Archives of Clinical Trials and Case Reports

Cervera-Bonilla S, et al., 2022-Arch Clin Trial & Case Report

Pilonidal Cyst with Malignant Degeneration: A Case Report

Sergio Cervera Bonilla¹, Manuela M. Ramirez Marin², Liliana Cuevas Lopez^{3*}, Margarita Tamayo Buendia⁴, Maddy Mejia Cortes⁵ and Rocio Marino Gonzalez⁶

Abstract

Introduction: Pilonidal cyst is a frequent surgical pathology with low risk of malignancy. Here, we present a case of a chronic pilonidal cyst with transformation to skin squamous cell carcinoma.

Case presentation: A 67-year-old male patient with a chronic presentation of injury in the sacrococcygeal region with occasional bleeding that got worse with progressive growth and purulent discharge. Initially was treated as a soft tissue infection with oral antibiotics but did not solve. A skin biopsy reported a squamous cell carcinoma, extension studies showed no secondary disease of the lesion. The patient was treated as if he was diagnosed a squamous cell carcinoma of the anal margin. Treatment was initiated with Nigro protocol chemoradiotherapy and wide local resection of the lesion with oncological margins of 1cm with success.

Discussion: The pilonidal cyst is an episodic inflammatory disease in the sacrococcygeal zone that usually responses to antibiotic treatment and surgery if it's necessary, malignant degeneration is rare, might it be related with chronic inflammation that affects repair mechanisms and create a predisposition for malignancy. The optimal treatment is in bloc surgical resection with tumor-free margins and complementary treatment depends on the histology.

Conclusion: This is an uncommon case of a Pilonidal

Universitario San Ignacio-Pontificia Universidad Javeriana, Bogotá, Colombia

²Third-year resident in dermatology, Pontificia Universidad Javeriana, Bogotá, Colombia

³Fellow Oncologic Surgery, Hospital Universitario San Ignacio-Pontificia Universidad Iaveriana. Bogotá. Colombia

⁴Dermatologist, Hospital Universitario Sar Ignacio-Pontificia Universidad Javeriana Bogotá. Colombia

⁵Dermato-pathologist, Hospital Universitario San Ignacio-Pontificia Universidad Javeriana Bogotá, Colombia

^oClinical Oncologist, Hospital Universitario San Ignacio-Pontificia Universidad Javeriana, Bogotá, Colombia

*Corresponding Author: Liliana Cuevas Lopez, Fellow Oncologic Surgery, Hospita Universitario San Ignacio-Pontificia Universidad Javeriana Bogotá, Colombia

Receiving Date: 08-05-2022

Accepted Date: 08-22-2022

Published Date: 09-15-2022

Copyright® 2022 by Cervera-Bonilla S, et al All rights reserved. This is an open access article distributed under the terms of the Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium, provided the original author and source are credited.

cyst with transformation to skin squamous cell carcinoma managed with Nigro protocol chemoradiotherapy and wide local resection. This case will give surgeons another tool to treat this condition.

Keywords: Pilonidal cyst; Squamous cell carcinoma; Malignant transformation.

Introduction

Pilonidal cyst is a common surgical pathology. It is estimated to have an incidence of up to 26 per 100,000 inhabitants [1,2]; nevertheless, it has a very low risk of malignancy, which has been reported only in recurrent disease [3]. It is diagnosed in people over 50 years of age and is more prevalent in the male sex [4]. The most commonly associated non-melanoma skin cancer is squamous cell carcinoma [3,5]. In which patients should we suspect malignant transformation? Based on this question, we present a clinical case managed in an interdisciplinary manner with satisfactory result.

Case presentation

A 67-year-old male patient was referred to the dermatology service with a clinical picture of several years of evolution consisting of an injury in the sacrococcygeal region with occasional bleeding. Four months earlier, it had a rapid, progressive growth associated with purulent discharge, which was treated as a soft tissue infection with oral antibiotics. On physical examination, multiple bridged comedones were found in the posterior cervical, axillary, and bilateral inguinal regions (Figure 1a). In the sacrococcygeal region and upper part of the intergluteal fold, a tumor lesion of approximately 7 x 4cm was identified, which was erythematous, suppurative, lobulated, indurated, with a keratotic center surrounded indurated area, painful on palpation, and fistulas with purulent discharge (Figure 1b). A skin biopsy was performed that reported a well-differentiated infiltrating squamous cell carcinoma, and chest and abdominal tomography showed no extension of the lesion. The patient was assessed by a clinical oncologist who, given the findings of the physical examination, considered it to be squamous cell carcinoma of the anal margin. Treatment was initiated with Nigro protocol chemoradiotherapy, with adequate tolerance without adverse events reported by the patient, achieving a good clinical response (Figure 1c); the lesion was reduced toward the sacrococcygeal region, allowing the realization of a wide local resection of the lesion with oncological margins of 1 cm.

