Case Report

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Adenomyoepithelioma of Breast in an Adolescent Girl: A Rare Clinical Presentation

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Abstract

The growing awareness of female breast cancer has led to increased sensitivity toward pathological breast conditions in pediatric patients. Palpable breast masses arising in adolescent and pubertal girls are uncommon. The most common benign breast lesions are fibroadenoma and benign phylloides tumors. The mammary tumors with myoepithelial component are very rare in pubertal girls. We report a case of adenomyoepithelioma in a 15 year old girl and discuss the problems of diagnosis of breast masses in adolescent girls.

Keywords: Adenomyoepithelioma; Myoepithelial Tumors; Adolescent Girls; Breast Mass.

Introduction

Tumors that occur in pubertal and prepubertal breast are uncommon and predominantly benign, most common being fibroadenoma and benign phylloides tumor. Other benign solid neoplastic breast lesions include fibroma, hemangioma, papilloma, lymphangioma and lipoma [1]. Regardless of histological type, 10-40% of the clinically detected breast masses in adolescents resolve completely [2]. However, because of increased awareness of breast cancer, paediatricians and surgeons are evaluating increasing number of pediatric patients with palpable breast masses [1].

We report a case of adenomyoepithelioma in a 15 year old girl and discuss the problems of diagnosis of breast masses in adolescent girls.

Case Report

A 15 year old female presented with a painful right breast lump since one month. On local examination, 7x7x6.5 cm, freely mobile, tender mass was present in upper outer quadrant of right breast. There was no nipple discharge or skin involvement and the left breast was normal. There was no axillary lymphadenopathy. There was no history of breast cancer in any of the family members. Her hemoglobin was 10.8 gm%, total white cell count was 8000/cumm with normal differential count. Pathological findings: On fine needle aspiration cytology, the diagnosis of cystosarcoma phylloides was offered. Gross examination revealed an encapsulated 7x7x6.5 cm tumor with bosselated external surface. The cut surface was solid, grayish white, lobulated and firm, without hemorrhage and necrosis (Figure 1a and 1b). Microscopically, the tumor was comprised of lobules, showing balanced proliferation of glandular and myoepithelial elements. The small round to oval glands were lined by inner cuboidal epithelial cells and outer polygonal clear myoepithelial cells and basement membrane (Figure 2a). The epithelial cells had deeply stained eosinophilic cytoplasm and hyperchromatic nuclei and myoepithelial cells had vacuolated cytoplasm with round to oval pale nuclei (Figure 2b). The cytological atypia and mitotic activity was absent and the fibrovascular stroma showed sparse chronic inflammatory infiltrate. The reticulin stain highlighted the characteristic lobulated appearance of adenomyoepithelioma with proliferation of ductal and myoepithelial cells (Figure 3).

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Fig. 1a: A 7x7x6.5 cm, capsulated tumor with bosselated external surface

Fig. 1b: The cut surface was solid, grayish white, lobulated and firm, without hemorrhage and necrosis.

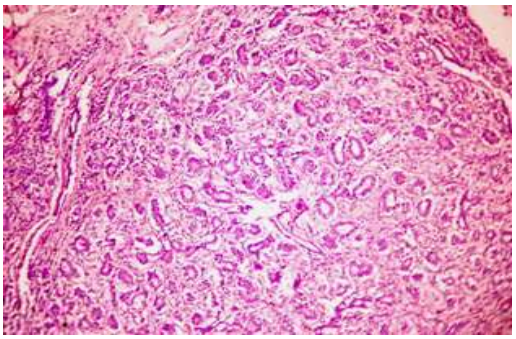

Fig. 2a: Adenomyoepithelioma comprised of round to oval glands with minimal stroma (H & E, X100)

