Carcinoma cuniculatum of foot: case report

Carcinoma cuniculatum del pie: reporte de un caso

Ignacio Arzac Ulla¹, Capelli Lucia², Gustavo Fiorentini¹, Graciela Carabajal³

Keywords:

Carcinoma, verrucous /surgery; Carcinoma, verrucous/pathology; Foot/pathology; Foot diseases/ surgery; Amputation/methods; Case reports

ABSTRACT

Sixty-year old male patient, with diabetes mellitus background who has consulted for a slightly painful tumor-related injury in the plantar region of the left foot, which presents progressive and slight growth and has six years of evolution. The patient referred a traumatic background previous to the appearance of the lesion. With a presumptive diagnosis of perforating foot ulceration, a prophylactic scheme of sulfamethoxazole/trimethoprim joined with a ciprofloxacin antibiotic was started. A consultation was sent to the dermatology department in order to dismiss a carcinoma cuniculatum diagnosis. A incisional biopsy was performed. Anatomical pathology confirmed carcinoma cuniculatum. All therapeutical options described in literature were evaluated. In this case, infrapatellar amputation was decided as the best choice available since local resection of the tumor and transmetatarsal amputation would not be enough.

Descriptores:

Carcinoma verrugoso/cirugía; Carcinoma verrugoso/patología; Pé/ patología; Doenças do pé/cirugía; Amputación/métodos; Informes de casos

 ¹ Seccion Pie y Tobillo, Hospital Churruca, Buenos Aires, Argentina.
² Servicio de Dermatologia, Hospital

Churruca, Buenos Aires, Argentina.

³ Servicio de Anatomia Patologica, Hospital Churruca, Buenos Aires, Argentina.

> Corresponding author: Ignacio Arzac Ulla Uspallata 3400, 1437 Buenos Aires, Argentina.

Conflicts of interest:

Received on: 10 /11/2014

Accepted on: 06/01/2015

RESUMEN

Paciente de sexo masculino de 60 años de edad, con antecedentes de diabetes mellitus que consultó por una lesión tumoral levemente dolorosa, de crecimiento leve y progresivo, localizada en la región plantar de pie derecho, de seis años de evolución.El paciente refería un antecedente traumático previo a la aparición de la lesión. Con el diagnóstico presuntivo de mal perforante plantar, se inició un esquema antibiótico profiláctico con ciprofloxacina y trimetoprima – sulfametoxazol y se realizó una interconsulta con el Servicio de Dermatología para descartar el diagnóstico de un carcinoma cuniculatum. Se realizó una biopsia incisional por punch. El resultado de la anatomia patologica fue de Carcinoma cuniculatum. Se evaluaron todas las opciones terapéuticas descriptas en la literatura. En este caso se decidió que la mejor alternativa es la amputación infrapatelar ya que la resección local del tumor y la amputación transmetatarsiana serían insuficientes.

INTRODUCTION

The term carcinoma Cuniculatum was first used in 1954 by Aird, to describe a well-differentiated neoplasm of slow growth, located at the foot with a tendency to local recurrence, which rarely metastasize.⁽¹⁻⁴⁾ It can also affect the oral cavity (oral florid papillomatosis), the genital region (Buschke Loewenstein tumor) and the hairless skin (carcinoid papillomatosis).^(5,8-10)

The most common site of this tumor is the plantar aspect of the foot. Usually starts as a flat lesion, then becomes verrucous, acquiring of cauliflower aspect. It can ulcerate and drain its fetid contents to the outside.⁽⁵⁻¹⁰⁾

The differential diagnosis includes verruca vulgaris, reactive epidermal hyperplasia and diabetic foot ulcers.⁽³⁻¹⁰⁾

Treatment consists of wide local surgical excision and, in certain cases, amputation. $^{\scriptscriptstyle{(8-10)}}$

CASE REPORT

Sixty-year old male patient, with diabetes mellitus background who has consulted for a slightly painful tumor-related injury, in the plantar region of the left foot which presents progressive and slight growth and has six years of evolution. A verrucous, keratotic, exophytic, ulcerated, yellowish-white lesion of 5x4cm (Figures 1, 2) was evident at the physical exam. The patient referred a traumatic background previous to the appearance of the injury.

A foot radiograph was requested and osteomyelitis was discarded. A NMR showed an edema of the soft tissue surrounding the 2nd and 3rd metatarsal and the proximal phalangeal area of the second finger, and signs of atrophy with adipose replacement in the plantar muscles of the foot. The image was compatible with an osseous inflammatory/infectious process with soft tissue compromise (Figures 3, 4).

With a presumptive diagnosis of perforating foot ulceration, a prophylactic scheme of sulfamethoxazole/ trimethoprim joined with a ciprofloxacin antibiotic was started. A consultation was sent to the dermatology department in order to dismiss a carcinoma cuniculatum diagnosis. A incisional biopsy was performed. Anatomical pathology showed an epidermal proliferation characterized by a noticeable acanthosis, crests with rounded ends, slim forked papillae and marked pleomorphism of the basal membrane, compatible with carcinoma cuniculatum (Figure 5).



