# Lymphangioma of breast and axilla in an adult female: A rare occurrence

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## ABSTRACT

Lymphangioma is a developmental condition caused by the failure of lymphatic channels to communicate with the venous system or sequestrated lymphatic channels. Breast and axilla are rare sites for lymphangioma. Very few cases of lymphangioma of breast and axilla have been reported in the literature. Here, we present the case of a 23-year old female presenting with swelling in the right breast and axilla for ten months. Ultrasonography (USG) showed a multiloculated anechoic mass lesion in breast and axilla. Magnetic resonance imaging (MRI) showed multiloculated septate mass homogenously hyperintense on T2 and hypointense on T1. The USG and MRI findings were suggestive of lymphangioma and confirmed on cytology and histopathology. The location and imaging findings were typical of breast lymphangioma.

**Keywords:** *Breast, Axilla, Lymphangioma, Lymphatic channels.*

Lymphangioma is a benign lymphatic tumor of developmental in origin. Approximately, 90% of lymphangiomas commonly presentin the second decade of life [1]. It is a vascular malformation rather than a true neoplasm resulting from the failure of lymphatic channels to connect with the venous system or sequestered lymphatic tissue which failed to communicate with lymphatic channels [1]. Subsequently, these lesions enlarge over time by the collection of fluid and turn cystic [2].

We present a case of lymphangioma of breast and axilla in an adult female with typical radiological, cytological and histopathological findings with a review of the literature. Since the breast and axilla are a rare sites of lymphangioma in adulthood, we are reporting this case because of its rarity.

## CASE REPORT

A 23-year old female presented with a painless swelling in the right breast and axilla for ten months. There was no relevant family and past medical history.

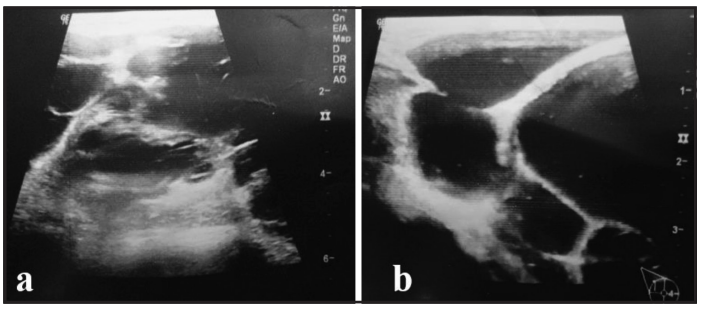


**Figure 1: USG showing a multiloculated anechoic mass lesion in outer quadrant of the right breast measuring 10 x 8 cm and is septate.**

The general physical examination (GPE) showed all vitals within normal limits. On examination, there was a lump in the right breast involving the right upper outer and inner quadrant. No palpable lymph nodes were detected.

Ultrasonography (USG) showed a multiloculated anechoic mass lesion in outer quadrant of the right breast measuring 10 x 8 cm and was septate (Fig. 1). The mass was compressing the breast tissue of inner quadrant. Areola and rest of breast tissue were normal in architect. The possibilities of hydatid cyst and lymphangioma were kept on USG. A magnetic resonance imaging (MRI) of the right breast and axilla showed a

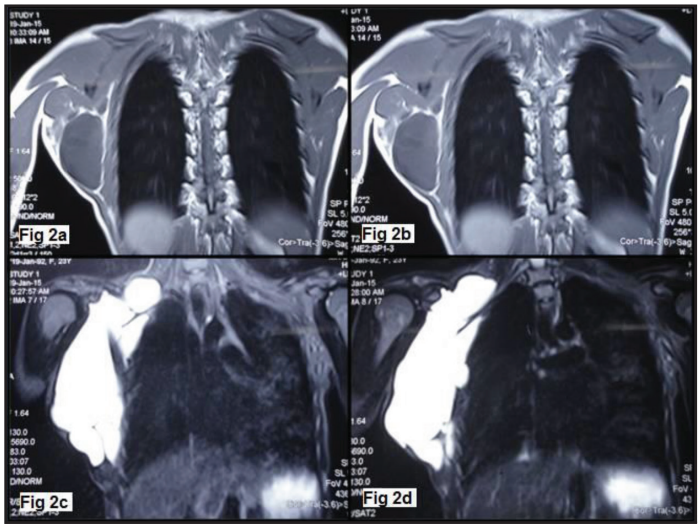


**Figure 2: (2a& 2b) Coronal cut showing hypointense well defined multiloculated septate mass lesion present in right breast and axilla measuring 12 x 6 x 18 cm (T1 weighed); (2c & 2d) Coronal cut showing hyperintensemultiloculated cysts (T2 weighed).**

