An Unusual Breast Mass in a 13-year-old Girl: a Case Report

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Case Report

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Abstract

Background

In puberty breast intraductal papilloma was rare, but the pathological changes of intraductal papilloma combined with intraductal fibroadenoma in the duct lumen were even less reported. Herein we reported a case of adolescent benign breast disease that contained the two features of neoplasm in the same duct and discussed its clinical course.

Case presentation

A 13-year-old adolescent female presented to our hospital with spontaneous bloody discharge of the left nipple. On clinical examination, a 2.5cmx1.5cm mass could be palpable under the left nipple, and a single-hole bloody discharge could be seen by squeezing the mass. Surgical resection of the mass and part of the breast duct was confirmed by postoperative pathology as intraductal papilloma and intracanalicular type fibroadenoma, with cystic changes of ductal epithelium hyperplasia. After 15 months of follow-up, there was no evidence of recurrence.

Conclusions

Intraductal fibroadenomatosis may be a new pathological subtype in which multiple histologic characteristics can be seen in the same duct. Surgical resection is the only effective treatment. Its etiology is still unknown, and it remains to be accumulated more cases for further exploration.

Introduction

Intraductal papilloma (IP) is a common benign breast disease, of which the typical clinical symptom is a unilateral bloody/serous nipple discharge and/or a palpable mass [1]. It can be seen in women of any age, but it usually occurs in 35–55 years old [2]. It is rare in adolescence or childhood. When IP combined with intracanalicular type fibroadenoma (or intraductal fibroadenoma, short for IF), such pathological change was much rare and was named by Chung et al as an unusual type. IP with admixture of IF or other benign pathological components occupying the same duct and some adjacent ductules, was given the name of intraductal fibroadenomatosis [3]. We reported a case of spontaneous unilateral nipple discharge in a 13-year-old adolescent female who was pathologically diagnosed as intraductal fibroadenomatosis with overlap features of IP and IF in the same duct, and discussed its clinical course.

Case Report

The patient was a 13-year-old Chinese girl who presented to the doctor with spontaneous bloody discharge of the left nipple for more than 20 days. Her personal history was remarkable for breast development for one and a half years, and menarche more than one month ago. Moreover, she sustained use of growth hormone due to short stature in the past six months. On clinical examination, a central area
palpable mass at 3 o’clock of his left breast was found with clear boundaries and measured approximately 2.5×1.5 cm. Bloody discharge could be seen by squeezing the mass from one single hole. Laboratory data, including blood count, chemistry and hormone levels, were within the normal limits. Ultrasound sonography showed an intracystic mass under the left nipple, measuring 23.6 × 15.8 × 8.7 mm in diameter (Fig. 1A). The mass seemed to be located within the lumen of a dilated duct with the width of 2.2 mm. The surrounding of mass was noted on color Doppler imaging. Fiberoptic ductoscopy showed a lobulated neoplasm at the opening of the grade II milk duct, which protruded into the lumen and did not completely block the duct (Fig. 1B). There was capillary hyperplasia on the surface with rupture and bleeding. There was no other neoplasm in the proximal and distal duct of which the wall was smooth.

Surgeons first considered to be a benign intraductal papilloma, and carried out removal the mass and part of the breast duct connected to it by methylene blue staining (Fig. 1C). On gross examination, the cut surface showed clear boundaries and coatings, and a thin cystic cavity is formed around it, measuring approximately 18 × 15 mm in diameter (Fig. 1D).

Microscopically, a dilated duct was found in the excised tissue, of which ductal epithelium proliferated to form cystic changes. Fibroadenomatous polypoid structures were seen within the cystic lumens (Fig. 2A). The neoplasm consisted of multiple polypoid papillary lesion and leaf-like fashion. Characteristic delicate fibrovascular stalks found in the polypoid papillary lesion was IP. The leaf-like fashion with a hypocellular and prominent myxoid stroma protruded into the lumens of the ducts, reminiscent of IF. Mitotic figures were very rarely seen. Overall, the main features were of IP and IF, and two features were in equal proportion (Fig. 2B), with cystic changes of ductal epithelium hyperplasia. The patient was discharged 2 days after surgery without any postoperative complications. So far, the patient has been routinely followed up for more than 15 months after surgery, and no recurrence has been found.

