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Case Report

Cutaneous and lip squamous cell carcinomas in an albinism patient: A case report

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ABSTRACT

Introduction and importance: The incidence of squamous cell carcinoma (SCC) in the population aged under 30 years reaches only 1% of total cases. We report the first case from Papua, Indonesia, with a double primary SCC in a patient aged just 25 years, with albinism as a risk factor. This case report can hopefully enrich existing knowledge of such tumors.

Case presentation: A 25-year-old Papuan female patient came to the oncology clinic of Jayapura Regional General Hospital with a tumor on the left lower lip and a skin tumor on the right temporal side of the face. The patient noticed that the tumor on the lower lip appeared a few weeks earlier than that on the right temporal skin. Both tumors had grown gradually for 5 years. Both tumors were painless, but for the last 3 months, the tumor had bled easily. The patient was born with oculocutaneous albinism (OCA) without other syndromic or systemic disorders.

Clinical discussion and conclusion: In this patient, we suspected double primary SCCs considering the location of the tumor, which theoretically spreads distantly; the size of the lesion at less than 2 cm; the depth of the lesion at less than 4 mm; and the well-differentiated cytology. Another consideration was that patients with OCA have a 1000-fold risk of developing skin cancer and an increased risk of recurrence. Therefore, continual evaluation and screening are necessary.

1. Introduction

Cutaneous squamous cell carcinoma (SCC) is the second most common skin cancer after basal cell carcinoma. SCC of the lip accounts for 25% of the total cases of oral cancer. The pathogenesis and risk factors of SCC are multifactorial, including ultraviolet exposure, immunosuppression, human papillomavirus (HPV) infection, trauma, genetic disorders, and age over 60 years [1–5].

Multiple SCCs can occur if a new primary SCC is formed elsewhere or the SCC metastasizes. SCC metastasis can be lymphogenic and hematogenic, influenced by several risk factors such as tumor size, tumor depth, and tumor differentiation [6].

The incidence of SCC cases in the population aged under 30 years reaches only 1% of the total cases. We report the first case from Papua, Indonesia, with double primary SCCs in a patient aged just 25 years, with albinism as a risk factor. This case report can hopefully enrich existing relevant literacy. This report was written following the 2020 Surgical Case Report guidelines [2,3,7].

2. Presentation of case

A 25-year-old Papuan female patient visited to the oncology clinic of Jayapura Regional General Hospital using a public transportation, presented with a tumor on the left lower lip and a skin tumor on the right temporal side of the face. The patient noticed that the tumor on the lower lip had appeared a few weeks earlier than the tumor on the right temporal skin. Both tumors had grown gradually for 5 years. Both tumors were painless, but for the last 3 months, the tumor had bled easily. The patient was born with oculocutaneous albinism (OCA) without other syndromic or systemic disorders. She was a homemaker with minimal sun exposure, no smoking history, and no history of arsenic exposure, radiation, burns, scars, or other relevant medical conditions. Previously, the patient had never had any examination or treatment. No family history existed of similar disease.

On physical examination of the lips, a tumor measuring 1 × 2 cm was found on the left lower lip, accompanied by ulceration and blood. The tumor was reddish with necrotic tissue at the edges. The margin of the tumor was irregular and extended beyond the inferior margin of the

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patient's left lower lip. On palpation, the tumor was spongy, fixed to the orbicularis oris muscle, and brittle and bled easily. There was no significant pain.

On physical examination of the right temporal area, a tumor measuring 1.5 × 1 cm was found with ulceration and blood. The tumor was reddish with necrotic tissue at the edges. The tumor margins appeared irregular. On palpation, the tumor felt solid and fixed, bled easily, and was painless. No tumors were found in other locations, and no enlarged lymph nodes were found.

The patient lived in a rural area far from the location of the surgical procedure. Considering this, along with her risk factors, clinical presentation, and financial capabilities, we performed a one-staged procedure surgery with general anesthesia. In the temporal region tumor, a wide excision was performed. In the left inferior labialis tumor, a V-shaped wide excision was performed. The defect was sutured primarily, layer by layer, using 4.0 polydioxanone and 5.0 nylon threads. After the surgery, the patient was given antibiotics and painkillers. She went home in good condition 1 day after surgery.

The two excised tissues showed the following histopathological results: a low grade keratinizing squamous cell carcinoma, well-differentiated. Based on the staging of the American Joint Committee on Cancer 8th Edition, it was T1N0M0 with stage I [8]. During the surgical procedure, treatment, and follow-up to day 5, no complications occurred and the patient was satisfied. At follow-up 12 months after surgery, no new tumors were found. The surgical procedures and treatments were performed by oncology surgeons in a hospital setting.

3. Discussion

One of the risk factors for cutaneous and lip SCC is older age. A cross-sectional study by Czerninski et al. on the incidence of oral cancer in Israel showed that 75.5% of oral cancer cases from 1970 to 2006 were among those aged 53 years and over. Only 19 of the 4337 patients (0.4%) were under 20 years of age. Another study by Xiang et al. extracting data from eligible published article from 1978 to 2012 described that the incidence of cutaneous SCC increases with age, reflecting the harmful effects of cumulative sun exposure [9,10].

In this case, tumor growth had been noticed since the patient was 20 years old. A risk factor that may be the reason for tumor growth at a relatively young age is the patient's congenital OCA. OCA is a condition where there is a disruption in the biosynthesis of melatonin so patients with OCA have a 1000-fold risk of developing skin cancer. A retrospective review from 64 cases by Mabula et al. showed that OCA in dark pigmented people is associated with a greater risk of SCC [11,12].

The spread of SCC can occur through various mechanisms such as infiltration of surrounding tissues, shelving, skating, and metastasis through lymphatic and blood flow. In this patient, two different sites of tumor growth existed, which we suspected were the growth of two primary tumors concurrently. This was supported by the location of the tumor, which theoretically spreads distantly; the size of the lesion at less than 2 cm; the depth of the lesion at less than 4 mm; and well-differentiated cytology [13,14].

Several previous reports and reviews have shown that patients with multiple primary SCCs generally have risk factors such as a history of HPV infection, currently taking immunosuppressant drugs, xeroderma pigmentosum, and albinism [15–17].

The recurrence rate of SCC is influenced by several factors, including relatively young onset. In addition, OCA, exacerbated in this case by the fact that the patient was dark pigmented, is a condition that increases the risk of recurrence. On 6-month and 1-year evaluations after surgery, no new tumor lesions were found, but continual education and follow-up are still necessary and recommended for patients [11,13].

As in Papua, there is no any genomic testing available that could confirm which albinism type presented in this case. However, we encouraged for the future work to conduct such test.

4. Conclusion

This is the first multiple SCC case to be reported from Papua, Indonesia. Its rareness and attractive patient presentation are the reasons the authors report this case. This case report will hopefully contribute to increasing the existing knowledge.

Ethical approval

No ethical approval is needed for this type of publication in our institution. However, the patient has given consent to this publication.

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This case report received no specific grant.

Author contribution

Jan Frits Siauta: study concept or design, surgeon, data collection, data analysis or interpretation, writing the paper, Carolus Aldo Windura: data collection, data analysis or interpretation, writing the paper, Louis Kartono Putra: data collection, data analysis or interpretation, writing the paper.

Please state any conflicts of interest

None.

Registration of research studies

1. Name of the registry:
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

Carolus Aldo Windura.

Consent

Written informed consent was obtained from the patient for publication of this case report.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.104556.

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