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An Uncommon Case Report of Accidental Benign Breast Adenomyoepithelioma

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Abstract

Adenomyoepithelioma is a rare tumor characterized by the biphasic proliferation of epithelial and myoepithelial cells in breast lobules and ducts. Clinical and radiological investigations are challenging; thus, the diagnosis is mostly histopathological and confirmed by immunohistochemical analysis. Most cases are benign, although a few malignant cases have been reported. Treatment remains controversial owing to the lack of high-volume data and the absence of prospective studies. In this article, we report the case of a 19-year-old female who presented with a lump in her left breast, which was diagnosed as a benign adenomyoepithelioma. The patient underwent local excision of the lump because it was small and showed no signs of malignancy on workup. We conducted a one-year follow-up and observed no relapse. We report this rare case to encourage physicians to consider this etiology as part of the differential diagnosis while evaluating breast lumps.

Keywords: Adenomyoepithelioma, Histopathological, Malignancy, Metastasis

Introduction

Adenomyoepitheliomas are uncommon breast lesions with simultaneous proliferation of both epithelial and myoepithelial cells which was first described by Hamprel in 1970[1].It is a rare condition that poses both clinical and diagnostic challenges, with the majority of reported cases being benign; however, it may show malignant transformation, recurrence, or even metastasis[2]. Local excision is the best treatment, and histopathological analysis is the gold standard for diagnosis[2].

Here, we report a case of benign breast adenomyoepithelioma in a 19 year old female which was an incidental finding.

Case Report

We report a 19 year old female, in generally good health, with no significant family history, who presented with a painless mass in her left breast, which she noticed incidentally while bathing a week before her visit. On physical examination, a solitary lump of size 4 × 2 cm at the 10 o'clock position of the left breast was palpable, which was non-tender, mobile, and had a smooth surface with well-defined margins. There were no inflammatory signs or palpable lymphadenopathy, and the rest of the examination was unremarkable.

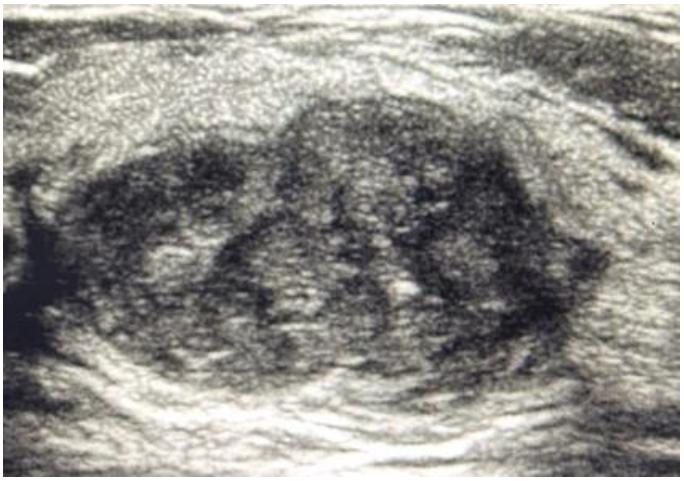

Figure 1: An ultrasound image of left breast showing well defined hypoechogenic oval lesion measuring 3.7*1.8*2.2 cm present in the 9 o'clock position

Ultrasonography revealed a well-defined hypoechoic oval lesion measuring 3.7 × 1.8 × 2.2 cm at the 9 o'clock position in the left breast. No calcific echogenic foci, liquefaction, or acoustic enhancement was observed (Figure 1). BIRADS-3 (Probably benign). Fine needle aspiration cytology was inconclusive and showed only fibroadenoma changes.

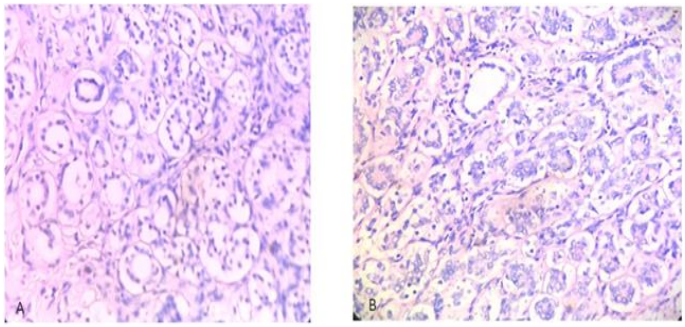

Figure 2 (A & B): Showing tubules with prominent myoepithelial cells at places forming aggregates.

