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Adult-Onset Cystic Lymphangioma of the Submandibular Region: A Rare Clinical Entity and Surgical Management

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Abstract: Cystic lymphangiomas are congenital malformations of the lymphatic system that predominantly arise in infancy. Adult-onset presentation in the submandibular region is exceedingly rare and may clinically mimic a spectrum of salivary, vascular, and soft-tissue lesions, complicating diagnostic accuracy. We report the case of a 34-year-old female who presented with a gradually enlarging right submandibular swelling of four months' duration. Magnetic resonance imaging demonstrated a multiloculated cystic lesion suggestive of a lymphatic malformation, and cytology yielded proteinaceous fluid with mature lymphocytes. Complete surgical excision was performed, and histopathology confirmed cystic lymphangioma. The postoperative course was uneventful, with no recurrence noted at early follow-up. This case highlights the diagnostic challenges of atypical adult presentations and emphasizes the role of imaging and histopathology in definitive diagnosis, as well as the importance of complete surgical excision for preventing recurrence.

Keywords: *Lymphangioma, Cystic Hygroma, Submandibular Swelling, Lymphatic Malformation, Adult Onset, Case Report.*

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I. INTRODUCTION

Lymphangiomas are benign, hamartomatous proliferations of lymphatic vessels that arise from developmental sequestration or failure of lymphatic channels to establish normal communication with the central lymphatic system[1]. They most commonly present during the first two years of life, with adult-onset cases comprising less than 1% of all lymphangiomas[2]. Approximately three-quarters occur in the head and neck region, often involving the posterior cervical triangle[3].

Cystic lymphangioma, also known as cystic hygroma, represents the macrocystic variant and is characterized by multiloculated cystic spaces filled with lymphatic fluid[4]. Presentation in the submandibular region of an adult is exceptionally rare and may be mistaken for a variety of benign or malignant neck masses, including salivary gland neoplasms, vascular malformations, branchial cleft anomalies, lipomas, and metastatic disease[5].

We describe a rare case of adult-onset cystic lymphangioma arising in the submandibular region, focusing on diagnostic evaluation, surgical management, and early outcome. This report adds to a limited body of literature documenting adult presentations of this classically pediatric lesion.

II. CASE REPORT

A 34-year-old woman presented to the oral and maxillofacial surgery department with a complaint of swelling and discomfort along the right lower facial region for the preceding four months. She reported that the swelling had gradually increased in size, although she occasionally noted transient decreases in volume without identifiable triggers. The swelling was not associated with pain, fever, difficulty swallowing, trauma, or recent infection. She denied systemic illness, significant dental history, or a family history of similar lesions.

Clinical examination revealed a unilateral fullness of the right submandibular region producing mild facial asymmetry. The overlying skin was normal in color and texture, with no visible vascular markings. On palpation, the mass was soft, compressible, nonpulsatile, and non-tender, with well-defined margins. It extended anteroposteriorly from just posterior to the oral commissure to the angle of the mandible. The regional lymph nodes were non-palpable, and salivary gland function appeared clinically normal. No abnormalities were appreciated in the temporomandibular joints or muscles of mastication.

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Magnetic resonance imaging of the neck demonstrated a well-circumscribed, multiloculated cystic lesion with high signal intensity on T2-weighted sequences and low signal intensity on T1-weighted images. The lesion occupied the submandibular soft tissue space without erosion of the mandibular cortex or infiltration into adjacent musculature. No significant enhancement of solid components was noted following contrast administration. These findings were suggestive of a macrocystic lymphatic malformation.

