Case Report

Sebaceous carcinoma of the lip: Comparing normal lip and cheek anatomy with the imaging features of a rare cutaneous malignancy

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Abstract

Sebaceous carcinoma is a rare cutaneous malignancy, commonly affecting the eyelids. This case highlights a patient who presented with sebaceous carcinoma of the right upper lip with extensive involvement of the soft tissues of the head and neck. As part of the initial investigation, ultrasound was requested. This case demonstrates the ultrasound features of sebaceous carcinoma as well as revising the normal ultrasound anatomy of the upper lip and muscles of the cheek.

Keywords: Ultrasound, facial anatomy, sebaceous carcinoma

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Introduction

Sebaceous carcinoma is a rare malignancy of skin adnexa.¹ There are two subtypes: ocular and extraocular. The former is the third most common type of malignancy of the eyelids, the majority arising in the meibomian glands. The extraocular subtype accounts for 20% of sebaceous carcinoma,¹ affecting in particular the scalp, face and parotid gland.

This case highlights a patient who presented with sebaceous carcinoma of the right upper lip. Although there are a few case reports of intraoral sebaceous carcinoma,^{2,3} there is nothing in the literature that describes the ultrasound (US) features of the disease at this anatomical subsite. This case demonstrates the US features of sebaceous carcinoma as well as revising the normal US anatomy of the upper lip and muscles of the cheek.

Case summary

An 81-year-old Caucasian male with Type II diabetes and hypertension was referred to the Oral and Maxillofacial Surgery Department with a two month history of a firm upper right lip mass. The swelling was first noticed by the patient after he had sustained a small abrasion whilst shaving. He had attended his general medical practitioner on several occasions and had been prescribed two courses of antibiotics. The swelling did not respond to antimicrobial therapy and the patient was subsequently referred for a surgical opinion.

On clinical examination the patient was systemically well. His upper right philtrum was erythematous and indurated, the overlying skin was intact. No other soft tissue asymmetry

was identified on the face. There was no palpable cervical lymphadenopathy. The clinical diagnosis was that of a chronic inflammatory process exacerbated by the patient's diabetes. US was requested as an initial investigation.

High frequency US was performed of the upper lip and cheek. This demonstrated a poorly defined hypoechoic irregular mass in the right naso-labial region, with no internal vascularity, involving skin, subcutaneous fat, orbicularis oris and the intraoral mucosa (Figure 1). It extended posteriorly towards the buccal commissure, involving buccinator muscle, modiolus and the zygomaticus muscles and the subcutaneous fat.

US-guided fine-needle aspiration biopsy was performed. The cytopathology suggested cells of salivary gland origin which could be either reactionary or malignant.

Contrast enhanced magnetic resonance imaging (MRI) was subsequently carried out for assessment of disease extent. This revealed an extensive aggressive infiltrative disease process involving the soft tissues of the right upper lip, buccal space, retromolar trigone (Figure 2), pterygopalatine fossa and apex of the infratemporal fossa, in addition to those muscles already described on the US.

Incisional biopsy was carried out and histopathology demonstrated sebaceous carcinoma. The patient was discussed in local and regional Head and Neck Multi-Disciplinary Meetings, and was referred on for palliative radiotherapy.

Discussion

Sebaceous Carcinoma (SebCA) is a malignancy of skin adnexa. Twenty per cent of SebCA are extraocular, the

Figure 1. Transverse section US showing disease (D) invading orbicularis oris muscle (o) and labial mucosa (lm)

Figure 2. T1 weighted post contrast axial MRI demonstrating disease involvement of the right orbicularis oris, the upper lip, buccal subcutaneous tissues up to the intraoral labial mucosa and retromolar trigone (white arrows)

most common site being the parotid, although cases in the scalp, face and more rarely the trunk, genitals and oral cavity have been reported.¹⁻⁶ The mean age of presentation is 57 years.² There is no clear gender predilection for extraocular SebCA.³ There is a positive association with Muir-Torre Syndrome, an autosomal dominant condition that manifests with multiple benign and malignant sebaceous carcinomas, keratoacanthomas and gastrointestinal visceral cancers.³

Extraocular SebCA is reported to present with yellow/tan coloured nodules which may be ulcerated. Our patient presented with an erythematous upper lip swelling, which is not typical. Diagnosis of SebCA is often delayed as it may appear inflammatory in nature as was the case in our patient.