Discussion

Nipple discharge is not common in adolescence, especially bloody discharge. When bloody nipple discharge is associated with breast masses, intraductal lesions are generally considered clinically. For such a young patient, IP would be considered first [4]. We reviewed the previous literature and found that there were few reports of IP in adolescence or childhood [5]. Our case should be one of the few reports about IP that occurred in adolescence. Especially, this was not a common type of IP, but a rare pathological type with overlap features in the same duct [6]. IP could be combined with other pathological features such as fibroadenoma, benign phyllodes tumor or ductal adenoma. Chung et al. first proposed the pathological concept of intraductal fibroadenomatosis [3]. The number of such cases were so small, only occasional and case reports. Whether this was a new pathological subtype remained unknown.

Due to the small number of such cases, the cause of the disease was unknown. Looking back on this case, it was found that the patient had used growth hormone therapy in the past six months. Breast diseases are rare in childhood and adolescence, most lesions being fibroadenomas and papillomas [7].
The previous literature found that GHR mRNA level in intraductal papilloma specimens was significantly higher than that in normal breast tissue [8]. We speculated whether the cause of this disease was related to growth hormone levels included endogenous high Levels of growth hormone or additional exogenous growth hormone. We tested growth hormone receptors through immunohistochemistry, and unfortunately those epithelial cells were negative (data not shown). This meant that the cause of the disease has little to do with the growth hormone receptor pathway. In conclusion, the etiology of these diseases was still unknown and further studies were needed.

**Conclusions**

To conclude, intraductal fibroadenomatosis may be a new subtype of pathology, which are seen in adolescence or childhood, with clear boundary of breast mass in central area or/ and unilateral nipple discharge as the clinical manifestations. Surgical resection is the only effective way of treatment. Its etiology is still unknown, and it remains to be accumulated more cases for further exploration.

**Abbreviations**

IP: Intraductal papilloma. IF: intracanalicular type fibroadenoma or intraductal fibroadenoma.

**Declarations**

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**Author`s contribution**

Tianzhu Long proposed the manuscript idea and finished the writing. Yuzhi Yao collected data. Quan Shi put forward the guidance of pathological diagnosis. Yuanxuan Cai, Quan Li and Hongmin Ma did the operation, and Hongmin Ma made suggestions for revision. Benjie Fu was responsible for the follow-up work. All authors read and approved the final manuscript.

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**Availability of data and materials**

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**Ethics approval and consent to participate**
Ethical approval and patient consent were acquired and recorded in the patient medical record with her and her mother as guardian signature.

**Consent for publication**

Not applicable.

**Author Disclosure Statement**

No competing financial interests exist.

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**Figures**
Figure 1

13-year-old Chinese girl with 20-day history of spontaneous bloody discharge of the left nipple. A. Ultrasound sonography demonstrated an intracystic well-circumscribed mass located within the lumen of a dilated duct. The surrounding of mass was noted on color Doppler imaging. B. Fiberoptic ductoscopy showed a lobulated mass protruded into the lumen. C. Surgical excision of the lesion and proximal part of the duct by methylene blue staining. D. On gross examination, the cut surface revealed a well-circumscribed and encapsulated nodule, measuring 18 × 15 mm in diameter, which looked whitish to yellowish in color.
Figure 2

Microscopic examination of the IF and IP of the breast (H&E stains). A. Low power view (×10) showed that fibroadenomatous polypoid structures were seen within the lumens. Red arrow pointed to the dilated duct. Yellow arrow and blue arrow pointed to intermingled intraductal proliferations. Bar = 2000 μm. B. Middle power view (×40) showed that IF and IP were seen in the same duct. Two features were in equal proportion, with cystic changes of ductal epithelium hyperplasia. Yellow arrow pointed to IP. Blue arrow pointed to IF. Bar = 500 μm.

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