Figura 1. Exophytic and ulcerated plantar lesion on the right foot



Figura 2. Exophytic and ulcerated plantar lesion on the right foot



Figura 3. MRI: soft tissue swelling and atrophy of the plantar muscles



Figura 4. MRI: soft tissue swelling and atrophy of the plantar muscles



Figura 5. Histology: epidermal proliferation characterized by marked acanthosis and pleomorphism of the basal membrane

All therapeutical options described in literature were evaluated. In this case, infrapatellar amputation was decided as the best choice available, since local resection of the tumor and transmetatarsal amputation would not be enough considering the huge extension of the tumor. Unfortunately, the patient refused such intervention even though he was warned about the risks that his denial would imply.

DISCUSSION

The term verrucous carcinoma was introduced by Ackerman in 1948.^(1,4,6,10) During 1954 Aird et al were

the first in English literature to describe an unusual verrucous carcinoma in the plantar region which they later denominated carcinoma cuniculatum (CC).^(2,3,5,10-12). This tumor more frequently affects male patients, over 50 years old.^(5,7,10,13) It's clinically presented as an exophytic mass with a cauliflower aspect that drains a fetid material through cavities that resemble burrows. (5,8,14,15) This tumor has a low level of malignancy; it's locally aggressive and able to produce methastasis in a 5%.^(1,5,14) The lesion gradually increases its size to invade deep tissues, creating mass destruction and eventually invading the bone.^(5,8,11,15) At a histological level an exophytic/endophytic pattern of growth can be observed. It presents areas of hyperkeratosis and parakeratosis. The crests contain keratin cysts that are projected to the dermis. Anaplasia is mild and the mitotic index is low.^(8,16) Multiple therapeutical options have been described. Wide local resection is the treatment of choice. Whenever the tumor produces foot deformity or invasion of the metatarsal bones, amputation should be considered. It must be considered that a relapse implies a bad prognosis.⁽¹⁴⁾ Other treatments proposed are cryosurgery, curettage, and Mohs surgery.^(8,9,11,14) The latter is the procedure of choice when the tissue should be preserved.⁽¹⁴⁾ Radiotherapy has proved not to be successful⁽⁸⁾. Long term prognosis of CC (as long as it is correctly treated) is favorable, with rates of healing higher than 99%.(10,14)

REFERENCES

- Affleck AG, Leach IH, Littlewood SM. Carcinoma cuniculatum arising in focal plantar keratoderma. Dermatol Surg. 2007; 33(6):745-8.
- Aird I, Johnson HD, Lennox B. Epithelioma cuniculatum: a variety of squamous carcinoma peculiar to the foot. Br J Surg. 1954;42(173):245-50.
- Coldiron BM, Brown FC, Freeman RG. Epithelioma cuniculatum (carcinoma cuniculatum) of the thumb: a case report and literature review. J Dermatol Surg Oncol. 1986;12(11):1150-5.
- Floristán MU, Feltes RA, Sáenz JC, Herranz P. [Verrucous carcinoma of the foot associated with human papilloma virus type 18]. Actas Dermosifiliogr. 2009;100(5):433-5. Spanish.
- Halpern J, Harris S, Suarez V, Jeyaratnam R, Smith AG. Epithelioma cuniculatum: A case report. Foot Ankle Surg. 2009;15(2):114-6.
- Ho J, Diven DG, Butler PJ, Tyring SK. An ulcerating verrucous plaque on the foot. Verrucous carcinoma (epithelioma cuniculatum). Arch Dermatol. 2000;136(4):547-8, 550-1. Review.
- Kao GF, Graham JH, Helwig EB. Carcinoma cuniculatum (verrucous carcinoma of the skin): a clinicopathologic study of 46 cases with ultrastructural observations. Cancer. 1982;49(11):2395-403.
- Miller SB, Brandes BA, Mahmarian RR, Durham JR. Verrucous carcinoma of the foot: a review and report of two cases. J Foot Ankle Surg. 2001;40(4):225-31. Review.
- 9. Mirigliano E, LaTour R, Abramczuk JW. Squamous cell carcinoma

of the foot mimicking osteomyelitis: a case report. J Foot Ankle Surg. 2011;50(4):480-5.

- 10. Shantharam Shetty M, Ajith Kumar M, Jagadish P. Recalcitrant plantar wart - suspect verrucous carcinoma. Foot Ankle Surg. 2008;23(1):54-6.
- 11. Schein O, Orenstein A, Bar-Meir E. Plantar verrucous carcinoma (epithelloma cuniculatum): rare form of the common wart. Isr Med Assoc J. 2006;8(12):885.
- 12. Shenoy AS, Waghmare RS, Kavishwar VS, Amonkar GP. Carcinoma cuniculatum of foot. Foot (Edinb). 2011;21(4):207-8.
- 13. Schwartz RA. Verrucous carcinoma of the skin and mucosa. J Am Acad Dermatol. 1995 Jan;32(1):1-21; quiz 22-4. Review. Erratum in: J Am Acad Dermatol 1995;32(5 Pt 1):710.
- 14. Spyriounis K, Tentis D, Sparveri IF, Arvanitis T. Plantar epithelioma cuniculatum. A case report with review of the literature. Eur J Plast Surg. 2004;27(5):253-6.
- 15. Verma S. A verrucous carcinoma of the foot on an injection site: a case report. Int J Low Extrem Wounds. 2005;4(4):252-4.
- 16. Steffen C. Dermatopathology in historical perspective: epithelioma cuniculatum (Aird). Am J Dermatopathol. 2006;28(5):451-61.