CASE REPORT

Bullosis diabeticorum in a morbidly obese woman in Haiti

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ABSTRACT

Bullosis diabeticorum is a rare skin complication of diabetes mellitus, characterized by sudden onset bullous lesions with no history of trauma. It predominantly affects men and has an acral asymmetric presentation. Here, we report a case of bullous disease in a diabetic patient with morbid obesity, the first described in Haiti. A 40-year-old woman, with a strong history of diabetic for five years, poorly controlled and morbidly obese presented to our emergency for bullosis lesions in her limb. She had a prior presentation about two years ago and, approximately a week before this actual presentation, the same symptomatology occurred suddenly without any trauma. After evaluation and screening, the diagnosis of bullosis diabeticorum was kept. She was initially treated with antibiotics due to signs of superinfection. However, as soon as the symptoms improved, antibiotics were discontinued. An antiseptic lotion and topical antibiotic, neomycin, were used along with daily dressings. About a week after, her blood glucose came to control, signs of infection disappeared as did the bullous lesions and surgical evaluation was performed to ensure proper wound evolution. She was educated by a nutritionist, and our team emphasized the importance of regular follow-up at the hospital. Diabetic bullous disease is very rare and easy to confuse with other diabetic skin complications. A good clinical history is essential to make the diagnosis, and management requires good therapeutic education to avoid the burdensome complications of diabetes.

KEYWORDS: bullosis diabeticorum; diabetic skin complication; diabetic foot; diabetes management

INTRODUCTION

Bullosis diabeticorum is a rare skin complication of diabetes mellitus [1-4] and is characterized by the spontaneous appearance of bullous, non-painful, and non-inflammatory lesions of unknown etiology [2,5]. With a prevalence of 0.16–0.5%, it predominantly affects men and has an acral and distal asymmetric presentation [1,2,6]. At the present time, there is no known etiology of this disease; hypotheses are in favor of a multifactorial etiology whose predominant element is poor control of glycemia. In Haiti, non-communicable diseases, including diabetes, are on the rise and complications are not trivial. To our knowledge this is the first of case of bullosis diabeticorum described in the country. The aim of this study is to present the clinical and management of bullosis diabeticorum in our limited context as a rural environment.

CASE PRESENTATION

The patient was a 40-year-old woman, known with diabetes for five years, poor adherence to metformin (500 mg daily) therapy, and was not followed up by a healthcare professional. She was morbidly obese (BMI, 42 kg/m^2) and had a disability in her lower right limb, which limited her everyday movement. She presented to the emergency room (ER) with lower left limb enlargement, pain, and erythema—of which she had a history.

The patient had a strong family history of diabetes mellitus. Her history included surgery on her right femur following a fracture sustained by a road traffic injury in 2019. She is on her second hospitalization for the current symptomatology.

A chart review revealed that she had a prior presentation about two years ago, with a symptomatology that would have started with the appearance of bullous lesions on the anterior leg that progressed to the dorsum of the foot (Figure 1), followed by a sudden fever, concomitant with the increasing volume of the limb, and a sensation of heat. She came to the hospital for management and was admitted for approximately five weeks with near-complete resolution. Approximately a week before this actual presentation, the same symptomatology occurred suddenly, with bullous lesions, followed by an increase in the volume of the limb and redness without any trauma. Given the progression of the bullous lesions and the onset of pain, she decided to return to the hospital after spending about five days in an outside hospital (Figure 2).

