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Case report

Rare benign mixed tumour of the upper lip: A case report

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HIGHLIGHTS

- CS are rare mixed tumours originating in the sweat glands.
- CS are slow-growing, asymptomatic, subcutaneous, or interdermal nodules ranging from 0.5 to 3.0 cm.
- CS are observed in patients above 35 years with a male predilection.
- CS require complete excision, adequate surgical margin to enable diagnostic evaluation and prevent recurrence.

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ABSTRACT

Background: Chondroid syringomas (CS) are rare benign mixed tumours. Clinical differentiation can be misleading due to the silent presentation, with only histopathological findings confirming the diagnosis.
Case report: A 23-year-old Caucasian gentleman presented with an eighteen month history of increasing size of his exophytic upper lip mass.
The initial clinical impression was thought to be related to the skin. Following a punch biopsy, histopathology confirmed appearance in keeping with part of a chondroid syringoma with subsequent excision of the lesion.
Discussion: CS present as a slow-growing, asymptomatic, non-tender, nonulcerated, smooth, firm subcutaneous, or intradermal nodule and can range from 0.5 to 3.0 cm, predominantly occurring in the head and neck region in patients aged above 35 years with a male predication. The most effective diagnostic method is microscopic examination. The gold standard treatment modality is by complete excision with a margin of normal tissue in order to examine the histopathologic features and prevent recurrence.
Conclusion: CS should be included as a differential diagnosis of facial subcutaneous skin lesions in middle aged male patients. Careful evaluation, with a view of total excision and adequate surgical margin will enable diagnostic confirmation, whilst maintaining the aesthetic and functional unit.
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1. Background

Chondroid syringomas (CS) are rare benign mixed tumours that exhibit both epithelial and mesenchymal components, originating from the sweat glands [1]. Occurrence rates have been reported to be 0.01%–0.098% of all primary skin tumours [2], typically occurring in the head and neck region [2,3] between the ages of 20–60 years with a male predominance [4].

CS of the upper lip is a rare clinical entity, and to our knowledge there has have only been four reported cases in the English literature to date [5–8].

Clinical differentiation can be misleading and difficult due to the silent presentation, with only histopathological findings confirming the diagnosis. Complete excision is necessary in such cases.

2. Case report

A 23 year old gentleman presented with an eighteen month history of increasing size of his exophytic upper lip mass. A 23-year old Caucasian gentleman was referred by his general practitioner to our oral and maxillofacial (OMF) unit regarding an exophytic polypoid mass on his right upper lip. The lesion was asymptomatic and had been slowly increasing in size over an eighteen month period, Fig. 1.

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The gentleman's main concern was to exclude sinister causes and also address the aesthetic appearance. He denied any family history of similar lesions and this was the only solitary episode relating to the swelling. He was medically fit and well with no known allergies. He did not smoke and alcohol consumption was minimal.

On clinical examination, he presented with an oval, well circumscribed 15 mm diameter spheroidal swelling of his right upper lip. The lesion was fixed to the skin, smooth with a reddish hue. The lesion was in close proximity to his vermilion border. Following a punch biopsy, histopathology confirmed an appearance which was in keeping with a diagnosis of chondroid syringoma. The mass was subsequently excised

2.1. Investigations

At the time of presentation it was thought to be related to the skin and although at that stage, the nature of the lesion was uncertain, but working differential diagnosis of sebaceous cyst and dermoid cyst was made. A punch biopsy was carried out under local anaesthetic, 2 ml Lignospan *(2% lidocaine and 1: 80 0000 epinephrine)*, to exclude any sinister pathology and confirm diagnosis.

Histological finding demonstrated myxochondroid areas together with duct-like structures, suggestive of chondroid syringoma.

2.2. Treatment

Following discussions with the patient regarding the total excision, he wished to have the procedure carried out under general anaesthetic due to his nervous predisposition and anxiety of having the procedure carried out under local anaesthetic.

The lesion was then excised with a margin of healthy tissue which extended beyond the vermilion border of his upper right lip, Fig. 2.

Assessing the defect, a local flap reconstruction would have resulted in significant collateral scarring and deformity in a prominent facial region. The decision was then taken to allow healing via secondary intention. Topical chloramphenicol was applied to the wound bed, along with *Jelonet* and a *Hydrocell* non-adhesive foam dressing sutured in place for a week, to avoid opportunistic infections. Fig. 3.

The excised specimen was sent for histopathology, subsequently confirming an appearance in keeping with the diagnosis of chondroid syringoma, Fig. 4.

