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Cavernous Lymphangioma of Unusual Location: A Case Report

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I. Introduction

Lymphangioma is a benign tumor involving the proliferation of lymphatic vessels. The commonly affected regions are head and neck, presenting two-thirds of cases at birth and 90% by the second year of life, and some victims may not manifest lifelong. [1,2] Lymphangioma formed along the tissue planes or penetrate in adjacent tissues, become canalized and accumulates fluid in them. These are classified into three types, namely Capillary lymphangioma, Cavernous lymphangioma, and Cystic hygroma. [2] Another variety being Hemolymphangioma shows vascular as well as lymphatic component.

These hamartomatous lesions contain clear lymph fluid, but some may present clinically as transparent vesicles containing RBCs because of extravasation. Mostly superficial, presenting as a swelling or a mass, but some may extend deeply involving the connective tissue. Amongst these, Cavernous lymphangioma contains dilated sinusoidal endothelium-lined vascular channels devoid of erythrocytes and may appear as subcutaneous nodules, with a rubbery consistency. [3] These are certainly demarcated from capillary lymphangioma. The overlying skin does not show any lesions or changes.

II. Materials and Methods

Cavernous lymphangioma of cheek region is discussed in the present article. The lesion presented with vestibular obliteration on the ipsilateral side and extending to the left maxilla. Diagnosis was made based on radio imagning and histopathology. Both clinical and histological features for proper management concerning this hamartomatous entity are incorporated in the present article.

III. Case Report

A 30-year-old male reported to our department with complaint of slow-growing mass in the right cheek region for ten years giving an unaesthetic appearance. There were no associated symptoms like pain and discharge. He noticed a gradual increase in size for three years. There was no history of trauma or any past illnesses. He complained of mild restriction during mouth opening.

Extra-oral examination revealed facial asymmetry concerning the right middle third of the face, round to oval solitary diffuse swelling extending from the right lateral ala of the nose to preauricular region anteroposteriorly and from right inferior orbital rim to the right commissure area supero-inferiorly. The lesion was measured about 3 × 3 cms in diameter with no change in the color of overlying skin and evident obliteration of the right nasolabial groove [Figure 1].



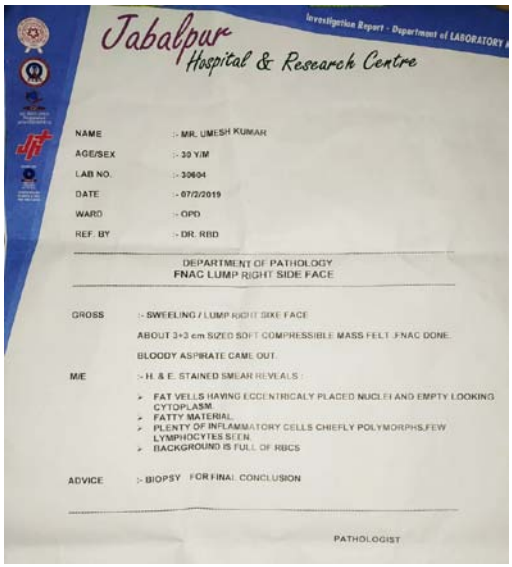
Figure 1: Pre OP Profile View

On palpation, the swelling was diffuse, soft in consistency; mobile, not fixed to skin, non-tender on palpation, no localized rise in temperature, non-reducible, and compressible, non-pulsatile. Intra-oral examination revealed vestibular obliteration in the right upper region posteriorly.

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Fine-needle aspiration cytology (FNAC) revealed the presence of fat cells having eccentrically placed nuclei and empty looking cytoplasm. Fatty material with plenty of polymorphs and few lymphocytes were present [Figure 2].


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DEPARTMENT OF PATHOLOGY
FNAC LUMP RIGHT SIDE FACE

GROSS :- SWEELING / LUMP RIGHT SIDE FACE
ABOUT 3×3 cm SIZED SOFT COMPRESSIBLE MASS FELT. FNAC DONE.
BLOODY ASPIRATE CAME OUT.

M/E :- H. & E. STAINED SMEAR REVEALS :
- FAT VELLS HAVING ECCENTRICALY PLACED NUCLEI AND EMPTY LOOKING CYTOPLASM.
- FATTY MATERIAL.
- PLENTY OF INFLAMMATORY CELLS CHIEFLY POLYMORPHS,FEW LYMPHOCYTES SEEN.
- BACKGROUND IS FULL OF RBCS

ADVICE :- BIOPSY FOR FINAL CONCLUSION

PATHOLOGIST

Figure 2: FNAC Report

Under general anesthesia, intraorally 2.5-3 cms maxillary vestibular incision taken. The Lesion was exposed, followed by dissection in the subcutaneous plane [Figure 3]. The facial vein was seen passing through the tumor and hemostasis achieved by ligating the parent vessel and circumscribed 3 X 3 X 3 cms tumor excision was done [Figure 4].