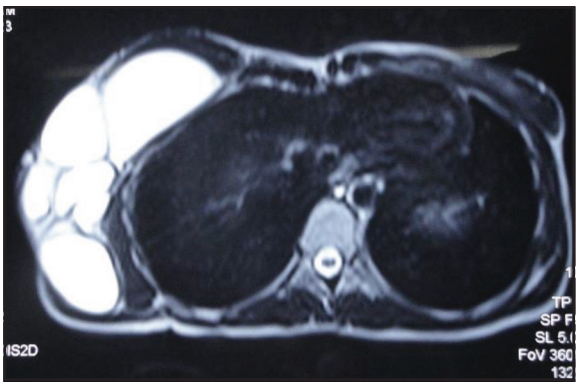


**Figure 3: Sagittal cut showing hyperintensemultiloculated cysts with hypointenseseptae (T2 weighed).**

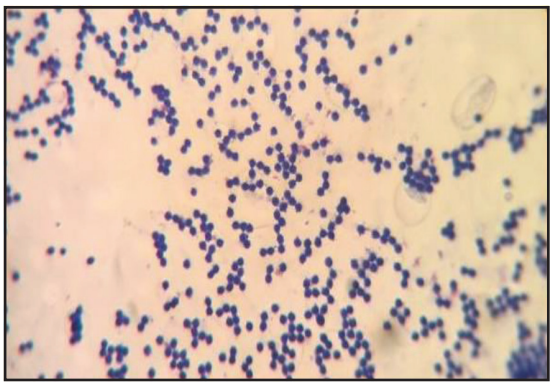

**Figure 4: The breast fluid smears showing mature lymphocytes. (Giemsa, x40).**

well-defined multiloculated septate mass lesion present in the right breast and axilla measuring 12 x 6 x 18 cm. Anteriorly and medially, it extended along the anterior chest wall lifting the right pectoralis major muscle and right breast. Right pectoralis minor muscle passed from within the substance of mass. Inferiorly, it extended along the lateral wall. Posteriorly, it extended between the serratus anterior and latismusdorsi muscle causing its displacement. Laterally, it bulged through axilla in the subcutaneous plane. Superiorly, it extended above the dome of axilla encasing the neurovascular bundle and reaching/ extending in the supraclavicular region in station V of the neck. The mass was heterogeneously hyperintense on T2, hypointense on T1. Septae within it were hypointense. No solid component was seen. Few fatty areas appeared between the locules of mass which appeared to be the continuation of subcutaneous fatty tissue. Surrounding subcutaneous tissue, underlying bones/ribs and scapula were normal. Related muscle showed masseffect; however, no signal changes were seen. On postcontrast, septae showed enhancement (Fig. 2 and 3). The impression on MRI was a multiloculated cystic mass lesion in the right breast with an extension on axilla with a diagnosis of lymphangioma of the right breast and axilla.

A 10 ml clear fluid was aspirated from the breast swelling on performing fine-needle aspiration (FNA). Wet smears were examined for cholesterol crystals; however, no cholesterol crystals were seen. On Giemsa staining, the breast fluid smears showed mature lymphocytes (Fig. 4). The cytological report was consistent with lymphangioma and the breast was signed out. Diagnostic vacuum-assisted biopsy was performed without complete excision of the mass and submitted for histopathological examination (HPE). HPE showed large-caliber lymphovascular spaces with smooth muscles in-wall (Fig. 5). HPE diagnosis of vaso-formative lesion favoring cystic lymphangioma was signed out. After six months follow-up, there was no new complaint and on examination and no new swelling developed in the right breast and axilla.

## DISCUSSION

Lymphangioma of the breast in an adult female is extremely rare and very few cases have been reported in the literature. Lymphangioma is developmental in origin and presents in the second decade of life [1]. Regarding the etiology of lymphangioma, some authors believe that it is a hamartomatous malformation resulting from the inability of the lymphatic system to communicate with the venous system. Another closely related opinion is that lymphangiomas are sequestered lymphatic tissue which failed to communicate with the lymphatic channels [3,4]. Subsequently, these lesions enlarge over time by the collection of fluid. These cystic spaces, in turn, communicate with large dermal lymphatics and subepidermal vesicles, but not with the regional lymphatic system [2].Review of literature shows that lymphangiomas arising from the breast in adulthood have been reported by Almohawes [5], Gómez-Calvo et al [6] and Kyueng-Whan Min et al [7].

