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Breast Enlargement in Male Infants and Children: Two Unusual Causes

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1. Abstract

1.1. Aim

Breast diseases are relatively rare in male and considered to be very rare in male children. We report two children with two rare diseases affecting the breast.

1.2. Results

Two patients, an infant and a child presented with bleeding from the nipple and abreast mass respectively. The male infant was found to have juvenile papillomatosis of the breast and the child was found to have galactocele of the breast. Both patients were treated surgically and made an excellent recovery.

1.3. Conclusions

Although very rare, juvenile papillomatosis and galactoceles should be included in the differential diagnosis of breast masses in infants and children. Although, these are benign lesions, they should be excised and the final diagnosis is based on histopathological examination.

2. Introduction

Breast pathology is relatively rare in male ant it is even considered very rare in infants and children [1,2]. Juvenile papillomatosis of the breast ("Swiss cheese disease") is a rare, benign localized proliferative condition of the breast which occurs almost exclusively in women under 30 years of age [3-6]. It is considered very rare in infants and children. The usual presentation is with a breast mass and may cause a bloody nipple discharge. Patients with this lesion often have a family history of breast carcinoma, and rarely carcinoma may coexist with the lesion at the time of diagnosis [6,7].

A galactocele is a rare benign breast lesion usually occurring in females during or following lactation [8,9]. Galactocele is an extremely rare disease in infants and children was found exclusively in male [10,11]. Galactoceles in children can be either cystic or pseudotumor. Galactoceles although rare should be included in the differential diagnosis of breast masses in infants and children.

We report two, an infant and a child with two rare diseases affecting the breast, a male infant with juvenile papillomatosis of the breast and a child with galactocele of the breast.

3. Case Reports

3.1. Case No. 1

A 7.5 months old mal child presented with bleeding from the right nipple of 3 weeks duration (Figure 1). The mother noticed drops of blood from the right nipple. There was no pain and clinically there was blood stain in the cloth close to the right breast. He was also found to have right breast selling which was firm and not tender. The left breast appeared and felt normal. Ultrasound of the right breast revealed a retromammary intraductal multiseptated cystic mass measuring 10x4 mm in size and 2mm deep to the overlying skin in the subareolar region with minimal septal vascular flow (Figures 2a and 2b). He was diagnosed as a retromammary multiseptated cystic mass. The mass was excised and histopathology revealed a mass measuring 1x 0.7x 0.5 cm in size (Figures 3a and 3b). Microscopic examination showed breast tissue with variable sized cysts, few small intraductal papillomas and duct ectasia. There was also fibro adenomatoid change, dense stromal sclerosis and eosinophilic proteinaceous cyst content. Two ducts showed organizing hematoma. There was no atypia or malignancy. This was consistent with juvenile papillomatosis. Postoperatively, he did well and was discharged home in a good general condition. He was seen in the clinic five months postoperatively doing well with no recurrence.

