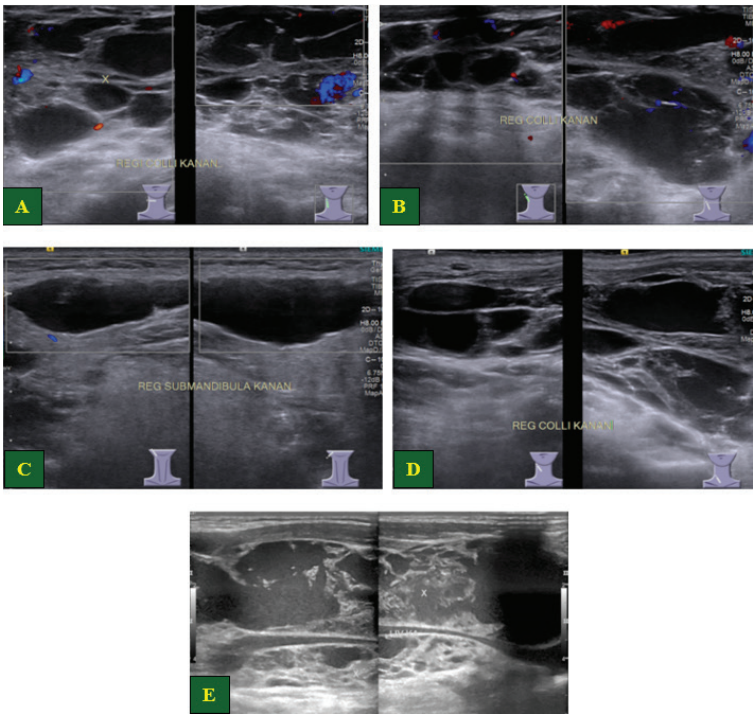

Fig. 3 Multiple Septated Anechoic Lesions on The Right Neck (A, B), Right Submandibular (C), and Right Supraclavicular (D, E) Region at US Examination

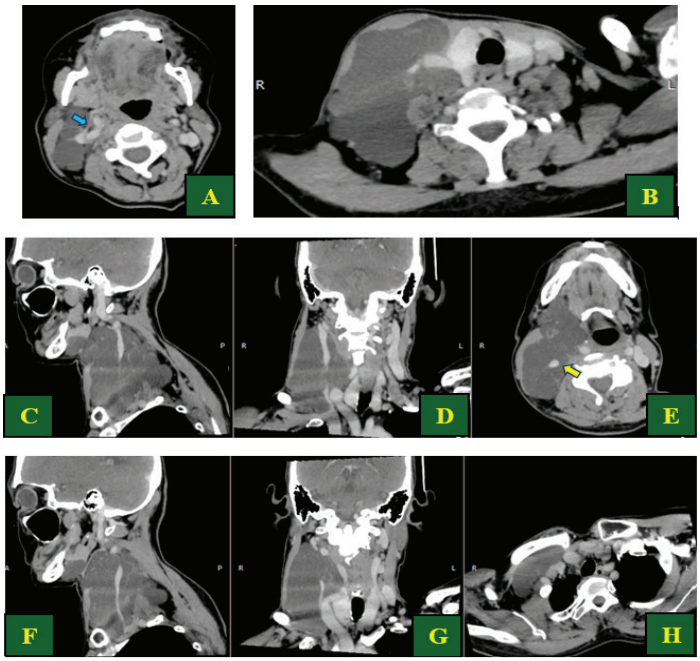

Fig. 4 Clustered cystic lesions in the right submandibular, upper jugular anterior and posterior, mid and lower jugular, upper and lower posterior triangle (C, D, F, G), medial and lateral supraclavicular (H) region, which encased the right jugular vein (E, yellow arrow), compressed right common carotid artery, internal and external carotid arteries (A, blue arrow) to the medial side, and slightly deviating trachea to the left side (B) were discovered by CECT of the neck

