SHORT REPORT

Surgical management of gluteal metastatic cutaneous Crohn's disease

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Abstract

Metastatic cutaneous Crohn's disease is a rare entity first described by McCallum et al. in 1976. It is diagnosed when histologically characteristic granulomata are seen at a site not contiguous with inflammatory disease in the gastrointestinal tract. We herein report presentation, diagnosis and management of a 28 year old lady with disabling, symptomatic cutaneous Crohn's of the buttocks and natal cleft refractory to Infliximab therapy. To the best of our knowledge only four other adult cases have been reported in the literature of metastatic cutaneous Crohn's disease of the buttock area distant from a flexure or area of skin apposition. The differential diagnosis in this case was Hidradenitis Suppurativa. A good cosmetic result and excellent symptom control were achieved with extensive debridement, wide local excision, vacuum assisted closure and delayed skin grafting.

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1. Introduction

Metastatic Crohn's disease is diagnosed when the histologically characteristic granulomatous inflammation of Crohn's disease occurs at a site not contiguous with disease in the gastrointestinal tract. Such a complication is extremely rare.1,2 In the literature it has been most commonly reported in skin flexures and sites of apposition.3–5 Regardless of the site, the majority of reported cases illustrate the relative ineffectiveness of medical treatment in managing moderate to severe disease.6 We herein report a case of cutaneous metastatic Crohn's disease in the gluteal region which was managed with aggressive surgical debridement, vacuum assisted closure and delayed skin grafting.

2. Case report

A 28-year-old female with a three year history of ileocolonic Crohn's disease presented with pain and discharge from the perianal and gluteal regions. Problems in the gluteal region and perineum had been a prominent feature since time of
diagnosis. The early management of her Crohn’s disease included steroids, immune modulators and several courses of antimicrobial therapy for perianal abscesses. Monthly infusions of Infliximab at a dose of 5 mg/kg were used for a two year period but had been stopped just prior to this admission due to extensive cellulitis of the gluteal region. During the six months prior to referral to our service, she had been experiencing considerable pain, pruritus and persistent pustular ooze from the buttock and natal cleft. This was not considered to be a reaction to Infliximab as mild symptoms had predated treatment with this agent. She was not symptomatic from the point of view of terminal ileal Crohn’s disease. Response to several outpatient courses of steroids, immune modulators and antimicrobial agents was limited and transient at best. Quality of life was severely affected by these symptoms and they became troublesome enough to necessitate inpatient admission. She was afebrile with a C Reactive protein elevated at 56 and a white cell count of 8.6. On examination extensive areas of erythematous nodular pustulant excoriation were present on both buttocks and in the natal cleft (Fig. 1). Multiple malodorous discharging sinuses were also present. Cultures of discharge from the affected area grew Coagulase Negative Staphlococcus, mixed anaerobes and Staphlococcus aureus, thought by microbiology colleagues to be of uncertain significance and potentially commensal. The anal canal had no evidence of Crohn’s disease and perianal fistulae were excluded by consultant radiologist review of Magnetic Resonance Imaging. Thus this pathological process was separate to the anal canal. The differential diagnosis was cutaneous metastatic Crohn’s disease versus Hidradenitis Suppurativa. Metastatic Cutaneous Crohn’s has previously been reported in a symmetrical distribution.\(^7\) The absence of necrotic tissue and insidious nature of onset precluded suspicion of Fournier’s gangrene, a rapidly progressive condition, rare in female patients.\(^8\) Pyoderma gangrenosum was also excluded as there was no characteristic ulceration, pain was not the predominant presenting feature and the lesions were not present in the characteristic lower limb or hand distribution. Understanding the inherent risks and after consultation with the Plastic Surgery service, the patient was keen to proceed with surgical management due to the disabling and persistent nature of symptoms. Wide local excision of the diseased areas with extensive debridement of all chronic sinuses and abscesses was performed (Fig. 2). A Vacuum Assisted Closure (VAC) dressing was applied by the Plastic Surgery service and a Foley catheter and faecal management system were inserted (Fig. 3). One week later, the VAC dressing was changed and the wound inspected under anaesthesia. A split thickness skin graft was then applied from a donor site on the left thigh. The skin graft took successfully. The wound was observed to be clean with the commencement of satisfactory healing evidenced by formation of granulation tissue. The VAC dressing was reapplied and the faecal management system was removed. The wound was dressed daily and exposed to air

Figure 1  Erythematous nodular excoriation of the gluteal region and natal cleft.

