

CASO CLÍNICO

Bullosis diabeticorum – una lesión poco común

Márcio Luís Duarte^{1,2*}, Thaiza Alonso Sima³, Lucas Ribeiro dos Santos⁴, André de Queiroz Pereira da Silva², José Luiz Masson de Almeida Prado².

Bullosis diabeticorum – an uncommon lesion

Resumen: *Introducción:* El bullosis diabeticorum forma parte del espectro de manifestaciones cutáneas de la diabetes mellitus, descrito por Kramer en 1930 y nombrado como bullosis diabeticorum por Cantwell y Martz. Es una enfermedad conocida, pero bastante rara (0.5 a 2% de la población diabética) siendo dos veces más común en hombres. Es una lesión espontánea y no está relacionada con trauma o causa fisiológica evidente, como infección, 3 que no causa dolor o se asocia a signos flogísticos. Se asocia principalmente a las extremidades, pudiendo ser una lesión única o múltiples lesiones. Puede ocurrir en la diabetes mellitus tipo 1 (DM1) y en el tipo 2. Las lesiones típicamente se curan espontáneamente de 2 a 6 semanas, pero pueden ocurrir en el mismo lugar nuevamente. El pronóstico es bueno, sin tratamiento específico necesario o seguimiento diagnóstico con biopsia. Se relata un caso de esta rara patología con documentación por resonancia magnética.

Palabras clave: Magnética, Pie diabético/diagnóstico, Resonancia, Vesícula/etiología, Vesícula/ patología.

Abstract: *Bullosis diabeticorum is part of the spectrum of cutaneous manifestations of diabetes mellitus, described by Kramer in 1930 and named bullosis diabeticorum by Cantwell and Martz. It is a known disease, but quite rare (0.5 to 2% of the diabetic population) and is two times more common in men. Bullosis diabeticorum is a spontaneous lesion and not related to trauma or obvious physiological cause, such as infection that does not cause pain or is associated with inflammatory signs. Occurs at the limbs and may be single or multiple. It can occur in diabetes mellitus type 1 (DM1) and type 2, usually in the advanced stages. The lesions heal spontaneously typically in 2 to 6 weeks, but they may happen again on the same site. The prognosis is good with no need of special treatment or further diagnosis with biopsy. We report a case of this rare complication with magnetic resonance imaging.*

Keywords: Blister/ etiology, Blister/pathology, Diabetic Foot/diagnosis, Magnetic Resonance

1. Mestre em Saúde Baseada em Evidências pela UNIFESP, São Paulo, São Paulo, Brasil.
2. Radiologista do setor de Radiologia Musculoesquelética da WEBIMAGEM, São Paulo, São Paulo, Brasil.
3. Dermatologista e Mestre pela UNIFESP, São Paulo, São Paulo, Brasil
4. Professor e Endocrinologista da Faculdade de Ciência Médicas de Santos, Santos, São Paulo, Brasil.

*Correspondencia: Márcio Luís Duarte / marcioluisduarte@gmail.com
WEBIMAGEM, Avenida Marquês de São Vicente 446, São Paulo, São Paulo, Brasil. CEP: 01139-020.

Apoyo financiero: no hay.

Introduction

Bullosis diabeticorum is part of the spectrum of cutaneous manifestations of diabetes mellitus^{1,2}, described by Kramer in 1930¹ and named bullosis diabeticorum by Cantwell and Martz³. It is a known disease, but quite rare (0,5 to 2% of the diabetic population)⁴, underdiagnosed in most cases², and is two times more common in men^{4,5}.

Bullosis diabeticorum is a spontaneous lesion, inflammatory and not related to trauma or obvious physiological cause, such as infection⁴ that does not cause pain or is associated with inflammatory signs^{4,6}. The lesions appear rapidly, mainly in the acral region and limbs, varying in size and, may be single or multiple^{2,4}. It can occur in diabetes mellitus type 1 (DM1) and type 2, usually in the advanced stages⁴.

We report a case of diabetes diabeticorum, a rare dermatologic lesion despite diabetes mellitus has a high incidence and prevalence worldwide, with a magnetic resonance imaging (MRI) study for evaluation of local infection and osteomyelitis.

Recibido: 12-08-2019
Aceptado: 22-10-2019

Case report

A 74-year-old male patient refers a bullous lesion in the right hallux for one day, painlessly, without signs of inflammation. Refers type 2 diabetes mellitus (DM2) for ten years treated with diet. Started treatment with januvia and metformin two years ago. The glycaemia, even with drug treatment, remains at a level between 140-180 mg/dl. Denies surgeries and trauma.

The blister developed spontaneously with a diameter of 2.5 cm in the right hallux, without phlogistic signs, no erythematous base, apparently composed of dark serous contents. It was tense with negative Nikolsky sign. The patient had no other bullous cutaneous lesions around. The blister burst and spontaneously resolved without complications and without specific medication two days after physical examination (Figure 1).