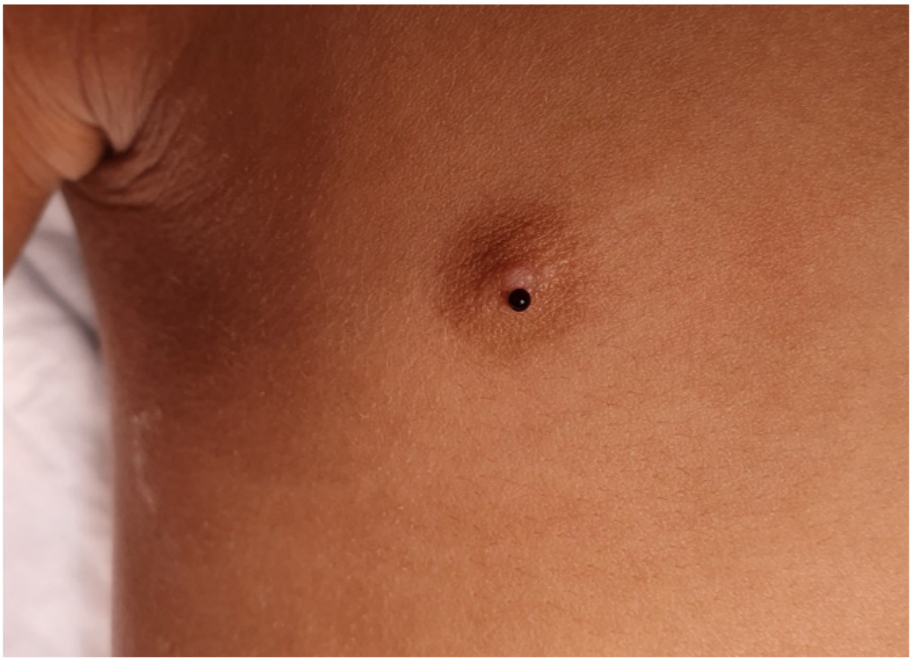
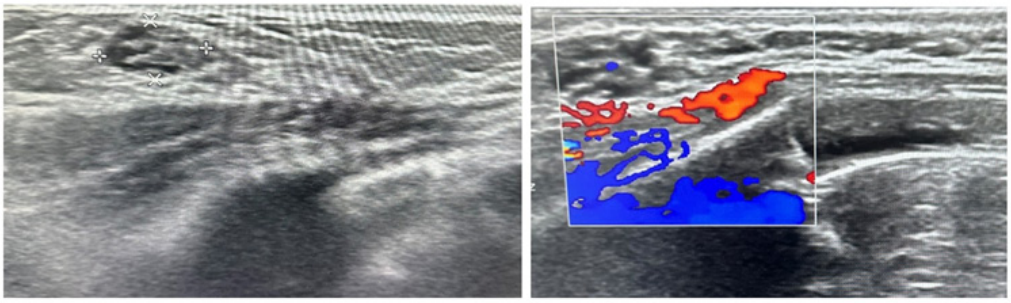
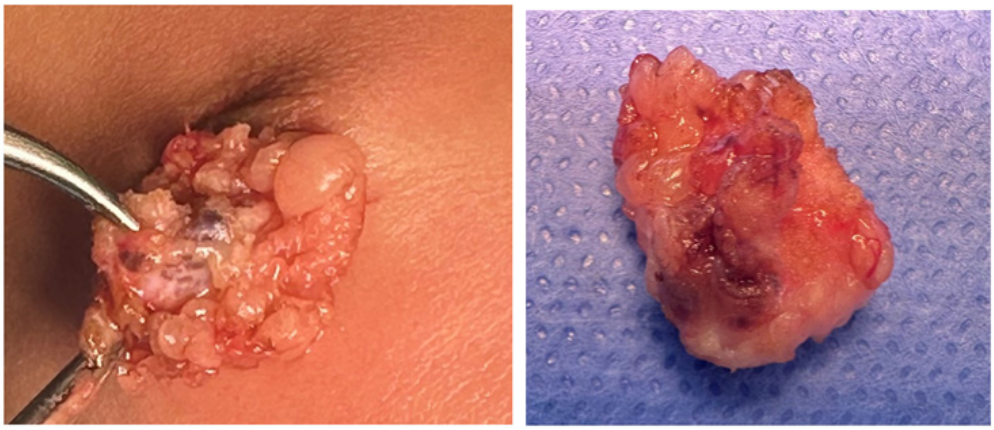


Figure 1: A clinical photograph showing bleeding from the nipple.



Figures 2a and 2b: Ultrasound of the right breast showing a retromammary intraductal multiseptated cystic mass with minimal septal vascular flow.



Figures 3a and 3b: Clinical intraoperative photographs showing a right breast mass being excised. Note the associated cystic changes.

3.2. Case No. 2

A 5-year-old male was referred to our hospital with swelling of the right breast. The swelling started small in size and increased gradually over the last two years. The swelling was painless. Clinically he was found to have right breast swelling which was 5x6 cm in size (Figure 4). The swelling was soft inconsistency and not tender. His investigations including his Hb, WBC, HTC and platelets were normal. Ultrasound of the right breast revealed a cystic swelling measuring 5.4x5.8 cm in size without increased vascularity. His abdominal ultrasound was normal. His hormonal profile including FSH, LH, testosterone, 17-OH progesterone, DHEAS and alpha-fetoprotein were normal. The swelling was excised and histopathological examination showed a cyst measuring 4x2x0.7 cm in size (Figures 5a and 5b). Microscopic examination revealed breast tissue showing multiple cysts filled with eosinophilic material with crystals in some places. The cysts were lined by cuboidal or flattened epithelium. There are ulcerations in places which are replaced by granulation tissue with many pigmented macrophages. The wall was fibrous containing some ducts, clusters of inflammatory cells and

some small blood vessels. This was consistent with galactocele. Postoperatively, the patient did well and was discharged home in a good general condition. He was followed up in the clinic and there was no recurrence 2 years postoperatively.

