

## Trichoblastic Carcinoma of the Upper Lip

COMPAORE Bertrand<sup>1</sup>, MOSSE Wilfried<sup>1</sup>, SEKA Evrard<sup>1</sup>, KIETGA Gael<sup>1</sup>, AGBANGLANON Patricia<sup>1</sup>, AKIMANA Gloria<sup>2</sup>, BOUTAYEB Salwa<sup>3</sup>, HARMOUCH Amal<sup>4</sup>, KEBDANI Tayeb<sup>1,5</sup>, BAKKALI Hanane<sup>1</sup>, BENJAAFAR Nouredine<sup>1,5</sup>

<sup>1</sup>Department of Radiotherapy, national Institute of Oncology, Rabat, Kingdom of Morocco

<sup>2</sup>Department of Medical Oncology, National Institute of Oncology, Rabat, Kingdom of Morocco

<sup>3</sup>Department of physical medical, National Institute of Oncology, Rabat, Kingdom of Morocco

<sup>4</sup>Anatomy pathology center of Kenitra, Kingdom of Morocco

<sup>5</sup>Department of Medicine and Pharmacy, Mohamed V University of Rabat, Kingdom of Morocco

\*Corresponding author: CB Ghislain, Department of radiotherapy, National Institute of Oncology, Rabat, Kingdom of Morocco, Tel: +212698912746; E-mail: compaorebertrandghislain@gmail.com

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### Abstract

**Introduction:** Trichoblastic carcinoma (TC) is a rare malignant tumour of pilar origin very often confused with basal cell carcinoma. It has un-codified and multidisciplinary care.

**Observation:** Moroccan patient, 69 years old, Farmer, non-smoking and without a history of pathology, presenting with trichoblastic carcinoma of the upper lip, resected twice, classified PT3N0M0 who received adjuvant interstitial brachytherapy.

**Discussion:** The discussion focused on the histological diagnosis of trichoblastic carcinoma and on the place occupied by interstitial brachytherapy in its management.

#### Keywords

Lip cancer; Trichoblastic carcinoma; interstitial brachytherapy

### Case Report

Moroccan patient, 69 years old, Farmer, non-smoker and without a history of pathology, presenting swelling of the upper left lip, measuring 3 cm approaching the midline, evolving for 14 months without other associated signs.

An initial biopsy of the lesion identified a desmoplastic trichoepithelioma without evidence of malignancy. The patient underwent a tumour resection a month later without lymph node dissection. Macroscopically, the lesion was budding, measuring 1.6 x 0.9 x 0.4 cm. Histologically, the lesion was unencapsulated dermal nodular without connection to the epidermis, infiltrating the dermis to the striated muscle. This lesion consisted of cell lobules and cords with basaloid-looking tumour cells, atypical with numerous mitoses, without retraction. The stroma was pilar, sometimes with calcifications and images of perineural sheaths suggesting trichoblastic carcinoma. The right limit of excision was tumoural.

The patient underwent surgical revision of the lesion without lymph node dissection a month later. Macroscopically, the lesion was pseudocystic and measured 2.1 x 1.5 x 0.4 cm. Histologically, the lesion was dermo-hypodermic, not connected to the epidermis made up of basaloid cells organized in small clumps without peripheral retraction with fibrous and inflammatory stroma evoking a residual trichoblastic carcinoma with internal and external borders which passed into the tumour zone.

A regional and remote extension workup, including head and neck CT scan and an AP chest x-ray, returned to normal. The patient was classified as PT3N0M0 (AJCC 8th edition 2017) with tumour surgical limits and perinervous sheaths, which motivated the performance of high dose rate interstitial brachytherapy.

#### Treatment

The patient received high dose rate interstitial brachytherapy

### Introduction

Trichoblastic carcinoma (TC) is a rare malignant tumour of pilar origin [1]. It is cancer that has locoregional but also general aggressiveness, with a high risk of recurrence [2]. Its diagnosis is difficult because it is very often confused with basal cell carcinoma (BCC) [2]. Its management is not codified because of its recent nosological individualization and the rarity of reported cases. We report a case of trichoblastic carcinoma treated at the National Institute of Oncology in Rabat.

(12 Grays / hour) and activity of 10 curies through the use of an iridium-192 source. The photons were delivered through non-radioactive plastic tubes used as conduction vector.