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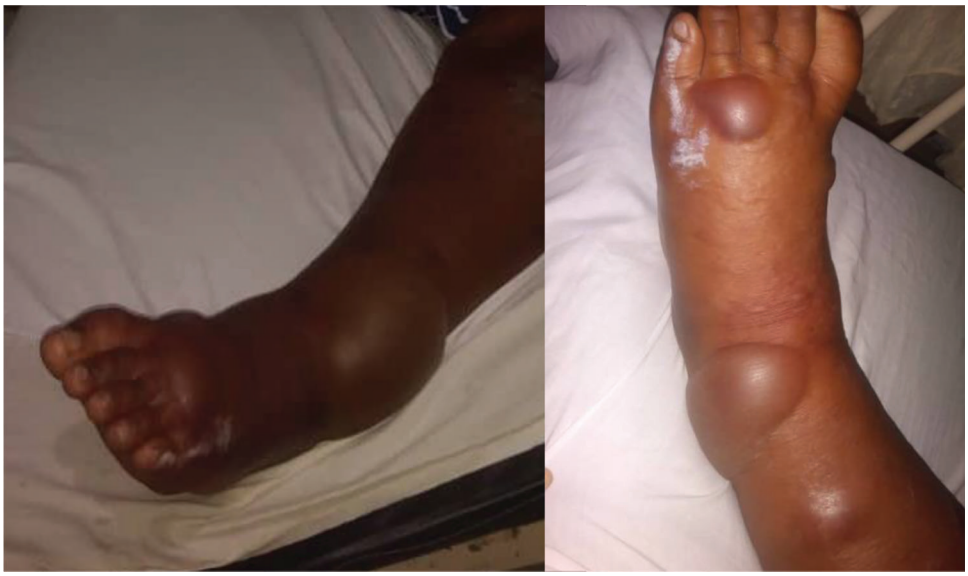

Fig. 1. Presentation of bullous lesions two years after the first hospitalization.

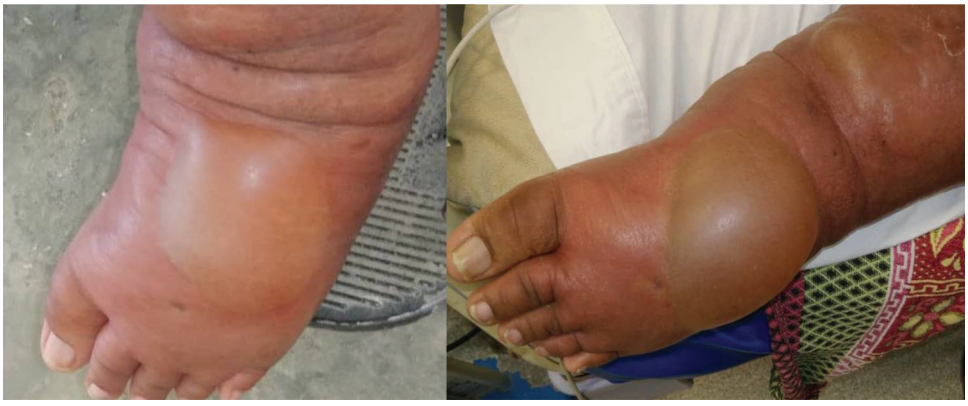

Fig. 2. Presentation of the lesions during the second hospitalization.

Upon arrival, her vital signs were as follows: blood pressure (BP), 140/95 mmHg; heart rate (HR), 107 beats/min; oxygen saturation (SpO_2), 94% on room air; respiratory rate (RR), 20 cycles/min; and blood glucose, 247 mg/dl. The findings of the first ER evaluation were within normal limits, except for the examination of the lower limbs, which revealed marked asymmetry with shortening of the right lower limb and increased volume of the left limb, with marked redness and bullous lesions localized on the anterior left leg and the dorsum of the foot (Figure 2).

She was transferred to the internal medicine service unit for further management. Upon arrival, her vitals were as follows: BP, 141/79 mm Hg; HR, 104 beats/min; RR, 24 cycles/min, SpO_2 93% on room air; temperature, 37.8°C, and blood glucose, 465 mg/dl. Physical examination of the limb revealed enlargement with bullous lesions on the inner thigh and anterior leg. These were accompanied by oozing lesions on the dorsum of the left foot, mild pain on palpation, high temperature, erythema, decreased mobility, and pulse that was difficult to evaluate. The results of the blood sample (taken in the emergency department) for laboratory tests received during his admission to internal medicine are shown in Table 1. Based on these findings, the following diagnoses were made: bullosis diabeticorum complicated by cellulitis vs erysipelas associated with diabetic decompensation and uncontrolled hypertension in a morbidly obese patient.

