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De Novo Ectopic Hidradenitis Suppurativa Masquerading as Post Operative Wound Infection: Case Report and Literature Review

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Abstract: **Case Report**

Verneuil's disease is a chronic inflammatory condition characterized by recurrent, deep, painful nodules and abscesses in intertriginous areas. We present the case of a 23-year-old female patient who developed de novo ectopic Verneuil's disease following gluteal lipofilling. We also conducted a literature review of post surgical cases of ectopic de Novo cases. A 23-year-old patient with no prior medical history presented 20 days after gluteal lipofilling with a painful, inflamed subcutaneous mass on the outer aspect of the right buttock, accompanied by fever. Following diagnostic evaluation, we confirmed the diagnosis of Verneuil's disease based on histopathological findings. Our literature review identified eight cases of postoperative Verneuil's disease, particularly at amputation stumps. Only one patient developed de novo lesions after undergoing plastic surgery. The postoperative period may serve as a precursor to ectopic Verneuil's disease in some patients, given the inflammatory context.

Keywords: Verneuil's Disease, Hidradenitis Suppurativa, Gluteal Lipofilling, Ectopic Hidradenitis, Postoperative Complications.

INTRODUCTION

Hidradenitis suppurativa (HS) also known as acne inversa is a chronic inflammatory condition characterized by deep-seated painful nodules and abscesses occurring in recurrent episodes in intertriginous areas [1].

Clinically, HS can mimic infection, especially in atypical locations in postoperative settings. We present a case of de novo lesions after surgery. To the best of our knowledge, based on a literature review, this is the first case describing De novo ectopic hidradenitis suppurativa following fat grafting.

Methods

Most of the patients were obese patients who had lesions on their amputation stump. We conducted a search in order to identify case reports and articles about ectopic HS in a post operative setting. The search was made in English-language using PubMed using MeSH with the following keywords (Hidradenitis suppurativa) AND (Ectopic), (Hidradenitis suppurativa) AND (Atypical), (Hidradenitis suppurativa) AND (Unusual), (Hidradenitis suppurativa) AND (De novo), (Acne inversa) AND (Ectopic), (Acne inversa) AND (Atypical), (Acne inversa) AND (Unusual), (Acne inversa) AND (De novo). The search yielded 281 results.

Inclusion Criteria

The study only includes articles with cases of patients with postoperative HS lesions in the surgical site, both in patients with known HS and patients with no history of HS.

After excluding duplicates, and cases that didn't involve a surgical site, we found seven eligible contributions reporting on a total eight patients.

Case Presentation

A 23 years old patient, with no medical history was operated in our ward for liposuction of the abdomen flanks and back with concomitant fat graft of the buttocks; we grafted 1560 ml per buttock. She was then discharged 2 days later without issues, on antibiotics, low molecular weight heparin and antalgics.

20 days after her operation, she presented with a fever. On presentation, she was alert and fully responsive, normotensive with a 39° fever and tachycardia at 110 bpm. On clinical examination, inspection of the buttocks revealed a painful erythematous plaque along with an inflammatory subcutaneous 3 cm nodule on the external part of the right buttock (On the 8th subunit of the buttock). No further anomalies were found at this stage.

Initial management included antipyretics, topical anti-septic and broad spectrum antibiotics (Cefalosporin + Metronidazole) after blood cultures were started.

At this stage, we suspected septic cytosteatonecrosis. Her lab work showed a hypochromic anemia at 8.9 g/l and a normal white blood cell count.

C-reactive protein (CRP) was at 70.91 mg/l. The patient underwent a local drainage of the abscess under local anesthesia. About 200 ml of sanguineous purulent exudate was removed and then sent to the lab for bacteriological examination. The cavity was then packed with gauze.

On her second day of admission, her vitals became normal while the right buttock remained inflamed and tender. During her dressing change, more purulent exudate came out. After consulting with internal medicine, we decided to order an MRI of the area to assess the extent of the collection and study the underlying tissues. The exam came back negative, showing no collection and no damage to the fasciomuscular tissues. It did however show inflammation of the fatty tissue.

