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Nodulocystic hidradenoma of the breast: a case review with imaging findings

Relevance. *A nodulocystic hidradenoma is a neoplasm of sweat glands which is very rare in the breast. Its diagnostics is difficult; its imaging characteristics are not well studied and documented. Breast hidroadenomas present a diagnostic challenge to both pathologists and radiologists. Malignant transformation of benign nodular hidradenomas has also been reported. Complete surgical resection is a choice of treatment for this lesion.*

Although we have found twenty-seven cases of clear cell hidradenomas of the breast reported in the literature till today, only four of those cases were diagnosed by both mammography and breast ultrasound. In two reported cases of nodular hidradenoma, false positive biopsy has led to a mastectomy. Malignant counterpart of hidradenoma is called hidradenocarcinoma. To our knowledge, this paper is the first to report MR findings of nodulocystic hidradenoma of the breast.

Case report. *We present a case of nodulocystic hidradenoma of the breast with mammography, tomosynthesis, ultrasound, and MRI results in a 24-year old woman.*

Conclusions. *Nodulocystic hidradenoma should be considered in differential diagnostics of a breast lump. However, there is no particular radiologic feature that could strongly predict the diagnosis of nodular hidradenoma. FNA and core biopsy may sometimes lead to a misdiagnosis, so the excision is usually needed for a definite histopathologic diagnosis.*

Keywords: *Nodulocystic hidradenoma, mammography, tomosynthesis, ultrasound, magnetic resonance imaging (MRI).*

Background. A nodulocystic hidradenoma is a rare cutaneous adnexal-type neoplasm originating from sweat glands. It can occur anywhere in the body but is very rarely seen in the breast.

There is a lack of awareness among breast radiologists about this lesion due to its very low incidence and common removal without imaging assessment. Thus, the imaging characteristics of this tumor are not well established and documented. We present the first case of nodulocystic hidradenoma of the breast with mammography, tomosynthesis, ultrasound, and MRI results in a 24-year old woman, along with a literature review.

Case report. A 24-year old woman applied to the breast imaging center complaining of a palpable painful lump persistent for a few years. Over the last few months, it was growing in size with discoloration of the skin.

She had a strong family history of breast cancer with several female relatives with breast cancer from both paternal and maternal sides. The patient was not tested for breast cancer gene mutations.

Clinical data. During the physical examination, slight skin discoloration was noticed. On palpation, a mobile hard lump was detected in the upper outer quadrant of the left breast, mildly tender to palpation. Ipsilateral axillary lymph nodes were not enlarged on palpation.

Diagnosis. Targeted ultrasonography showed a complicated cyst measuring 3 centimeters in the left breast at the site of a palpable finding (Figure 1).

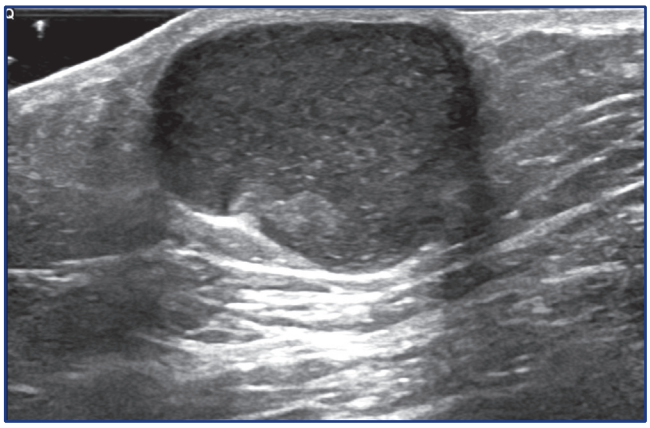

Figure 1 - Ultrasound image demonstrating a complicated cyst in the left breast at the site of a palpable finding.

Ultrasound-assisted core biopsy revealed a low-grade papillary neoplasm of unknown origin but resembling of a papillary lesion of urothelial origin.

The patient then underwent digital breast tomosynthesis and contrast-enhanced breast MRI. CC and MLO views demonstrated a round high density circumscribed mass sized 2.5 centimeters with associated marker clip in the left breast at 3 o'clock located 15 centimeters from the nipple (Figure 2).

