Cystic Lymphangioma of the Breast: Magnetic Resonance Imaging Features

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Key Words
Cystic lymphangioma · Magnetic resonance imaging · Cystic spaces

Summary
Background: The aim of this study was to report magnetic resonance imaging (MRI) features of cystic lymphangiomas of the breast. Methods: MRI of the breast was performed using a phased-array double breast coil with a 1.5-T MR scanner. Routine T1 and T2 and post-contrast sequential imaging was performed. Results: The MRI characteristics of cystic lymphangioma in the breast are described. Conclusion: MR imaging provides for multiplanar evaluation and diagnosis of cystic lymphangioma of the breast, which is a rare occurrence.

Introduction
Cystic lymphangiomas, otherwise known as cystic hygromas, are congenital malformations of the lymphatic system usually seen in children below 2 years of age (90%) and rarely in adults [1]. Common locations include neck (70%), axilla (20%), and abdomen (10%). Cystic lymphangiomas of the breast are extremely rare, and very few reports exist on their imaging appearance [2, 3]. They are asymptomatic and grow very slowly. In this report, we describe the magnetic resonance imaging (MRI) findings of cystic lymphangioma involving the entire breast.

Case Report
A 23-year-old woman presented with a history of swelling in the left breast. She had first noticed the swelling 5 years back but was not evaluated as she never experienced any discomfort. Recently, the swelling had become painful and rapidly increased in size, which prompted her to seek medical advice. Physical examination revealed an enlarged left breast with stretched shiny skin. There was no discoloration or puckering of the skin or nipple retraction. No enlarged lymph nodes were palpable in the axilla. The right breast was normal. Sonography showed a multicystic septated lesion involving the entire breast and extending to the axillary tail. Low-level echoes were seen in the fluid within the cysts (fig. 1). Doppler evaluation revealed no significant vascularity. MRI demonstrated a large multiloculated and multiseptated mass involving the entire breast and extending to the axillary tail. The cyst spaces appeared hypointense on both T1- and T2-weighted fat-suppressed images, which indicated the presence of highly proteinaceous fluid or hemorrhagic components (fig. 2). The septae appeared hypointense on both T1- and
T2-weighted imaging. On dynamic post-contrast images, the intervening septae showed considerable enhancement. The cross-junctions of the septae showed enhancing nodules. The fluid-filled spaces had regular margins and did not show any intraluminal enhancement (fig. 3).

A diagnosis of cystic lymphangioma was offered. Aspiration was carried out under ultrasound guidance. The fluid obtained was hemorrhagic and microscopy revealed few lymphoid cells. Biopsy from the lesion revealed cavities that were lined by flattened endothelium and contained lymphoid cells, which was in keeping with the radiologic diagnosis.

**Discussion**

Cystic lymphangiomas are rare lymphatic malformations resulting from retarded development or obstruction of primordial lymphatic channels. These commonly occur in the neck, and involvement of the breast is extremely rare. Cystic lymphangiomas have been classified into simple, cystic, and cavernous types. Simple lymphangiomas are composed of...
small capillary-sized, thin-walled vessels with considerable connective tissue. Cavernous lymphangiomas are made up of dilated lymphatic channels in a lymphatic stroma containing lymphoid aggregates. Cystic lymphangiomas consist of well-defined cyst-like spaces lined by endothelial cells and filled with clear lymph fluid [4]. These result from sequestration of either lymph sacs or lymph spaces that fail to connect or communicate with the main central lymphatic channels [5] and occur along lymphatic drainage routes. Lymph vessels of the adult mammary gland originate from interlobular connective tissue and walls of the mammary ducts and communicate with cutaneous lymphatic plexuses around the nipple, the subareolar plexus and finally pass to the axillary nodes [6]. Localized cystic lymphangiomas of the breast can occur in the upper outer quadrant, the tail of Spence or in the subareolar space [6–12]. In our case, the entire breast was affected. Mammography and ultrasound have been used to diagnose cystic lymphangioma of the breast [6–14]. On mammography, they appear as diffuse or localized dense masses with lobulated margins. No macro- or micro-calcifications are identified. Ultrasound shows these lesions to be cystic with multiple septations. The cyst contents are usually clear, but may show low-level internal echoes that represent internal hemorrhage or proteinaceous content. MR imaging reveals a well-defined multiseptated and multicystic fluid-containing lesion involving the entire breast and extending to the axillary tail. The lesion can also be unilocular. The cystic spaces are usually filled with clear fluid that appears hypointense on T1- and hyperintense on T2-weighted imaging. The fluid at times appears hyperintense on both T1- and T2-weighted sequences, suggesting the presence of hemorrhage or increased protein levels which may indicate inflammation [9, 14]. Cystic lymphangioma of the neck with hemorrhage has shown complex MR imaging signal intensities [15].

Lymphangiomas are benign and have never been known to harbor malignancy. Cytological studies of fluid aspirated from lymphangiomas show scanty cells predominantly of lymphoid origin [16]. Surgery is the choice of treatment. Complications include infection, hemorrhage, and fistula formation. Grossly, cystic hygromas are multiloculated and multicystic, having cysts of varying sizes with intervening septae. Microscopically, the cyst walls are lined by a single layer of flattened epithelium.

The differential diagnoses include simple cysts and fibrocystic disease, lymphocele, hemATOMA, and hemangioma. Fibrocystic disease is generally bilateral and confined to the breast disc [2, 3]. Hematomas are localized, unilocular and may show hemorrhagic residua. Hemangiomas are also localized and enhance on contrast administration [2]. Rarely, cavernous hemangiomas may involve an entire breast and thus closely mimic cystic hygromas. Hemangiomas show intense enhancement after gadolinium administration, but some may even show gradual filling in of contrast on delayed scans up to 30 min later. Phleboliths may be identified on mammograms [6]. Cystic hygromas always diffusely involve the breast, extend to the axillary tail and are relatively avascular.

In conclusion, we have described the MR imaging features of cystic lymphangioma of the breast, a rare congenital malformation of the lymphatic system characterized by the presence of non-communicating lymphatic channels, resulting in the formation of multiple cystic spaces. Though MR imaging may be expensive and not freely available, it is very helpful in characterizing the lesion and identifying its extent, because of its multiplanar imaging ability.

Conflict of Interest

The authors declare no conflict of interest.

References