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Original Research Paper

General Surgery

AXILLARY AND BREAST LYMPHANGIOMA: AN UNUSUAL CAUSE OF BREAST AND AXILLARY LUMP IN ADULT PATIENTS

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ABSTRACT Lymphangioma is a benign congenital lymphatic tumor of the lymphatic system due to developmental anomalies. Lymphangioma is commonly seen in the infant and pediatric age groups. In this study, we presented a rare case of axillary and breast lymphangioma in an adult female patients. Patient 24 -year-old lady presented with a large swelling around 15 x 10 cm in the breast and axilla since 6 months of duration. Clinical and radiological examinations were suggestive of breast and axillary lymphangioma. Patient underwent complete surgical resection and diagnosis confirmed on histopathological examination. Lymphangioma of the breast and axilla in adulthood is very rare and very few cases have been reported in the literature. Evaluation for breast cystic lymphangioma should be considered for a prompt diagnosis and definitive treatment to prevent recurrence and complications First line of treatment is surgery with safe margins.

KEYWORDS : Lymphangioma, Breast and axillary lymphangioma, Lymphatic malformation

INTRODUCTION

Lymphangioma is a benign congenital lymphatic tumor of the lymphatic system due to developmental anomalies. Lymphangioma is also known as cystic hygroma. Lymphangioma is commonly seen in the infant and pediatric age groups [1]. Lymphangioma uncommonly seen in adults and around 150 adult cases reported to them have appeared in the literature [2]. Common locations are the cervical and facial regions followed by less frequently in the axillary, inguinal, retroperitoneal, and thoracic regions. Causes of lymphangioma, is mainly congenital in etiology, involving malformation of lymphatic pathways, aberrant lymphatic growth, and tissue sequestration during the developmental phase. Some literatures have suggested the role of trauma or viral infection in their development [3]. In this study, we presented a rare case of axillary and breast lymphangioma in an adult female patient.

CASE REPORT

Our patient, a 24 -year-old lady, presented with a complaint of swelling over the left side of breast and axilla since 6 months of duration. The swelling may arise after 3 months of post-natal period after normal vaginal delivery. She was actively breast feeding.

Patient noted that swelling initially in the breast and later it is extending to the axilla. . She denied similar kinds of history in family. She was not a known case of any chronic illness. She had irregular menstruation since she delivered baby. On examination, breast asymmetry was noted and a large swelling was present in the left side of the breast extending to the apex of the axilla, size around 15 x 10 cm. which was irregular in shape, normal overlying skin, normal temperature and tenderness, smooth surface and soft in consistency. (Figure- 1)

On ultrasonography- It was showing a large multisepted cystic lesion around 18 x 8 cm in the anterio-lateral chest wall suggestive of veno-lymphatic malformation and MRI was showing a multiloulated well circumscribed cystic lesion measuring 10.2 x10.3 x16.5 cm in anterolateral aspect left chest wall with multiple thick septation within the lesion and it was appeared fluid signal intensity on all sequences. (Figure-2)

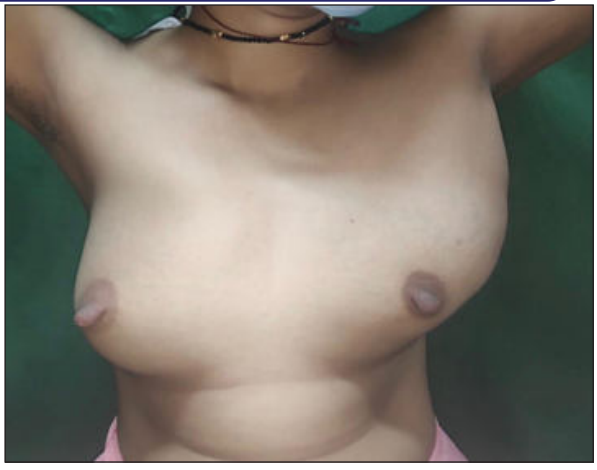

Figure 1- On Inspection On Hand Rising- There Was Asymmetry Of The Breast Seen With Large Swelling Present In The Left Side Of The Breast And Axilla