Breast MRI showed a round mass with circumscribed margins with a rim of solid enhancement up to 0.6 cm thick. Kinetic assessment of the solid component demonstrated its fast initial enhancement followed by plateau enhancement on the delayed portion of the curve (Figure 3).

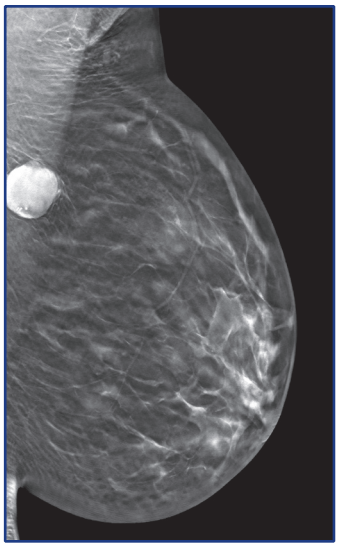

Figure 2 - Mammography, tomosynthesis demonstrating a round high density circumscribed mass with associated marker clip.

Clinically nodulocystic hidradenoma grows slowly; the reported duration of clinical symptoms ranges from 2 months to 15 years, with a mean duration of 2-3 years.

Such tumors may develop at any age but typically they develop between the third and seventh decades of life, usually peaking during the fifth decade [7].

Nodulocystic hidradenomas range in size from 0.7 to 8.0 cm, with an average size of 2.9 cm reported across the studies.

This tumor is more common among women than in men [8]. In our literature review, just six (22%) of the twenty seven cases were male.

Clinical findings often include a palpable mobile hard lump, usually tender on palpation, skin discoloration or ulceration, and nipple discharge if the tumor is located in the nipple areolar area [9, 10].

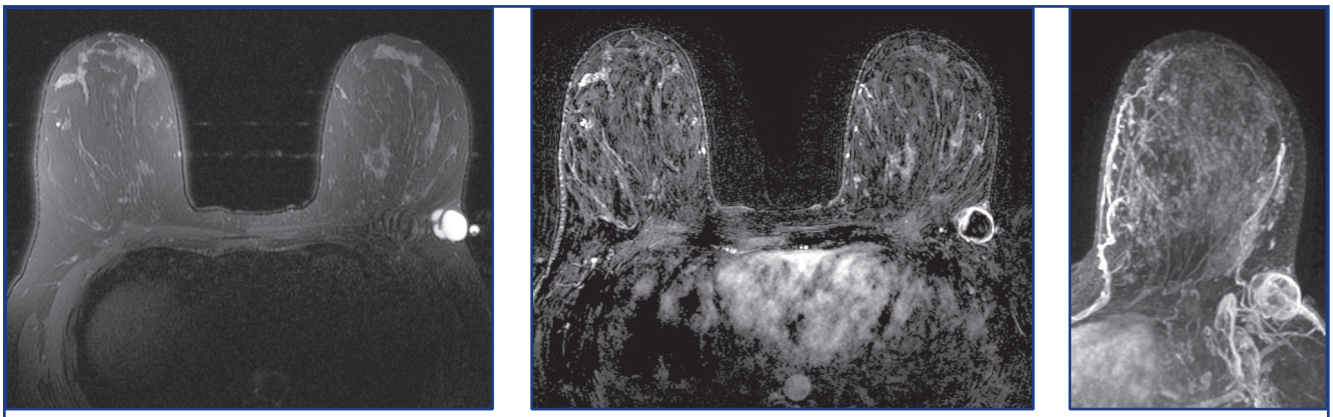

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Figure 3 - Axial T2-weighted image (3-1) of a hyperintense mass. Axial T1-weighted (3-2) and MIP images (3-3) of a round mass with circumscribed margins with a rim of solid enhancement.

A patient then underwent surgical excision of the lump. Final histopathologic results confirmed nodulocystic hidradenoma of the breast.

Pathology

Microscopic description: a relatively circumscribed intradermal cystic proliferation consisting of ribbons of monomorphous keratinocytes with eosinophilic cytoplasm and vesicular nuclei. No atypical cytomorphology or necrosis. Focal ductal differentiation, clear cells, and a vague papillary architecture. Stromal sclerosis and compressed fibrous tissue were also present.

p63 and CK5/6 expressions were noticed.