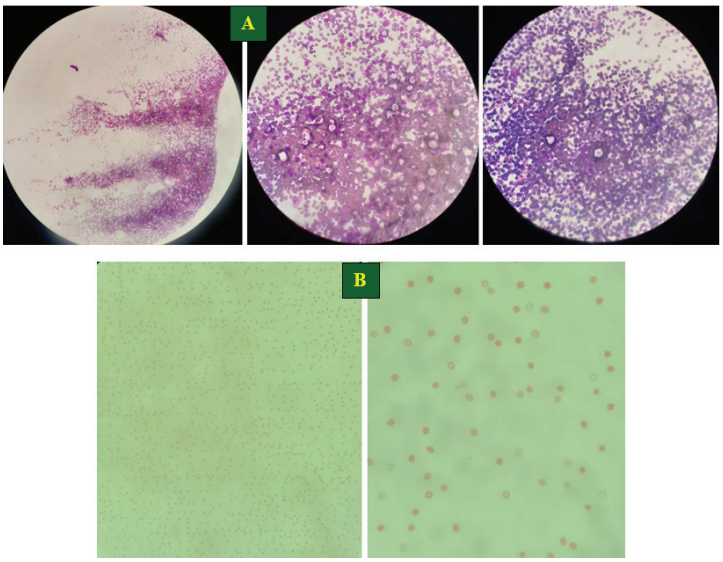

Fig. 5 A. The FNAB Analysis Revealed Widespread Distribution Of Erythrocytes, A Few Macrophages, and A Distribution of Lymphocytes (Diff-Quick, X100). B. The Microscopic Cytology Smear Contained A Scattering of Lymphocytes and A Few Plasma Cells. (Left: PAP X40; Right: PAP X400)

right neck, in the right submandibular and supraclavicular regions (Fig. 3).

CECT of the neck revealed a clustered, non-enhancing, indistinctly demarcated cystic lesion in the right submandibular, upper jugular anterior and posterior, mid and lower jugular, upper and lower posterior triangle, medial and lateral supraclavicular region measuring approximately 9.55 x 6 x 10 cm, which encased the right jugular vein, and compressed the right common carotid artery, internal and external carotid arteries to the medial side, and slightly deviating the trachea to the left (Fig. 4), which was consistent with the features of cervical cystic lymphangioma.

Following neck US and CECT examinations, FNAB was performed using a G25 needle, making three punctures in the swollen area, yielding cystic fluid aspirate material combined with blood. Diff Quick stain was used to create a smear on three slides. Microscopic analysis revealed a large distribution of erythrocytes, a few macrophages, and dispersed lymphocytes, suggesting an acellular cystic lesion (Fig. 5A). Then, a cytological analysis using Papanicolaou staining revealed a scattering of inflammatory cells and lymphocytes, along with a small number of plasma cells (Fig. 5B). In the

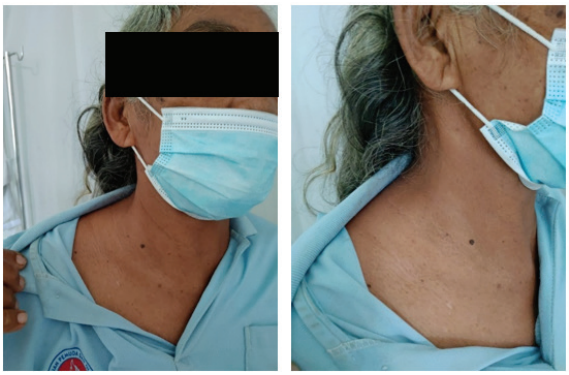

Fig. 6 The Tumor on The Patient's Right Neck Appeared to Have Decreased Following Two Bleomycin Injections

preparation, no cancerous cells were detected. As a result, the cytologic examination reveals the presence of lymphangioma characterized by cystic fluid with mature lymphocyte infiltrates and erythrocyte scattering.

Sclerotherapy with bleomycin injection was then performed on this patient, because the cervical lymphangioma has encased the right jugular vein and surgery is not an appropriate treatment option. The tumor on the patient's right neck had reduced after two bleomycin injections administered over a month (Fig. 6).

