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Case Report

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Cutaneous Crohn's Disease as the First Presentation: A Natal Cleft Lesion in a Middle-Aged Woman

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ABSTRACT

Background: Cutaneous Crohn's disease, particularly as an initial presentation without gastrointestinal involvement, is exceedingly rare. Natal cleft lesions can mimic other inflammatory or infectious conditions, delaying diagnosis.

Case Presentation: A 43-year-old woman presented with a perianal fistula and a friable natal cleft lesion. Endoscopic and radiologic investigations were unremarkable. Histopathology of the lesion revealed non-caseating granulomas and plasma cell infiltration, consistent with cutaneous Crohn's disease. Infectious causes were excluded. The patient responded well to metronidazole and anti-TNF therapy.

Conclusion: This case underscores the need for heightened clinical suspicion of metastatic Crohn's disease in atypical cutaneous presentations, especially in the natal cleft region.

Keywords: Cutaneous Crohn's Disease, Natal Cleft, Perianal Fistula, Granulomatous Dermatitis, Anti-TNF Therapy

Introduction

Crohn's disease is a chronic granulomatous inflammatory condition that primarily affects the gastrointestinal tract. Extraintestinal manifestations, including cutaneous involvement, are well-documented but relatively rare. Metastatic Crohn's disease refers to granulomatous skin lesions that occur at sites distant from the gastrointestinal tract. Natal cleft involvement is an uncommon presentation and may be the first clinical sign of Crohn's disease.

Case Presentation

A 43-year-old female was referred to gastroenterology for evaluation of a newly diagnosed perianal fistula.

Investigations revealed:

- Upper and lower endoscopies: unremarkable
- Terminal ileum and random colon biopsies: normal
- Magnetic Resonance Enterography (MRE): unremarkable
- A 6 cm friable, protruding lesion was noted at the 12 o'clock position from the anal verge near the sacral bone

Biopsy findings:

- Focal ulceration with adjacent marked epidermal hyperplasia
- Superficial epidermis showing sheets of plasma cells and scattered non-caseating granulomas
- Granulomas extended to deep dermis, particularly around adnexal structures, with lymphoplasmacytic infiltration
- Special stains (GMS, Ziehl-Neelsen, Giemsa, Warthin-Starry) and microbial cultures: negative
- STD swabs and serology for HIV, Hepatitis B, and Hepatitis C: negative

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A diagnosis of cutaneous Crohn's disease was made based on clinical, histopathologic, and negative infectious findings.

Treatment:
- Metronidazole for 3 months
- Initiation of anti-TNF therapy
- Follow-up at 3 months showed significant improvement in the excision site and decreased drainage from the fistula

Discussion

Metastatic Crohn's disease (MCD) is an uncommon dermatologic manifestation of Crohn's disease, involving granulomatous inflammation of the skin distant from the gastrointestinal tract. It most commonly presents on the legs, genitalia, and intertriginous areas, but natal cleft involvement is rarely reported.

The histological hallmark of MCD is the presence of sterile, non-caseating granulomas within the dermis, frequently accompanied by plasma cells and lymphocytic infiltration. These findings are similar to those seen in gastrointestinal Crohn's disease, although they occur without direct mucosal involvement.

Differential diagnoses include infections (e.g., tuberculosis, fungal diseases), sarcoidosis, hidradenitis suppurativa, and neoplasms. Diagnosis is often delayed due to nonspecific clinical presentations and overlapping features with these conditions.

Management includes topical or systemic corticosteroids, immunosuppressants (e.g., azathioprine), antibiotics such as metronidazole, and biologics like anti-TNF agents. In this case, a combination of metronidazole and anti-TNF therapy led to a marked improvement, consistent with current literature.

Literature Review

A review of the literature reveals limited reports on natal cleft involvement in cutaneous Crohn's disease. Palamaras et al. and Tolkachjov et al. describe cutaneous Crohn's as a rare but significant extraintestinal manifestation [1,2]. In a study of 15 patients with MCD by Tolkachjov et al., the genital region was the most commonly affected, but involvement of the intergluteal cleft was infrequent. The rarity of these presentations often leads to misdiagnosis or delayed treatment.

The presence of non-caseating granulomas and exclusion of infectious etiologies are essential for diagnosis. Special stains and cultures help rule out differential diagnoses [3]. Therapeutic success has been reported with systemic immunosuppressants and biologic therapy, particularly anti-TNF agents [4].

Conclusion

This case illustrates a rare presentation of cutaneous Crohn's disease involving the natal cleft as the first manifestation in a patient with an otherwise unremarkable GI workup. High clinical suspicion, appropriate histopathological evaluation, and exclusion of infectious causes are crucial. Early treatment with antibiotics and biologics can lead to substantial improvement.

Patient Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying clinical information.

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