# Bilateral intraductal papillomas arising in ectopic axillary breast tissue synchronously with right breast intraductal carcinoma

Ahmad A. Al-Boukai, MD, FRKSU.

### **ABSTRACT**

تصف هذه الدراسة مريضة تبلغ من العمر 45 عام والتي تبين لديها ورم في الثدي الأيمن. لقد كشف الفحص السريري وجود كتلة غير مؤلمة بقطر 3 سم في الثدي الأيمن إضافة لتورم في كلا الإبطين، دون وجود تضخم للعقد اللمفاوية. وأظهر تصوير الثدي بالأشعة والموجات فوق الصوتية وجود ورم بعلامات خبيثة في الثدي الأيمن حليمي صغير متعدد داخل الأقنية الغدية. تمت مناقشة و ظهور هذه الأنسجة في كل من تصوير الثدي بالأشعة و الموجات فوق الصوتية. إن الهدف من الدراسة هو مناقشة المظاهر الشعاعية لهذه الموجودات الشعاعية و تسليط الضوء على ظهور أورام حليمية داخل الأقنية الغدية في نسيج ثدي هاجر بكلا الإبطين و بشكل متزامن مع الورم الخبيث في الثدي الأيمن و التركيز على أهمية الفحص الشعاعي و الخبيث في الثدي الأيمن و التركيز على أهمية الفحص الشعاعي و الأمواج فوق الصوتية في كشف هذه الحالة.

This study describes a case of 45-year-old woman with a mass developed in the right breast. Physical examination revealed a 3 cm painless mass in the right breast as well as diffuse bulge in both axillary regions, though no significant lymphadenopathy was evident. Mammogram and ultrasound examination were carried out and showed malignant mass in the right breast and ectopic breast tissue in both axillae with multiple intraductal papillomas. Both mammographic and sonographic appearances of these lesions are described and discussed. The purpose of this study is to highlight the importance of occurrence of bilateral ectopic axillary breast with intraductal papilloma synchronously with right breast malignant mass and emphasize the importance of thorough mammographic and sonographic examination of the axilla in detecting such pathology.

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From the Department of Medical Imaging and Radiology, King Khalid University Hospital, College of Medicine, King Saud University, Riyadh, Saudi Arabia.

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Address correspondence and reprint request to: Dr. Ahmad A. Al-Boukai, Department of Medical Imaging and Radiology (40), King Khalid University Hospital, PO Box 7805, Riyadh 1147, Kingdom of Saudi Arabia. Tel. +966 (1) 4671155. Fax. +966 (1) 4671746. E-mail: a\_boukai@hotmail.com

The term "ectopic breast tissue" describes presence of accessory, supernumerary breast, and/or aberrant breast tissue. This is usually occurred as a result of failure of regression of the milk line remnant during embryogenesis. This line develops in the sixth week of gestation as an ectodermal thickening extending from the axilla to the groin along both sides of the ventral surface of the embryo. Under normal circumstances, this milk line regresses except in the thoracic region, where it forms the mammary ridge, from which the normal breast tissue develops. The failure of regression of this line may occur at any level from the axilla to the groin region leading to the formation of ectopic breast tissue.1 It has been postulated that a genetic factor has a role in this development.<sup>2</sup> Ectopic breast tissue may undergo same physiological and pathological processes affecting the normal anatomic breasts and frequently responds to hormonal stimulation associated with menses, lactation, or pregnancy.<sup>3</sup> Engorgement and discomfort may occur as well. Cases of pathologic conditions reported in ectopic breast tissue include phyllodes tumors,<sup>4</sup> fibroadenoma, <sup>5</sup> leiomyosarcoma, cystosarcoma, ductal and lobular carcinomas, mastitis, and Paget's disease. Among others is the reported case of advanced cancer in one breast occurring concurrently with mastopathy in ectopic axillary breast bilaterally.8 Also of interest is the occurrence of intraductal papillomas in ectopic vulvar breast tissue which was traditionally believed to be papillary hydradenoma of the vulva.9 The unique occurrence of bilateral ectopic axillary breast with intraductal papilloma synchronously with right breast malignant mass in this patient prompted this paper, as to highlight the importance of thorough mammographic and sonographic examination of the axilla in detecting such pathology.