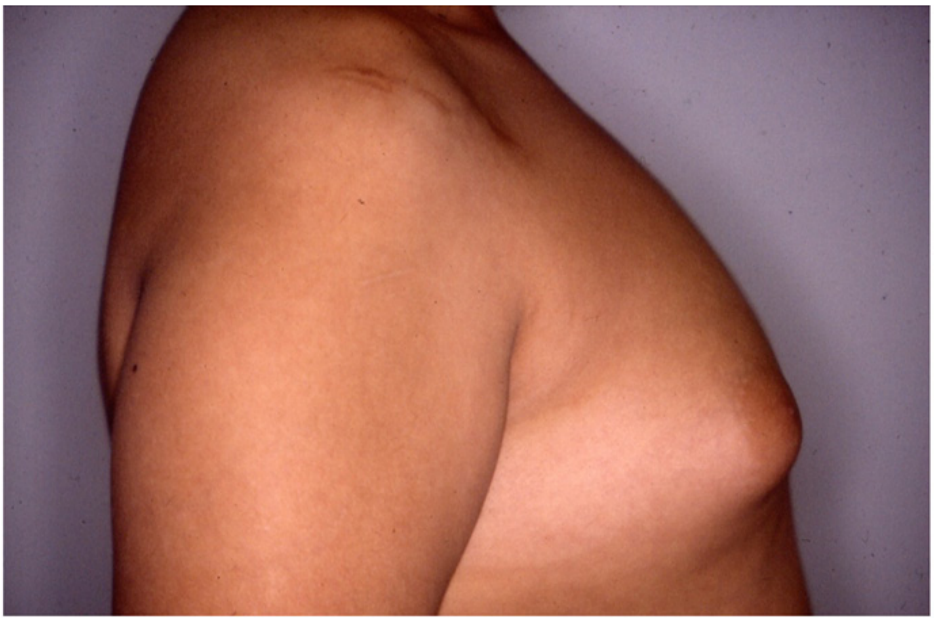
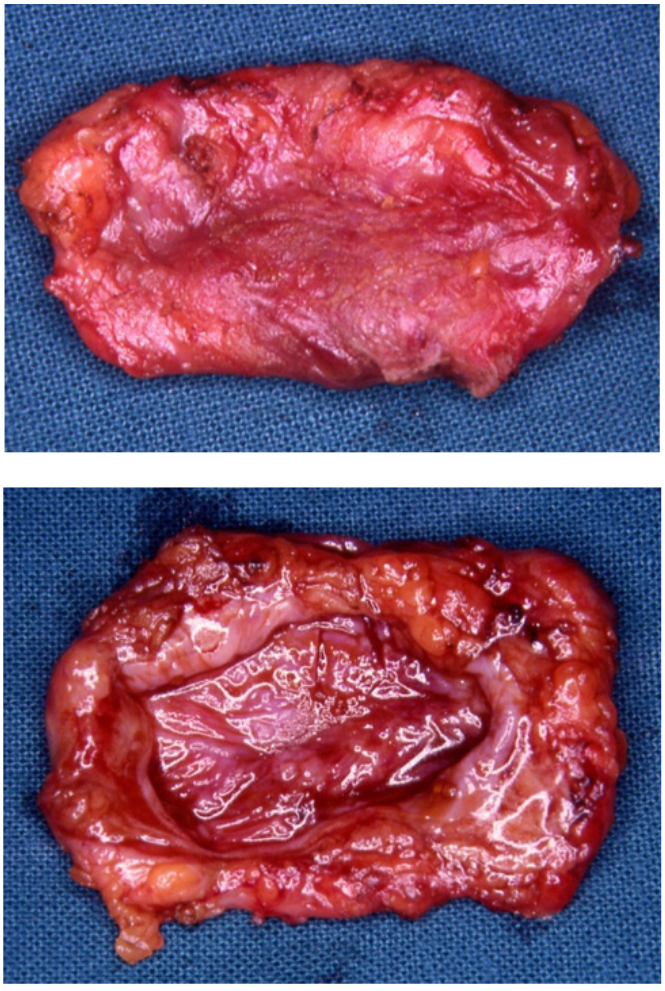


Figure 4: A clinical photograph of a child showing a right breast mass.



Figures 5a and 5b: Clinical intraoperative photographs showing excised right breast mass in a child. Note the cystic cavity within the excised mass.

4. Discussion

The incidence of breast diseases in children is very low and commonly seen in girls. Among adolescent girls, fibroadenoma is the commonest pathology seen and these make up around 50% of breast masses seen in girls. Although fibroadenoma is not uncommon among adolescent girls, breast diseases among male children are very rare. West et al treated 74 children and adolescents with palpable breast masses, 16 (21.6%) of them were males and gynecomastia was the cause in 50% of them [12].