The implantation of these plastic tubes was performed in a brachytherapy operating room, under local anesthesia based on 1% lidocaine along the routes provided for the rigid vector needles, under optimal aseptic conditions. After measuring (**Figure 1**) we pierced the skin using rigid needles (**Figure 2**) which will serve as a vector for implanting the plastic tubes in a single plane.

The implantation rules (length and location of the sources) were made while respecting the conditions of the Paris system with a safety margin of 5 mm on either side of the tumor.

Treatment planning based on catheter coordinates was performed as the basis for interstitial dosimetry performed on an ONCENTRA Brachy planning system (**Figure 4**). The dose calculation was performed according to the Paris system with a prescribed dose at a fixed percentage of 85% of the basal dose.

After dosimetry, we had a radioactive length of 4 cm and a radioactive width of 2.2 cm. A dose of 5 grays was administered per delayed session by an iridium-192 source projector of the FLEXITRON type. Two fractions spaced 6 hours apart were performed per day for a total dose of 40 Grays in eight fractions and a 6-day spread with the goal of achieving an EQD2 = 50 Grays and a BED = 60 Grays.

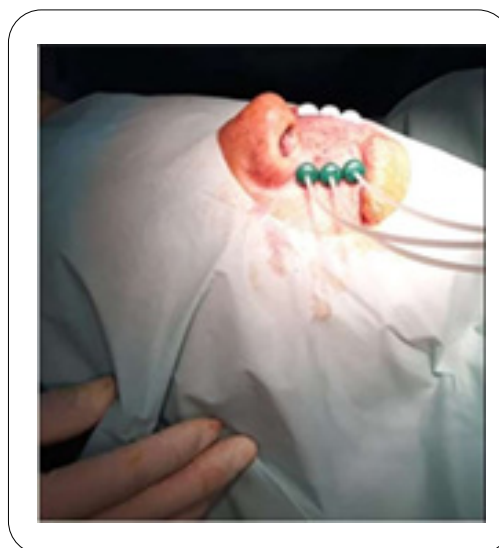
We did not perform nodes prophylactic irradiation or prophylactic cervical lymph node dissection.

Monitoring of the patient during treatment to reveal grade 2 radiodermatitis from the 4th fraction of brachytherapy treated with emollients.

The first follow-up after brachytherapy took place three months after the last fraction, the patient was in good local control without any sign of progression, neither of acute nor chronic complications. After a 9-month follow-up, the patient is in good locoregional control.



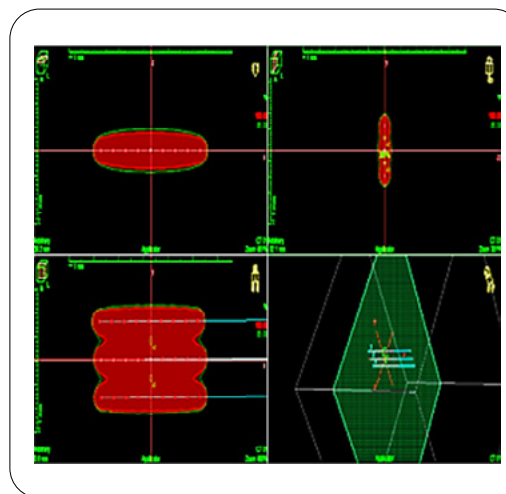
**Figure 2** Placement of vector needles.



**Figure 3** Installation of 3 non- radioactive plastic tubes.



**Figure 1** Measurements of the path of the vector needles.



**Figure 4** Interstitial brachytherapy dosimetry

## Discussion

Trichoblastic carcinoma accounts for approximately 8% of adnexal cancers of the skin [3]. It is a nosological entity that has recently been individualized and which is still not recognized as such by all pathologists. This explains why they are underdiagnosed; in the literature around thirty cases have been described [4]. It most often occur de novo, developed at the expense of the hair sheath epithelium but sometimes from trichoblastoma or following radiotherapy [5]. The epidemiological characteristics of my patient were similar to those found in the literature. In fact, trichoblastic carcinoma occurs mainly in elderly people with an average age of 70 years [6,7] and a male predominance objectified by a sex ratio of 2 [2]. It preferentially affects the face (60%) and the scalp, but it can also affect the torso, back, shoulders, and extremities [4]. Trichoblastic carcinoma has clinical and histological similarities with basal cell carcinoma. If it is impossible to differentiate them clinically, it remains possible, but difficult to do so histologically, because, in 95% of cases, TCs can be designated as a BCC during the first histologic study after a biopsy [2]. This difficulty is more marked when it comes to differentiating it from an infiltrating nodular-type BCC, for which 6% of cases are considered to be TCs [3].

