Case report

Clinical management of myoid hamartomas of the breast: A case report and literature review

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ABSTRACT

Background: Myoid Hamartoma of the breast (MHB) is an extremely rare benign breast lesion composed of mammary ducts and lobules, fibrous stroma, adipose tissue, and smooth muscle. Due to its rarity, the clinical management of MHB is not well described. Surgical excision is the most common form of management. This study reviews the current literature on the clinical management of MHB and describes a case report of a young patient presenting with MHB managed with surveillance and shared-decision making.

Materials and methods: A healthy 23-year-old female presented with a one-year history of a palpable left breast mass. Her right breast exam was normal. Ultrasound of the left breast revealed a 2.7 cm × 1.4 cm × 2.4 cm lobulated mass at the one o’clock position. The mass caused slight discomfort to palpation but otherwise had no associated skin changes. Ultrasound-guided biopsy revealed a left breast myoid hamartoma. Management options were presented to the patient, and she elected to observe the mass with surveillance imaging.

Results: There have been no reported cases in the literature of malignant transformation of MHB. Rather than rely on reflexive surgical excision of MHB, our review suggests that surveillance and routine imaging may be an appropriate form of clinical management in patients who present with a favorable clinical and histopathological profile which includes: a low MIB-1 proliferative index, low breast cancer risk assessment score, lesion size less than 1.2 cm, and radiological-pathological concordance.

Conclusion: Further research is needed to determine the clinical significance and threshold levels of these clinical and histopathological factors in patient care. However, given current trends to minimize over treatment in breast pathology, we pose that observation of MHB can be performed when favorable clinical criteria is met.

1. Introduction

Breast hamartomas are extremely rare benign breast tumors composed of various proportions of all components of the breast, ducts and lobules, stroma, and adipose tissue, in a disorderly fashion. They account for approximately 0.7%–5% of all breast lesions [1, 2]. Myoid Hamartoma of the breast (MHB), a histological subtype of breast hamartomas, is a benign proliferative lesion characterized by the additional presence of smooth muscle [1, 3, 4]. MHB was first described in 1973 by Davies and Riddell [5]. Its incidence rate is unknown, with few documented cases to date. Although various theories have attempted to explain the origins of MHB lesions, the histogenesis remains unclear [3, 4, 6]. One theory suggests that MHB originates from mutated mesenchymal stem cells capable of differentiation into stromal cells, adipocytes, and smooth muscle cells [6]. Rosen et al. proposed that the majority of MHB are tumors with adenosis and leiomyomatous metaplasia of myoepithelial components [4]. Further, increased expression of High Mobility Group AT-Hook 2 (HMGA2) protein rearrangements in MHB and other benign breast lesions suggest MHB may be genetically related to other types of benign breast tumors such as lipomas, myolipomas, chondroid hamartomas, and hamartomas of the breast [6].

MHB commonly presents clinically as a firm, mobile, well-defined, non-tender mass in postmenopausal women with a mean age of diagnosis of 41 [1]. As a result, their clinical presentation is easily misdiagnosed for other benign breast lesions such as fibroadenomas or lipomas [1, 4, 7, 8]. Diagnosis may be made using core biopsy, however some argue that definitive diagnosis is only possible following excisional biopsy [8]. Due to the extreme rarity of the disease, recurrence rates,
potential for malignant transformation, and clinical management of MHB have not been well described. There are three documented cases of recurrence of MHB in the literature, and zero cases of malignant transformation of MHB have been reported [1, 9, 10]. Given the lack of data regarding long-term outcomes, surgical excision is the treatment of choice [3, 4, 8]. We present an interesting case of MHB managed with surveillance with a literature review on the diagnostic workup and management of this rare breast disease.

2. Case description

An otherwise healthy 23-year-old female presented with a 1-year history of a stable palpable left breast mass. Her right breast exam was normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal. The patient stated that the mass did not regress so she brought it to the attention of her physician. She was worked up with an ultrasound normal.

The patient underwent an ultrasound-guided biopsy which revealed a left breast myoid hamartoma. Hematoxylin and eosin (H&E) stain of the breast core biopsy of this patient revealed mammary glandular tissue with fibroadenomatoid changes mixed with areas of intersecting fascicles formed by spindle cells (Figure 2A). Immunohistochemical analysis with desmin confirmed the smooth muscle nature of the spindle cells seen in myoid hamartomas (Figure 2B). Ki-67/MIB-1 proliferation index, a measure of cell proliferation, revealed a low index of less than 1% nuclear staining. Management options were presented to the patient. Observation with short interval imaging, similar to the management of benign proliferative lesions, versus surgical excisional biopsy were discussed. The patient elected to observe the mass with short interval imaging. Informed consent was then obtained from the patient for the purpose of this study.

3. Discussion

The clinical presentation of MHB confers a wide range of differential diagnoses from benign to malignant, requiring further radio-imaging and surgical workup [1, 3, 4]. Diagnostic workup of MHB includes imaging with mammogram and ultrasound and definitive diagnosis with core biopsy or excisional biopsy when the core is inconclusive [4, 8]. MHB appear as well-circumscribed masses on a mammogram, often with a thin pseudo-capsule that is radio-opaque seen surrounding a portion of the mass [7]. Ultrasonography reveals a solid hypoechoic mass with only partial or no posterior acoustic shadowing and irregular hyperechoic lines or bands interspersed within the mass [1, 7]. Microscopically, they show mass forming disorganized growth of all components of the breast tissue, in varying proportions, in addition to smooth muscles cells [8]. The smooth muscle cells on H&E stain will characteristically show bundles of spindle cells in intervening fascicles with eosinophilic cytoplasm and elongated nuclei without atypia. On immunohistochemistry, they stain positive for smooth muscle actin, desmin, and vimentin and are negative for cytokeratin and S-100 protein [1, 8].