Juvenile papillomatosis is a rare, benign breast condition [13-16]. It is considered an epithelial proliferative disorder with cysts and papilloma's that arises within the small ducts and lobules of the breast. It is characterized by multiple cysts and dilated ducts separated by areas of dense stroma. Juvenile papillomatosis of the breast was first described in 1979 by Kiaer et al. and they called it "extreme duct papillomatosis." [17]. They reported three female patients aged 11, 14 and 17 years with severe ductal papillomatosis of the breast. Two of the patients developed breast cancer 11 and 27 years later. Since then, the association of juvenile papillomatosis (up to 28%) with breast cancer within the family of affected patients was established. Rosen et al. [18], in 1980 were the first to describe Juvenile papillomatosis as a distinct clinicopathologic entity [3]. They also coined the terms "Juvenile Papillomatosis" or "Swiss cheese disease" because macroscopically it resembles cheese from the Swiss Emmental region. In 1982, they studied the relationship of juvenile papillomatosis with a family history of breast cancer and in 1990, they published a review of a series of 41 patients with juvenile papillomatosis [4].

Juvenile papillomatosis which is also so-called Swiss cheese disease, is a rare benign breast disease commonly seen in young girls and women less than 30 years of age. The age at diagnosis of juvenile papillomatosis ranges from 10-48 with an average of 23 years old. Our patient was only 7.5 months old at the time of diagnosis. Clinically, patients with juvenile papillomatosis present with a firm breast mass which is characterized by dilated ducts and cysts. The size of the breast mass is variable ranging from 1-8cm [3-7]. The usual presentation is with a breast mass but may cause a bloody nipple discharge like in our patient [13,18].

Juvenile papillomatosis is characterized by a multinodular cystic breast mass and sometimes calcifications. The histological features of juvenile papillomatosis are diverse and include multiple intraductal papilloma's, cysts formation, papillary ductal hyperplasia, duct ectasia, perifocal sclerosing adenosis, and calcification [4-6]. In some cases, this is associated with calcification.

Although juvenile papillomatosis is considered a benign lesion, follow-up is recommended for the patient and family since there is an association with a family history of breast carcinoma and increased risk of development of breast carcinoma. A family history of breast cancer is seen in a substantial number of patients with juvenile papillomatosis. Bazzochi et al. [5], found a positive family relationship of 33% in these series. Rosen et al [3,4], found an even greater association (58%). Munitiz et al. [18], reported a case of breast cancer associated with juvenile papillomatosis of the male breast [19]. The treatment of Juvenile papillomatosis is localized excision which is curative. These patients and their family should be followed up taking in consideration the increased incidence of breast cancer in the members of the family.

A galactocele is a benign breast condition defined as cystic dilatation of the mammary gland containing milk [8, 9,12]. Galactocele is an uncommon condition and usually seen in lactating woman [1,12]. Galactocele is most commonly found in young fertile women during or after breastfeeding [12,20]. It is an extremely rare cause of breast enlargement in infants and children [21-26]. Galactocele in infants and children are usually unilateral but bilateral cases have also been reported [27-29]. Among 320 infants and children reviewed, Pettinato et al and Bower et al identified only 3 cases of galactoceles in male children aged 12 months, 21 months and 6 years [11, 20].

The exact aetiology of galactocele in infants and children is still unknown. Several theories were proposed but none of them gives a satisfactory explanation. Transplacental hormonal transmission was suggested as a possible cause that may lead to stimulation of breast parenchyma, resulting in the development of galactocele [9,21]. This may explain the development of galactoceles in male infants but it does not explain why galactocele develops in male children only, nor does it explain the occurrence of galactocele in older children. Another theory suggested an inflammatory reaction secondary to trauma in an already existing quiescent small cyst leading to its sudden enlargement as a cause for galactocele [8]. There were no associated abnormalities in the majority of reported cases, but in the literature, there were 5 patients with galactocele reported in association with hyperprolactinaemia and one case associated with congenital hypopituitarism [29-31]. Visvanathan suggested that in children galactocele is a mass of inspissated secretory material which results from the partial resorption of the secretory contents as a sequence of cessation of the secretory activity of the cysts [32].

In conclusion, although very rare, juvenile papillomatosis and galactoceles should be included in the differential diagnosis of breast masses in infants and children. Although, these are benign lesions, they should be excised and the final diagnosis is based on histopathological examination.

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