Turkish Journal of Pediatric Disease
Türkiye Çocuk Hastalıkları Dergisi

A Rare Presentation of Behcet's Disease Detected Postoperatively

Postoperatif İzlemde Saptanan Nadir Bir Behçet Hastalığı Olgusu

Yusuf Alper KARA[1], Fatma Tugba GUVENC[1], Asuman GURKAN[2], Derya ERDOGAN[1]

[1] SBU, Ankara Dr. Sami Ulus Maternity and Child Health and Diseases Training and Research Hospital, Pediatric Surgery, Ankara, Turkey
[2] SBU, Ankara Dr. Sami Ulus Maternity and Child Health and Diseases Training and Research Hospital, Department of Dermatology, Ankara, Turkey

ABSTRACT

Behçet's disease is a rare vasculitic disorder that presents as a triple-symptom complex of recurrent oral aphthous ulcers, genital ulcers and uveitis. Although the common symptoms may be present in patients with Behçet's disease, delayed wound healing can also occur due to environmental factors. A 16-year-old-boy operated for a non-communicating hydrocele presented to the emergency room shortly after discharge with a complaint of recurrent dehiscence and serous leak from the incision, despite being treated once. After detailed investigations, the patient was diagnosed with Behçet's disease and treated with colchicine and the wound and genital ulcerations were healed. Hence, Behçet's disease should also be considered when diagnosing and treating patients with deficient wound healing, a history of surgical interventions and different ulcer lesions after surgery.

Key Words: Behcet syndrome, Skin manifestations, Surgery

ÖZ

Behçet hastalığı; tekrarlayan aftöz ülserler, genital ülserler ve üveit üçlü semptom kompleksi ile, nadir görülen bir vaskülitik bozukluktur. Behçet hastalığı klasik semptomlarıyla bulgu vererek ortaya çıkabildiği gibi, çevresel faktörlere bağlı yara iyileşmesinde gecikme de görülebilmektedir. Nonkommunike hidrosel nedeniyle opere edilen 16 yaşında erkek hasta postoperatif erken dönemde insizyonunda akıntı ve açılma nedeniyle acil servise başvurdu, yara yeri revizyonu sonrası tekrar yara yerinde açılma ve yeni ülsere alanların gözlenmesi üzerine hastada Behçet hastalığı düşünüldü. Detaylı tarama sonrasında hastaya Behçet hastalığı tanısı koyuldu ve kolşisin tedavisi başlandı ve insizyonda iyileşme ve genital ülserlerde düzelme görüldü. Çocuk hastalarda cerrahi sonrası iyileşmeyen insizyonlar ve farklı ülserasyonlar bulunan olgularda tanı koymada ve tedavi yapılırken Behçet hastalığı da mutlaka akılda tutulmalıdır.

Anahtar Kelimeler: Behçet hastalığı, Cilt bulguları, Cerrahi

INTRODUCTION

Behcet's disease (BD) is a rare vasculitic disorder that presents as a triple-symptom complex of recurrent oral aphthous ulcers, genital ulcers and uveitis (1). The onset is insidious, with the peak age of onset being young adulthood (25–30 years); however, 4%–26% of cases have been reported in children aged <16 years (2). Although the common symptoms may be present in patients with BD, delayed wound healing may also occur due to environmental factors. Here we present the case of a 16-year-old boy diagnosed with BD after undergoing hydrocelectomy.

KARA YA : 0000-0002-1265-0997
GUVENC FT : 0000-0002-9464-9737
GURKAN A : 0000-0001-9708-296X
ERDOGAN D : 0000-0003-1730-4177

Conflict of Interest /*Çıkar Çatışması:* *On behalf of all authors, the corresponding author states that there is no conflict of interest.*

Financial Disclosure / *Finansal Destek*: The authors declared that this case has received no financial support.

***Confirmation / Onay*:** The written consent was received from the patient who was presented in this study.

How to cite / *Atıf Yazım Şekli* : Kara YA, Guvenc FT, Gurkan A, Erdogan D. A Rare Presentation of Behcet's Disease Detected Postoperatively. Turkish J Pediatr Dis 2020;14:536-537.

Additional information / *Ek Bilgi*: In this study, 26 to 29 September 2019 Date of Izmir - Turkey at the International Congress of Gynecology and Pediatrics (International Maternity and Child Diseases Congress) was presented as a Poster Presentation.

Correspondence Address / Yazışma Adresi :
Yusuf Alper KARA
Ankara Dr. Sami Ulus Maternity and Child Health and Diseases Training and Research Hospital, Pediatric Surgery, Ankara, Turkey
E-posta: dryakara1024@gmail.com

Received / Geliş tarihi : 26.02.2020
Accepted / Kabul Tarihi : 12.05.2020
Online published : 05.10.2020
Elektronik yayın tarihi
DOI: 10.12956/tchd.681974

CASE REPORT

A 16-year-old boy presented to the hospital with a complaint of swelling on the left scrotum. Physical examination revealed the presence of a left non-communicating hydrocele, and hydrocelectomy was planned. Under general anaesthesia, left hydrocelectomy (Winkelmann's procedure) was performed via a scrotal raphe incision that was closed with subcuticular sutures. There were no postoperative complications at the 24-hour follow-up until discharge.