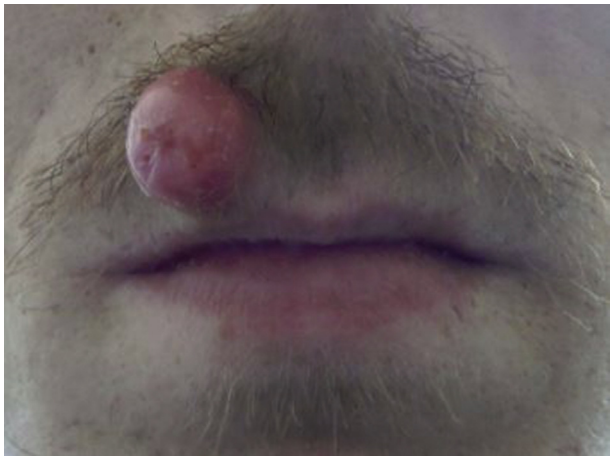

Fig. 1. The lesion at presentation.

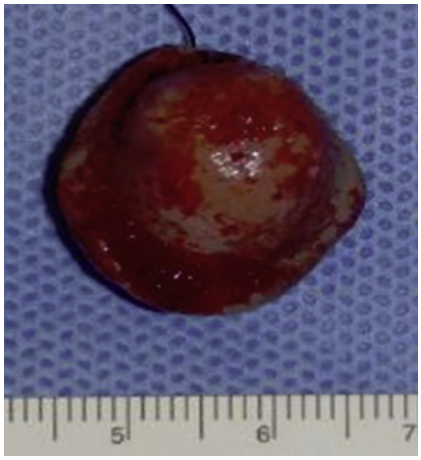

Fig. 2. Excised lesion.

2.3. Follow-up

The gentleman had an uneventful recovery with a satisfactory cosmetic and functional outcome, and did not require any further flap reconstruction, Figs. 5 and 6).

3. Discussion

CS present as slow-growing, asymptomatic, non-tender, non-ulcerated, smooth, firm subcutaneous, or intradermal nodules and can range from 0.5 to 3.0 cm [9–11], and may also exhibit a purplish or reddish hue [1,10]. The lesion is commonly mobile and distinct from the surrounding tissues [11,12]. However Sunguar et al. reported a benign case where rapid growth, ulceration and necrosis was evident at the tumour site [10]. They predominantly occur in the head and neck region with varying rates; 66% [3] – 80% [13] of cases. In descending order of frequency, the areas affected involve the nose, cheek, upper lip, scalp, forehead, and chin [1,3]. Other reported sites include the chest, abdomen, scrotum, penis, foot, eyelid, orbit, vulva, axilla and brain [1,9,11].

CS have been observed in patients aged above 35 [3,13,14], with a male predilection and a male-to-female ration of 2:1 [3,14].

Historically, Billroth in 1859 first identified these lesions as

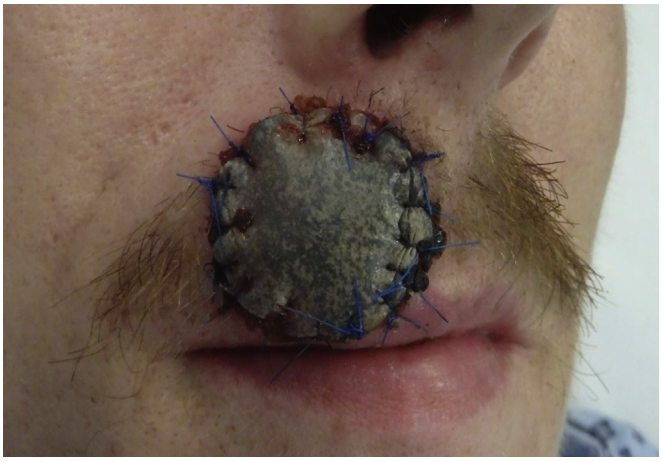

Fig. 3. Topical chloramphenicol applied to the wound bed, along with *Jelonet* and a *Hydrocell* non-adhesive foam dressing sutured in place for a week, to avoid opportunistic infections.

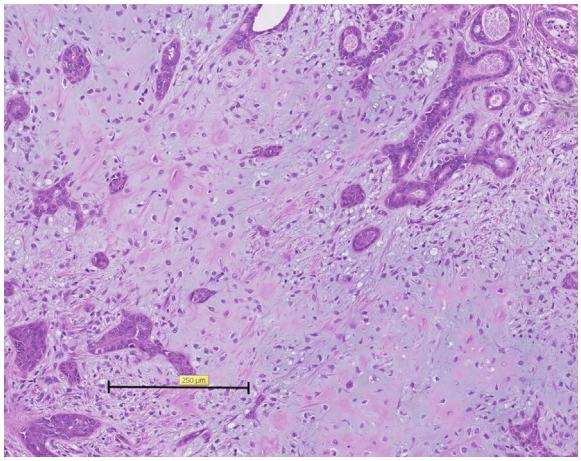

Fig. 4. Haematoxylin & eosin stain (x10) demonstrating myxochondroid areas together with duct-like structures in keeping with chondroid syringoma.

benign mixed tumours of the skin having the same histopathologically properties of the mixed tumours of the salivary glands [11]. In 1961, they were subsequently given the term by Hirsch and Helwig who proposed that they may originate either from apocrine or eccrine sweat glands [9]. CS may evolve from entrapped ectopic cell rest of embryologic origin stimulated to proliferate by an unknown factor [1].

