Case Report

Apparent cyclic vaginal bleeding in a child: factitious disorder

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Abstract. A 20-mo-old girl was brought to our department by her mother because of breast enlargement. She was diagnosed with premature thelarche. One month later, she returned to our hospital with a complaint of vaginal bleeding. During the subsequent 6 mo, her vaginal bleeding recurred every month while her breast development disappeared. We performed laboratory tests and imaging. At the end of 6 mo, we realized that her mother's menstrual bleeding and the patient's blood staining were concurrent. The mother confessed applying her vaginal flow to her daughter's underwear. Factitious disorder should be included in the differential diagnosis of unexplained vaginal bleeding in childhood.

Key words: prepubertal vaginal bleeding, factitious disorder

Introduction

Vaginal bleeding (VB) is an uncommon problem in childhood. Early vaginal bleeding (premature menarche) associated with other pubertal symptoms such as breast development (thelarche) may be a sign of true or pseudoprecocious puberty. On the other hand, many cases of isolated vaginal bleeding are related to local problems such as vulvovaginitis,

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presence of a foreign body, sexual abuse, urethral prolapse, and sarcoma botryoides (1). VB can be a source of great concern for both pediatricians and family, and should be investigated carefully. Here, we describe a case of apparent cyclic vaginal bleeding, later diagnosed as a factitious disorder.

Case

A 20-mo-old girl was brought to our department by her mother because of breast enlargement. Physical examination revealed Tanner stage 2 bilateral breast development, with no pubic hair. Body weight was 10.5 kg (25^{th} percentile) and height was 82 cm (25^{th} - 50^{th} percentile). Basal and GnRH-stimulated plasma FSH levels were increased, and LH levels were prepubertal (Table 1). The plasma estradiol level was < 10 pg/ml. The bone age was consistent with age. She was diagnosed with premature

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	FSH (mU/ml)		LH (mIU/ml)		- Peak LH/FSH
	Basal	Peak	Basal	Peak	- reak Ln/rSn
First test	2.03	22.03	< 0.06	4.82	0.21
Second test	1.65	20.30	< 0.06	5.04	0.24

Table 1.	GnRH	test	results
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thelarche based on clinical and laboratory findings. We advised monitoring.

One month later, she presented with a complaint of vaginal bleeding. Physical examination showed Tanner stage 2 bilateral breast development. There was no pubic hair or skin pigmentation. There was no active vaginal bleeding, laceration, abrasion, or urethral prolapse. Genital examination was repeated by a gynecologist. Routine laboratory and coagulation tests were normal. There was no hematuria or bleeding diathesis. Repeated basal and GnRH-stimulated gonadotropin levels were not in the pubertal range (Table 1). The plasma estradiol level was less than 10 pg/ml. Pelvic ultrasound and magnetic resonance imaging (MRI) demonstrated a $7 \times 20 \times 25$ mm uterus, a $20 \times 15 \times 20$ mm (3.1 ml) right ovary, and a 20 \times 10 \times 25 mm (2.6 ml) left ovary; endometrial thickness was not observed, ovarian follicular microcysts with maximum size of 10 mm were present, and there was no tumor or foreign body. We decided to follow the patient without treatment. However, during the subsequent 6 mo, she presented every month with a complaint of recurrent vaginal bleeding, although breast development regressed, growth velocity was normal, and bone age did not accelerate.

At the end of 6 mo, after excluding all possible medical explanations for vaginal bleeding, we suspected that our case might be a "factitious disorder." On her final presentation, we asked her mother for her menstrual pattern, and realized that apparent vaginal bleeding in the patient and menstrual bleeding in the mother were concurrent. The mother was referred to a psychiatrist, and confessed that her own vaginal flow was applied to her daughter's underwear. On the first visit, the mother brought her daughter to the hospital for a complaint of breast enlargement. She was advised to take her daughter to the doctor 3–6 mo later, but returned earlier because she was told to do so in the event of any vaginal bleeding or pubic hair growth.

Discussion

We initially thought that GnRH given for the stimulation test on admission might have caused a burst of ovarian estrogen secretion and vaginal withdrawal bleeding; however, this idea lost validity because our patient's complaint of bleeding was sustained.

