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Clinical characteristics and conservative treatment of perineal groove

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ABSTRACT

Purpose: Perineal groove is a rare congenital anomaly of the perineum, and only a few papers describing a small number of cases have been reported in the medical literature. This study aimed to evaluate the clinical characteristics and proper management of perineal groove.
Methods: We performed a retrospective review of 26 pediatric patients who were diagnosed with perineal groove between January 2012 and October 2018 at our institution.
Results: Perineal groove was extremely prevalent among the females: 25 of the 26 patients were girls, and only one patient was a boy. All the patients presented with an unusual lesion at the anus or perineum, but no symptoms related to this anomaly. The median age at the first visit to our clinic was 1.5 month (range, 0.3–11.4 month). Two types, complete and partial, were used to categorize the appearance of perineal groove. In a sample group, 55% (12/22) of the patients had complete perineal groove while 45% (10/22) had partial perineal groove. One patient underwent an anoplasty at another hospital following the diagnosis of an imperforate anus. One male and 13 female patients were followed beyond the age of two, and 10 patients (71%) showed a natural healing process.
Conclusion: Perineal groove manifested as two types of appearance and showed excellent results with conservative treatment in our study. A natural healing process can be expected in the long-term follow-up. Perineal groove must be differentiated from other defects to avoid unnecessary surgical treatment.
Levels of Evidence: Therapeutic Study, Level IV.

Perineal groove is a congenital anomaly of the perineum that is characterized by a groove extending from the posterior fourchette of the vagina to the anterior edge of the anus with normal formation of the vestibule, urethra, and vagina [1]. The incidence of this anomaly is unclear, but it is known to be rare. There are a limited number of studies on perineal groove, and most of them are confined to case reports [2–4], so it is unlikely that pediatric surgeons and pediatricians are fully aware of, and therefore understand, this defect well.

In the current study, we collected and analyzed a large number of cases and conducted a review of the relevant literature with the aim of providing the clinical characteristics and proper management of perineal groove.

1. Material and methods

We performed a retrospective review of 26 pediatric patients who were diagnosed with perineal groove between January 2012 and October 2018 at our institution. Each patient was diagnosed via a physical examination depending on the gross appearance of the lesion. They underwent digital rectal examination or insertion of Hegar dilators to exclude other anorectal malformation. We did not use specific radiologic evaluation tools, but a simple abdominal film was usually taken to screening for sacral anomalies and stool impaction in the rectum.

We recommended that the patients visit our outpatient clinic every year even if they did not have any specific symptoms. We took a picture of most of the patients' lesions and compared them with previous photos to evaluate the clinical course of perineal groove.

This study was approved by the Institutional Review Board of our hospital. This research did not receive any specific grants from funding agencies in the public, commercial, or not-for-profit sectors. There are no conflicts of interest to declare.

2. Results

The patients' characteristics are summarized in Table 1. Perineal groove was extremely prevalent among the female patients, with 25 girls and only one male among the 26 patients. All the patients presented with an unusual lesion at the anus or perineum, but no symptoms related to this anomaly. All the patients were born in good health without a history of admission. Their mean gestational age was 39.4 ± 1.2 weeks, and their mean birth weight was 3.27 ± 0.36 kg.

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Table 1
Characteristics of patients.

Characteristics	
Cases (n)	26
Duration	Jan 2102–Oct 2018
Sex (male:female)	1:25
Birth weight (mean ± SD, kg)	3.27 ± 0.36
Gestational age (mean ± SD, weeks)	39.4 ± 1.2
Median age (month) at first visit to clinic (range)	1.5 (0.3–11.4)
Type	
Complete	55% (12/22)
Partial	45% (10/22)
Follow-up more than 2 years	54% (14/26)
Median age (year) at follow-up (range)	2.9 (2.0–5.7)
Healing process (+)	71% (10/14)

Their median age at the first visit to our clinic was 1.5 months (range, 0.3–11.4 months). One patient had a perineal hemangioma, one patient had congenital pulmonary valve stenosis, and one patient had plagiocephaly.

The physical appearance of perineal groove was categorized into two types, complete and partial. In complete perineal groove, a sulcus extends from the posterior fourchette of the vagina to the anterior edge of the anus, but in the partial type, the sulcus is incomplete and does not reach the posterior fourchette of the vagina (Fig. 1). The appearance of the sulcus was evaluated in 22 female patients by gross photography, and 55% (12/22) were found to have complete perineal groove and 45% (10/22) had partial perineal groove. We had one male patient with perineal groove, and he did not present with any other abnormalities (Fig. 2).