Table 1. Initial patient's screening results at the admission.

	Patient values	Normal range
Hb (g/dl)	11.5	12-15
WBC (count per mm3)	12,600	4,000-11,000
K (mmol/l)	3.93	3.5-5.5
Na (mmol/l)	144	135-145
Ca (mmol/l)	2.25	2.13-2.55
Creatinine (mg/dl)	1.1	0.5-1.1
Urea (mg/dl)	238	10-30
Cholesterol (mg/dl)	133	120-200
Triglyceride (mg/dl)	127	30-150
LDL (mg/dl)	103.7	10-130
Sedimentation rate (mm/h)	35	0-29
CRP (mg/l)	192	9-10

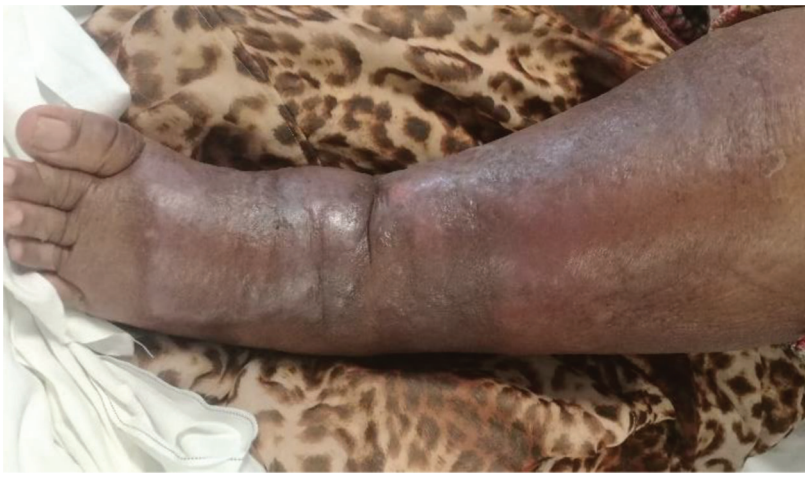

Fig. 3. Disappearance of bullous lesions after two weeks of management.

She was rehydrated with 0.9% NaCl solution at a rate of 2 L afloat and then 1 L of 0.9% NaCl + 30 mEq of Kcl at 40 drops per min. This was followed by insulin therapy of 10 IU insulin R quarter in die (QID) for blood glucose level >200 mg/dl, replaced about 24 hours later with Neutral Protamine Hagedorn (NPH) insulin at 0.8 UI/Kg and insulin R per the blood glucose scale; then ceftriaxone 1 g IV q 12 h, replaced by clindamycin 600 mg IV bis in die (BID), dicloxacillin 500 mg PO QID, enalapril 10 mg PO BID, amlodipine 5 mg PO q day, and prophylactic heparin 5000 U s/c BID. Wound care was performed using an antiseptic solution (bacterisol) BID and a topical antibiotic (neomycin).

About a week after her blood glucose came to control, signs of infection disappeared (WBC:6,800, no fever), as did the bullous lesions (Figure 3). Antibiotics were discontinued, and surgical evaluation was performed to ensure proper wound evolution while the dressings and limb elevation continued. The patient was educated by a nutritionist on managing her diabetes and obesity, and our team emphasized the importance of regular follow-up at the hospital.

DISCUSSION

It has been projected that 30% of patients with diabetes will have skin manifestations during their lifetime [5,7-9] Patients with type 1 diabetes often present with autoimmune manifestations, whereas those with type 2 diabetes present with skin infections [3]. Although discovered more than 80 years ago by Professor Kramer in 1930, it took about 37 years for this condition to be described in the medical literature by Cantwell and Martz under the name by which it is known today: bullosis diabeticorum [4-6,10,11]. It is a very rare skin condition, with only a hundred case descriptions in the medical literature and is characterized by sudden onset bullous lesions with no history of trauma, most of whom have poorly controlled type 2 diabetes [2,11,12]. Our patient was admitted for not being followed up by a healthcare professional for her diabetes, and unfortunately HBA1C was not available at the center, and the patient could not afford to have it done elsewhere. She self-medicated with metformin at irregular doses when she experienced discomfort, knowing that she was diabetic and obese. Kang et al. reported observations of Wilson disease over 11 years, leading to the conclusion that there is a correlation between bullosis diabeticorum and uncontrolled hyperglycemia in diabetic patients [2,13].