Since premature thelarche may progress to central precocious puberty (CPP), we evaluated our patient in this regard. Her clinical and hormonal features included normal growth velocity and bone age, and prepubertal levels of GnRH-stimulated LH were not compatible with early puberty. Furthermore, her breast enlargement disappeared during follow-up.

Repeated measurements revealed that basal LH levels were prepubertal. The peak LH values were 4.82 and 5.04 mIU/ml in the first and second GnRH tests. These LH values could be considered pubertal. The peak LH response to GnRH that defines the onset of puberty is above 3.3-6.9 mIU/ml in girls (2–4). In these studies, LH levels were determined by immunochemiluminometric assay (2–4). It is known that an elevated gonadotropin response to the GnRH test is not diagnostic of CPP, and is not associated with progression to true precocious puberty in girls younger than 3 yr. GnRH-stimulated LH was found to be > 5 mU/

mL in 36.4% of girls with idiopathic premature thelarche (5). The patient's age should be taken into consideration when GnRH stimulation test results are interpreted in small children. The peak LH/peak FSH ratios in our patient were 0.21 and 0.24. A high peak LH/peak FSH ratio (> 1) is an indicator of CPP (5, 6). One study suggested that a peak LH/peak FSH ratio higher than 0.66 can be diagnostic of CPP (7). We interpreted the GnRH stimulation tests as prepubertal due to predominant FSH responses and the patient's age.

Ovarian volumes were of pubertal size on ultrasound and MRI (> 2 ml). At two years of age, the maximum ovarian volume is 1.5 ml (+2 standard deviation) (8). The cut-off value for pubertal ovarian volume is in the range of 1 to 3 mL (9). We thought that large ovarian volumes might be due to follicular microcysts in the ovary. There was also no endometrial echo. These findings were not compatible with vaginal bleeding. The patient's pelvic ultrasound and MRI demonstrated ovarian follicular microcysts, which can be observed in girls with isolated premature thelarche (10, 11).

Factitious disorder imposed on another (previously known as Munchausen syndrome by proxy) is a form of child maltreatment caused by a caregiver. In 85% of cases, the disease is initiated by the mother of the child, and less often by the father or other caregivers. This situation leads to unnecessary and invasive medical procedures and/or treatment (12). Females and males are victimized equally (13, 14). The problem presents most often in small children up to 2 yr old and seldom older than 6 yr (12, 15). Perpetrators may fabricate, exaggerate, and/or induce physical, psychological, behavioral, and/or mental health problems in the child. A reported case series of 117 children who suffered falsely reported or induced illness showed that 25% of patients had their illness simulated, 50% had illness induced, and 25% had illness simulated and induced. The most common presentation involves hemorrhage or bleeding. Described forms of bleeding usually

are hematuria, hematochezia, melena, or hematemesis. Less common sites include the ears, nose, and respiratory tract (13). In the literature, unexplained vaginal bleeding in a 6-mo-old girl proved to be Munchausen syndrome by proxy, when blood found in the baby's vagina was found to match the mother's blood type but not the baby's (16).

Our patient's family had a low sociocultural environment. Because she was living in a crowded and troubled family environment, and wanted to be away from home and have an excuse to come to the hospital, the mother fabricated the complaint of vaginal bleeding. Parents with a sick child might obtain secondary gain by controlling the environment and making new friends at the hospital. Parents might derive satisfaction from such a situation (17). The psychiatrists thought that the mother obtained secondary gain from her child's situation to escape from some of the undesirable aspects of her life such as doing housework or caring for elderly people in her family.

The psychiatric problem of the mother was not determined. According to some researchers, psychiatric assessment of these mothers usually yields normal results (18). However, another study reported that a large proportion of perpetrators have chronic somatoform disorders (15). The psychiatrists decided that the child did not need to be removed from her mother.

The mother was quiet and cooperative at routine visits, and did not display any suspicious behavior. Perpetrators have typical characteristics, and convey a sense of selfsacrifice; they are interested in medical details, highly helpful to the doctors, and are calm when presented with the seriousness of their child's medical condition (16).

In conclusion, we suggest that factitious disorder should be included in the differential diagnosis of unexplained vaginal bleeding in childhood. The physician should see the actual blood and determine the blood type when a patient has a report of unexplained bleeding. **Conflict of Interest:** The authors have no potential conflicts of interest to disclose.

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