Intergluteal Contour Deformity in Hidradenitis Suppurativa

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Hidradenitis suppurativa (HS) is an uncommon chronic disorder with a significantly negative impact on the quality of life of affected patients. HS is typically localized on the axillary and inguinal folds, less commonly on the breast and at perianal/perineal areas, whereas gluteal involvement is generally considered atypical (1). Extensive HS involvement may interfere with everyday activities, such as walking or hugging, due to pain in the groin or axillary area, or sitting in case of gluteal localization.

Current clinical classifications of HS are not specific and present serious limitations (2). Recently, a new classification, considering 5 phenotypes that include regular, frictional furuncle, scarring folliculitis, conglobata, syndromic and ectopic types, has been proposed (2). Frictional furuncle and scarring folliculitis types share gluteal involvement.

Based on the observation of intergluteal contour deformity in some of our patients with HS and in cases from the literature (1, 3, 4), we considered whether this finding may represent a common sign of HS and if it may have a correlation with specific HS localization and disease severity.

CASE REPORTS

A total of 74 patients with HS (37 males (M)/37 females (F)) were evaluated from November 2016 to February 2018. Twenty-five (34.2%; 13 M/12 F) showed involvement of the gluteal and/or perianal/perineal areas, respectively, in 14 (7 M/7 F) and 17 (8 M/9 F) patients (6 patients showed both localizations). Of these, 8 (10.8%; 3 M/5 F) showed a deformity of the intergluteal contour. This consisted of an irregularly bending serpiginous outline of the intergluteal fold showing contractures and skin induration corresponding to multiple and opposed retractive scars. In some cases the skin surrounding the contour deformity appeared hyperchromic (**Fig. 1**). All 8 patients clinically fitted into frictional furuncle type from van der Zee & Jemec (2) recent classification and their HS severity was rated Hurley II–III; disease mean duration was 12.5 years (range 3–25 years). None of them showed a positive family history for HS, but 6 (75%) had a past history of acne. Obesity was present in 2 cases (25%). All patients reported a sedentary lifestyle similar to those without gluteal involvement matched for age, sex, disease severity and body weight.

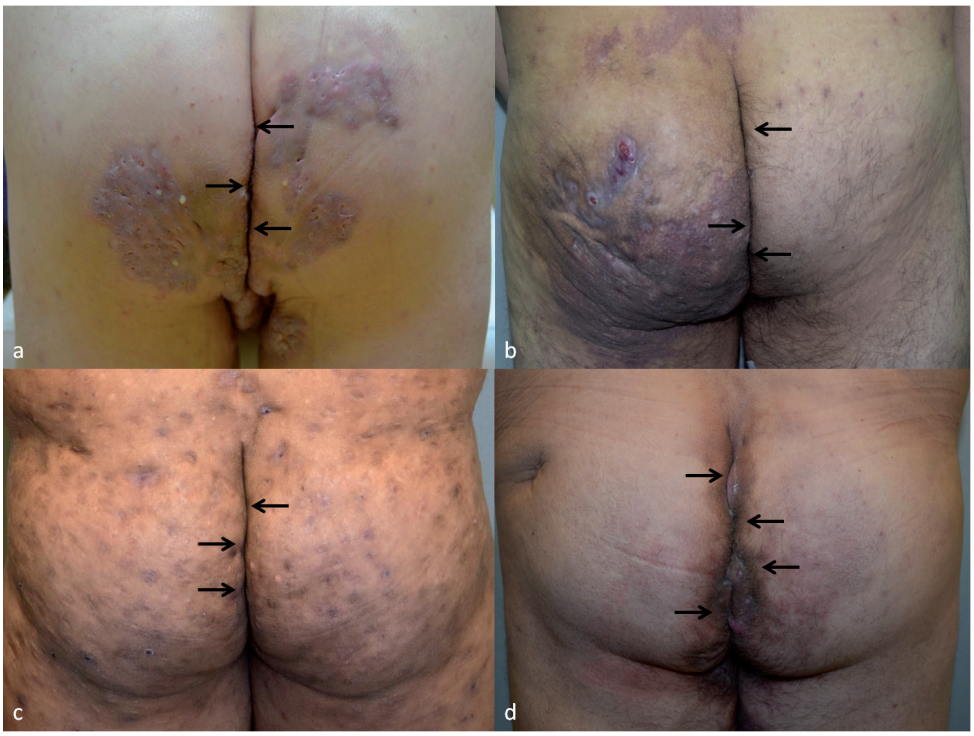

Fig. 1. Multiple intergluteal contour deformity (arrows) resulting from dermal contractures and induration in patients with Hurley II–III long-lasting disease.

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DISCUSSION

Distortion of tissue architecture of anatomical areas such as the umbilicus and the groin has been reported, so far not regarding the intergluteal fold (3). In our series of 74 patients with HS, we have observed intergluteal contour deformity in 8 out of 25 cases showing gluteal and/or perianal/perineal involvement and we believe that this phenomenon should be regarded as the result of repeated healing processes, probably due to chronic and relapsing inflammatory events associated with localized deep subcutaneous abscesses (5). Chronic tissue repair followed by the onset of new underlying inflammatory phenomena may eventually result in fibrotic tissue and opposite retracting scars. A triggering role of mechanical stress (friction) also seems likely (6), despite in our cases obesity and occupation did not play a role. The prevalence of gluteal and perianal/perineal involvement in our study was respectively 76% and 61.5% in males and 92% and 75% in females. These data are higher compared with a previous study (50% and 56% in males and 25% and 33% in females for buttocks and perianal and perineal localizations, respectively), but confirm how the gluteal and the perianal/perineal involvement is more common in males (1). Of note, in that study the overall M:F ratio was 1:4 compared with 1:1 in our cohort and 2:3 from a recent Italian survey (7).

The differential diagnosis of intergluteal contour deformity includes complications by inflamed pilonidal cyst (PC) and perianal abscesses management. PC is probably an under-reported condition that may have many similarities with HS, including relapsing inflammation. It may occur isolated or in association with HS (one-third of patients) or with follicular occlusion syndromes (8, 9). PC is usually found in the sacro-coccygeal region and is frequently associated with obesity, trauma, hirsutism and hyperhidrosis (10). With time, it may be followed by the formation of a sinus tract and repeated inflammatory processes followed by scar formation may cause an intergluteal fold deformity generally limited to the upper portion of the fold. A similar deformity may be observed following surgical management of PC (11). Idiopathic perianal abscesses are generally located in the perianal region, in the inter-sphincteric space, and are classified as superficial or deep. Approximately 90% of perianal abscesses are the result of infection of the cryptoglobular glands. Perianal tracts are generally associated with advanced disease. Perianal abscess may also represent a complication of Crohn's disease (CD) (12) and since an association between CD and HS has been reported (13), cases of HS exclusively affecting the perianal region may be difficult to differentiate from cutaneous CD. The management of recurrent perianal abscess may cause retracting scar which may result in intergluteal contour deformity limited, however, to the lower portion of the fold.

In conclusion, although the involvement of the gluteal and perianal/perineal area – whose real incidence is probably underestimated – has been repeatedly reported, not enough emphasis has been dedicated to intergluteal contour deformity, which is not seen in all patients, but in those with long-standing severe disease; in our series, all the affected patients were classified Hurley stage II–III and the mean disease duration was 12.5 years. The recognition of this simple but evocative sign may enhance the diagnostic HS capability of non-HS specialists, residents in dermatology/surgery/proctology, young practitioners or GPs with special interest in dermatology and all physicians who are not familiar with HS.

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