Fig 1 Preoperative Extraoral Picture

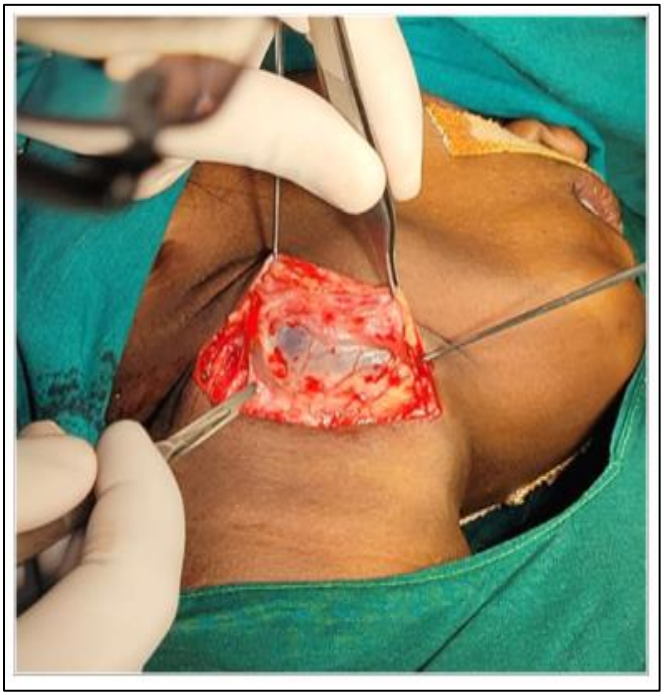

Fig 2 Exposure of Cystic Lesion

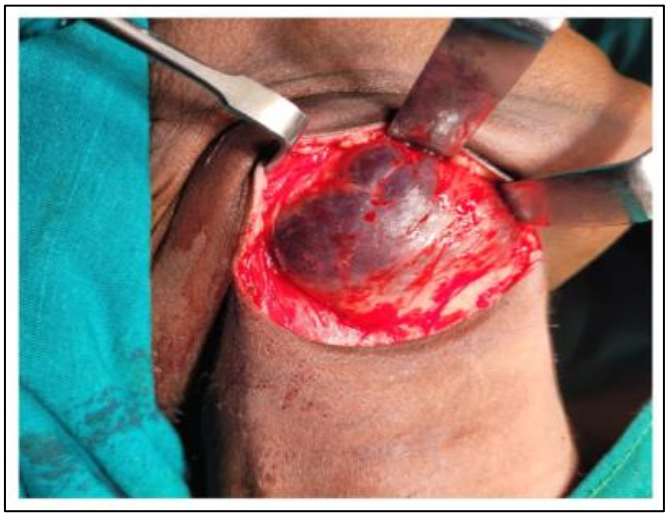

Fig 3 Intraoperative Clinical Picture

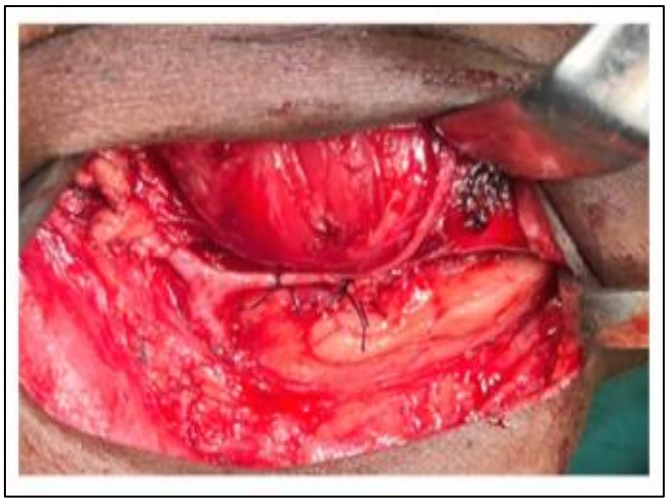

Fig 4 Preservation of Marginal Mandibular Nerve

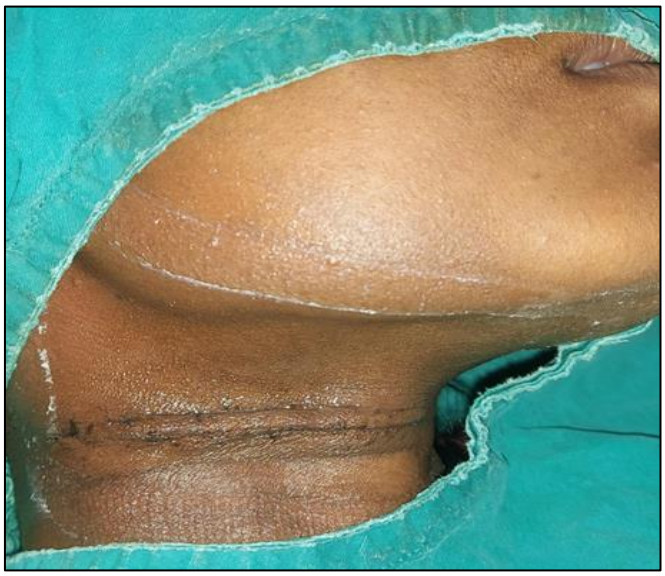

Fig 5 Closure-Subcuticular

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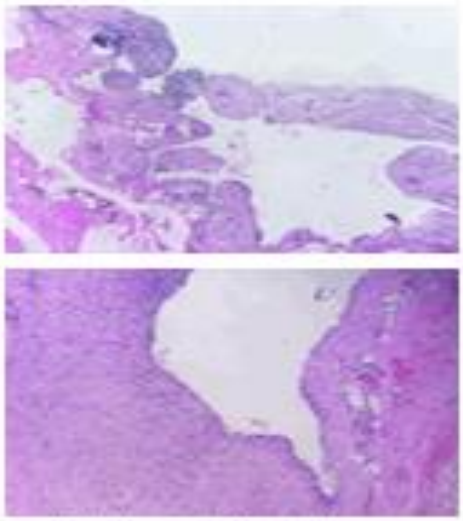
HISTOPATHOLOGY REPORT

