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Single Case

Unusual Presentation of Cutaneous Spindle Cell Squamous Cell Carcinoma: A Case Report

Erika Wernheden^a Hannah Trøstrup^a Anette Pedersen Pilt^b

^aDepartment of Plastic and Breast Surgery, Zealand University Hospital, Roskilde, Denmark; ^bDepartment of Pathology, Zealand University Hospital, Roskilde, Denmark

Keywords

Cutaneous spindle cell squamous carcinoma \cdot Case report \cdot Squamous cell carcinoma \cdot Surgery

Abstract

Cutaneous spindle cell squamous cell carcinoma (SpSCC) is a rare and often aggressive subtype of squamous cell carcinoma (SCC), which usually appears in sun-exposed areas, in areas that have received prior ionizing radiation, or in immunosuppressed individuals. SpSCCs are histologically characterized by keratinocytes infiltrating the dermis as single cells with elongated nuclei rather than as cohesive nests or islands and, in contrast to conventional SCC, are lacking features of keratinization. Immunohistochemical studies are useful to distinguish SpSCC from other spindle cell neoplasms, such as spindle cell/desmoplastic melanoma, cutaneous leiomyosarcoma, and atypical fibroxanthoma. We present a rare case of a patient with SpSCC in the gluteal region with regional lymph node metastasis. The patient was treated with wide excision of the tumor, inguinal lymph node dissection, and adjuvant radiotherapy. Cutaneous SpSCC is clinically similar to conventional SCC but can demonstrate more aggressive behavior. This case is rare since it was localized in the gluteal region of an otherwise healthy man.

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Introduction

Spindle cell squamous cell carcinoma (SpSCC) is a rare subtype of squamous cell carcinoma (SCC) which accounts for 1% of SCC [1]. The most common locations are the oral cavity, larynx, sinonasal areas, and pharynx [1, 2]. Primary cutaneous lesions are very rare. Cutaneous SpSCC is a rare type of SCC in which keratinocytes infiltrate the dermis as single cells with elongated nuclei rather than as cohesive nests or islands and, in contrast to conventional SCC, it is lacking features of keratinization [3, 4]. It must be differentiated from other spindle cell lesions, such as spindle cell/desmoplastic melanoma, cutaneous leiomyosarcoma, and atypical fibroxanthoma (AFX).

Clinically similar to conventional SCC, SpSCC presents as an ulcerated or exophytic mass. It is more often seen in older Caucasians and is usually confined to sun-damaged sites, such as the head and neck area, or areas that have received prior ionizing radiation [2–5].

Due to the lack of differentiation, the tumor may exhibit more aggressive characteristics with increased risk of recurrence and metastasis [4]. The aggressive behavior has been described primarily in SpSCCs in areas that have received prior radiation and in immunosuppressed individuals, whereas SpSCCs unassociated with radiation have shown more indolent behavior [6–8]. As with SCCs in general, grade of differentiation, tumor depth, and perineural growth are of great prognostic significance [6, 9].

We present a rare case of SpSCC in the gluteal region with regional lymph node metastasis.

Case Presentation

A 65-year-old man with no comorbidities presented to the department with a 2×3 cm large, well-circumscribed elevated tumor with central ulceration located in the right gluteal region. The tumor had developed over several years with increasingly rapid growth over the previous 6 months. The patient's general practitioner had performed a curettage biopsy and referred the patient to the Department of Plastic Surgery for complete excision. The primary biopsy was interpreted as partly residual of an adnexal tumor and partly consisting of changes that were in line with a condyloma with dysplasia, transferring to invasive low-differentiated carcinoma with characteristics of desmoplastic SCC.

Physical examination also revealed an enlarged lymph node in the right inguinal area. Ultrasound-guided fine-needle biopsy of the enlarged lymph node, as sentinel node biopsy is not a routine procedure in the treatment of SCCs, and wide excision of the tumor was planned in an outpatient setting.

At the time of excision, the tumor had grown to 3×3.5 cm (Fig. 1). The tumor was excised with a 1-cm distal margin. The histology following the complete resection of the tumor showed a central area with papillomatosis and partly ulcerated surface, and transition from hyperplastic and dysplastic squamous epithelium to invasive low-differentiated SCC with a dominating spindle cell component (Fig. 2). Focally, perineural carcinosis was observed but no intravascular growth. The tumor thickness was 9 mm and focally involved the subcutaneous fat. No remnant of the previously described adnexal tumor was observed.

Fine-needle biopsy of the inguinal lymph node was without signs of malignancy, but since histopathology revealed a high-risk tumor, it was decided to excise the lymph node.

The excised lymph node showed infiltration of malignant epithelial tumor tissue, partly with necrosis. There were islands of epithelioid tumor cells as well as spindle cells.