Figure 3: Exposed Lesion



Figure 4: Lesion excised

Histopathologic examination of the excised specimen showed numerous dilated, sinusoidal spaces of varying sizes within the deeper connective stroma. The vascular areas consist of walls of variable thickness and lined by a single layer of endothelial cells. Some areas show eosinophilic material with lymphocytes and few RBCs [Figure 5].

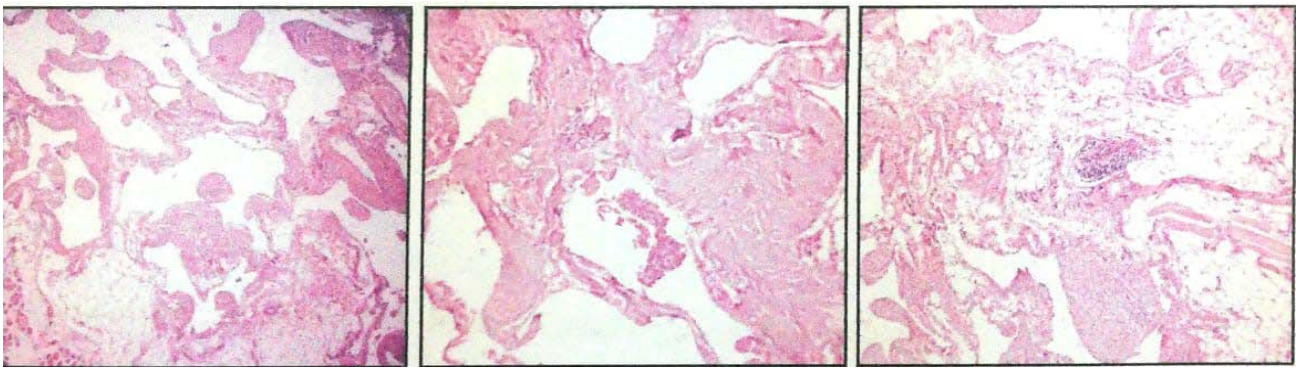


Figure 5: H & E stained section shows numerous dilated, sinusoidal spaces of varying sizes within the deeper connective stroma. The vascular spaces consist of walls of variable thickness and lined by single layer of endothelial cells. Some spaces show eosinophilic material with lymphocytes and few RBCs. Vascular channels are infiltrating into adjacent muscle.

IV. Discussion

Oral lymphangiomas usually involve anterior tongue, causing macroglossia, lips, and buccal mucosa. Clinically they appear as nodular or elevated masses and may resemble surrounding mucosa. Histopathologically multiple intervening lymphatic channels are contained within a loose fibrovascular stroma. Lymphangioma is usually confused with hemangiomas very often. The absence of valves and the presence of numerous erythrocytes in hemangiomas is a characteristic feature for differentiation.

The origin of this abnormality explained on the basis of three theories. The first theory entails about any blockage or disruption in the growth of primitive lymph channels during embryogenesis; the second states that

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the primary lymphatic sac does not communicate the venous system, while the third hypothesized, that lymphatic tissue was laid down in the faulty region during embryogenesis.

Poor aesthetics becomes the primary concern in patients with cavernous lymphangioma of the cheek. In this case, the lesion was present on the right cheek region with unaltered speech or breathing. Differential diagnoses include hemangioma, lipoma, amyloidosis, neurofibroma. Lymphangioma accounts for about 6% of all tumors, is benign vascular malformation. Lymphangioma of the cheek is a rare entity that often shows slow progressive enlargement of the lesion resulting in swelling over the affected region. Whereas, it can be associated with syndromes like Turner's syndrome, Noonan's syndrome, trisomies, cardiac anomalies, fetal hydrops. Treatment depends on the size and location of the lesion, proximity to anatomic structures, and infiltrating into the surrounding tissues, further complicating treatment. Macrocystic lesions are localized and can be excised easily, while microcystic lesions are diffuse and are difficult to remove into Completely excised mass with two cms of safe margin, involving complete depth removed from the adjacent unaffected mucosa. No after-surgery complications like wound dehiscence and scarring were noticed figure [6]. Regular follow up at definite time intervals are being done for assessing the recurrence.



Figure 6: Post OP Profile View

There are ample of techniques for management of this benign lesion like surgical resection, radiotherapy, cryotherapy, electrocautery, sclerotherapy, administration of steroids, embolization, ligation, laser surgery, and radiofrequency tissue ablation technique. [4] A definitive diagnosis and intervention help to reduce the functional, psychological disturbances, and cosmesis.

Surgery is considered to be the gold standard and is the treatment of choice for every surgeon, but the absence of capsule and infiltrating nature of the tumor makes complete removal next to impossible, and therefore chances of recurrence are more often. Lymphangiomas did not response to sclerosing agents like hemangiomas.

V. Conclusion

The Differential diagnosis in cases of soft tissue swelling of the buccal mucosa should involve the lymphangioma. As earlier diagnosis will help in better treatment options for the patient. In the present study, surgical excision seems to be the safe and effective treatment of lymphangioma, preventing further recurrence in the future.

Conflicts of Interest: Jabalpur Hospital and Research Center, Jabalpur (M.P.).

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