Case Report

# Away from the milky way: An extremely rare case of pulmonary ectopic breast

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## ABSTRACT

Ectopic breast is defined as the presence of breast tissue outside the pectoral region. In this article, we are reporting an extremely rare case of pulmonary aberrant breast tissue. A 79-years-old Caucasian woman with the history of hypertension, diabetes mellitus and ischemic heart disease, presented to the emergency department for worsening dyspnea on exertion and pleuritic chest pain over the last few days. Both thoracic CT scan and the gross evaluation of tumor after its removal by thoracotomy were in favor of a soft tissue tumor such as pulmonary lipoma, whilst surprisingly the histological examinations revealed the mass to be ectopic breast tissue. Although it is extremely rare, this diagnosis should be considered in the evaluation of pulmonary masses.

**KEY WORDS:** Ectopic breast, intra-thoracic, mass, pulmonary

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## INTRODUCTION

During the fifth week of gestation, ectodermal thickening is differentiated to form milk-line or mammary ridges on the ventral side, which are extended bilaterally from the axilla to the groin.[1] Normally, these ridges persist in the pectoral region allowing for the development of breast tissue and regress in non-pectoral areas. Any disruption in the embryonic regression of these ridges may result in the formation of ectopic breasts through this line.[2-6]

According to the prevalence rate of ectopic breast tissue among the general population (0.2-6%),[1,7-11] this phenomenon may not be uncommon. However, the prevalence of ectopic breast (supernumerary and/or aberrant) outside the mammary ridges is exceptionally uncommon. The face, chest wall, back, buttocks, and thighs are just some of the various places where ectopic breast tissue (EBT) have been found.[8,12-17] To date, there are only three cases of EBT documented in internal organs.[17-19] We report an extremely rare case of pulmonary aberrant breast tissue.

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## CASE REPORT

A 79-year-old Caucasian female with a medical history significant for ischemic heart disease, hypertension, and diabetes mellitus, was found to have a pulmonary mass visualized on high resolution computed tomography (HRCT) of the chest in 2002 during a workup for dyspnea. Based on the HRCT, the mass appeared cystic in nature but the patient was lost to follow-up over the last ten years.

Recently, she presented to the emergency department for worsening dyspnea on exertion and pleuritic chest pain over the last few days. Initial chest radiography revealed a large cystic mass in her left pleural space [Figure 1].

On physical examination, vital signs were within normal limits and pertinent findings during pulmonary auscultation revealed decreased breath sounds on the left side along with left-sided dullness with percussion. The results of the initial laboratory tests at this time, which included complete blood count (CBC), chemistry panel, and liver function, were normal. A Thoracic CT scan was

requested by the surgical team and it described the mass as a well-defined concentric thick wall cavity in the left upper lobe with smooth inner surface without any calcification and with a region of interest (ROI) of -124 HU, suggesting a pulmonary lipoma [Figure 2].

Due to size of the probable tumor and the patient's symptoms, the surgical team recommended a thoracotomy under general anesthesia in the right lateral decubitus position. The left lower lobe of the lung was collapsed due to the large size of the tumor and its compression effect. After pneumolysis, the mass was observed to occupy the entire left upper lobe. Upon visualization of numerous, enlarged lymph nodes in the mediastinum, lobectomy of the left upper lobe, lymphadenectomy, and contrapleural costal resection were performed.

The solitary, encapsulated mass was surgically removed. Grossly, the mass appeared to have features of a benign soft tissue tumor (i.e. lipoma) measuring 240 × 130 × 100 mm [Figure 3].