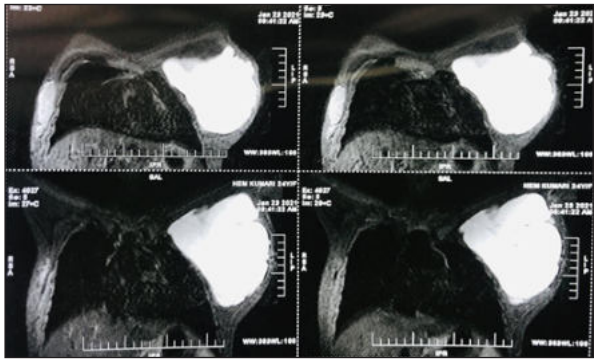

Figure 2- MRI Image Showing-well Circumscribed Cystic Lesion In The Anterolateral Aspect Left Chest Wall And Appearing Fluid Signal Intensity.

She underwent surgical intervention intra-operatively it was large, thin walled, and cystic in nature with loosely adhered to surrounding structures, it was looking like arising from the left

axilla and extending inferiorly till the lower border of the breast and medially under the pectoralis muscle, posteriorly just above the chest wall and laterally till the latissimus dorsi muscle. During the procedure we easily dissected from the surrounding structures but at the apex it was encircling the axillary vein and extending above the vein, hence we punctured and aspirated its content then we secured the axillary vein and then we excised it completely.

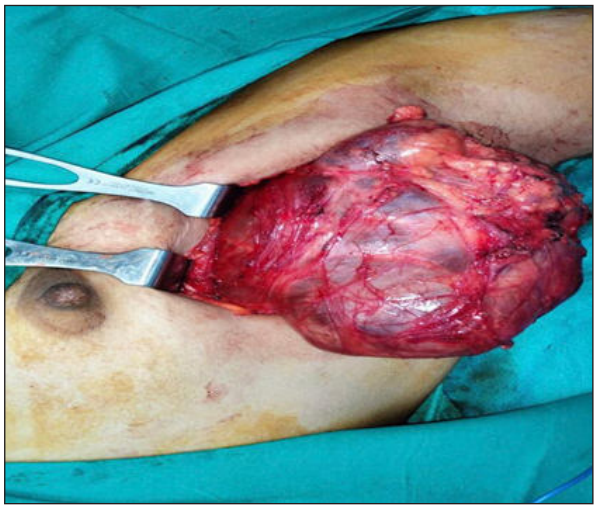

Figure 3- Intra Operative Image Showing- Large, Thin Walled And Cystic Mass In The Left Breast And Axillary Region

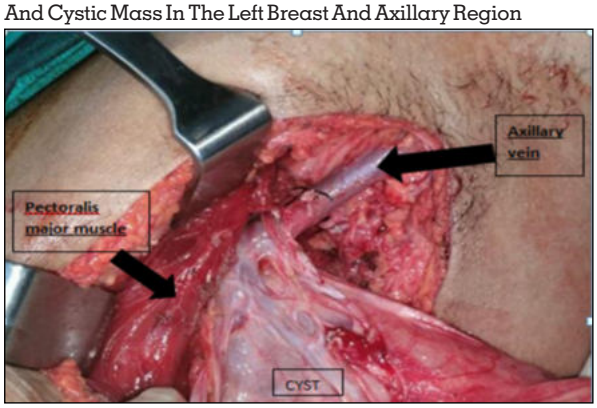

Figure 4- Intraoperative Image Of Cyst After Opening Showing – At The Apex Of The Axilla It Was Encircling The Axillary Vein And Extending Above The Vein

On histopathological examination, this finding was consistent with lymphangioma which showed cyst wall lined flattened lining endothelial cells. The sub-epithelium showed smooth muscles, irregular dilated vascular channels of variable size, mixed adipose tissue, and mild lymphoid aggregates. Post-operative period was uneventful. Regular follow-up was done till 6 months post-surgery and there was no sign of recurrence and seroma formation was noted.