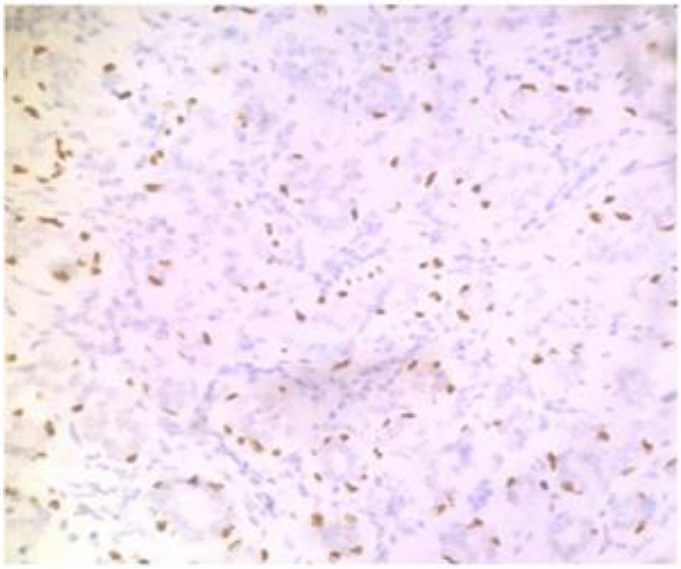

Figure 3: P63+ve, Myoepithelial cells. Magnification 400X, objective lense.

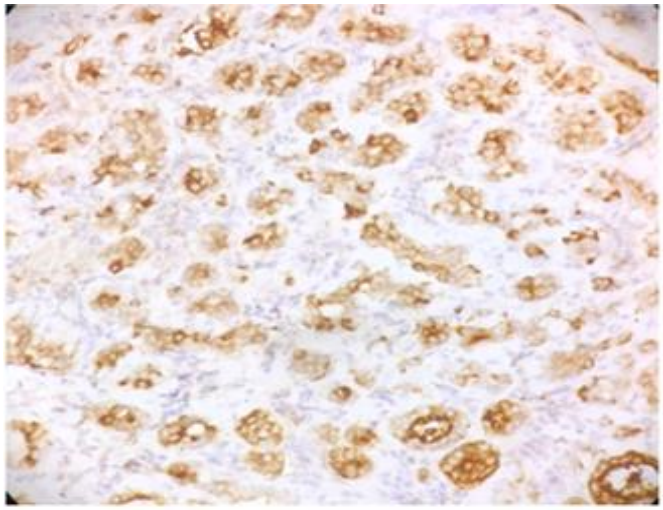

Figure 4: Epithelial Membrane Antigen positive in epithelial component.

Since the tumor was small in size and there were no signs of malignancy, both clinically and diagnostically, local excision of the breast lump was performed and sent for histopathological examination, which confirmed clear surgical margins. The specimen was grossly brownish-white in color with a brownish cut surface. Microscopic examination revealed that the tumor was composed of epithelial and stromal tissues. The epithelial elements were composed of ducts and tubules surrounded by prominent myoepithelial cells (Figure 2 A & B, Figure 4).

The Immunohistochemical markers especially P63 were positive in the myoepithelial component (Figure 3), and all findings were consistent with adenomyoepithelioma of the breast. At present, the patient shows no signs of tumor recurrence within follow-up period of 1 years.

Discussion

Adenomyoepithelioma, first reported by Hamper in 1970[1], is an uncommon tumor characterized by biphasic growth of epithelial and myoepithelial cells. They are divided into solid, papillary, and tubular subtypes [3]. With a median age of 56.75 years [5], these tumors can manifest between the ages of 16 and 86[4]. Although bilateral lesions are less common, they have been reported occasionally [6].

When cytological atypia, an infiltrative growth pattern, and a high mitotic index (local >3/10 high-power field (HPF)) are present, adenomyoepithelioma, which is often benign, must be categorized as malignant. Hematogenous metastasis is the most common method of cancer spread and can occur in the lung, brain, bone, or even the thyroid [8,9]. Numerous cases of hematogenous metastasis have been reported. Both groups have been linked to reports of distant metastases and local recurrences [10].

Imaging methods are inconclusive; MRI and mammography features are non-specific and infrequently demonstrate microcalcifications. A solid oval hypoechoic mass with irregular borders is visible on ultrasound [2]. From a pathological standpoint, the core biopsy diagnosis of AMEs is challenging. When a suspicion arises, immunochemistry clarifies it by highlighting the distinct qualities of each component: 1) Cytokeratin 5/6 antibodies, calponin, p63, smooth muscle actin, smooth muscle myosin, caldesmone, cd10, and S100 protein are positive in the myoepithelial portion; 2) Low molecular weight keratin, cytokeratin antibodies, and AE1/AE3 are positive in the epithelial portion [11].

The size, location, focality, and related pathology all influence surgical options. It is usually advised to perform a wide local excision with distinct margins; however, as certain AME instances exhibit quick local recurrences, the required margin is unknown [8]. In cases of malignancy, the efficacy of hormonal treatment, radiation, and chemotherapy is not very high [11]. Chemotherapy was utilized in 26% of instances, hormone therapy in 8% of cases, and radiotherapy in 36% of cases in a recent case series that used a sizable US database and 110 cases for analysis [12]. However, the overall survival (OS) of the population did not improve.

Conclusion

Owing to its uncommon and often benign nature, adenomyoepithelioma can be difficult to diagnose because of its inconsistent imaging appearance, requiring immunohistochemical investigation. Because there are no prospective studies or large volumes of data, the treatment remains controversial. These tumors have a high propensity for local recurrence and the potential for malignant transformation; hence, the primary course of treatment should involve complete excision with wide margins. After surgery, patients should undergo routine follow-up visits.

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