MHB are considered benign proliferative lesions without atypia, suggesting that their aggregate relative risk of breast cancer may be similar to that of other benign proliferative lesions without atypia, such as fibroadenoma, sclerosing adenosis, or intraductal papilloma. The relative risk (RR) for the development of breast cancer in these lesions is 1.76 (1.58–1.95) [4, 11]. This is consistent with the literature, which has reported zero cases of malignant transformation of MHB and very few cases of recurrence since its discovery in 1973, suggesting that the RR of MHB is very low [1, 9, 10]. For other benign proliferative lesions without atypia, surveillance is viewed as an appropriate treatment modality; however, for MHB, surgical excision has been the treatment of choice [4]. This suggests that reflexive surgical excision of MHB may not be the optimal treatment for all patients who may present with an otherwise favorable clinical and histopathological profile. However, we recognize that surgery may be indicated in patients with MHB for cosmetic reasons.

Prior studies have demonstrated that histopathological and patient factors can be used to evaluate clinical management for other benign proliferative lesions without atypia. One study by Symbol et al. determined that the atypia rate for intraductal papillomas less than 1.2 cm in size was significantly lower than the atypia rate in tumors greater than 1.2 cm in size (16% vs. 36% respectively, p = .008) [12]. As a result, in benign proliferative lesions such as intraductal papillomas, the authors suggest that close clinical and radiographic follow-up is a safe treatment modality for patients with lesions less than 1.2 cm [12]. Similarly, for MHB a size threshold may be an appropriate tool to help physicians assess whether surgical excision is an appropriate treatment.

MIB-1 is a monoclonal antibody that recognizes Ki-67 antigen in formalin-fixed, paraffin-embedded tissue [13]. MIB-1 is used to measure the proliferative activity of cells useful for predicting good and poor prognosis in breast cancer [13, 14, 15]. Nakagomi et al. determined that a MIB-1 proliferative index of less than 10% predicts a good prognosis for patients. However, a MIB-1 proliferative index greater than 30% predicted a poor prognosis [14]. They also found that the survival rate was inversely correlated with the MIB-1 proliferative index [14]. Further,
A- HE - low mag 5x

B – HE – 20x

Figure 2. Pathology of the left breast mass. (A) Higher magnification of the breast core biopsy demonstrating breast tissue (ducts and lobules) with fibroadenomatoid changes (blue arrows) admixed with areas of intersecting fascicles formed by spindle cells (red arrows), showing morphologic and architectural pattern of smooth muscle. (B) High magnification of the spindle cells. Immunohistochemical analysis with desmin, confirms the smooth muscle nature of the spindle cells (Inset). Close relation of the smooth muscle with breast duct is again identified (blue arrow).

Jansel et al. and Querzoli et al. found that the MIB-1 proliferative index is an important prognostic factor of clinical behavior in patients with breast cancer [15, 16]. These findings suggest that the MIB-1 proliferative index may be useful in clinical decision-making.

The Breast Cancer Surveillance Consortium (BCSC) benign breast disease (BBD) model is a tool used to estimate ‘women’s risk for breast cancer at 5- and 10-years who have benign breast lesions using clinical and histological features assessed from biopsy, imaging, and patient demographics [17]. A study by Tice et al. found that the BCSC BBD accurately estimates ‘women’s risk of breast cancer using breast density and benign breast diseases [18]. The BCSC BBD model and other risk assessment tools such as the Tyer-Cuzick or Gail Model provide useful information to physicians in determining prevention and treatment. A complete clinical and histopathological profile including lesion size, MIB-1 proliferative index, and breast cancer risk assessment may provide physicians the ability to determine whether surgical excision is indicated for the treatment of MHB.

The current literature has recommended that surgery is the treatment of choice for MHB because of concerns over potential malignant transformation and the inability to reach a definitive diagnosis by core biopsy alone [3, 4, 8]. First, there have been no documented cases of malignant transformation of MHB in the literature and only one documented case of neoplasia within a MHB [19]. Mathers et al. found foci of lobular neoplasia within the MHB and high-grade ductal carcinoma in situ in the contralateral breast of a 44-year-old woman [19]. These findings suggest that while physicians should always be cautious of the potential of a hidden malignancy or malignant transformation of MHB, surveillance of the lesion may be an appropriate treatment for patients with a favorable and concordant clinical and pathological profile. Second, although some researchers suggest that definitive diagnosis is only possible with surgical excision, recent literature by Georgian-Smith et al. on the surgical excision of mammary hamartomas suggests that if there is a radiologic-pathologic concordance, further surgical excision is not needed [20]. Their findings applied to MHB, a subtype of mammary hamartomas, provide further evidence for surveillance in patients whose imaging and biopsy findings both provide a favorable prognosis.

4. Conclusion

Given the paradigm shift in the local treatment of breast cancer and breast lesions toward more conservative approaches to care, our paper suggests that surveillance may be a safe treatment modality in a sub-cohort of patients with MHB who present with a favorable and pathological profile. A low MIB-1 proliferative index, low breast cancer risk assessment score, lesion size less than 1.2 cm, and radiological-pathological concordance are clinical criteria to consider observation of MHB with surveillance imaging. Further research is needed to determine the clinical significance and threshold levels of these factors in patient care.

Declarations

Author contribution statement

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The authors declare no conflict of interest.

Additional information

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