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| RESEARCH ARTICLE

Trichoblastoma: Case Report and Review of the Literature

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| ABSTRACT

Trichoblastomas are benign adnexal skin tumours that occur preferentially in hairy areas. Clinically, trichoblastomas appear as well-limited nodules or papules, which may be confused with basal cell carcinomas. A positive diagnosis is made on the basis of anatomopathological examination. The gold standard of treatment is still surgery, involving complete removal of the tumour with sufficient margins, up to 5-10 mm in aggressive forms, to avoid any recurrence or malignant degeneration. This paper reports a case of giant labial trichoblastoma, highlighting the diagnostic difficulties and the importance of rigorous management.

| KEYWORDS

Trichoblastoma, Surgery, Differential diagnosis.

| ARTICLE INFORMATION

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1. Introduction

Trichoblastomas are adnexal skin tumours, long considered rare [1]. They belong to the benign tumours with pilar differentiation, including the entity of trichoepitheliomas according to the WHO classification, and their preferential locations are the hairy regions [2,3]. Their main differential diagnosis is basal cell carcinoma. Anatomopathological examination plays an essential role in establishing the diagnosis, although the distinction between the two entities proves difficult in certain cases [4-7].

We report a case of giant labial trichoblastoma and stress the importance of correct diagnosis and management.

2. Clinical case

Patient aged 82, operated on for an inguinal hernia twenty years ago, chronic smoker, who has had a nodule on the upper white lip for five years, progressively increasing in size. At the time of consultation, there was a 2 cm long, non-pediculated nodule involving the right white hemi-lip and philtrum. The lesion was painless, non-pulsatile, with telangiectasias. On palpation, it was a hard nodule, immobile in relation to the two planes, with infiltration of the entire upper lip in depth, extending to the endobuccal mucosa and sparing the right lateral third of the lip. Palpation of the cervical lymph nodes was free. The patient's oral condition was poor and he was partially edentulous. No endobuccal lesions were found on clinical examination. A biopsy was performed, which came back in favour of a trichoblastoma.

At the multidisciplinary consultation meeting, no further investigations were requested. The decision was made to remove the tumour surgically without additional radiochemotherapy.

The tumour was clinically giant and infiltrative, so we performed a transfixing surgical excision with 5mm lateral margins, followed by reconstruction at the same time using a heterolabial Estlander flap combined with a contralateral Webster flap. The

definitive anatomopathological examination confirmed the diagnosis of trichoblastoma of complete resection. At three months post-operatively, the patient underwent a commissuroplasty under local anaesthetic to improve the mouth opening. The medium-term outcome was favorable (Fig 1-5).

Figure 1 : Clinical aspect of trichoblastoma with preoperative drawing of the Estlander heterolabial flap

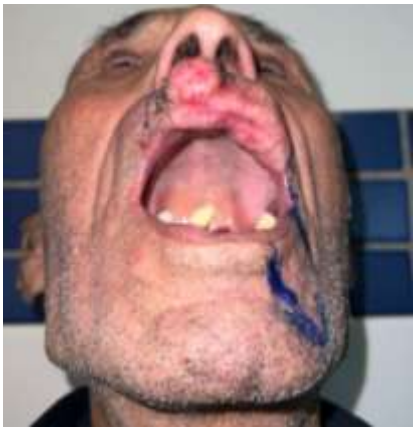

Figure 2 : Clinical aspect of endobuccal infiltration of trichoblastoma

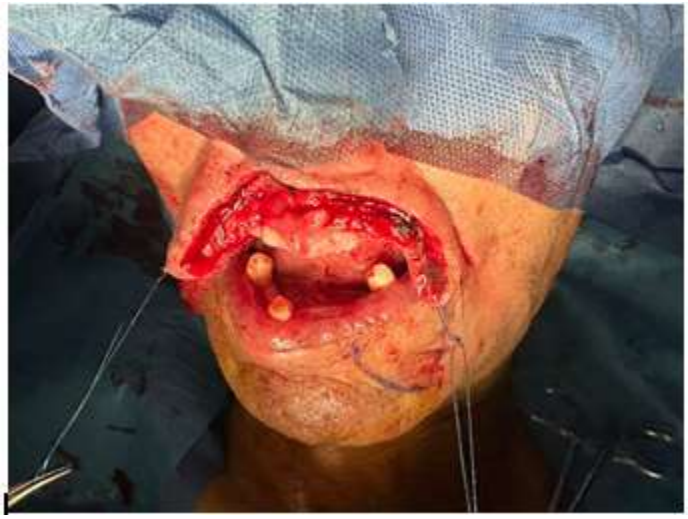

Figure 3 : Intraoperative aspect of surgical removal of trichoblastoma

Figure 4 : Immediate intraoperative appearance after reconstruction using a heterolabial Estlander flap combined with a contralateral Webster flap