On the 4th postoperative day, the patient presented to the emergency room with a complaint of dehiscence and serous leak from the incision (Figure 1). Daily wound dressing was performed for 12 days, but there was no sign of healing. Wound culture taken during the first presentation to the hospital was negative. The incision was repaired with non-absorbable sutures. Two days after the incision repair, the patient presented to the hospital again with a complaint of wound dehiscence. Moreover, he had ulcerations on the scrotum because of recurrent wound dressing (Figure 2). Because of the genital ulcerations, it was thought that the patient had a rheumatological disorder. Therefore, he was consulted to the rheumatology and dermatology departments and was subsequently hospitalised. An aphthous lesion occurred in his oral mucosa during the hospital stay (Figure 3). A deeper medical history taken from the patient revealed that he previously had a recurrent aphthous lesion in his mouth and erythematous lesions consistent with erythema nodosum on the skin of his leg. Dermatological, rheumatological and ophthalmological consultations were performed. There was no sign of uveitis on the ophthalmological examination. Clinical findings supported the existence of BD, although the pathergy test was negative. Local wound care and oral colchicine were prescribed. During the follow-up period, the wound was healing and genital ulcerations were gradually disappearing. There were no surgical complaints during the 12-month follow-up in the rheumatology department since the diagnosis of BD.

DISCUSSION

BD is a multisystem disease characterised by mucocutaneous and ophthalmological manifestations. Recurrent oral aphthae and genital ulcerations, erythema nodosum, papulopustular lesions, thrombophlebitis and positive pathergy test results are diagnostic factors for BD (3). In the present case, the patient had oral aphthae, genital ulcerations and a history of erythema nodosum, which led to the diagnosis of BD.

Hydrocele surgery is one of the most frequent surgical approaches in the inguinal region in children. After surgery, there is a 0.6%–1.4% rate of dehiscence of the incision (4, 5). This is an unexpected complication, unless the patient has a connective tissue disease or a lack of general hygiene. The most expected causes of this complication are wound infections, age younger than 12 months, median abdominal incision, and a history of emergent surgery. Other causes include concomitant systemic diseases, such diabetes, anaemia, and vasculitis (4).

In the present case, the patient's lesions and history were suggestive of BD. After the investigations, the diagnosis became clearer and proper treatment was administered to the patient. After the treatment, the incision healed and there were no other complications. Moreover, other lesions, such as oral aphthae and genital ulcers, healed.

Recurrent wound dehiscence is a rare entity in children. In patients with a similar history, the causes mentioned in the present case must be investigated and eliminated. This is the first report on a case of BD occurring after a surgical approach with wound dehiscence in the English literature. BD should also be considered in patients with deficient wound healing, a history of surgical interventions and different ulcer lesions after surgery.

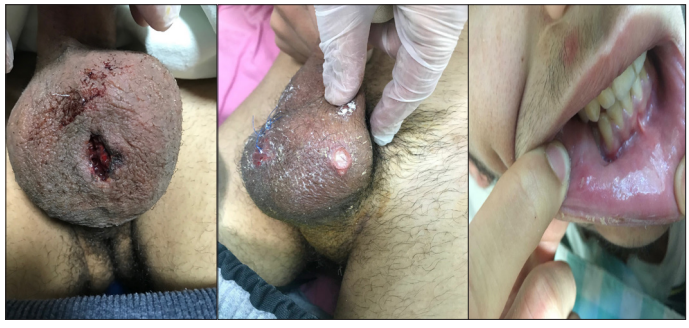

Figure 1 Figure 2 Figure 3

REFERENCES

1. Davatchi F, Chams-Davatchi C, Shams H, Shahram F, Nadji A, Akhlaghi M, et al. Behcet's disease: epidemiology, clinical manifestations, and diagnosis. Expert Rev Clin Immunol. 2017;13:57-65.
2. Koné-Paut I. Behcet's disease in children, an overview. Pediatr Rheumatol Online J 2016;14:10.
3. Davatchi F, Shahram F, Chams-Davatchi C, Shams H, Nadji A, Akhlaghi M, et al. Behcet's disease in Iran: analysis of 6500 cases. Int J Rheum Dis 2010;13:367–73.
4. Shanmugam VK, Fernandez S, Evans K, McNish S, Banerjee AN, Couch KS, et al. Postoperative wound dehiscence: Predictors and associations. Wound Rep Reg 2015;23:184–90.
5. Van Ramshorst GH, Salu NE, Bax NMA, Hop WCJ, van Heurn E, Aronson DC, et al. Risk Factors for Abdominal Wound Dehiscence in Children: A Case-Control Study. World J Surg 2009;33:1509–13.