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Axillary Cystic Lymphangioma Presenting in Pregnancy

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Lymphangiomas are benign tumors that are composed primarily of lymphatic vessels. They are classified into three pathologic groups: the lymphangioma simplex, the cavernous lymphangioma, and the cystic lymphangioma or cystic hygroma (1). Cystic lymphangiomas most commonly affect infants. Occurance in adults is rare, with fewer than 90 cases reported in the literature.

A 24-year-old female presented with a large left axillary mass, three months after delivery. She first noticed the mass as a small swelling in her left axillary fossa during the 7th month of her pregnancy. Within a 6-month period, the swelling enlarged gradually into a huge mass, 10x12x17cm in diameter. She also reported having received a vaccination against tetanus in her left arm two weeks before she first noticed the swelling.

Physical examination revealed a non-tender fluctuant mass, 10x12x17cm in diameter, situated between anterior and posterior axillary lines. The mass was not attached to the skin and had no signs of inflammation. Ultrasonod revealed a large cystic mass with internal echoes and debris at the dependent part of the cyst (Fig. 1A). Color doppler ultrasonography did not show any flow within the mass.

Computed tomography showed that the cystic mass was encapsulated and the attenuation value of the cyst content was 3 Hu (Fig. 1B). MRI examination predicted the benign nature of the mass, which was well-demarcated from the surrounding tissues. Hyperintensity of the mass in both T1 and T2 -weighted images (Fig. 1C, 1D), and T2-hypointense debris indicated that the cyst content was hemorrhagic. Total excision was performed under general anaesthesia. Pathology revealed a 17cm unilocular cystic mass, with a thin wall and a

serohemorrhagic content. Pathologic findings were compatible with cystic lymphangioma.

Most cystic lymphangiomas are detected in childhood, 50 to 60 percent being evident at birth and 90 percent appearing before 2 years (2). Sevent-five percent occur in the neck, 20% occur in the axilla, and the remaining 5% arise in locations such as the mediastinum, retroperitoneum, bone, kidney, colon, liver, spleen, and scrotum (3).

Presentation of the tumor for the first time in adult life is rare, and fewer than 90 cases have been reported in literature, most of which were located in the cervical region (2, 4). Our review of the literature revealed only two cases of adults with axillary cystic lymphangiomas (5, 6).

The most widely accepted theory about the development of cystic lymphangiomas is that they arise from sequestrations of the primitive embryonic lymph sacs (3). The etiology in the adult population is controversial. Some authors believe that the adult cases are due to delayed proliferation of congenital or acquired lymphoid rests after stimuli such as respiratory infection or local trauma (4). Others propose that adult cystic lymphangiomas are the result of trauma rather than of congenital origin (3, 7).

Our case is remarkable in that it is the second reported case of cystic lymphangioma presenting in the gravidic-puerperal cycle. The first case was a 15-year-old patient with a giant axillary cystic lymphangioma, diagnosed during her second pregnancy (5).

In our country, pregnant women are routinely vaccinated against tetanus infection. The history of

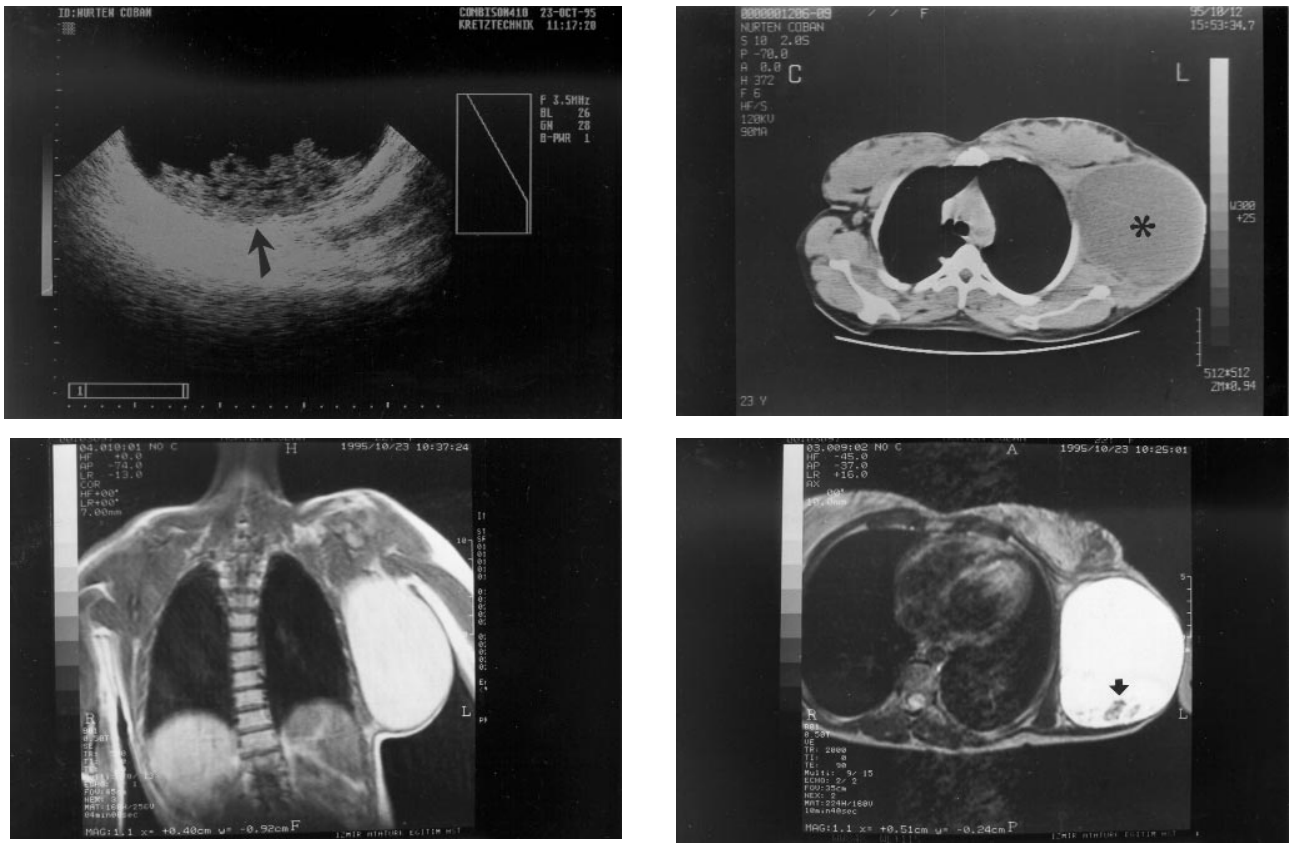


Figure 1. Axillary cystic lymphangioma. A) Ultrasonography reveals a cystic mass with debris (arrow) at the dependent part of the cyst. B) CT appearance of the cystic mass (asterisk). C) T1W coronal and D) T2W axial magnetic resonance images of cystic lymphangioma. Hyperintensity of cyst contents in both T1W and T2W images and T2-hypointense debris (arrow in D) indicate the hemorrhagic character of the cyst.

vaccination in the same arm two weeks before the appearance of the swelling suggests that vaccination might have been the etiologic factor in the development of the tumor. We suggest that the tetanus antigen-mediated reactions or an accompanying local infection might have interfered with local lymphatic circulation,

leading to the formation of a gradually enlarging cystic tumor.

Imaging methods have proven useful in tumor characterization and treatment planning. The benign nature and relation of the tumor to the surrounding

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