DISCUSSION

Lymphangiomas are one of the rare benign tumors of the lymphatic system which is due to lymphatic malformations resulting from lymphatic dilatation of endothelial lining caused by congenital weakness of the wall, blockage of lymphatic channels, or proliferation of lymphatic vessels. Lymphangiomas are usually found in infants prior to the age of 1 year, and about 90% of the lesions occur in children younger than 2 years [1]. Lymphangiomas are considered a congenital malformation and account for 6% of all pediatric soft tissue tumors. The most common sites of occurrence are the posterior triangle of the neck and it is rarely found in the axilla, mediastinum, groin, retroperitoneal space, and pelvis [4]. Lymphangioma of the breast and axilla in an adult female is extremely rare and very few cases have been reported in the literature [5]. On the basis of microscopic characteristics, three types of lymphangiomas are described as capillary, cavernous, and cystic. Capillary lymphangioma is composed of small, capillary-sized lymphatic vessels. Cavernous is composed of dilated lymphatic channels in a lymphatic stroma containing lymphoid aggregates. Cystic lymphangioma is largely filled with straw-colored, protein-rich fluid. Cystic types have the potential for extensive infiltration of surrounding tissues and lead to surgical difficulties [6]. For diagnosing of lymphanigoma, the following modalities are available- imaging studies, FNAC and confirmatory diagnosis are made on histopathology. Mammography identified a round/lobulated lesion with increased opacity, whereas ultrasonography showed a multiloculated hypoechoic cystic mass, with variable sized septa with solid components [7].Computed tomography (CT) or magnetic resonance imaging (MRI) provides more accurate images and a more in-depth assessment of the tumor. On MRI, cystic lymphangiomas are seen as septated masses with a low T1- weighted and high T2-weighted signal intensity [8]. FNA is usually not diagnostic, since it cannot evaluate the architecture of these lesions. Therefore, surgical excision with histopathological diagnosis remains the mainstay of diagnosis. Part et al. [5] suggested immunohistochemical investigations in their study, which are able to distinguish between hemangioma and lymphangioma. Lymphatic endothelial markers are CD31, CD34, podoplanin, LYVE-1, and PORX-1. Otherwise, the vascular endothelial marker is VIII-associated antigen. The treatment of choice for breast lymphangioma is complete surgical excision. Ensuring safe margins is the importance to prevent recurrence; however, obtaining safe margins may be difficult due to the tendency of lymphangioma to infiltrate the surrounding soft tissue. However, in our case surgical excision was felt to be particularly uncomplicated with thin encapsulation of the mass, weak adhesions to the surrounding tissues, with minimal neurovascular sacrifice except in apex where it was densely adhere to axillary vein. Different treatment options include incision and drainage, sclerotherapy, steroids, radiotherapy, and carbon dioxide laser [9]. However, these are associated with a high rates of recurrence. Thus complete surgical excision is needed for low probability of recurrence[1]. Long term follow-up has been suggested as an important element of management, as recurrence has occurred as late as 6 years after removal [10].

CONCLUSION

In conclusion, lymphangioma is an uncommon tumor in adulthood. Breast and axillary lymphangioma is even rarer and very few cases have been reported in the literature. Symptomatically presents as a soft, painless, and cystic mass in the breast and axillary region. MRI is useful in diagnosing and can help to determine the extent of the tumor. Final diagnosis can be achieved by histopathological examination of the operative specimens. This is a benign tumor that can be treated with different methods. First line of treatment is surgery with safe margins.

Evaluation for breast cystic lymphangioma should be considered for a prompt diagnosis and definitive treatment to prevent recurrence and complications

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