The histological diagnosis of TC in our patient was made on an excisional piece, one month after a biopsy which found desmoplastic trichoepithelioma without a sign of malignancy. This diagnosis was confirmed on a second excisional piece after revision surgery indicated by the presence of a tumour surgical boundary after the first excision, thus highlighting the aggressive nature of the TCs. It is recognized in the literature, a degeneration of trichoepithelioma in trichoblastic carcinoma without specified the time of this transformation [2,3,4]. However, TC can be mistaken for trichoepithelioma if it is low grade [8] or if the amount of material to be examined is insufficient, which may be the case during a biopsy.

Like BCCs, TCs are made of basophil cells [9,10], but TCs are larger, less well-limited, and often shoot deep into muscle planes as shown in the images (Figures 5 and 6). Mitosis is common (Figure 7) in cancer cells. Tumour lobules are made of basophilic cells with characteristic trichilemmal-like keratinization (Figure 8). The key to diagnosis is the image of blue lobules on the periphery and red in the center. Some lobules can be centered by true cysts which are often calcified (Figure 9). Finally, at the periphery of the lobules, there is no palisade as clear as in the BCC whose diagnosis is based on the demonstration of a palisade arrangement of the tumour cells on the periphery of the tumour masses with the phenomenon of retraction of the stroma. Adjacent as well as rare mitoses and rare pleomorphisms [11].

We did not observe locoregional or distant metastases. Indeed, TC is better known for its local aggressiveness and its great ability to recur, however in high-grade TC, one can have lymph node 5% [2], bone or visceral metastases 10% [12].

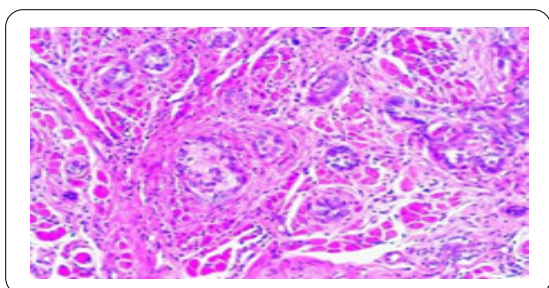


Figure 5 Muscle infiltration (G\*40)

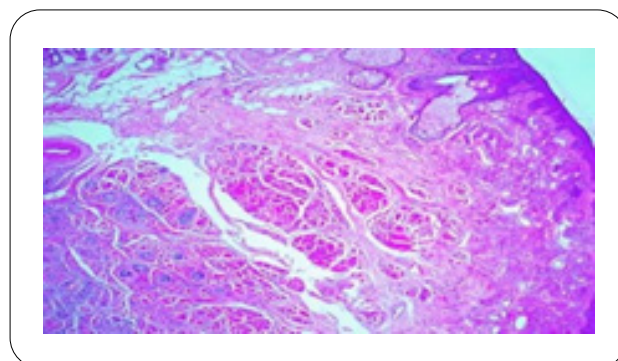


Figure 6 Muscle infiltration (G\*40)

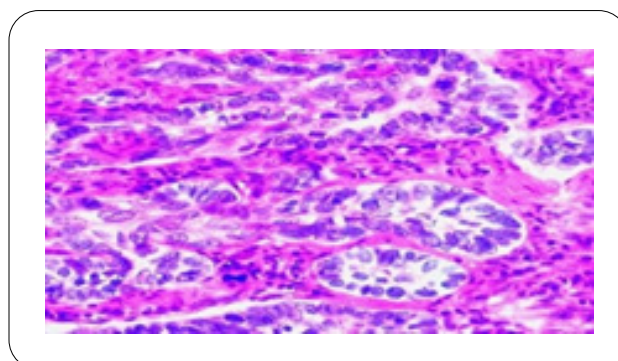


Figure 7 Characteristics of cancer cells (G\*40)

