Asian Journal of Surgery xxx (xxxx) xxx

Contents lists available at ScienceDirect

Asian Journal of Surgery

journal homepage: www.e-asianjournalsurgery.com

Letter to Editor

# Preputial balanitis xerotica obliterans shortly after hand, foot and mouth disease in a 5-year-old boy: Coincidence or consequence?

*Keywords:*
Balanitis xerotica obliterans
Hand
Foot and mouth disease
Children

*Dear Editor,*

Balanitis xerotica obliterans (BXO), a genital form of lichen sclerosus (LS) in males, is a progressive, sclerosing, inflammatory dermatosis affecting the foreskin, frenulum, glans penis, meatus, and urethra. BXO was considered a disease of adulthood until 1962 when Catterall and Oates reported the first case of isolated BXO of the prepuce in a 7-year-old child.[1] The exact cause and etiology of BXO remain unknown. However, autoimmune dysregulation, various infections, genetic predisposition, chronic irritation through urine exposure, and hormonal influences have been associated with the development of BXO.[2] Recent studies indicate that BXO is more common than previously thought to cause acquired phimosis and meatal stenosis.[3] However, despite an increasing rate of diagnosis of BXO in children, little is known regarding the potential for associated comorbidities or an association with various human viruses. To the best of our knowledge, no cases of hand, foot, and mouth disease (HFMD) associated with BXO have been reported so far. A 5-year-old boy was referred to our department with an 8-month history of a painless progressive inability to retract the foreskin of the glans penis. The condition worsened to almost complete occlusion over the last 15 days. His father reported that the inability to retract the foreskin over the glans penis occurred ten days after the appearance of the HFMD. HFMD diagnosis was based on clinical features in the form of initial fever and trouble swallowing, followed by oral erosions and vesicles affecting the palms and soles. The illness resolved spontaneously after a week. The family history was negative for any metabolic and immune-related disorders. On physical examination, whitish, non-retractable foreskin with acquired scarring phimosis was observed (Fig. 1). After preoperative preparation, the boy underwent circumcision. The meatus itself was normal with no stenosis. The circumcised foreskin was submitted for histopathological examination. Histopathological examination of the biopsy sample was consistent with BXO (Fig. 2A and B). HPV testing was not done.

The etiology of BXO remains unknown. Various microorganisms (acid-fast bacilli, spirochetes, and viral agents such as human papillomavirus/HPV/and hepatitis C/HCV/) have been linked with BXO.[2,4] The presence of HPV by a polymerase chain reaction (PCR) in some pediatric patients with confirmed BXO[4] does not prove the association between HPV and BXO. However, it may indicate that HPV infection may be superimposed on BXO. A possible association of HCV with BXO has also been investigated but without clear evidence.[4] HFMD is an infectious disease caused by the *Picornaviridae* family members, such as Enterovirus 71 (EV71) and Coxsackievirus A16 (CVA16). These infections typically affect children with underlying immune or metabolic disorders.[5] Since the current literature is sparse, we can only speculate whether immune dysfunction or other mechanisms may underlie these two entities.

Nevertheless, the treatment of preputial BXO with circumcision is well established as the preferred therapeutic modality, as confirmed in our case. In conclusion, the association between BXO and HFMD has not been previously reported. Further studies should reveal whether it is a real association or a coincidence.

**Fig. 1.** Preputial appearance characterized by whitish cicatricial phimosis and scarred foreskin.

https://doi.org/10.1016/j.asjsur.2021.02.014

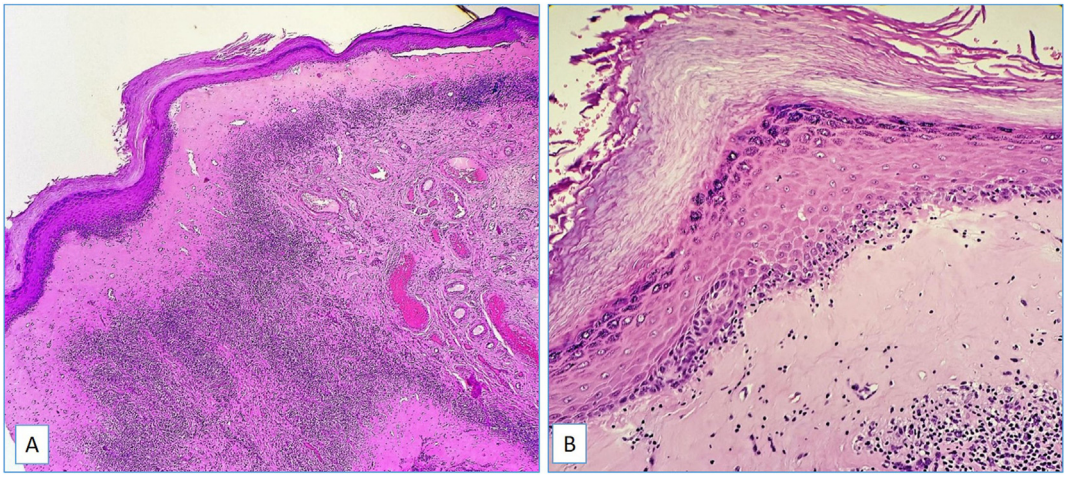


**Fig. 2.** A–B: Hematoxylin and Eosin (H&E) slide of the preputial biopsy showing a markedly thickened surface epithelium with deep sclerosis/hyalinized stroma (collagen), and lymphocytic infiltrates in the lamina propria (A, 5× magnification); Overlying squamous epithelium exhibited marked hyper- and parakeratosis (B, 20× magnification).

## Declaration of competing interest

The authors have no conflict of interest associated with the current manuscript.

## Acknowledgment

The authors thank the patient's father for permitting us to report this peculiar case.

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9 February 2021
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