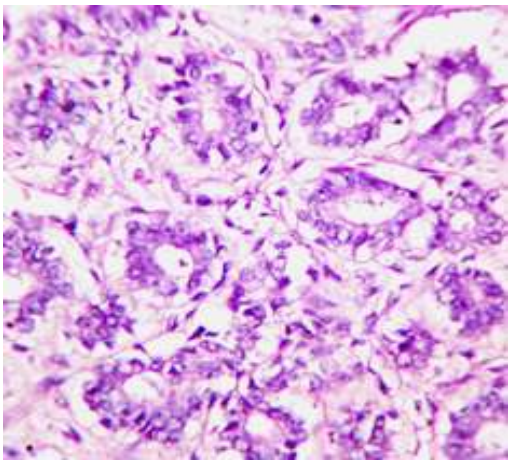

Fig. 2b: The glands are lined by inner ductal epithelium and outer myoepithelial layer (H & E, X400)

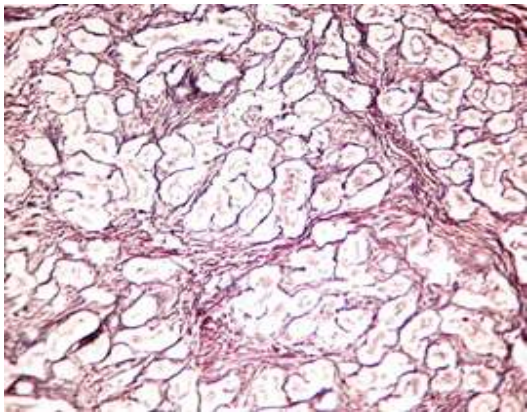

Fig. 3: Reticulin stain to demonstrate the lobulated architecture of tumour

Discussion

Myoepithelial cells are normally present in mammary and salivary gland. The tumors derived from myoepithelial cells are commonly described in salivary gland. The mammary tumors with myoepithelial components are infrequent. World health organisation (WHO) [3] has classified myoepithelial lesions into four categories- myoepitheliosis, adenomyoepitheliosis, adenomyoepithelioma and malignant myoepithelioma.

Adenomyoepithelioma of breast is a rare tumor characterised by biphasic proliferation of both epithelial and myoepithelial cells [3-9]. The gland forming epithelial cells and myoepithelial cells are present in variable proportions. Myoepithelial cells are spindle to ovoid to polygonal in shape with clear to eosinophilic cytoplasm [7-9].

This tumor was first described by Hamper [4] in 1970 and the exact etiology of breast adenomyoepithelioma is still not known. Choi et al [5] proposed that adenomyoepithelioma was derived from a long standing underlying disease, such as adenosis and fibroadenoma. Clinically, adenomyoepithelioma present in females of 40–60 years, as a palpable, non tender and centrally located mass which may or may not be associated with serous nipple discharge. Rapid enlargement of mass is highly suggestive of malignant change [4]. Our case of adenomyoepithelioma had unusual clinical presentation as a painful mass in a 15 year old pubertal girl.

Accurate diagnosis of adenomyoepithelioma can be difficult based solely on radiological observation; therefore histological examination is required for precise diagnosis. Proliferative nature of

adenomyoepithelioma differentiates it from fibroadenoma, tubular adenoma and sclerosing adenosis [6,7]. Moreover, lobular pattern of growth rules out fibroadenoma. Tubular adenoma shows epithelial overgrowth with closely packed tubules, inconspicuous myoepithelial cells and little intervening stroma. Sclerosing adenosis with extensive myoepithelial cell proliferation may cause difficulty in diagnosis, but it has multiple nodular areas comprised of compressed and distorted tubules (more marked in centre of nodules) with dense and hyalinised surrounding stroma. Immunostaining for smooth muscle actin, calponin and p63 can support the myoepithelial differentiation with no reactivity in the epithelial cells [6].

The treatment of benign adenomyoepithelial tumor is complete surgical excision. An additional rim of normal breast tissue should be excised if the tumor has satellite nodules, infiltrating margins, or malignant features. The tumor has potential for local invasion and recurrence after excision [7]. Recurrence is associated with incomplete removal of the original tumor. The prognosis of patients with adenomyoepithelioma of the breast is usually good [6].

Conclusion

Adenomyoepithelioma is an unusual breast neoplasm which should be considered in the differential diagnosis of solid mass lesions in the breast in adolescent or pubertal girls. As this tumour has risk for recurrence, malignant transformation and metastasis, accurate diagnosis with close follow-up is necessary.

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