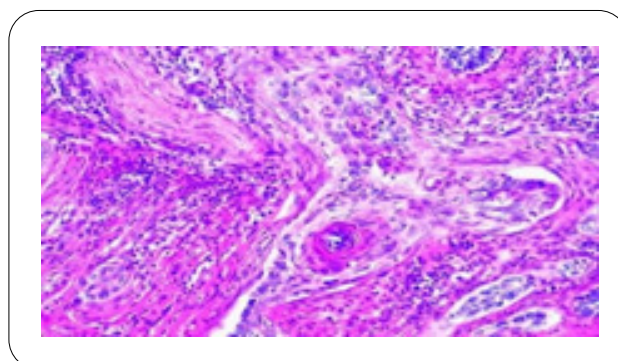


Figure 8 Keratinization (G\*40)

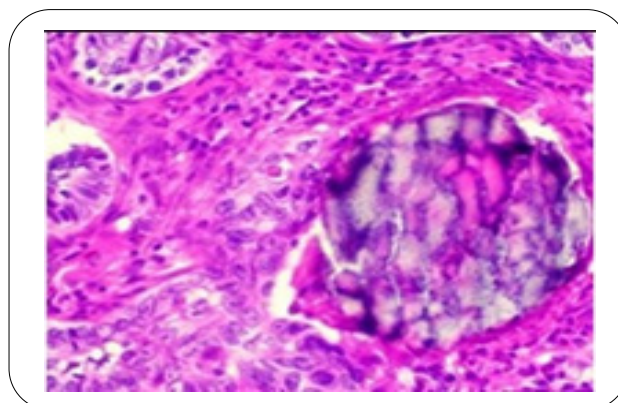


Figure 9 Central calcification of tumor masses (G\*40)

Treatment of TC is not codified due to the rarity of reported cases and their recent nosological individualization. Surgery is considered to be the gold standard in this treatment. The swelling representing only the visible part of the TC, it is necessary to perform surgery with

sufficient margins to hope to completely eradicate the tumour. This margin is estimated between 0.5 to 3 cm, depending on the authors. Our patient underwent two surgical excisions after which, we had tumour surgical limits. In fact, tumour margins are found in 67% of cases after the first surgery [2] and in the event of incomplete excision, these lesions recur and may extend further [13]. It often takes several rounds of excision to remove the entire tumour [13]. A third surgical revision would expose our patient to a cosmetic problem with the related psychological consequences, which motivated us to perform radiotherapy. The choice of radiotherapy technique was made according to the characteristics and location of the tumour.

The characteristics of the tumour (PT3N0M0 with EPNs and tumour limits) would have recommended external radiotherapy with irradiation of the lymph node areas. But the localization of the tumour at the level of the upper lip which is a poorly drained area, having a low rate of lymph node metastasis, for a pathology which causes 5% of lymph node metastasis. The CT scan made after surgery conformed the absence of node metastasis so it was possible to perform interstitial HDR brachytherapy. This brachytherapy made it possible to deliver a high dose to the tumour bed and with margins while minimizing the side effects of external radiotherapy.

In the literature, we did not find a case of TC treated by high dose rate interstitial brachytherapy, however, we did find the case of a patient with a palpébro-eyebrow TC who received external adjuvant radiotherapy to surgery, 50 grays with a good local, regional and remote control after a two-year follow-up [14].

Our patient is currently at 9 months of follow-up after HDR interstitial brachytherapy. The examination of the patient shows good local and regional control with no signs of progression or recurrence. However, in the literature, the recurrence rate can reach 17% beyond 18 months [2].

## Conclusion

Trichoblastic carcinoma is a relatively rare cancer of hair origin that often occurs in older people. Its histological diagnosis may be difficult due to its similarities to basocellular carcinoma. It is a very aggressive locally cancer, with a great ability to recur. Its management is not codified due to the rarity of reported cases and its recent nosological individualization. Surgery with sufficient margins remains the gold standard treatment. To increase the local control rate and reduce the risk of recurrence, it can be supplemented by radiotherapy, the choice of the technic external beam or brachytherapy will depend on the characteristics and location of the tumour.

## Ethics

Patient informed consent was obtained for publication of this case

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## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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