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Immunohistochemically, a positive reaction for cytokeratin CK14 and P40 was observed as well as co-expression of vimentin (Fig. 3). Epithelial membrane antibody was not performed in this case.

As lymph node metastasis of SpSCC was observed, a subsequent PET-CT was performed, which revealed another 3 lymph nodes suspected for malignancy in the right inguinal area. Thus, the TNM staging for this patient was T2N1M0.

An inguinal lymph node dissection was performed, which revealed 3 lymph node metastases without perinodal growth. Postoperative complications of the lymph node dissection occurred in the form of a wound defect and the development of seroma, which required debridement. Following surgery, the patient received adjuvant radiotherapy to both the area of the primary tumor and the inguinal region.

Discussion

As shown in this case, SpSCC can exhibit aggressive behavior and increased risk for metastasis due to the lack of differentiation. In this case, the tumor showed rapid growth from first clinical examination until excision 3 weeks later and multiple regional metastases.

SpSCC is clinically similar to regular SCC, and the diagnosis relies on pathological examination. Typically, keratinocytes infiltrate the dermis as single cells with elongated nuclei rather than as cohesive nests or islands, and, in contrast to conventional SCC, SpSCC lacks features of keratinization. Immunohistochemical studies are useful tools in the distinction from other spindle cell neoplasms. SpSCC will stain positively with cytokeratin and epithelial membrane antibody, and variably with vimentin [6, 8, 10].

SpSCC usually occurs in sun-damaged areas, in areas that have received prior ionizing radiation, or in immunosuppressed individuals. This case is rare since it was localized in the gluteal region of an otherwise healthy man.

Differential diagnoses of SpSCC include AFX, spindle cell melanoma, and different variants of sarcoma. SpSCC is different from AFX because the latter lacks typical squamous differentiation and is negative for cytokeratins. In contrast to SpSCC, spindle cell melanoma is positive for S-100. Foci showing squamous differentiation and a nested growth pattern are useful in distinguishing between SpSCC and sarcomas [11]. There are no separate treatment guidelines for SpSCCs compared to other cutaneous SCCs. The treatment consists of surgery, and adjuvant radiotherapy may be considered in certain settings, such as positive margins or perineural growth. Treatment of regional nodal metastases typically includes lymph node dissection combined with radiotherapy [9], as performed in the presented case. The role of sentinel node biopsy in cutaneous SCC is to date unclear and is not routinely performed [12]. As shown in this case, fine-needle biopsy is not always reliant in revealing SCC metastasis, and excision of complete lymph nodes is, as a consequence, often necessary.

Conclusion

SpSCC is a rare and often aggressive subtype of SCC. SpSCC typically occurs in sun-damaged areas and is often, and in the most aggressive cases, related to prior skin damage, such as burns and ionizing radiation, although, as shown in this case report, it can also occur in otherwise healthy individuals in areas of non-sun-exposed skin. As the pleomorphic spindle





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cells lack recognizable features of keratinization, immunohistochemical studies are useful to distinguish SpSCC form other spindle cell neoplasms.

Statement of Ethics

The patient has given his written informed consent to the writing of this article and publishing of pictures. The research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

E.W.: conception and design, acquisition of data, drafting the work. H.T.: conception and design, acquisition of data, critically revising the work. A.P.P.: acquisition of data, critically revising the work. All authors have given final approval of the version to be published and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Fig. 1. Spindle squamous cell carcinoma prior to excision.



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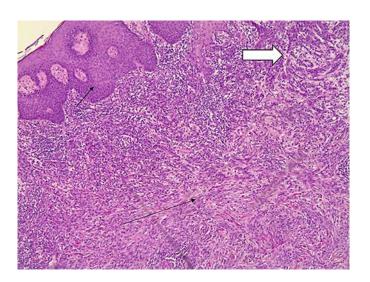


Fig. 2. Hematoxylin-eosin stain of section from completely excised skin tumor located on the right gluteal region. Short black arrow: epidermis with hyperplasia. White arrow: poorly differentiated squamous cell carcinoma in upper dermis. Long black arrow: spindle cell carcinoma in dermis

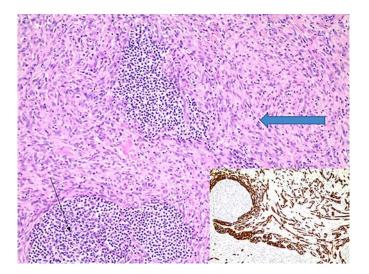


Fig. 3. Hematoxylin-eosin stain of section of lymph node from right inguinal region (×10). Black arrow: lymphatic tissue. Blue arrow: metastatic spindle cell carcinoma. Lower right corner: metastatic spindle cell carcinoma, immunohistochemical positive reaction cytokeratin 14, confirming epithelial origin.