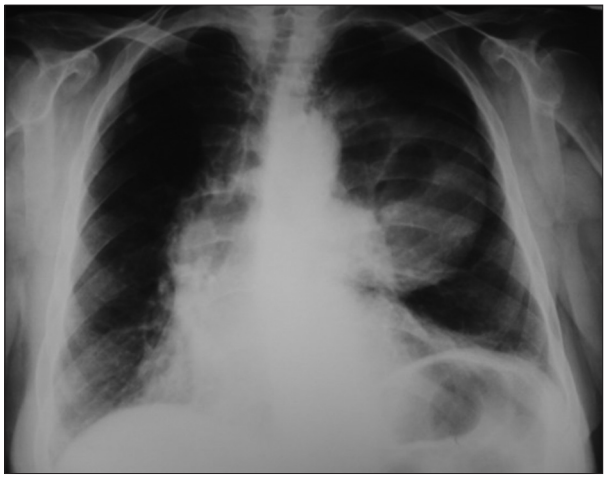

**Figure 1:** Chest radiography of the patient before thoracotomy

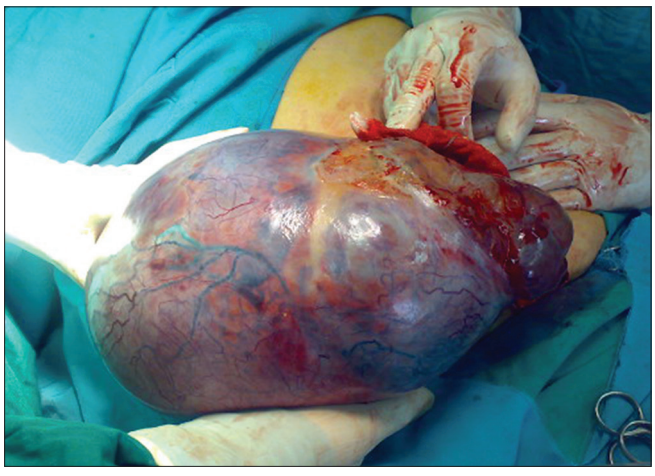

**Figure 3:** Gross appearance of the pulmonary mass

Histological examinations revealed papillary and tubular structures, similar to breast tissue. No mitosis, necrosis, or cells with atypical features were detected via histologic evaluation, suggesting the benign nature of the tumor [Figure 4].

Formalin-fixed, paraffin-embedded sections of the sample were stained for estrogen receptor (ER) and progesterone receptor (PR) using immunohistochemistry (IHC). For IHC investigation, we utilized FLEX monoclonal mouse anti-human antibodies: 1D5 at dilution 1:60 for ER, and PgR636 at dilution of 1:300 for PR from Dako-Denmark (A/S production svej 42 DK-2600 Glostrup, Denmark) according to the manufacturer's protocol. All slides which were stained on an automated system were negative for both ER and PR.

Post-operatively, the patient was monitored for six days and was discharged to home without any surgical or hospital complications. During a 6-month follow-up appointment,