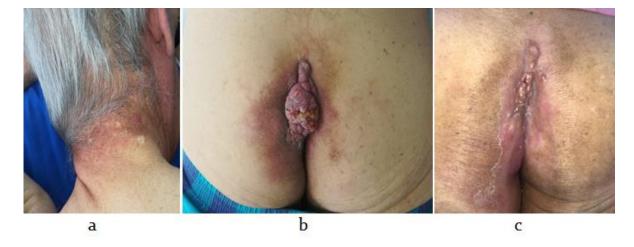


Figure 1: (a) Multiple bridged comedones in posterior cervical region (b) Lobed exophytic tumor (c) post-management with radiotherapy and chemotherapy.

The pathological anatomy showed a skin specimen with a macroscopic description of an indurated plaque on the surface, with a depressed center of 4 x 6cm; in the sclice, a fistulous path was revealed from the epidermal surface to the center of the tumor lesion (Figure 2a). The histological report described a fistulous tract lined with keratinizing squamous epithelium, with a dilation located in deep dermis that showed

markedly hyperplastic squamous epithelium and basal atypia infiltrating the dermis, as well as the cyst surrounded by chronic inflammation and extensive fibrotic changes, without lymphovascular or perineural invasion and with tumor-free margins (Figure 2b). The patient had a satisfactory evolution after resection and did not present recurrence in the three-year follow-up period (Figure 2c).

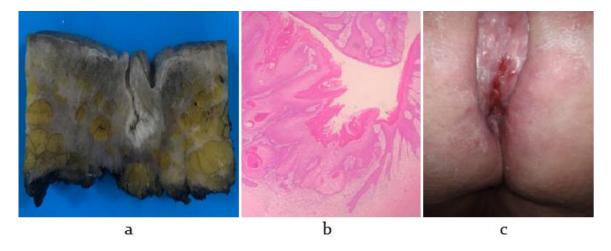


Figure 2: (a) Surgical specimen (b) Histological image showing markedly hyperplastic squamous epithelium with basal atypia infiltrating the dermis (c) Post-surgical result.

Discussion

pilonidal cyst episodic is an inflammatory disease in the sacrococcygeal region [6]. Its development is attributed to intrusion of hair into the subcutaneous cellular tissue, which triggers a chronic inflammatory response [7]. It has been suggested that it is part of the spectrum of follicular occlusion diseases, a chronic, condition with systemic autoinflammatory basis that includes dissecting cellulitis of the scalp, acne conglobata, hidradenitis suppurativa, and pilonidal cyst [8,9], which is compatible with the initial clinical picture presented by our patient. Although the pilonidal cyst is an extremely frequent pathology, its malignant degeneration is rare with an estimated incidence of up to 0.1% [10]. The

mechanism by which it occurs is similar to that of chronic scars and Marjolin ulcers due to recurrent and chronic inflammation. The release of free radicals by inflammatory cells causes damage to the genetic material. In addition, chronic inflammation affects repair mechanisms, creating a predisposition for malignancy [11,12]. In most cases, it develops into squamous cell carcinoma, which has an aggressive and recurrent behavior [13,14].

The indicated treatment is in bloc surgical resection until the presacral fascia, with tumor-free margins [15]. Adjuvant radiotherapy and chemotherapy have been recommended, observing a decrease in local recurrence [12,14]. Early resection reduces morbidity and mortality, but this risk must always be considered. Experts recommend

conducting a pathological study in all longstanding pilonidal cysts, in patients older than 50 years, and in case of atypical findings during clinical evaluation [16].

Conflicts of interests

The authors do not have existing conflict of Interest

Ethics approval

Written informed consent was obtained from the patient for participated and publication for this case report and accompanying images.

Availability of data and material

The data of the clinical history is true and rests in the clinical history system of the San Ignacio University Hospital

Authors' contributions

Each one of the authors contributes with the elaboration and correction to finish the document.