US is a useful initial investigation in the assessment of soft tissue swellings of the head and neck region. US of the face is not routinely performed by sonographers or radiologists and this case was a useful reminder of the complex anatomy of the upper lip and cheek. Useful anatomical landmarks to identify are the orbicularis oris muscle, the intraoral mucosa and the dentition (Figure 3).

Figure 3. Transverse section US demonstrating normal anatomy of the upper lip: orbicularis oris (o), labial mucosa (lm) and the hyperechoic teeth (t)

Figure 4. Transverse section US of right cheek: masseter (*), buccinator (^), zygomaticus major (z) muscles, the modiolus (m) and the intraoral mucosa (im)

Figure 5. Transverse section US demonstrating depressor anguli oris (da) and the lower teeth (t)

The orbicularis oris muscle may be referred to as the oral sphincter muscle and occupies the entire width of the lips. It has no direct skeletal attachment, but merges with the modiolus.

The buccal commissure is commonly referred to as the corner of the mouth. Deep to the cutaneous tissues is the modiolus. The modiolus is a focus of multiple muscle tendons which end in the skin, mucous membrane of the upper lip or lateral to the corner of the mouth.⁷ Contributing muscles include buccinator, risorius, zygomaticus muscles, and the levator and depressor muscles of the upper and lower lips, respectively.

Figure 4 shows the normal anatomy of the commissure in transverse section. The modiolus is identified. Lateral to this the zygomaticus major muscle is well demonstrated and may be followed superiorly to its site of attachment on the temporal process of the zygomatic bone. The zygomaticus muscle pulls the corner of the mouth superiorly and laterally. The muscle appears wider towards the modiolus as it may be inseparable from the levator anguli oris and the minor zygomaticus muscles, whose function is to not only raise the corner of the mouth, but also the upper lip and to widen the nostril. The depressor anguli oris

(Figure 5), responsible for pulling the corner of mouth downward, runs inferiorly from the modiolus to the lower border of the mandible, where it interdigitates with the platysma muscle.

In this case US was a very useful initial investigative tool as it identified disease involvement of skin and muscle early on in the patient's care pathway. This information ultimately changed the patient's management from local surgical excision to planning for complex soft tissue reconstruction. Indeed, the treatment modality of choice for SebCA is surgical excision. However, this case also highlights the limitations of US. Although initially comparable to MRI in its assessment of disease invasion of superficial soft tissues of the face, the deep extent of disease cannot be identified on US. Contrast-enhanced MRI demonstrated disease spread into the retromolar trigone (Figure 2), pterygopalatine fossa and parapharyngeal space, none of which are easily visible on transcutaneous US. These findings changed the management again to that of palliative radiotherapy as the disease was considered unresectable.

Although an inflammatory process was a differential diagnosis, malignancy needed to be excluded. US was again useful as it facilitated guided FNA of the mass.

However, the cytology was misleading in this case as salivary cells were reported suggesting minor salivary gland tissue had been inadvertently sampled. An incisional biopsy revealed sebaceous carcinoma. Pathology, normally the 'gold standard' in reaching a definitive diagnosis, may often be confusing, as it can be difficult to distinguish between benign conditions, such as basal cell carcinoma, and malignant tumours and thus often requires multiple immunohistochemistry tests, including epithelial membrane antigen markers.¹

The behaviour of ocular and extraocular SebCA may be aggressive with local recurrence of approximately 40%¹ reported and distant metastases in 25% of cases.⁴ This may be due to the fact that SebCA can be multi-centric. Thus, in our patient the extensive disease may in fact have been amalgamation of multiple foci of disease as opposed to a large solitary tumour.

Conclusion

This case revises the normal US anatomy of the upper lip and cheek and applies it to the imaging of a patient with a rare cutaneous malignancy. This report highlights the advantages and disadvantages of US:

- US may be useful in aiding the early surgical planning for patients with extensive dermatological disease.
- US guided biopsy is straightforward to perform on superficial pathology involving the soft tissues of the face, although in this case the cytological findings were misleading.
- US may be considered comparable to MRI (an expensive, time-consuming procedure) in its assessment of disease invasion of superficial muscles and subcutaneous tissues of the face. However, US is very limited in its assessment of deep structures within the head and neck region, but may be useful in deciding what further investigations are appropriate.

Although the US findings of an infiltrative mass involving skin and muscle are shared by a variety of inflammatory and malignant conditions, the authors suggest that SebCA should be included in the list of differential diagnoses if further foci of disease are identified separate from the main body of disease.

DECLARATIONS

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