**Case Report.** A 45-year-old woman presented with painless right breast mass and bilateral axillary fullness. No family history of breast cancer. She discovered the mass; incidentally, one month prior to the presentation. Physical examination revealed a 3 cm hard mass in the

upper inner quadrant of right breast. No skin changes, nipple retraction nor discharge. No other masses could be felt on either side. However, soft tissue swelling in both axillae was felt and appeared of diffuse nature with no definite focal masses or enlarged lymphadenopathy. These axillary masses were thought to be benign lesions such as lipoma, and the patient was referred to mammography for evaluation of the right breast mass. Mammogram showed an irregular, ill-defined area of asymmetric added density in the deep portion of the upper inner quadrant of the right breast, no associated skin thickening nor architectural distortion (Figures 1a & 1b). No micro-or macro calcification. Both axillae showed areas of glandular parenchyma, which represent accessory breast tissue with multiple low density radio-opaque nodular lesions; some of these lesions are surrounded by radiolucent halo (Figures 2a & 2b). She underwent ultrasound examination, which confirmed the presence of bilateral axillary accessory breast tissue

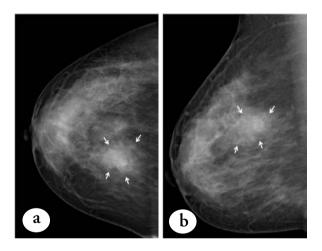


Figure 1 - Mammogram showing the a) craniocaudal and b) oblique mediolateral views of the right breast demonstrating an ill-defined asymmetrical area of an added density in the deep portion of the inner upper aspect (small white arrows), no architectural distortion nor microcalcification.

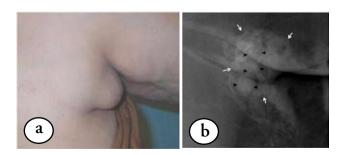
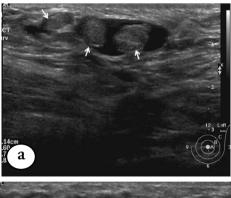
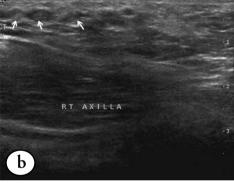


Figure 2 - This is a localized view of the a) left axillary accessory breast and b) the corresponding coned mammographic image showing glandular parenchyma (white arrows) with multiple low density radio-opaque nodular lesions (small black arrows) that represent intraductal papillomas. Note the hallo around these lesions.

and showed multiple dilated ducts with multiple pedunculated intraductal soft tissue nodules that are fixed to the wall. These are seen more clearly on the left side and are consistent with intraductal papillomas (Figures 3a & 3b). The biopsy confirmed benign etiology of these lesions. The right breast mass appeared as an irregular hypoechoic focal mass with ill-defined outline and indistinct margins (Figure 4), no areas of cystic degeneration or microcalcification. No other lesions





**Figure 3 -** Ultrasound examination showing both axillae demonstrating a) multiple dilated ducts with b) multiple pedunculated soft tissue nodules which appeared fixed to the wall and consistent with intraductal papillomas. Changes more clearly on the left side (white arrows pointing to these papillomas).



**Figure 4 -** Ultrasound showing the right breast mass, which appeared as an irregular hypoechoic focal mass with ill defined outline and indistinct margins (white arrows).

to suggest multicenteric or multifocal process. These findings raise the possibility of malignant mass (Breast Imaging Reporting and Data System-5). Fine needle aspiration and true-cut biopsy were accomplished, and the specimen showed invasive malignant epithelial cells consistent with infiltrating ductal carcinoma, modified as Scarff-Bloom-Richardson (SBR) grade III. An extensive workup, including thoracic and abdominopelvic computed tomographic studies, as well as isotope bone scan were carried out to look for any deposits in the bones, abdomen or chest. None of these examinations revealed any distant metastases.

