Rare disease

Bloody nipple discharge in infancy—report of two cases

Marta Nascimento, Alexandrina Portela, Filipa Espada, Marcelo Fonseca

Department of Pediatrics, Hospital Pedro Hispano, Matosinhos, Portugal

Correspondence to Dr Marta Nascimento, marta.r.n.nascimento@gmail.com

Summary

Bloody nipple discharge (BND) in infancy is an exceptionally rare finding. We report the cases of two children who are 9 and 29 months old. The first case presented with 1 month of bilateral intermittent blood-stained nipple discharge with no other symptoms. The second case presented with 15 days of intermittent right BND and a small palpable mass, without obvious signs of inflammation. The coagulation and hormonal tests were within the age-appropriate reference ranges. Ultrasound examination was normal. Cytological evaluation of nipple discharge showed no malignant cells. Both patients had spontaneous symptoms resolution. BND in paediatric age is usually benign and self-limited and often related to mammary duct ectasia. Unnecessary invasive procedures or treatments should be avoided.

BACKGROUND

Breast hypertrophy with or without milky discharge is relatively common in newborns and infants of both sexes. This benign phenomenon is linked with placenta-transmitted maternal and fetal hormones and relates to the hormonal adaptation process that occurs in the first months of life.^{1 2} In contrast, bloody nipple discharge (BND) is an extremely uncommon entity in childhood and relates mostly to benign processes such as mammary duct ectasia. However, this is a distressing condition to both parents and physicians because of its association with breast carcinoma in adulthood.² This may lead to unnecessary diagnostic approach and treatment. The authors describe two cases of BND in infancy.

CASE PRESENTATION

Case 1

A 9-month-old white girl presented with bilateral intermittent BND of 1 month duration. Both the parents denied any previous episode of breast or bloody vaginal discharges, recent drug ingestion, breast manipulation or trauma. The child was exclusively breastfed during her first 6 months and afterwards with formula-feeding. Her medical history was uneventful. Familial history was negative for bleeding diathesis, breast carcinoma or endocrine disorders. Her growth curves were all at the 90th percentile. On examination, both breasts showed no signs of inflammation, engorgement or hypertrophy. Pressure on the areolar area resulted in a bilateral bloody discharge (figure 1). The remainder of the physical examination was unremarkable, with normal female genitalia.

Case 2

A 29-month-old white girl was referred to our clinic with a 2-week history of intermittent right BND. There was no history of previous breast discharge, trauma, manipulation or drug ingestion. The child medical history was uneventful. She was in good health and her growth curves were at the 50th and 75th percentile for weight and stature, respectively. Physical examination showed a palpable small

mass under the right breast without signs of inflammation and bloody discharge from the right nipple with areolar pressure. Further physical examination was normal.

INVESTIGATIONS

The results of blood cell count and coagulation tests were normal. Hormonal serum levels, on such as prolactin, oestradiol, thyrotropin, thyroxine, follicle-stimulating hormone and luteinising hormone, were within the age-appropriate reference ranges. Culture of nipple discharge was negative. In patient 1, cytology showed, on a background of red blood cells, macrophages, lymphocytes, neutrophils and ductal epithelial cells. In patient 2, cytological study showed haemosiderin-laden macrophages and ductal epithelial cells. Bilateral breast ultrasonography showed no abnormalities in both cases.

OUTCOME AND FOLLOW-UP

In patient 1 the nipple discharge diminished gradually and resolved spontaneously within 3 months. Follow-up until the age of 3 years was uneventful.

Figure 1 Bloody nipple discharge with no signs of infection, engorgement or hypertrophy.

In patient 2 the nipple discharge ceased spontaneously over a period of 6 months. After a follow-up of 7 months, there had been no relapse.