We decided to keep the patient on general antibiotics and daily dressing change. We received negative blood cultures and later a negative culture of the exudate, respectively on days 4 and 5.

The patient’s vitals remained stable and the dressings were clean but the 8th subunit of the right buttock remained inflamed, painful to the touch and indurated. A biopsy of the cutaneous and fatty tissues was performed. Histopathologic findings showed acute and chronic inflammation in the dermis, including chronic periadnexal inflammation, focal dermal granulation tissue, and fibrosis. The patient was then diagnosed with hidradenitis suppurativa (HS)

The patient was discharged under erythromycin 3% gel and Amoxicillin-Clavulonic Acid for a week. She was monitored until remission; she was then put on Doxycycline and scheduled for a checkup after six months.

At said check up, the patient was still in remission, with no local relapse. The general assessment showed no papules nor nodules elsewhere.

She however, complained of an esthetic concern with the look of the 8th subunit of her right buttock. The examination

shows a subcutaneous depression with indurated skin (Figure 1).

Figure 1: Aspect before secondary surgery

We suggested a second local fat graft to help with this issue. After explaining the benefits and risks of this procedure, we got the patient's consent. She was then scheduled for a secondary liposuction of the abdomen, flank and back to harvest enough fat to graft on her right buttock

By the end of the liposuction, we made subcisions in the dermis of the retracted area. The harvested fat was first washed with Xylocaine and Gentamicine and then filtered before we injected 600 ml of it in the 8th and 5th subunits of the right buttock (Figure 2).