HP No. :	EDH 751/24 (A,B)
CLINICAL DETAILS	Right submandibular mass lesion. MRI face-T2 hyperintense cystic lesion extending to adjacent floor of mouth and parotid region measures 4.7x2.9x2.6cm, in maximum dimension.
SPECIMEN	Right submandibular mass lesion.
GROSS DESCRIPTION	Received single unilocular cystic lesion measures 2x1x0.5cm, grey, wall thickness is 0.2cm, no solid areas. Part submitted.
MICROSCOPIC DESCRIPTION	Sections show fibrocollagenous wall lined by flattened endothelium with areas of erosion at places. Underlying stroma show lymphoid aggregates in sheets and nodules with areas of congestion. No granuloma / atypia / malignancy in sections examined.
IMPRESSION :	Right submandibular mass lesion- Suggestive of Cystic lymphangioma.



REMARK	Correlate clinically and with radiology.
NOTE	Blocks enclosed with report.

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Fig 6 Histopathology Report

Fine-needle aspiration yielded pale, straw-colored fluid. Cytologic examination revealed mature lymphocytes dispersed in a protein-rich background, with no evidence of atypia or infectious etiology. Hematologic investigations were unremarkable.

A provisional diagnosis of cystic lymphangioma was made. After obtaining informed consent, the patient underwent surgical excision of the lesion under general anesthesia. Intraoperatively, the mass consisted of thin-walled cystic cavities containing clear lymphatic fluid. The lesion was well encapsulated, allowing careful dissection from surrounding tissues. It was noted to lie superficial to the submandibular gland without direct glandular invasion. The mass was excised in toto without rupture.

Histopathologic examination confirmed the diagnosis of cystic lymphangioma, revealing numerous dilated lymphatic channels lined by attenuated endothelial cells. The cystic spaces contained eosinophilic proteinaceous fluid, and the surrounding stroma showed sparse lymphoid aggregates without dysplasia or malignancy.

The patient's postoperative course was uneventful, with satisfactory healing at one-week follow-up and restoration of facial symmetry. No evidence of recurrence was noted during the early postoperative period.

III. DISCUSSION

Lymphangiomas are believed to arise from congenital developmental abnormalities of the lymphatic system, particularly sequestration of lymphatic sacs that fail to regress or establish proper drainage pathways[6]. Although most lymphangiomas present in early childhood, delayed diagnosis or late proliferation due to infection, trauma, or hormonal changes has been suggested as a mechanism for adult-onset lesions[7]. Nevertheless, adult submandibular lymphangiomas remain exceptionally rare.

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Clinically, cystic lymphangiomas present as soft, fluctuant masses that may vary in size and occasionally enlarge rapidly due to infection or hemorrhage[8]. In adults, their nonspecific presentation means they often enter the differential diagnosis of a wide variety of cervical lesions, including vascular tumors such as hemangiomas[9], venous malformations[10], branchial cleft anomalies[11], lipomas[12], ranulas, and even metastatic lymphadenopathy.

Imaging plays a critical role in diagnosis. Ultrasound may reveal cystic spaces but lacks deep anatomical detail. MRI is considered the gold standard for evaluating lymphatic malformations, as it clearly delineates the multilocular cystic architecture, extent of disease, and relationship to surrounding structures[13]. The characteristic high T2 signal and low T1 signal with minimal post-contrast enhancement are strongly suggestive of cystic lymphangioma[14].

Histopathology remains the definitive diagnostic modality. The presence of dilated lymphatic channels, thin endothelial lining, and proteinaceous fluid distinguish lymphangiomas from vascular tumors and cystic neoplasms[15].

Management depends on lesion size, location, and symptoms. Complete surgical excision remains the treatment of choice for well-circumscribed, resectable lesions[16]. Recurrence rates may reach 10–20% following incomplete excision due to microscopic residual channels[17]. For unresectable or extensive lesions, sclerotherapy—particularly with agents such as OK-432 or bleomycin—has demonstrated excellent results[18], while in selected cases radiofrequency ablation, laser therapy, or combination therapy may be considered[19].

In the present case, the lesion was well circumscribed and anatomically favorable for complete excision. Early postoperative recovery was uneventful, and no recurrence was noted at short-term follow-up. Nonetheless, long-term surveillance is recommended due to known recurrence tendencies in lymphatic malformations.

IV. CONCLUSION

Adult-onset cystic lymphangioma of the submandibular region is exceedingly rare and poses significant diagnostic challenges due to its nonspecific presentation and overlap with more common cervical masses. MRI and histopathology remain essential for accurate diagnosis. Complete surgical excision is the treatment of choice for well-localized lesions and offers excellent prognosis. Clinicians should maintain a high index of suspicion for lymphatic malformations even in adult patients to prevent misdiagnosis and ensure timely intervention.

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