A MRI of the right foot was performed to evaluate the possibility of infection in surrounding tissues and osteomyelitis. The MRI demonstrated a skin blister in the plantar aspect of the distal phalanx of the hallux (Figure 2) and a discreet peripheral contrast enhancement, without any signs of infection.

With anamnesis, physical examination and MRI, it was concluded that the lesion corresponded to a bullosis diabeticorum.

Discussion

The pathogenesis of the disease is not clearly understood² – some theories relates metabolic disorders³ and also nephropathy⁷. In diabetic patients, there would be a greater tendency for blister forming mechanism for suction⁸. In other theories, microangiopathy explains the lesions with the histological finding of surface vessels hyalinosis⁹.

The liquid levels in the bullous would be explained by re-epithelialization, commonly observed in this type of injury⁴.



Figure 1: A) Blister in the right hallux, without phlogistic signs, no erythematous base, apparently composed of dark serous contents. (blue arrow). B) Ruptured blister without complications (blue arrow).

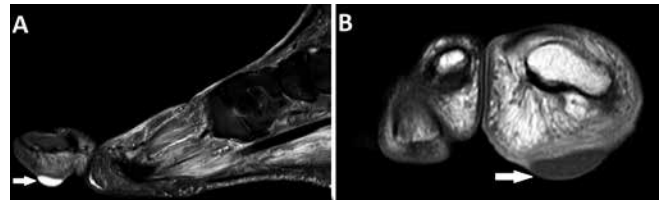


Figure 2: Sagittal T2 STIR MRI (A) and Coronal T1 weighted-image with contrast MRI (B) demonstrating skin blister in the hallux (white arrow), with discreet peripheral contrast enhancement, without any signs of infection (white arrow).

The histological finding of injury shows a nonacanthotic subepidermal blister with liquid levels⁴. Histological diagnosis is necessary immunohistochemistry to differentiate from subepidermal blisters^{4,10}.

The differential diagnosis includes:

- Bullous pemphigoid.^{2,11}
- Chemical or electrical burns.^{2,11}
- Drug-induced blister.^{2,6}
- Epidermolysis bullosa.^{2,6}
- Friction blister.^{2,11}
- Porphyria cutanea tarda.¹¹
- Pseudoporphyria.¹¹

The lesions heal spontaneously typically in two to six weeks^{2,9}, but they may happen again on the same site¹⁰. Secondary infection after rupture is a concern² with MRI being useful to elucidate this complication, such as osteomyelitis¹². The prognosis is good with no need of special treatment or further diagnosis with biopsy³.

We report a case of diabetes diabeticorum, a rare dermatologic lesion despite diabetes mellitus has a high incidence and prevalence worldwide, with a magnetic resonance imaging (MRI) study for evaluation of local infection and osteomyelitis, complications which were not confirmed by the imaging test.

Conclusion

We report a case of bullosis diabeticorum in the hallux characterized by MRI. In cases where there is a bullous lesion, bullosis diabeticorum is a diagnosis that needs to be remembered by the physicians and they must evaluate the possibility of local infection of the soft tissue and bone parts. Because of a self-limited course, usually painless, no specific treatment or invasive diagnostic procedures are required.

References

1. Kramer DW: Early warning signs of impending gangrene in diabetes mellitus. *Med J Rec* 1930; 132: 338-342.
2. Taylor SP, Dunn K. Bullosis Diabeticorum. *J Gen Intern Med.* 2017 feb; 32(2): 220.
3. Cantwell AR, Martz W: Idiopathic bullae in diabetics. *Arch Dermatol* 1967; 96: 42-44.
4. Derighetti, M et al. Bullosis diabeticorum in a Newly Discovered Type 2 Diabetes mellitus. *Dermatology* 2000; 200: 366-b367.
5. Gupta, V et al. Bullosis Diabeticorum: Rare Presentation in a Common Disease. *Case Reports in Endocrinology. Volume 2014, Article ID 862912, 3 pages.*

CASO CLÍNICO

6. Mota, A N C M, Nery N S, Barcaui C B. Caso para diagnóstico. *An Bras Dermatol*. 2013; 88(4): 652-654.
7. Bernstein JE, Medenica M, Soltani K: Bullous eruption of diabetes mellitus. *Arch Dermatol* 1979; 116: 324-325.
8. Bernstein JE, Levine LE, Medenica M: Reduced threshold to suction-induced blister formation in insulin-dependent diabetics. *J Am Acad Dermatol* 1983; 8: 790-791.
9. Toonstra J: *Bullosis diabeticorum: Report of a case with review of literature*. *J Am Acad Dermatol* 1985; 13: 799-805.
10. Larsen K, Jensen T, Karlsmark T, Holstein PE. Incidence of bullosis diabeticorum-a controversial cause of chronic foot ulceration. *Int Wound J* 2008; 5: 591-596.
11. Bello, F et al. 2 Cases of Bullosis Diabeticorum following Long-Distance Journeys by Road: A Report of 2 Cases. *Case Reports in Endocrinology Volume 2012, Article ID 367218*, 5 pages.
12. Kurdi AT. *Bullosis diabeticorum*. *Lancet*. 2013 Nov 30; 382(9907): e31.