No etiology has been attributed to bullosis diabeticorum. A multifactorial etiology has been proposed, along with many theories and hypotheses [2,5,6,11]. One hypothesis is that the underlying cause is a long-term vascular compromise characterized by the thickening of the basal membrane of capillaries, leading to tissue hypoxia, which is responsible for the formation of bullae [2,6,11,14]. This hypothesis has been corroborated by the observation of hyaline deposits in the walls of capillaries on anatomopathological examination, thus supporting the possibility of microangiopathy and diabetic neuropathy as causes of bullosis diabeticorum. The frequent presence of peripheral neuropathy in patients with bullosis diabeticorum may also explain the preferential localization of lesions in the distal limbs [6,15]. The diagnosis is essentially clinical, [6,10] and the sudden appearance of bullae in a known diabetic patient, especially one with type 2 diabetes, in the acral and/or distal regions of the limbs, without any history of trauma, is generally sufficient to make the diagnosis. Complementary examinations, including biopsy of lesions and direct and indirect immunofluorescence, are usually inconclusive [1,6,14]. These screenings made it possible to exclude other cutaneous pathologies of inflammatory origin, such as bullous pemphigoid which is an autoimmune disease, necrobiosis lipoidica diabeticorum, and rubeosis faciei, often accompanied by retinopathy as it is a form of microangiopathy; We can also mention Eruptive xanthoma, with lesions generally on the buttocks, elbows and knees; Acrochordon, a benign pathology whose lesions are made up of reddish and blackish patches; and finally Diabetic dermopathy, which is characterized by well-demarcated lesions, with small depressions at pretibial level [6,9,16,17]. Regarding our patient, there were no findings indicative of nephropathy or microangiopathy, as her renal workup was within normal limits, as were the results of fundus examination and absence of others types of lesions, apart from bullae on her limbs. The only anomalies observed were elevated sedimentation rate (35 mm/h) and CRP level (192 mg/l).

The management of bullosis diabeticorum is conservative and the primary goal is to minimize the risk of complications and discomfort [2,6,12,18,19]. With repeated wet dressings and prevention of secondary infection, the duration of

treatment varies between two and six weeks. It is recommended that the blisters remain intact, as this protects against infection [2,6,10] and major complications (e.g., osteomyelitis) encountered in some cases [4,20]. Antibiotics are recommended in cases with signs of superinfection and analgesics are used to reduce the discomfort often felt by these patients [1,5]. Our patient was initially treated with antibiotics because she had signs of infection, such as fever and hyperleukocytosis. However, as soon as the symptoms improved, antibiotics were discontinued. An antiseptic lotion and topical antibiotic, neomycin, were used along with daily dressings to reduce the risk of superinfection. In addition to wound care, our patient was treated not only nutritionally for decompensated diabetes and uncontrolled hypertension but also medically to reduce the risk of thromboembolic events related to her morbid obesity and her being on bed rest, as medical nutrition therapy is essential in the management of diabetic patients for glycemic control, weight management, and the improvement of cardiovascular risks [21].

CONCLUSION

To the best of our knowledge, this is the first case of bullosis diabeticorum documented in Haiti, where diabetes is increasing. Bullosis diabeticorum is not without complications, even though it remains a benign pathology. It has been established that the risk of recurrence is very high, as is the occurrence of vascular and neurological complications, such as diabetic foot, which carries the risk of amputation and osteomyelitis.

Statement on Participant Consent

The images were taken with the written permission of the patient, who was also informed that a case publication will be conducted. Information regarding the identity of the patients was kept confidential.

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Disclosure

The authors declare no conflicts of interest related to this study.

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