References

- 1. Farrell D, Murphy S. Negative Pressure Wound Therapy for Recurrent Pilonidal Disease: A Review of the Literature. J Wound Ostomy Continence Nurs. 2011;38(4):373-8. <u>PubMed | CrossRef</u>
- 2. Isik A, Idiz O, Firat D. Novel Approaches in Pilonidal Sinus Treatment. Prague Med Rep. 2016;117(4):145-152. PubMed | CrossRef
- 3. Mayol Oltra A, Boldó Roda E, Lozoya Albacar R, Morillo Macias V, Nobleja Quiles N. Squamous Cell Carcinoma over Pilonidal Chronic Disease. A New Therapeutic Approach. Int J Surg Case Rep. 2020;70:172-177. PubMed | CrossRef
- 4. Frost BM, Riddell AD, Austin S, Stephenson BM. Malignancy in an Old Pilonidal Sinus. Colorectal Dis. 2007;9(9):857. PubMed | CrossRef
- 5. Eryılmaz R, Bilecik T, Okan I, Ozkan OV, Coşkun A, Sahin M. Recurrent Squamous Cell Carcinoma Arising in a Neglected Pilonidal Sinus: Report of a Case and Literature Review. Int J Clin Exp Med. 2014;7(2):446-50. PubMed
- 6. Allen-Mersh TG. Pilonidal Sinus: Finding the Right Track for Treatment. Br J Surg. 1990;77(2):123-32. PubMed | CrossRef
- 7. Karydakis GE. Easy and Successful Treatment of Pilonidal Sinus after Explanation of its Causative Process. Aust N Z J Surg. 1992;62(5):385-9. PubMed | CrossRef
- 8. Pesce A, Capuzzo G, Cammisuli B, Musumeci ML, Micali G. Pilonidal Disease, Hidradenitis Suppurativa and Follicular Occlusion Syndrome: A Diagnostic Challenge. Eur Rev Med Pharmacol Sci. 2018;22(15):4755-4756. PubMed | CrossRef
- 9. Chintapatla S, Safarani N, Kumar S, Haboubi N. Sacrococcygeal Pilonidal Sinus: Historical Review, Pathological Insight and Surgical Options. Tech Coloproctol. 2003;7(1):3-8. <u>PubMed | CrossRef</u>
- 10. Chatzis I, Noussios G, Katsourakis A, Chatzitheoklitos E. Squamous Cell Carcinoma Related to Long Standing Pilonidal-Disease. Eur J Dermatol. 2009;19(4):408-9. <u>PubMed | CrossRef</u>
- 11. de Bree E, Zoetmulder FA, Christodoulakis M, Aleman BM, Tsiftsis DD. Treatment of Malignancy Arising in Pilonidal Disease. Ann Surg Oncol. 2001;8(1):60-4. PubMed | CrossRef
- 12. Michalopoulos N, Sapalidis K, Laskou S, Triantafyllou E, Raptou G, Kesisoglou I. Squamous Cell Carcinoma Arising from Chronic Sacrococcygeal Pilonidal Disease: A Case Report. World J Surg Oncol. 2017;15(1):65. PubMed | CrossRef
- 13. Nunes LF, Castro Neto AK, Vasconcelos RA, Cajaraville F, Castilho J, Rezende JF, et al. Carcinomatous Degeneration of Pilonidal Cyst with Sacrum Destruction and Invasion of the Rectum. An Bras Dermatol. 2013;88(6Supplı):59-62. PubMed | CrossRef
- 14. Parpoudi SN, Kyziridis DS, Patridas DCh, Makrantonakis AN, Iosifidis P, Mantzoros IG, et al. Is Histological Examination Necessary when Excising a Pilonidal Cyst? Am J Case Rep. 2015;16:164-8. PubMed | CrossRef
- 15. Humphries AE, Duncan JE. Evaluation and Management of Pilonidal Disease. Surg Clin North Am. 2010;90(1):113-24. PubMed | CrossRef

16.	Boulanger G, Abet E, Brau-Weber AG, Leclair F, Denimal F, Jean MH, et al. Is histological Analysis of Pilonidal Sinus Useful? Retrospective Analysis of 731 Resections. J Visc Surg. 2018;155(3):191-194. PubMed CrossRef