Unilateral galactocele in a male infant

Jednostrana galaktocela kod muškog deteta

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Abstract

Introduction. Galactocele, generally occurring in young women during or after lactation, is an extremely rare cause of breast enlargement in infants and children of exclusively male gender. Only 26 cases have been published so far, including two our cases. Case report. We described unilateral, cystic, breast enlargement, without any endocrinologic and other abnormalities in a 29-month-old boy. A typical clinical and histopathologic presentation of galactocele was followed with a complete excision. Conclusion. This was a 27th well documented case of galactocele in a male infant with typical clinical and histopathologic presentation. There are several hypotheses regarding etiology of the lesion, but it is likely to be multifactorial. Because of its extreme rarity, there are some difficulties in differential diagnosis and treatment options of galactocele in male infants.

Key words: breast neoplasms; child, preschool; diagnosis; histological techniques; magnetic resonance imaging.

Introduction

Galactocele has been described as a cystic enlargement of the mammary gland containing milk or milk-like fluid 1. It generally occurs in young women during or after lactation 2. Galactocele is a very rare cause of breast enlargement in infants and children 3,4. Because of its rarity, there are many difficulties in diagnosing, treatment and attempts to explain pathogenesis of galactocele in early childhood. We have recently described two new cases of galactocele in boys with a detailed analysis of cases published in medical journals in English and other languages 4. Cattani 5 reported the first case of a galactocele in male infants back in 1880 and so far now only 26 cases have been published.

We here presented another case of unilateral galactocele with a typical clinical and histopathologic presentation in a 29-month-old boy, without any endocrinologic and other genetic, clinical, and laboratory abnormalities.

Case report

A 29-month-old male infant was referred with a 16-month history of unilateral left-sided, slowly progressive breast enlargement (Figure 1). The child was born at term by normal vaginal delivery after an uneventful pregnancy. There was no history of nipple discharge, trauma, infection, maternal medication, contact with estrogen products, or familial breast problems. Clinical examination showed a cystic,
A painless, freely moveable lump of the left breast, 50 × 40 × 30 mm in size. The right breast was normal. The left-sided nipple and areola were larger with no signs of inflammation. The external genitalia were normal. Both testes were prepubertal, 1–2 mL in size. There were no signs of puberty or endocrinologic abnormalities. Laboratory investigations including complete blood count, blood glucose, serum electrolytes, renal and liver function tests, serum lipid profile, and urinalysis were all within the normal range. Endocrine evaluation revealed levels of luteinizing hormone, follicle-stimulating hormone, estradiol, cortisol, thyroid-stimulating hormone, prolactin and thyroxin within normal range. Chromosomal analysis revealed normal male karyotype 46, XY. Magnetic resonance imaging (MRI) demonstrated an unilateral, unilocular cystic breast mass 43 × 18 mm in size (Figure 2). The right breast had a normal MRI appearance. The cystic mass was then excised completely using semicircular intra-areolar incision (Figure 3). The postoperative course was uneventful.

Microscopic examination of the resected specimen revealed an unilocal, irregularly shaped cyst with smooth, focally rugged inner surface. Thin, fibroelastic wall of the cyst was lined by columnar epithelium. In some of epithelial cells, clear vacuoles were nicely visible as an expression of apocrine secretory process. Cyst wall was poorly defined from the surrounding mammary fibrous tissue containing rare terminal duct lobular unites (Figure 4).

Fig. 1 – Left-sided galactocele in a 29-month-old boy.

Fig. 2 – Magnetic resonance imaging revealing cystic lesion at the left side of the thorax.

Fig. 3 – Intraoperative view of excised galactocele.

Fig. 4 – Histopathology of galactocele with a poorly defined, thin fibrous wall and the surrounding mammary fibrous tissue containing one terminal duct lobular unit. Galactocele is lined by the columnar epithelium (hematoxylin and eosin, × 25); Some of the epithelial cells show clear intracytoplasmic secretory vacuoles (inset - hematoxylin and eosin, × 250).

Seven months after the surgery there were no signs of recurrence or any other pathological abnormalities (Figure 5).

Fig. 5 – Clinical appearance 5 months postoperatively.

Discussion

Galactoceles generally occur in young women during or after lactation because of the continuous production of milk with impeded evacuation which leads to engorgement and cystic dilatation of the mammary ducts. The pathogenesis of the condition is considered so simple that, with the exception of one of the major textbooks of pathology, Sternberg, it is not even mentioned in either Rosai, Silverberg or Anderson. Galactoceles paradoxically and extremely rarely may be the cause of breast enlargement in infants and children, exclusively of male gender.

The three major factors have been suggested to be important for the development of galactoceles in young boys: previous or present stimulation by prolactin, the presence of secretory breast epithelium, and ductal obstruction. Some authors suggest that galactoceles may develop after a local nipple trauma and inflammation which induce enlargement of small and silent retention cyst already formed in the neonatal period. It was also postulated that it could develop due to unsuccessful hollowing of ducts due to lesser sensitivity of male breast to the maternal hormones responsible for canalization of the buds and their transformation into lactiferous ducts. Some authors believe that etiology and pathogenesis of galactoceles in male infants are multifactorial.

We have recently described two new cases of galactoceles in young boys with analysis of 26 well documented cases in the world medical journals in English and other languages published until 2012. The analyzed articles were usually single case reports. There was only one article describing three and two articles describing two cases, respectively. Unilateral involvement predominates and only one third of patients had bilateral galactoceles. The median age of onset was 7 months, and the median age of clinical presentation 15 months (range from 2 to 72 months). Only one patient had breast enlargement since birth. The onset of the disease in the presented patient at the age of 16 months, and clinical presentation of unilateral galactocele at the age of 29 months, respectively, were in the expected age range.

The presented patient and the most of the previously reported patients were without any developmental and clinical abnormalities, including endocrinological. However, one previously reported 28-month-old boy with unilateral galactocele had persistent hyperprolactinemia and another 18-month-old infant with bilateral breast involvement had elevated insulin-like growth factor-1 because of growth hormone treatment of congenital hypopituitarism. In only 3 reported cases, all with unilateral galactocele, isolated congenital malformation was described: renal dysplasia, ventricular septal defect, and cleft lip.

Differential diagnosis includes lymphatic malformations, at the first place. The dilemma can be resolved by needle aspiration of cyst, which can obtain milk-like fluid suggesting the diagnosis of galactocele. Other diagnostic possibilities include hemangioma, ductal ectasies, hypertrophic mastitis, and also gynecomastia in older boys. Ultrasound examination of galactoceles may show well-defined subcutaneous fluid collection or a complex mass. MRI was performed as a diagnostic procedure in 4 cases only, including one of our previously published cases and also this one. There were no specific mammographic findings in children, but plain mammograms can reveal water-fat level, which is characteristic for galactocele in adults.

Most authors agree that the best treatment of galactoceles is surgical. Simple excision through the intra-areolar incision was curative in all reported cases, as was in this patient. The needle aspiration of cyst fluid may eliminate the need for the surgery but there are only three such cases reported in the literature.

Conclusion

Galactoceles are extremely rare condition in male infants. We presented a 27th case of galactocele in a male infant published in the world literature. The clinical presentation and the age of the patient are typical. There are several hypotheses regarding etiology of the lesion, but it is likely to be multifactorial. Because of its extreme rarity, there are some difficulties in differential diagnosis of galactoceles in the pediatric age. Surgical intra-areolar incision is the treatment of choice and was curative in all the reported cases.
REFERENCES


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