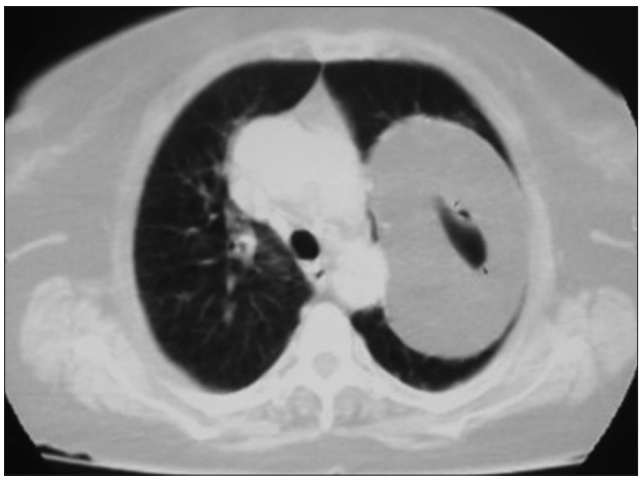

**Figure 2:** Chest CT before surgery (note the large mass in the left lung)

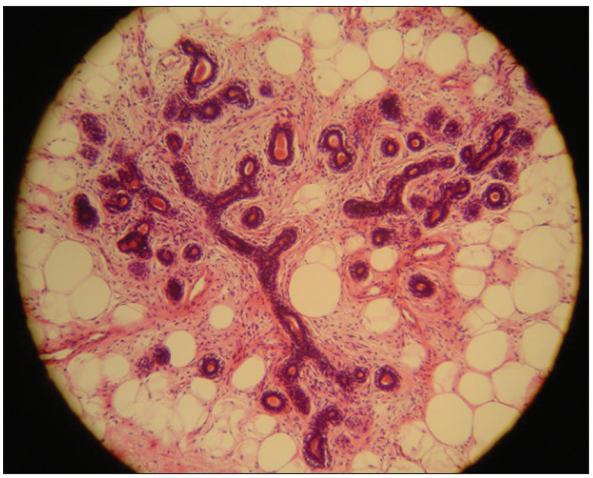

**Figure 4:** Histopathology of pulmonary ectopic breast (H and E, ×100)

the patient denied any symptoms of shortness of breath or dyspnea on exertion. Re-expansion of the left lower lobe significantly improved the patient's condition, and she is currently undergoing regular medical follow-up.

## DISCUSSION

EBT is residual tissue abnormally persisting during the embryological development against the gradual regression of mammary ridges in non-pectoral regions.[7] The axilla is the most frequently involved location for EBT (70%). The second most frequent place is the vulva. EBT may be accompanied with symptoms like discomfort or pain during pregnancy, lactation, or in the pre-menopausal period.[5,20]

According to de-Cholnocky, approximatly 4% of EBT cases may develop into a primary ectopic breast carcinoma.[21] Therefore, the conversion of EBT to malignancy should be considered in all diagnosed cases.

Yeniay *et al.*, reported a 71-year-old female with an abdominal aberrant breast tissue found incidentally in a piece of mesenteric biopsy during operation for a recurrent endometrial malignant tumor.[17] Another case of developed heterotropic breast tissue found in internal organs was reported by Sasaki *et al*. They reported a heterotopic breast epithelial inclusion of the heart incidentally found on a native heart of a 73-year-old man who received an orthotopic heart transplantation for ischemic cardiomyopathy.[18]

Sundaram in 1969 reported several pseudo-tuberculosis conditions of the lung including silicosis, flurosis, etc. One of the reported cases was of a 20-year-old woman with throat irritation and chronic cough, bilateral axillary lymph nodes, and streaky shadows on chest x-ray localized to the left upper lobe. She was treated as a case of smear-negative TB laryngitis with anti-TB therapies that resulted in no appreciable changes. The histopathologic examination of the removed axillary glands yielded the diagnosis of mature lobule of breast. Although the author proposed the diagnosis of accessory breast for the pulmonary findings in chest x-ray, no sampling was performed from the pulmonary inclusions in that case.

Our case is the only well-documented case of pulmonary ectopic breast worldwide. Although the results of ER and PR antibody studies did not add any confirmation to the diagnosis of ectopic breast, the obvious resemblance of the hisopathologic pattern of tumor to the breast tissue rejected any other possibility.

Unfortunately the concordance of ER and PR expression with the existence of breast tissue in samples was not investigated before. There are some studies around the concordance of ER and PR expression and existence of ectopic breast carcinoma.[22,23] In the study of Shabaik *et al.*, the results showed a sensitivity of 85.7% and specificity of 100% for ER, and a sensitivity of 80% and specificity of 100% for PR.[23] Three cases (16.6%) among the 18 different articles which investigated the ER, PR antibodies in ectopic breast cancer cases, were reported to have negative ER and PR results.[6,20,24]

As mentioned above, our patient did not recall any symptoms during pregnancy, lactation, or in the pre-menopausal period. This could be interpreted by the negative ER and PR status of the ectopic breast tissue. This infers that many of the cells may not contain receptors that are influenced by the alterations in hormone levels.

Generally despite the rare nature of pulmonary ectopic breast tissue, the clinicians should take into consideration this disease in evaluating the differential diagnoses of pulmonary masses.

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