One patient underwent an anoplasty at another hospital following the diagnosis of an imperforate anus. One male and 13 female patients were followed beyond the age of two. One patient had a symptom of intermittent pain on defecation, and other patients had no symptoms associated with this malformation. Ten patients (71%) showed a natural healing process, but none of the patients healed completely (Figs. 3, 4).

3. Discussion

Perineal groove is rare congenital anomaly. Its characteristics were well described by Stephens and Smith [1] as "normal formation of the vestibule, including urethra and vagina in its anterior; hypertrophic minoral tails which skirt the perineum and course posteriorly to join at the anus or surround it: a wet groove in the perineum between the fourchette and the anus". It was originally classified in the "miscellaneous" rare group in the 1970 international classification but was recently reclassified as "rare/regional variants" [5,6]. Previous studies of this malformation are limited: we located only a few papers reporting a small number of cases in the medical literature [2–4,7]. To the best of our knowledge, this study includes the largest patient cohort compared to previous reports, and it was therefore possible for us to analyze more accurately the clinical characteristics of perineal groove and its management.

In this study, we categorized the appearance of perineal groove into complete and partial types. The complete types in our study reflected the classic appearance described above. Mention of the partial type were rare in the literature, with only one previous study describing the types of perineal groove, and their classification was similar to ours [8]. As shown in Fig. 2, the partial type in our study was vastly different from the complete type as there was an incomplete sulcus from the anus that did not reach the vagina. The number of partial-type patients in our study was not small; instead, it accounted for nearly half our patients. In our opinion, the partial type of perineal groove resembles the appearance of anal fissures and should therefore be differentiated from this disorder. Awareness of partial-type perineal groove is important to diagnose this malformation properly.

In addition, we found a case of perineal groove in a male. Perineal groove has been established as extremely prevalent among females, with only one case of a male with perineal groove mentioned in previously published papers [9]. We have therefore added one more case to the study of males with perineal groove. Although the pathogenesis of perineal groove is not well known and several embryological hypotheses have been controversial, the confirmation of the occurrence of perineal groove in males suggests that it could be derived from an embryological developmental anomaly related to both sexes [10].

The incidence of combined anomalies in perineal groove is rare although a few studies have reported the association of hypospadias and bifid scrotum [9], anomalies of the urinary tract [11], and ectopic anus [12]. We could not find anomalies associated with perineal groove in this study.

Although perineal groove is known to be rare, its actual incidence is likely to be higher than suggested in previous reports [4,10]. In this

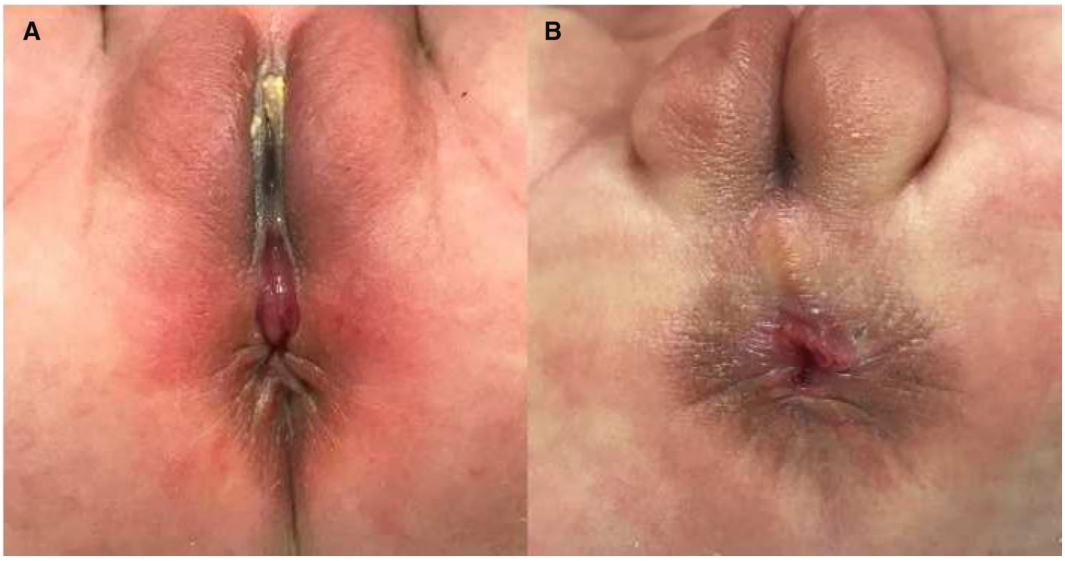

Fig. 1. Photographs of perineal grooves showing the complete type (A) in a five-day-old girl and the partial type (B) in a four-day-old girl.

