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Single Case

Cutaneous Metastases from Salivary Duct Carcinoma of the Submandibular Gland

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Keywords
Basal cell carcinoma · Carcinoma · Metastasis · Metastatic skin cancer · Skin cancer

Abstract
Salivary duct carcinoma is a rare and highly aggressive malignant neoplasm that frequently metastasises to other organs, but cutaneous metastasis is uncommon. There are only 6 cases reported in the literature with metastases to the skin and in all cases the tumour originates from the parotid gland. We present a case of skin metastases from the submandibular gland that was mistaken for basal cell carcinoma. To the best of our knowledge, this is the first reported case of salivary duct carcinoma arising from the submandibular gland with cutaneous metastases.

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Introduction

Salivary duct carcinoma (SDC) is a rare and highly aggressive malignant neoplasm that was first described in 1968 by Kleinsasser et al. [1]. This neoplasm arises most often in the parotid gland, followed by the submandibular gland and less frequently in minor salivary glands. SDC mainly affects middle-aged and older males [2, 3]. Histologically, the neoplasm resembles ductal carcinoma of the breast and is characterised by a cribriform growth pattern and intraductal comedonecrosis [1, 4].

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SDC has a high rate of mortality and distant metastasis, but cutaneous metastases from the malignancy is uncommon [5]. Skin metastases of an internal cancer are, in general, rare [6] and amongst women the neoplasms that most commonly have skin metastases are breast cancer, neoplasms of the large intestine, and ovarian cancer. In men tumours from the lung, large intestine, and kidney have the highest rate of skin metastases [7]. Cutaneous metastatic carcinoma has more than one clinical morphology, which indicates that it can mimic several benignant and malignant skin diseases.

Cutaneous metastases from SDC are unusual. There are only 6 cases reported in the literature and in all of the reported cases the carcinoma arises in the parotid gland [8–12]. We present a case of skin metastases from the submandibular gland. To the best of our knowledge, this is the first reported case of SDC arising from the submandibular gland with cutaneous metastases.

Case Report

In February 2016, a 78-year-old man presented with an enlarging mass on the right side of the neck in relation to the submandibular space. The mass was painless and had been enlarging for the previous month. An immobile soft tissue mass was noted at the submandibular space on the right side. Ultrasonically, a 16 × 19 mm hypodense tumour was visualised in relation to the posterior pole of the submandibular gland on the right side (Fig. 1). A fine needle biopsy and Tru-cut biopsy was performed which showed cells from adenocarcinoma, but it was not possible to classify the tumour further (Fig. 2). MRI showed a 19 × 17 × 12 mm process in the lateral part of the right submandibular gland, growing into the platysma muscle, and there was no lymphadenopathy. The patient underwent a neck dissection of level I, II, and III and removal of the submandibular gland on the right side. Surgical pathology revealed a 30-mm SDC with perineural invasion and 1 lymph node was positive for tumour cells. Postoperatively, the patient received radiotherapy at a total dose of 70 Gy delivered to the right submandibular area and level I, II, and III of the neck.

In January 2017 the patient presented with a skin tumour on the right side of his forehead (Fig. 3). Clinically, the tumour resembled a basal cell carcinoma and was treated with curettage by a dermatologist. The pathology report described islands or nests of basaloid cells associable with basal cell carcinoma. However, the wound never healed and the patient developed a 10 × 8 mm superficial ulcer at the site. An excision of the tumour with a 3-mm margin was performed and this time the pathology report described metastases from SDC arising from the submandibular gland. The specimens from the first curettage biopsy were examined and it was concluded that even this biopsy showed cells from an SDC and not basal cell carcinoma.

Discussion

Cutaneous metastases from SDC are rare. There are only 6 reports in the literature [8–12]. The skin manifestation of visceral malignancies ranges from subcutaneous nodule, erythematous patch and plaque, to firm papules and nodules. This indicates that skin metastases can mimic numerous skin lesions such as benign and malignant primary skin tumours, including epidermoid cyst, keratoacanthoma, and basal cell carcinoma. Furthermore, cutaneous metastases can mimic bacterial infection or they may present as an ulcer. Our patient

had an ulcer with small nodules and telangiectasia, which was mistaken for a basal cell carcinoma.

Around 10% of internal malignancies develop cutaneous metastases. Visceral neoplasms may metastasise directly to the overlaying skin or through hematogenous or lymphatic spread. Skin metastases are underestimated, underdiagnosed, and challenging. At the moment the variety in the clinical appearance of the metastases does not give a clue with regard to the origin of the primary tumour. The localisations of the cutaneous metastases might give a hint towards the target organ [13, 14].