DISCUSSION

Few data exist regarding the development and growth of the human breast during the first months of life. Physiological enlargement of the breast and milky discharge in newborns and infants is frequently observed and results from the precipitous drop in maternal hormones after delivery and high levels of fetal and newborn prolactin.^{1–3} However, BND in infancy is extremely rare with only 30 reported cases in the literature, in children of both sexes.^{1–6}

According to many authors, the most common underlying cause of BND is mammary duct ectasia which consists of duct dilatation surrounded by periductal fibrous tissue and inflammatory reaction. This duct dilatation causes a substantial disparity in relation to the other ducts, and its lumen is occupied by lipids and debris. Over time, phagocytic giant cells that surround the lipid material together with the histiocytes form a granulation tissue with ulceration of the ductal epithelium which will then be responsible for the BND. Commonly, it occurs during the perimenopausal years in the multipara and only rarely in the nullipara, in men or children.³ The specific aetiology of mammary duct ectasia remains unknown. Although many factors including maternal hormonal stimulation, congenital abnormalities of the nipple and duct system, infection, trauma or autoimmune reaction have been implicated, neither was confirmed.^{1 3 5} There is a wide range of age at presentation, with infants as young as 6 weeks presenting with the disease.² This condition is more prevalent in the male gender with a male-to-female ratio of 11:4 which contrasts to the reported female sex predilection in the adult population.^{1–6} This suggests a different aetiology of BND in the paediatric population.

The typical clinical presentation in children includes the presence of intermittent unilateral or bilateral BND, in the absence of any inflammatory features and with or without associated breast hypertrophy or palpable mass. These latter findings do not change the benign course of BND in children.^{3 5}

The initial workup of an infant with BND should include careful clinical examination to assess the nature of the discharge, signs of inflammation and presence of any breast lump. Gram stain, cell count and culture of the discharge followed by serum hormonal levels analysis (prolactin, oestradiol and thyrotropin) and breast ultrasound are recommended.⁷ Ultrasound examination is a useful diagnostic tool. Nevertheless, sonographic findings can vary in this condition and, although the presence of dilated ducts is a common finding, it is not a constant feature.³

In published reports, most cases of BND resolved spontaneously within 11 months which demonstrates the self-limiting character of this condition. Moreover, all reported cases of children submitted to total subcutaneous mastectomy showed histological findings compatible with

mammary duct ectasia.^{2 3} This suggests that an expectant line of management would be appropriate in this age group.

In our patients, the clinical presentation resembles that of the previously reported in the literature suggesting this benign process. Ultrasound did not reveal any abnormalities, normal hormonal and coagulation levels were obtained, and culture of the discharge was negative. Cytology showed ductal epithelial cells without malignancy which confirmed mammary duct ectasia. The nipple discharge ceased spontaneously in both patients.

Given the spontaneous resolution of the BND in the majority of the reported cases and in the light of malignancy being extremely uncommon in the paediatric population a conservative management with reassurance and periodic assessments would be appropriate. Invasive procedures, including biopsy or surgery, and further investigations should be considered only in case of a sonographic finding of a mass or abnormality other than mammary duct ectasia and if BND does not resolve in 6–9 months.

Although BND may be of great concern for both parents and physicians, it is a benign and self-limited process. It is often an expression of mammary duct ectasia which seems to be a variant of physiological breast development in childhood.

Learning points

- Bloody nipple discharge in childhood is a rare but benign and self-limiting condition and often associated with mammary duct ectasia.
- Mammary duct ectasia may represent a variant of breast development in the paediatric population.
- Unnecessary invasive investigations or treatments should be avoided.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Seo JY**, Kim SJ, Lee SJ, *et al.* Bloody nipple discharge in an infant. *Korean J Pediatr* 2010;**53**:917–20.
2. **Tan R**, Bosstraeten BV, Casteels K. Does bloody nipple discharge occur during normal breast development in infancy? *Pediatr Int* 2010;**52**:825–43.
3. **Sousa SG**, Carvalho L, Oliveira JG. Bloody nipple discharge in a breastfeeding boy. *J Paediatr Child Health* 2010;**46**:784–9.
4. **Djilas-Ivanovic D**, Boban J, Katanic D, *et al.* Bilateral bloody nipple discharge in a male infant: sonographic findings and proposed diagnostic approach. *J Pediatr Endocr Met* 2012;**25**:163–4.
5. **McHoney M**, Munro F, Mackinlay G. Mammary duct ectasia in children: report of a short series and review of the literature. *Early Hum Dev* 2011;**87**:527–30.
6. **Pampal A**, Gokoz A, Tansu S, *et al.* Bloody nipple discharge in 2 infants with interesting cytologic findings of extramedullary hematopoiesis and hemophagocytosis. *J Pediatr Hematol Oncol* 2012;**34**:229–31.
7. **Kelly VM**, Arif K, Ralston S, *et al.* Bloody nipple discharge in an infant and a proposed diagnostic approach. *Pediatrics* 2006;**117**:814–16.

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