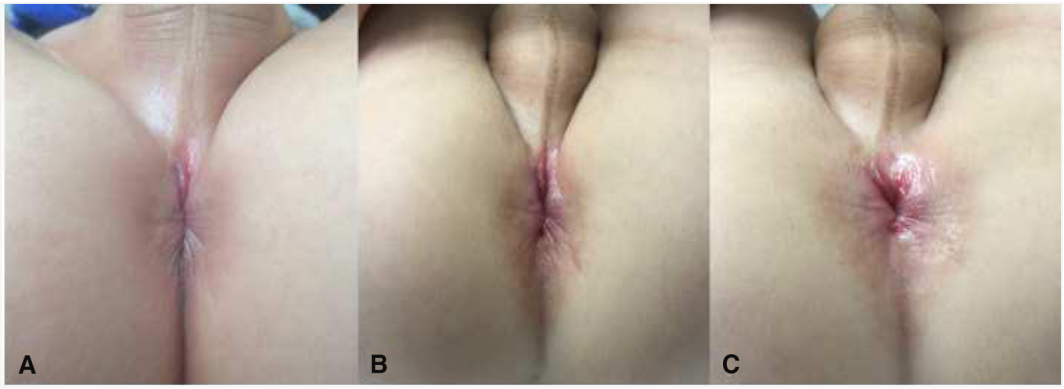

Fig. 2. A case of perineal groove in a male. The photographs were taken at ages two (A) and 4 years (B, C).

study, we reviewed 26 cases covering a seven-year period while Garcia-Palacios et al. [4] reported five cases over the course of 2 years. These incidences suggest that it could be more common than previously known.

The treatment of perineal groove in our study was mainly conservative, but a few studies have advocated for the surgical treatment of perineal groove to mitigate possible complications and cosmetic problems