When patients with previous history of internal malignancies present with a new skin tumour or lesion it should be considered that it could potentially be cutaneous metastases of the visceral neoplasm. This group of patients with previous cancer history should examine cutaneous, subcutaneous, and visible mucous surfaces routinely either by a doctor or through self-examinations. Furthermore, the skin metastases can also be the presenting manifestation of a previously undiagnosed cancer.

In conclusion, SDC is an uncommon aggressive malignant neoplasm. Skin metastases from this malignancy are unusual. To the best of our knowledge, cutaneous metastatic SDC emerging from the submandibular gland has not been reported previously.

Statement of Ethics

Patient consent for publication has been obtained.

Disclosure Statement

The authors have no conflicts of interest to disclose.

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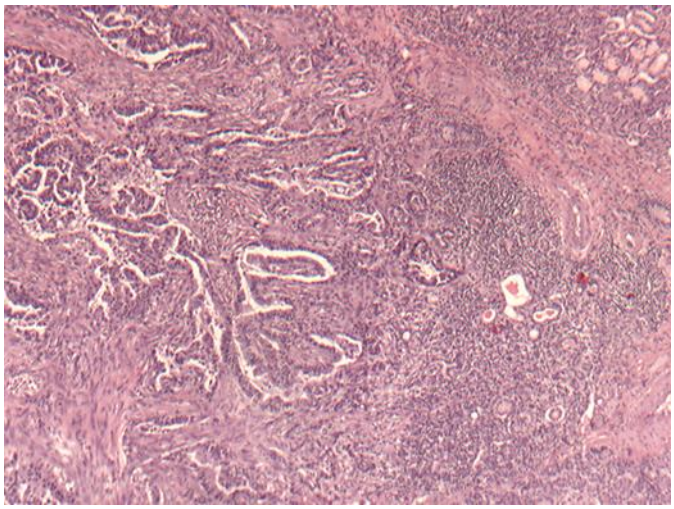

Fig. 1. Section from the submandibular gland showing extensive infiltrative growth of irregular glandular structures resembling a high-grade ductal carcinoma.

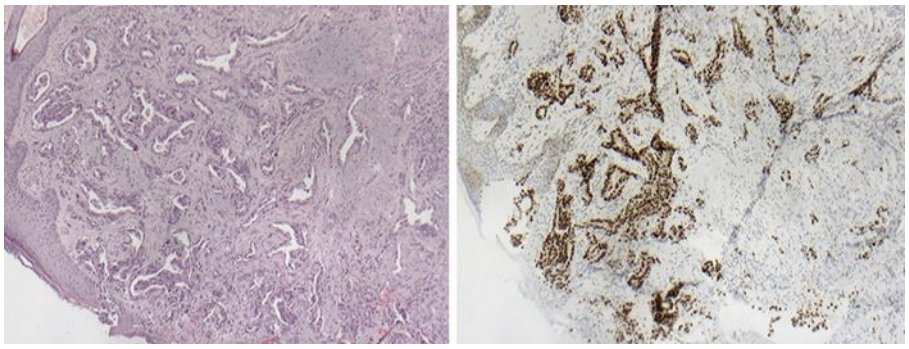

Fig. 2. Section from the skin showing infiltrative growth of irregular glandular structures with similar resemblance to high-grade ductal carcinoma and diffuse nuclear staining for androgen receptor.

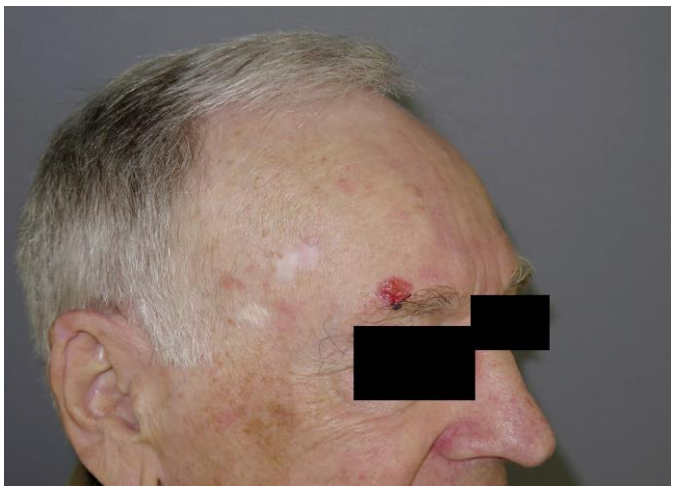

Fig. 3. A 78-year-old male patient presented with a superficial ulcerated skin tumour above the right eyebrow, measuring 10 × 8 mm. Clinically, the tumour resembled a basal cell carcinoma.