**Discussion.** Polymastia which refers to the presence of more than 2 breasts in human being indicate incomplete involution of the mammary ectodermal ridges, which are also known as the milk line, resulting in the formation of supernumerary/accessory mammary tissue from the redundant clusters of ectopic primordial breast cells. This occurs in 2-6% of females and 1-3% of males. 10 Approximately one third of affected individuals have more than one site of supernumerary breast tissue development. Though accessory breast tissue may be of no physiological significance, some may undergo similar changes affecting normal pectoral breast tissue, such as enlargement with the onset of puberty, pregnancy, or lactation, and can be the site of breast carcinoma. Accessory breast tissue can occur anywhere along the milk line. It is reported to occur in approximately 67% along the thoracic or abdominal portions of this line often below the inframammary crease on the left side of the body, and in 20% in the axilla. Other reported locations include anywhere along the milk line or in the buttock, back, face, and neck. The presence accessory breast tissue at any location other than along the milk line represents a migratory arrest of breast primordium during chest wall development. Accessory breast tissue has been classified according to the contents of the tissue into 8 types; this remains in use up to this date. Type I consists of a complete breast with nipple, areola, and glandular tissue; Type II consists of nipple and glandular tissue but no areola; Type III consists of areola and glandular tissue but no nipple; Type IV consists of glandular tissue only; Type V consists of nipple and areola but no glandular tissue; Type VI consists of a nipple only (polythelia); Type VII consists of an areola only (polythelia areolaris); and Type VIII consists of a patch of hair only (polythelia pilosa).11 Intraductal papillomas are common neoplasm with a relative incidence of 2-3%. They are essentially benign proliferation of ductal epithelium that project into the lumen of the duct and are connected to the epithelium by a fibrovascular stalk. They are often confined to a single breast, while bilateral lesions are reported in up

to 14% of cases. 12,13 Intraductal papillomas as their name implies, are contained within the duct, the duct around them can dilate and form a cystic structure. They are broadly classified into central and peripheral types. The central ones are usually subareolar in location within a major duct, while the peripheral lesions are less common and tend to be multiple. They believed to arise within terminal duct lobular unit and are associated with an increased likelihood of atypical changes and carcinoma. 13 This increased risk of breast cancer ranges from one and half to 2 times the risk of a woman who has non-proliferative changes.<sup>14</sup> Multiple papillomas are usually asymptomatic, discovered incidentally on mammographic, and ultrasound examination. The benign nature of these lesions is confirmed by histology which is usually composed of multiple branching fibrovascular cores lined by 2 layers of epithelial and myoepithelial cells.<sup>13</sup> It is therefore, a crucial issue for the radiologist to have a comprehensive and thorough mammographic and ultrasound analysis as to detect and evaluate such lesions. On mammography, these lesions may appear as multiple rounded or linear dense soft tissue masses with well circumscribed or partially circumscribed margi, 14,15 some of which demonstrate radiolucent halo around it, and occasionally they may produce non-specific cluster of microcalcifications. Shell like calcific deposits in the subareolar region with central lucency may be seen and are certainly due to partial calcification of a large ductal papilloma. Ultrasound findings primarily depend on the gross macroscopic appearance of the lesion. The features may vary from a pure tiny cystic lesion due to focal ductal ectasia to totally solid mass. Three basic patterns are described, intraductal mass with or without ductal dilatation, intracystic mass due to associated excess fluid production in an obstructed duct, and predominantly solid pattern with the intraductal mass totally filling the duct. 13-15 Further, imaging techniques may add in the differentiation between benign and malignant nature of such lesions, particularly the color Doppler and magnetic resonance imaging.

In summary, this is an unusual case of multiple intraductal papillomas in ectopic breast tissue of both axillae presenting synchronously with right breast malignant mass, which the mammographic and the sonographic findings played a role in highlighting this pathology. The awareness of such unique occurrence throughout radiologists and physicians will definitely aids in improving patient management and cares.

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# Related topics

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