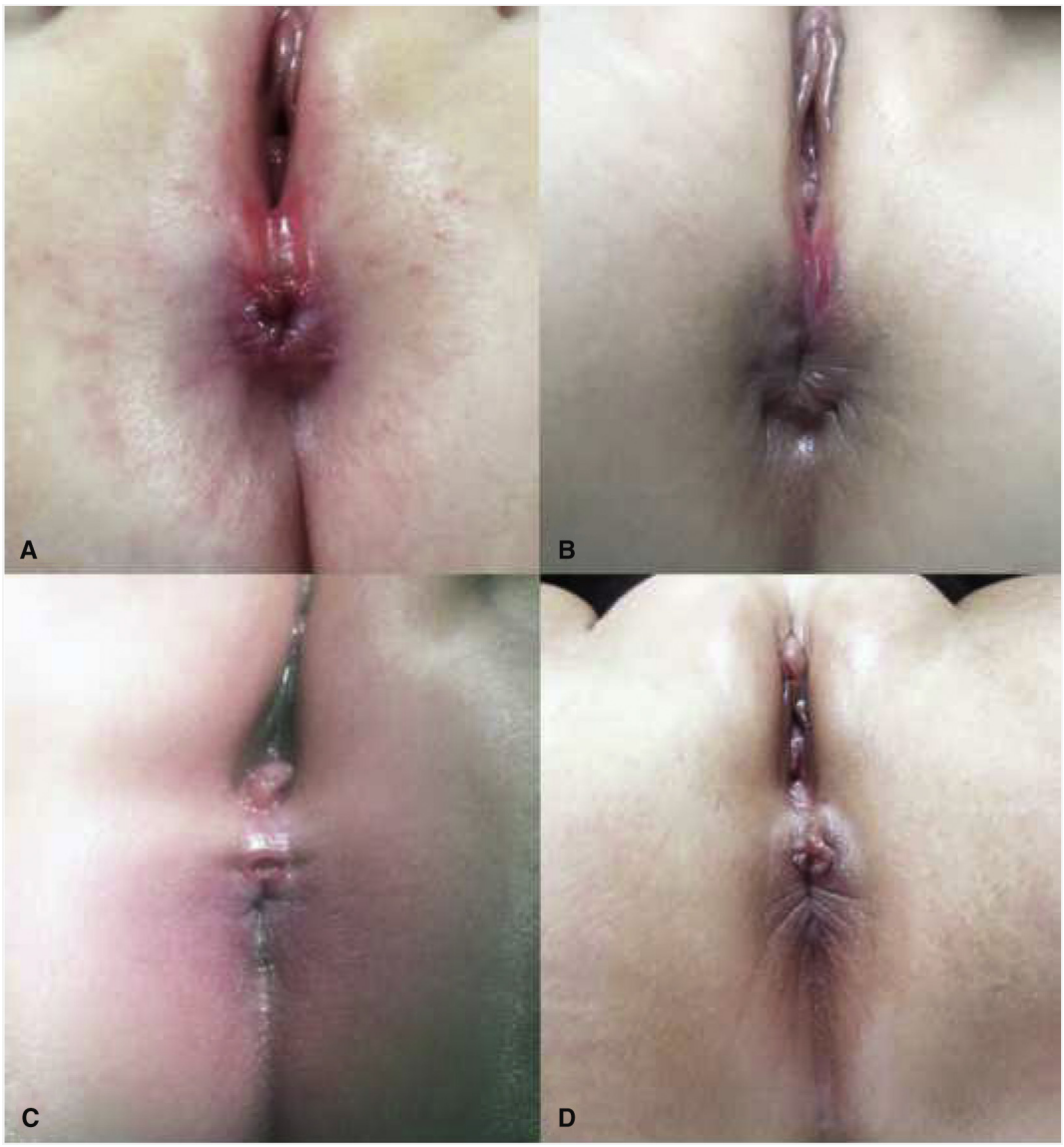

Fig. 3. Perineal groove showing a natural healing process. These photographs were taken at 6 months (A) and 3 years of age (B) in a girl, and 4 months (C) and 2 years of age (D) in another girl.

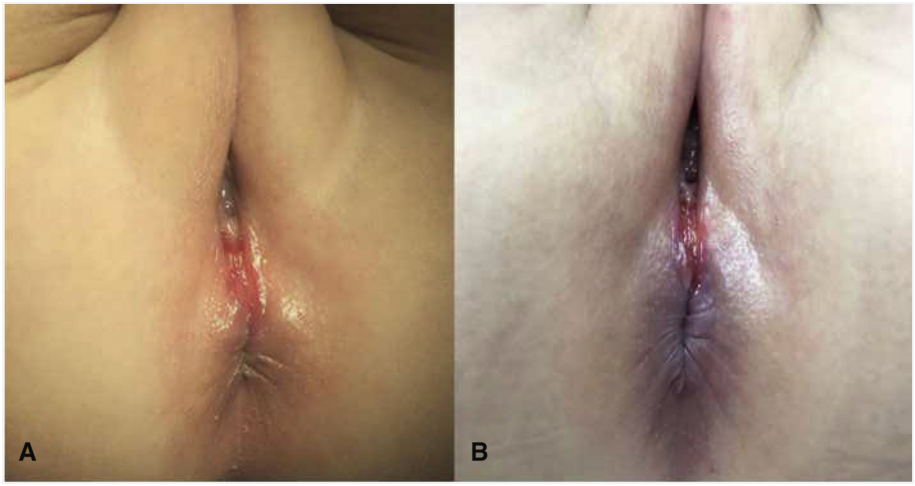

Fig. 4. Perineal groove showing no healing. These photographs of the same girl were taken at 1 year (A) and 3 years of age (B).

[13,14]. In this study, we followed 14 patients beyond that age of two and found only one patient who had mild symptom associated with perineal groove. However, healing did not take place in all the patients, and no one healed completely. Accordingly, we think the healing process may take longer than previously thought [2,7,15].

As shown in our results, perineal groove should be differentiated from the perineal cutaneous fistula of anorectal malformation. A differential diagnosis is occasionally not easy, so careful observation of the lesion is required to avoid unnecessary surgical treatments. Other disorders, such as anal fissure, and sexual abuse should be considered for a possible differential diagnosis [3,4,15].

In summary, we discerned two types of perineal groove based on appearance, and both responded excellently with conservative treatment. The incidence of this malformation could be higher than previously reported, and we confirmed a male case of perineal groove. Perineal groove must be differentiated from other possible disorders or sexual abuse to avoid unnecessary surgical treatment. A long-term natural healing process should be expected.

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