Discussion

Most often occurring before the age of two and uncommon in adults and the elderly, cervical cystic lymphangiomas are abnormalities of the lymphatic channels that arise when the lymphatic system is unable to connect with the normal jugular vein. It is most commonly seen in the head and neck area (75% in the posterior triangle of the neck and 20% in the submandibular triangle)[3] Endothelial cells line the lymphatic aggregates that comprise these tumors, and they develop into cysts of varying sizes that have the potential to bleed or infect only the lesion.[5] In both adult and pediatric patients, the majority of cervical lymphangiomas are unilateral and do not exhibit gender or side preference.[3]

The clinical presentation varies depending on the location.[9] Neck lymphangiomas often show no additional symptoms and are refractory. Sometimes they can result in symptoms from the compression of organs, such as dysphagia when the esophagus gets compressed and dyspnea when the trachea is affected.[5,10] According to the size and depth of the aberrant lymphatic vessels, lymphangiomas can be classified as either deep or superficial, or as either congenital or acquired. Based on depth, the types of lymphangiomas may be separated into superficial types, such as acquired lymphangioma, also known as lymphangiectasia, and lymphangioma circumscriptum. The two distinct and well-defined congenital abnormalities known as cavernous lymphangiomas and cystic hygromas are examples of the deep types of lymphangiomas.[2] Lymphangiomas can also be divided into three groups based on the size of the blood vessels that are involved: cavernous, which are found in the tongue and mouth; capillary (simplex), which are often seen in the subcutaneous tissue; and cystic, also known as cystic hygroma, which is most frequently found in the neck.[5] In contrast to the proteinaceous background, the cytologic results showed primarily mature cells, which are indicative of lymphangiomas.[8] The characteristic fluid of cystic lymphangiomas is clear, and the cytologic findings of lymphocytes, histiocytes, and proteinaceous debris are usual.

Although several established treatment approaches exist, surgical resection remains to be the most effective method for treating lymphangiomas. Recurrence rates can reach 88% when partial removal is used.[8] However, it could be challenging or impossible to completely remove head-neck lymphangiomas surgically.

Complete surgical resection may not be possible, or surgical problems may arise when cervical lymphangiomas are excessively large and located near the great vessels and upper airways. For lesions in the retro pharynx, tongue base, or oral floor, deformity and loss of function may occur, making surgical resection infeasible. There are additional therapeutic options, including laser applications, radiation therapy, cryotherapy, sclerotherapy, and electrocoagulation.[11] OK-432 (Picibanil) and Bleomycin are both effective sclerosing agents. These therapies may be chosen either individually or in conjunction with surgery.[12] Sclerotherapy or surgery are the usual treatments for lymphangiomas. Nevertheless, because they tend to invade neighboring muscles and neurovascular networks, total excision of lymphangiomas may not be possible, and sclerotherapy is typically only effective in macro cystic areas. The endothelial lining of vessels is irritated by intralesional sclerotherapy, which results in significant inflammation and fibrosis.[13] The recurrence rate following treatment is high, ranging from 22% to 27% following surgery and 57% following sclerotherapy. In the treatment of lymphangiomas, prior research has demonstrated that bleomycin and OK-432 are either as effective as or similar to surgical excision.[14] Furthermore, it appears that sclerotherapy with OK-432 and bleomycin has a lower risk of complications than surgery.[15]

Sclerotherapy, however, is most successful (shrinkage ≥60%) in macro cystic lesions, and the disease recurrence rate is around 9%. Sclerotherapy has also demonstrated a poor response in microcystic tumors.[7]

Repeated bleomycin injections are the preferred treatment for preventing the many recurrences that follow surgical excision since they have resulted in good results in follow-up observations over a 24-month period with no recurrences seen.[9]

The primary factors to consider when making a differential diagnosis of cervical lymphangiomas in an adult are lymph node neoplasms, dermoid cysts, and branchial cleft cysts. Characterizing the cystic components, assessing the size of lesions, and directing treatment choices are all made possible by imaging modalities, particularly US and CECT. Histopathology confirms the diagnosis, and the preferred course of therapy is total surgical resection.

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