On the basis of microscopic characteristics, three types of lymphangiomas are described as capillary, cavernous, and cystic [8]. 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. Cystic lymphangiomas are also classified into microcystic, macrocystic, and mixed subtypes, according to the size of their cysts [8]. Microcysticlymphangiomas are composed of cysts, each of which measures <2 $cm^3$while in macrocystic lymphangiomas, cysts measure >2 $cm^3$in volume. Lymphangiomas of the mixed type contain both microcystic and macrocystic components.

In the adult mammary gland, lymphatics originate from interlobular connective tissue and wall of mammary ducts, communicate with the cutaneous lymphatic plexus around the nipple and finally pass to the axillary lymph nodes. Lymphangiomas of the breast are often seen in areas of the jugular buds in the neck. Most common sites in the breast are upper outer quadrant, the tail of Spence and subareolar space [9]. Since lymphangiomais believed to be due to failure of communication with the main central lymphatic ducts, thus they have a tendency to occur along the lymphatic drainage routes being directed towards the axilla [5]. Lymphangioma is usually an asymptomatic smooth mass. However, rapid expansion can occur because of hemorrhage into

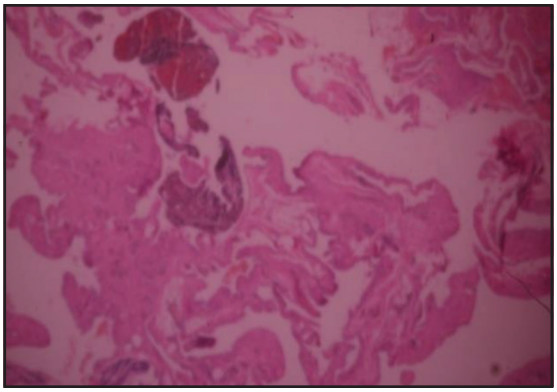

**Figure 5: HPE of biopsy from breast swelling showing large caliber lymphovascular spaces with smooth muscles in wall. (H&E, x40).**

the cyst, inflammation or trauma [5]. Breast is a very rare site of lymphangioma and differential diagnosis include simple cysts, fibrocystic disease, lymphocele, hematoma, abscess, hemangioma and post-surgical collection [1].

Diagnostic modalities are imaging studies, FNAC and confirmatory diagnosis is made on histopathology. Among imaging studies, USG is useful in young women with dense breasts and helps to differentiate solid from cystic masses [1]. Some authors suggest that when the lymphatic ducts in cases of lymphangioma are prominent on ultrasound, appearance can resemble duct ectasia, however, upon compression by the transducer, lymphatic ducts in lymphangioma are totally collapsed while they will not be affected by the compression in cases of duct ectasia [5]. On mammography, breast lymphangioma appears as round or lobulated density. However, the findings are non-specific since many cases occur high in axilla making visualization difficult [5].

MRI is proposed to be the best modality for the diagnosis and evaluation of lymphangioma extension [10]. Cystic lymphangioma appears as multiseptate masses on MRI, usually following the signal intensity of fluid, showing low signalintensity on T1 and high signal intensity on T2, with an enhancement of the septa. FNA fluid aspirates will show clear, yellowish, or straw-colored fluid, with plenty of lymphocytes. However, 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.

Treatment of lymphangioma is total surgical excision. Ensuring safe margins is of paramount importance to prevent recurrence; however, obtaining safe margins may be difficult due to the tendency of lymphangioma to infiltrate the surrounding soft tissue. Steroid injection and sclerosis which causes sclerosis and decrease the size of the lesion. Other options are carbon dioxide laser which vaporizes some of surface lymphatic vessels. Streptococcal lysine induces local inflammatory cytokines that increase endothelial cell permeability, lymph drainage and flow leading to shrinkage of cystic spaces. All these modalities are associated with a high recurrence rate and thus subsequent surgery becomes difficult. Hence they are used as an adjuvant to surgery. For poor surgical candidates, radiotherapy is the option considered [1,5].

## CONCLUSION

In conclusion, lymphangioma of breast and axilla in adulthood is rare and very few cases have been reported in the literature. Imaging helps in guiding to correct diagnosis, however, FNAC and histopathology remain the mainstay of diagnosis in most cases. Primary treatment is surgery with safe margins.

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