Due to the rare and unremarkable clinical presentation, CS can be overlooked and misdiagnosed as skin lesions such as dermoid or sebaceous cysts, neurofibromas, dermatofibromas, basal cell carcinomas, squamous cell carcinomas, histicoytomas, pilomatrixomas, and seborrheic keratosis [1,9,14].

CS can also be preceded by trauma, in areas such as the sole of the foot where ulceration may occur [11].

The most effective diagnosis is confirmed by microscopic examination [15], requiring the total excision of the lesion.

Although CS are usually benign, malignant variants have been reported.

It is therefore important to obtain an incisional biopsy if in doubt to ensure appropriate management. Malignant chondroid syringomas (MCS) may arise de novo or from an incompletely excised benign CS that has recurred and undergone malignant transformation [12,16]. It is of note that no cases of malignant degeneration or transformation of benign tumour have been reported [1].

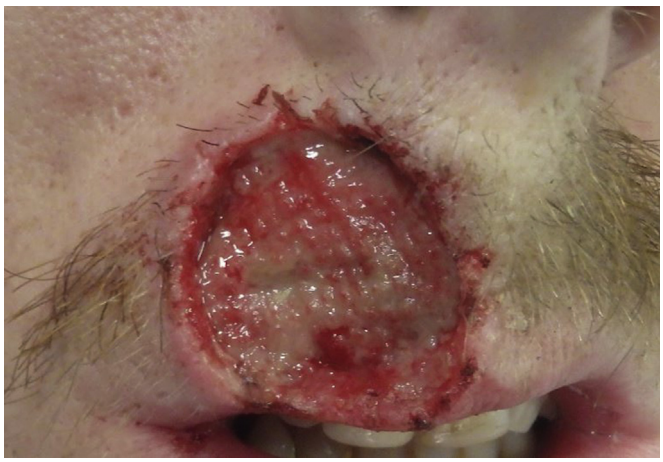

Fig. 5. One week post-op.

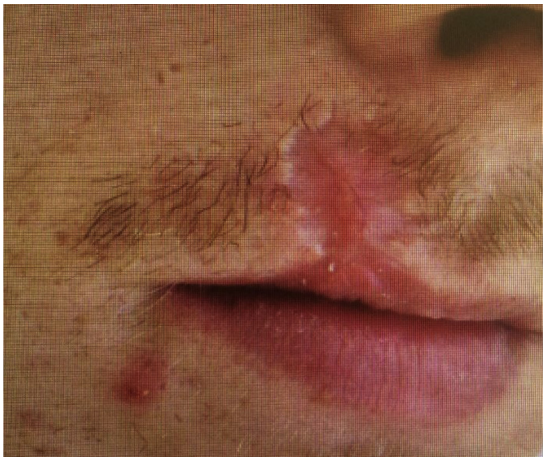

Fig. 6. Healed area 9 months post-op.

Microscopic findings are more aggressive and include nuclear pleomorphism, cytologic atypia, increased mitotic figures, infiltrative margin, satellite tumour nodules, and tumour necrosis [1,9]. Wide excision with a broad margin **is** the most reliable treatment to date [16].

Though adjuvant radiotherapy is often unsuccessful, skeletal metastasis have been shown to respond. Combination chemotherapy in patients with metastases is not reported to be beneficial [16].

4. Conclusion

CS of the upper lip are rare benign clinical entities, predominately occurring in middle aged male patients. Careful clinical and histological evaluation **is** required to obtain diagnosis, with a view of total excision and an adequate surgical margin to prevent recurrence. Healing via secondary intention could be considered to avoid unsightly scars from local flap reconstructions, and maintain the aesthetic and functional unit for a satisfactory patient outcome.

Ethical approval

None.

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Author contribution

Dr Sandra Girgis: Concept of case report, review of literature, critical revision and final approval.

Mr Gerrad Gillan: Critical revision and final approval.

Professor Kim Piper: Final approval.

Conflicts of interest

None.

Guarantor

Sandra Girgis.

Patient confidentiality

The report does not include any patient identifying features.

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