Postsurgical healing course – without complications.

Discussion. Nodulocystic hidradenoma is a benign dermal tumor which arises from distal excretory ducts of eccrine or apocrine sweat glands [1, 2]. It is also known as clear cell hidradenoma, eccrine acrospiroma, nodular hidradenoma, or solid-cystic hidradenoma [3]. It may contain varying quantities of solid and cystic components and comprises approximately one-third of all hidradenomas [4]. This neoplasm usually occurs in the head, neck, upper body, and extremities. Its occurrence in the breast is rare [5, 6].

The preferable location if in the breast is the nipple areolar complex (around half of the lesions) and the axillary tail [11, 12].

Differential diagnostics includes primary breast carcinoma [13], metastatic carcinoma (renal cell carcinoma) [14], adenomyoepithelioma [12], and other benign skin tumors (eccrine spiradenoma, syringomatous squamous tumor, papillary syringocystadenoma, cylindroma, keratoacanthoma, trichoblastoma, trichilemmoma, etc.) [15, 16].

Imaging of a clear cell hidradenoma has been poorly described in the literature.

We have found twenty one articles reporting twenty seven cases of both malignant and benign clear cell hidradenomas of the breast in the literature to date. Seven articles were published in pathology or dermatology journals, eleven – in surgical and oncologic journals, and only three articles were published in radiology journals. Most of the articles provided a detailed description of histopathologic findings with very few authors briefly describing the radiologic appearance of the lesion. Due

to its rare occurrence in the breast and active surgical approach, very few cases have been reported based on radiological findings. To our knowledge, only four cases of benign breast hidradenoma have been diagnosed by both mammography and breast ultrasound [9, 10, 17, 18].

The imaging characteristics of nodular hidradenoma are summarized below.

Ultrasound

On ultrasound, it usually presents as a well-circumscribed complex cystic and solid mass with a variable solid portion. The fluid part of the lesion can be complicated due to hemorrhage or clear. Although in our case this tumor manifested as a complicated cyst on ultrasound- homogeneous, low-level echoes, without a discrete solid component, and with an imperceptible wall. Some articles report hypervascularity on Doppler flow.

Mammography and digital breast tomosynthesis

All articles describe nodular cystic hidradenomas as well-circumscribed, usually high-density round or oval masses on mammography.

MRI

There have been a few case reports describing MRI characteristics of such tumors [19, 20]. In one case of plantar eccrine acrospiroma, MRI revealed a homogeneously enhancing solid nodule. In another case of recurrent eccrine acrospiroma of the thigh, MRI showed a cystic mass with an enhancing mural nodule. However, the MR findings of nodulocystic hidradenoma of the breast have not been described before.

In our case, MRI presented a complex circumscribed mass with rim enhancement suggestive of malignancy.

Often, such neoplasms present a diagnostic challenge for both radiologists and pathologists [15, 21]. In our case, core biopsy has yielded low-grade papillary neoplasm possibly of urothelial origin that required an additional urologic investigation – CT urography, urine and blood tests.

These tumors are often misdiagnosed cytologically as benign cystic lesion [22] or as ductal carcinomas [6, 14, 23].

There were two cases of nodular hidradenomas reported where false positive biopsy results led to mastectomy [24, 25].

The malignant counterpart, hidradenocarcinoma, is a very rare tumor that can arise de novo or from a pre-existing hidradenoma [21]. Malignant transformation of nodular hidradenomas also been reported in a few cases [26]. The malignant variants are typically aggressive with early metastases to lymph nodes, bones, and lungs [27, 28]. Benign and malignant versions are indistinguishable by imaging appearance [29-31].

Nodulocystic hidradenoma may recur after inadequate surgical excision [26, 15]. That is why a complete surgical excision with clear margins is the treatment of choice for these tumors.

Conclusions. To conclude, nodulocystic hidradenoma should be taken into account in differential diagnostics of a breast lump. However, there is no particular radiologic feature that could strongly predict the diagnosis of nodular hidradenoma.

FNA and core biopsy may sometimes lead to a misdiagnosis, so the excision is usually needed for a definite histopathologic diagnosis.

Awareness of these lesions among radiologists is essential for their prompt diagnostics and adequate management.

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