INTRODUCTION

Organ transplant recipients (OTR) are highly susceptible to developing skin neoplasms due to significant iatrogenic immunodeficiency. These tumors, occurring in more than 50% of transplant recipients, are often aggressive and multiple. They are mainly represented by squamous cell carcinomas (SCC) and basal cell carcinomas (BCC). Exceptionally, cases of sebaceous carcinoma (SC) in OTR have been reported. SC is an uncommon and potentially aggressive cutaneous tumor that usually occurs in adults, older than 60 years, on head, neck, and trunk. Herein, we describe the case of a patient presenting with SC and BCC 10 years after a kidney transplant.

CASE HISTORY AND EXAMINATION

The patient was a dark-skinned 55-year-old male with no personal or family history of neoplasia or previous irradiation. The patient received a kidney transplant in 2009. The protocol for immunosuppression was as follows: induction and maintenance with corticosteroids, mycophenolate mofetil, and cyclosporine. After 11 years post-transplant (2019), he developed a basal cell carcinoma of the nose, which was treated with surgical excision. In 2020, he presented with a tumor of the right ear. A clinical examination revealed a firm, 1 cm, erythematous, ulcerative-budding nodule located in the concha, suggesting SCC (Figure 1).
Histology revealed a multilobulated tumor of the deep dermis formed from small basophilic cells with sebaceous differentiation (Figure 2A, B). The diagnosis of SC was reached. Microsatellite instability was negative. The extension record was negative. Examination of the gastrointestinal tract, including endoscopy and colonoscopy, was performed in search of additional visceral tumors and excluded the possibility of Muir–Torre syndrome (MTS). Positron emission tomography was also performed, and no underlying malignancies were detected in any of these evaluations.

4 | MANAGEMENT AND FOLLOW-UP

The patient underwent wide local excision with lymph node dissection. He was followed up for 2 years after the operation with no evidence of recurrence. Short-term follow-up is still being performed.

5 | DISCUSSION

Sebaceous carcinoma is an aggressive adnexal tumor developed at the expense of the epithelial lining of the sebaceous glands. It is extremely rare, representing 0.7% of skin tumors. It may be sporadic or associated with Muir–Torre syndrome, a genodermatosis characterized by the association of sebaceous skin tumor, keratoacanthoma, visceral digestive, and urogenital neoplasms. Its main risk factors are advanced age, previous irradiation, UV rays, and immunosuppression. It has been described in OTR, yet such cases are exceptional. Indeed, the prevalence of SC with post-organ transplantation was 0.03% according to Sargen et al. A total of 102 cases were reported, 51% of which were kidney transplant recipients. This registry-based cohort study on 301,075 cases of OTR found a 25-fold increase in the overall risk for SC after transplantation. This risk increased over time suggesting the importance of chronic immunosuppression in the development of this tumor. Although CIs were similarly wide for the transplanted organ type, lung transplants appeared to be associated with the highest elevation in risk. Thus, in 60% of cases, it appears 5 years after transplantation. The occurrence of SCC or BCC post-transplant, as in our patient's case, was also associated with an increased risk of SC in this population. SC has traditionally been divided into two groups: the periocular variant comprising approx. 75% of cases and the extraocular or cutaneous form. The latter is the form described in OTRs. It commonly presents on the head and neck, where sebaceous glands are more concentrated. Approx. one quarter of extraocular tumors arise in other regions, including the trunk, thighs, and rarely the genitalia. Other exceptionally affected sites include the nasal vestibule, breasts, fingers, feet, and external auditory canal, as in our patient. The involvement of the outer ear is observed in only 3% of cases. There are less than fifteen known cases in the literature originating from the external auditory canal (EAC). This tumor is difficult to diagnose due to its clinical and histological polymorphism. The clinical appearance in OTRs is similar to that observed in the general population. It presents as a small, non-specific, slowly growing, ulcerative, erythematous to slightly yellow nodule resembling an SCC or BCC. Because of potential misdiagnosis, correct pathological identification is highly important. Histology reveals a multilobulated tumor of the deep dermis formed from small basophilic cells with sebaceous differentiation (delicate and multiple, cytoplasmic, foamy vesicles),
high mitotic activity, and nuclear pleomorphism. The treatment of SC requires extensive surgical excision with lymph node dissection, whether or not associated with adjuvant radiotherapy or chemotherapy. Our patient had no lymphovascular invasion and no metastasis; therefore, no other treatment besides excision was included. The prognosis depends above all on the early diagnosis. It is aggressive in 29% of cases and the risk of lymphatic and visceral metastasis is common.

6 | CONCLUSION

The high risk of skin cancer after organ transplantation is a major clinical challenge. Although it is exceedingly rare, SC should always be considered in the presence of any skin lesion occurring after a transplant. Regular dermatological monitoring is necessary in order to identify and treat it in an early stage.

AUTHOR CONTRIBUTION

Dr. Refka Frioui is the guarantor of the content of the manuscript, included the data and analysis. Dr. Faten Rabhi contributed to interpretation of data and revision of the manuscript. Dr. Faten Gargouri is the dermatopathologist who analyzed the histological images. Dr. Kahena Jabeur and Dr Mohamed Raouf Dhaoui contributed to analysis and interpretation of data, revised it critically for important intellectual content, and final approval of the version to be submitted.

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CONFLICTS OF INTERESTS

The authors declare that there are no conflicts of interest in this work.

DATA AVAILABILITY STATEMENT

All data generated are included in this published article.

ETHICAL APPROVAL

Informed consent was obtained from the patient.

CONSENT

A written informed consent was obtained from the patient to publish this report in accordance with the journal’s patient consent policy.

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