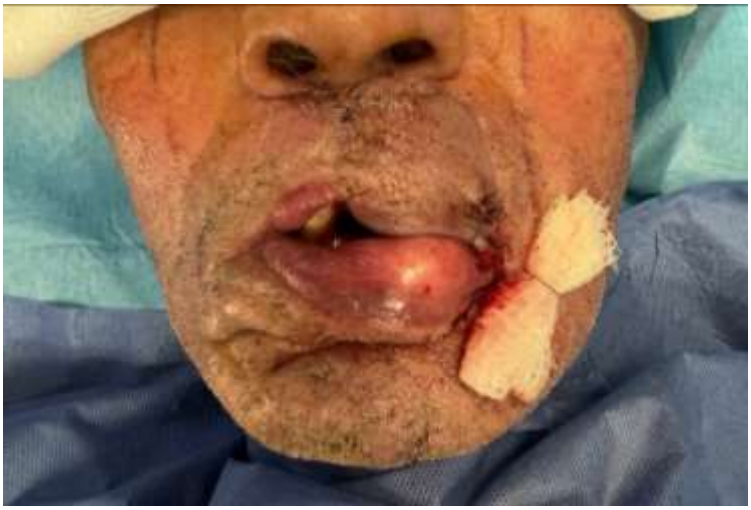

Figure 5 : Immediate intraoperative appearance after commissuroplasty, 3 months after the first operation

3. Discussion

Trichoblastomas are benign tumours, first described by Headington as follicular neoplasia. Over time and as new discoveries were made, the nomenclature evolved to retain the term "trichoblastoma" as the definitive term [8-13].

These benign tumours [14] are generally located in hairy areas, with a preference for the face and scalp, as well as the limbs, trunk and inguinal-perineal region [2].

Clinically, trichoblastoma presents as a papule or nodule, measuring 0.5 to 3cm in diameter, well limited, firm, with the presence of telangiectasias, pink in colour, sometimes pearly [15], causing it to be confused with basal cell carcinoma, with which it constitutes the main differential diagnosis; one study has shown that 2.6% of tumours considered to be basal cell carcinoma are trichoblastoma [1]. Other differential diagnoses include squamous cell carcinoma, trichilemmal carcinoma or skin metastases, and trichoadenoma in benign forms [7,16-18].

It is a dermal tumour, often localised. These trichoblastic tumours may be a benign secondary proliferation of a sebaceous nevus. Some forms are also distinguished by their large size, their polyploid nodular nature and the fact that they can invade the hypodermis and skeletal muscles, as in the case of our patient. The evolution of these so-called "giant" or "locally aggressive"

trichoblatomas remains poorly understood [2,19]. However, some trichoblastomas can degenerate into trichoblastic carcinoma [17].

The positive diagnosis is essentially a histological one of a tumour unrelated to the epidermis, with a characteristic image of invagination of spindle cells of the stroma within the epithelial lobules, suggestive of the papilla pilaris with the search for the PHLDA1 marker which is expressed in trichoblastoma and not present in other carcinomas [7,15,20-22].

Treatment is mainly surgical and is the "gold standard". It is based on a complete exeresis in order to limit the risk of local recurrence or even degeneration into a carcinoma, especially as the frequency of these risks is not well known [2,23]. Exeresis margins can be up to 5-10mm in giant aggressive forms [24,25]. In our case, and given the macroscopic appearance of the lesion, we opted for a transfixing excision with 5mm margins, which were sufficient for pathological examination.

4. Conclusion

Trichoblastomas are benign adnexal skin tumours. However, the giant and/or infiltrating forms are a challenge to manage and also a cause for concern about malignant degeneration.

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