Figure 2  Wide local excision and extensive debridement.

Figure 3  Vacuum Assisted Closure (VAC) dressing and faecal management system in situ.
for a period of one hour per day. Final histology of the skin and underlying subcutaneous tissue revealed florid chronic inflammation with associated deep sinus tracts. Characteristic non-caseating epithelioid granulomata were seen resulting in a diagnosis of cutaneous metastatic Crohn’s disease (Fig. 4). Rectal biopsies taken at the time of colonoscopy revealed rectal mucosa within normal limits. Infliximab was recommenced two weeks post operatively and the patient was subsequently discharged home. On follow-up, good healing was observed (Fig. 5). One year postoperatively the patient is free from troublesome symptoms, on Infliximab and has not required any further surgical intervention.

3. Discussion

Many types of cutaneous disorders are exhibited by patients with Crohn’s disease. These may include erythema nodosum, pyoderma gangrenosum, erythema multiforme and Hidradenitis Suppurativa. These cutaneous pathologies may exist in many diseases other than Crohn’s, however histological evidence of characteristic non-caseating granulomas such as those seen in this case, are confined to Crohn’s disease.

While Parks et al. described first described cutaneous Crohn’s disease in 1965, the concept of “metastatic” Crohn’s appeared in the literature in 1976 when McCallum et al. recognized the existence of non-contiguous cutaneous granulomata histopathologically pathognomonic for Crohn’s Disease. Diagnosis and management of this manifestation of Crohn’s can be challenging and it is easily missed in the absence of classical gastrointestinal symptoms. In our case, a diagnosis of Hidradenitis Suppurativa (HS) was also considered. HS is a chronic skin infection which often presents as an acute abscess and is often complicated by sinus formation and purulent discharge. It is primarily an infection of apocrine sweat glands and due to the high concentration of these glands in the perineal area disease at this site is common. HS was considered a potential diagnosis due to the fact that Crohn’s disease and HS frequently co-exist. A review of HS patients treated at the Cleveland Clinic over a ten year period revealed that 39% had a concurrent diagnosis of Crohn’s disease. This frequent dual pathology has the potential to confuse the diagnosis of cutaneous Crohn’s disease.

In our case, due to the disabling nature of symptoms and failure of medical therapy, surgical intervention was necessary regardless of diagnosis. Due to the relative rarity of the condition, no high grade evidence exists as to optimum management of cutaneous Crohn’s disease. On review of the literature, case reports and small series appear to reveal a trend towards inefficacy of medical treatment. Some cases of a favourable response to Infliximab therapy, however, have been reported. In our case, two years of monthly treatment with Infliximab failed to achieve any symptomatic control of clinical improvement. Indeed, in severe cases such as this one, successful medical therapy appears to be the exception rather than the rule. Williams et al. reported use of surgical debridement of areas of severe perineal metastatic cutaneous Crohn’s disease in five patients, all of whom, as in the case reported, had failed to improve after a variety of medical treatments. Four out of five patients had a good outcome with complete resolution of symptoms and satisfactory cosmetic results.

The distribution of disease in this case was unusual. The skin involved in cutaneous Crohn’s is generally affected by direct extension (perianal ulceration, fistulation or scar post surgery). Even in non-contiguous or metastatic cutaneous Crohn’s disease, the majority of reported cases have occurred in areas of skin apposition with flexures (retroauricular, vulva, submammary, groin). While an area of skin apposition (natal cleft) was also involved in our case, the skin of the left and right buttocks was extensively involved. To the best of our knowledge, only four cases of metastatic cutaneous...
Crohn’s of the buttock skin distant from flexures or areas of skin apposition have previously been reported in adults in the literature.2,17–19 This rare diagnosis of cutaneous metastatic Crohn’s disease affecting the buttocks and natal cleft was successfully managed using wide local excision, vacuum assisted closure and delayed skin grafting. A good cosmetic result and symptomatic control were achieved.

References