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Case Report

Squamous cell carcinoma arising from an epidermal cyst of the scrotum

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ABSTRACT

We present an 83-year-old man with squamous cell carcinoma in the lining of an epidermal cyst that had been in his scrotum for more than 23 years. The patient presented with a discharge from the lesion, and received oral antibiotics, followed by wide excision of the cyst. We found the excised cyst wall was uneven. Serial sections of the thickened part showed squamous cell carcinoma. The patient had an uneventful recovery after surgery. Malignant transformation in an epidermal cyst is rare, with only a few such cases reported in the English literature.

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1. Introduction

Epidermal cysts are common benign skin lesions, comprising 85% to 90% of all excised cysts [1]. Historically, epidermal cysts have been referred to as infundibular cysts, epidermal cysts, and epidermal inclusion cysts [2]. They present as well-circumscribed, firm subcutaneous nodule formed by downgrowth and cystic expansion of the epidermal squamous epithelium. A nodule is filled with keratinous debris and cholesterol or sebaceous materials. Grossly, it appears as a painless, soft lesion of variable size with intact overlying skin, and may develop anywhere in the body [3]. There are few reports of malignant transformation of this benign lesion in the English literature [1,3–10]. In this article, we report a scrotal epidermal cyst that underwent malignant changes 23 years after the initial detection of the lesion.

2. Case report

An 86-year-old man presented to our department with recent onset of a grayish-white discharge from a mass on the lateral aspect of the right scrotum. The mass, which had been present for 23 years, began to rupture 2 days previously, with continuous leakage of purulent material. Clinically, the mass was cystic and 4.1 cm in diameter. The overlying skin was slightly reddened. There were no

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palpable groin lymph nodes. He had no fever, chills, dysuria, or weight loss. The cystic contents were retrieved as much as possible. Empirical broad spectrum antibiotics were prescribed. The lesion improved with antibiotic treatment and an excision biopsy was performed 2 weeks later. Total excision of the tumor, including the elliptical covering skin, was performed under local anesthesia. The pathological examination of the excised specimen showed measuring $4.1 \times 3.1 \times 1.0$ cm cystic tumor with an area with fungating thickened wall. Microscopically, there was a large keratincontaining cyst lined by squamous epithelium. There was an area with thickened keratinizing squamous epithelium with atypical keratinocytes with pleomorphic hyperchromatic nuclei and frequent mitotic activity (Fig. 1). There were nests of atypical keratinocytes invading the stroma (Fig. 2). It was a well-differentiated keratinizing squamous cell carcinoma arising from an epidermal cyst. The resection margins were free of tumor. The patient recovered uneventfully.

3. Discussion

The nature of malignant transformation of the cystic lining of an epidermal cyst into squamous cell carcinoma is uncertain. It is well recognized that chronic inflammation, high exposure to ultraviolet light, human papillomavirus infection and mutation of the p53 gene in cutaneous tissue are predisposing factors for squamous cell carcinoma [11].

Previous case reports show that the frequency ranges from 0.011% to 0.045% [6,9]. In 1980, Bauer and Lewis reported 72

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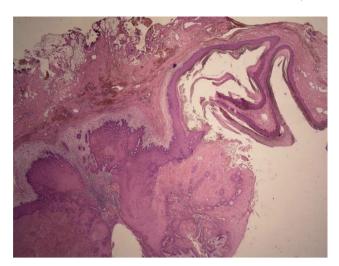


Fig. 1. The epidermal cyst has thickened squamous epithelium with proliferation of atypical keratinocytes and keratinization. There are nests of atypical keratinocytes infiltrating the stroma (hematoxylin and eosin, $40\times$).

carcinomas noted among 3300 cases of skin cyst, with an incidence of 2.2% [5]. However, Epstein discovered that many patients in the series cited by Bauer and Lewis were incompletely examined or the excised cystic lesion was not further examined microscopically [2,12].

In 1999, after reviewing 27 articles in the English literature during the past 55 years, López-Ríos et al found that only eight were well-documented with a microscopic description and photomicrographs corresponding to squamous cell carcinoma arising in an epidermal cyst [7,8].

In 2010, Anton-Badiola et al critically reviewed 19 well-documented cases among those case reports in the English literature, including his own case. The mean age of those patients was 43.2 years (range 21–80 years), and there were 13 men and 6 women [4]. Nine of these lesions were located on the head and neck, with the remaining 10 found on the trunk or extremities. The lesion ranged from 1.5 to 13 cm (mean 5.7 cm), with a duration of 2 to 480 months (mean 101 months). All patients had excision of the cyst. Seventeen cases had a normal gross appearance of the cyst wall and only two cases showed either a thickened cyst wall or granulation tissue within the cyst [4]. Therefore, routine histological examination of all excised cysts is required. Resection with adequate margins is the treatment of choice for a well-confined lesion [7].

In conclusion, this is one of a few case reports of squamous cell carcinoma arising from an epidermal cyst, particularly of the scrotum. Despite the rare occurrence of malignancy in an epidermal cyst, it is recommended that guidelines for treating cutaneous squamous cell carcinoma are followed [6].

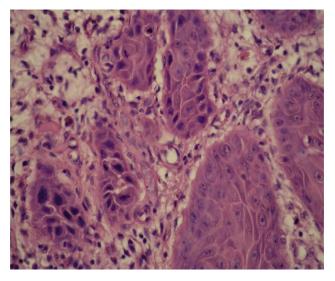


Fig. 2. Well-differentiated squamous cell carcinoma: nests of atypical keratinocytes with pleomorphic, hyperchromatic nuclei and mitoses (hematoxylin and eosin, $400 \times$).

This case emphasizes the necessity for routine histopathological examination of all excised cysts, especially in thickened areas or granulation tissue, to exclude malignancy [8].

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