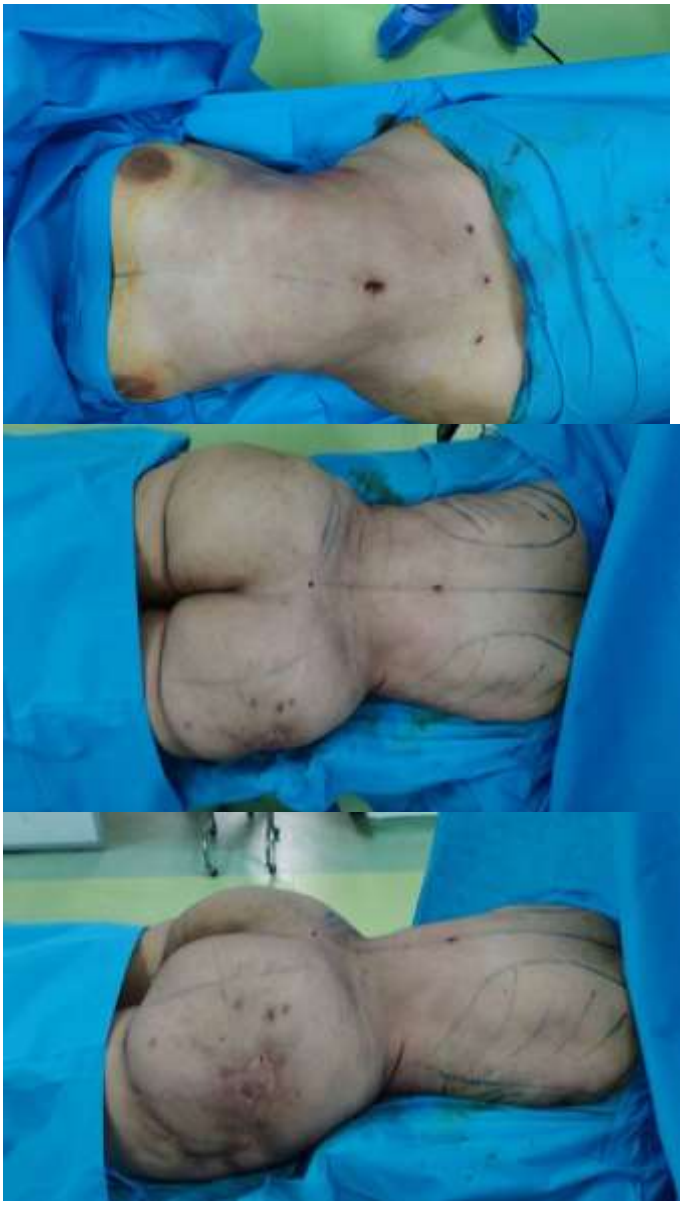

Figure 2: Aspect after second round of fat grafting

DISCUSSION

Hidradenitis suppurativa is characterized by an insidious onset of painful nodules and abscesses that progress to suppuration, fistulization, and scarring in anatomical regions with apocrine glands. The disease progresses in recurrent and painful flare-ups [2].

It was initially believed that HS was caused by an inflammation of apocrine glands but the pathogenesis of Hidradenitis suppurativa complex, and the literature is leaning towards the involvement of hair follicles [3].

While genetic and immunity factors are known contributors to the development of HS, multiple studies suggest that the influence of extrinsic factors also contribute to the onset of HS; mechanical stress in particular. The skin in certain areas of the body can be exposed to many types of stress including friction, pinching, pressure and pulling. The buttock area is prone to friction, which is the force resisting motion between skin and the outer surfaces.

In our case, not only was the buttock area prone to friction, it was under added pressure and local trauma after lipografting. The synergy of all these forces may have been the cause for the development of HS [4].

In fact, some studies suggest that continuous stress including pressure and friction, may lead to follicular occlusion and dilatation [5] and increased cutaneous micro-injuries, and a local immunodeficiency resulting in the release of cellular damage-associated molecular patterns prompting changes in skin microbiome which is both a cause preceding the HS lesions and an effect. All these factors lead to the creation of biofilm on the debris and bacterial colonization [6] subsequently causing recurrent infection and abscesses [7].

Research suggests that changes in the composition of the local microbiome may precede and contribute to development of lesions, rather than altering the local microbiome via secondary colonisation. The cutaneous microbiome is also influenced by host immunity; alterations in the local microbiome have been demonstrated [8, 9]

This explains both the onset de novo lesions in patients who didn't have a diagnosis of HS only in areas that were operated on and the koebnerization in the latter areas in patients with known HS. Our review of literature found eight cases of post operative HS (Table 1). Most of the patients were middle aged, obese and had HS lesions on their amputation stumps. Only one patient had de novo lesions after undergoing plastic surgery.

The clinical challenge of patients presenting with ectopic HS, especially de novo ectopic HS, is recognizing the diagnosis itself. Delays in proper treatment may occur in patients with de novo ectopic HS, given that these solitary lesions may be mistaken for postoperative infections. The treatment for ectopic HS is the same as the treatment of HS using an array of means such as topicals drugs, oral antibiotics, biotherapy, laser, and surgical deroofing or excision [10].

	De Novo	**Koebner**
Men	**Kluger *et al*.,** [11] Age:55-year-old, Diabetes type 2, hypertension, hypercholesterolemia, BMI 28.9, and Charcot-Marie-Tooth disease. Stump amputation (neurological disease) **De Winter *et al*.,** [12] Age: 44 years old, BMI = 31.3 Diabetes type II, hypopituitarism Amputation stump	**Kluger *et al*.,** [11] Age: 41 year-old, smoker, obese, 1st-degree relative with HS Post traumatic amputation stump **Fleischer *et al*.,** [13] 55-year-old, Crohn's Peristomal orifice
Women	**Darch *et al*.**, [14] Age: 33 years old C-section **Henderson *et al*.,** [15] Age: 24 years old Post traumatic amputation stump **Alhusain *et al*.,** [16] 35-year-old, psoriasis Submental liposuction with vaser	**Agiasofitou *et al*.,** [17] Age: 34 years old, smoker, obese C-section

CONCLUSION

Hidradenitis suppurativa is an inflammatory disease at its core which makes the postoperative period a precursor for ectopic lesions in patients with HS and disease naive ones.More studies are needed to understand the pathophysiology and potential preventive means to avoid the development of this condition.

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