

CASE REPORT

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Basal Cell Carcinoma Arising from a Port Wine Stain

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Abstract

Objective

In this study, we present the diagnosis, clinical approach, and treatment process of BCC developing from the PWS background.

Material and Method

This study is a case report of a patient who applied to our clinic in March 2023.

Results

Most of the BCCs appear in people over the age of 50 and those who have a long exposure to the sun. It's

possible to cure it with local wide surgical excision and adjuvant treatment. It has been documented in case reports that this phenomenon arises from PWS, which is a congenital vascular malformation.

Conclusion

Topical or radiation therapy that we use while treating various skin diseases can also increase the risk of BCC. The following presentation outlines the approach and treatment algorithm for BCC developing on the former known PWS.

Keywords: Basal cell carcinoma, hemangioma, port wine stain, skin tumor.

Introduction

Basal cell carcinoma (BCC) is the most common skin tumor (1). It is especially common in sun-exposed areas such as the head, neck, and trunk. The absence of a precursor lesion in this cancer requires caution against newly developing lesions in possible areas (2). There are three distinct classifications of BCC: superficial, nodular, and morpheaform. (3). In cases where a diagnosis is suspected, the primary method of investigation is dermoscopic examination. However, excisional biopsy is considered the optimal approach for differential diagnosis.

The most important risk factors are exposure to sunlight and its cumulative effect with advancing age. People with type 1 and 2 features on the Fitzpatrick skin scale have a higher risk (4,5). In addition, we know that this skin tumor is seen in people with congenital and acquired skin anomalies, as in our case. For example, BCCs are seen in multiple foci in genetically inherited syndromes such as the nevoid basal cell carcinoma syndrome (1). It can also be observed that BCC lesions are frequently seen in syndromes such as Gorlin syndrome, xeroderma pigmentosum, basex-dupre-christol, and rombo syndrome (6).

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Port wine stain (PWS), also known as nevus flammeus, is a capillary vascular malformation in the dermatome area of the 5th cranial nerve (7). The stain, which appears as a pink colour at birth, turns purple in later ages and creates a rough facial appearance. This congenital stain tends to be unilateral. It has also been reported to occur not only at birth but also at later ages, especially after trauma during adolescence. In congenital cases, additional investigations and imaging methods are required to exclude syndromes. The present study reports a case of BCC arising from a port wine stain.

Case Report

A 60-year-old male patient was referred to us for the evaluation of a biopsy by the Dermatology department with a lesion on the nose, which has existed for about 7-8 years, and has been growing rapidly for 8 months. The patient was examined, and it was observed that a congenital port wine stain was extending from the left facial half v1 and v2 of the trigeminal dermatome area to the left nasal half and the left temporal region. We also observed that the patient had a complete loss of vision in half of the face affected by the port wine

stain. The patient's photograph is shown in Figure 1.

In the first stage, an incisional biopsy was planned to confirm the diagnosis. The histopathology report resulted in basal cell carcinoma. Therefore, surgical excision was planned to extirpate the tumoral lesion. The patient was found to have polycythemia and high blood sugar during preoperative preparations. In addition, it was informed that the presence of polycythemia was not a problem in terms of anesthesia.

The patient was operated on under general anesthesia 3x2 cm tumor involving the nasal tip and supra tip region was excised with a 6 mm clean surgical margin. Clear margins were revealed by frozen section biopsy. The surgical reconstruction was planned with the defect covered with the paramedian forehead flap based on the defect dimension. The flap donor site was closed with -thickness skin graft from the groin area. Two weeks later, the flap was separated from the midline of the nasal dorsum, and the transferring portion of the flap was repositioned to the forehead. The distal end of the flap was sutured with 6/0 Prolene. The post-operative photograph is presented in Figure 2.



Figure 1

A 60-year-old male patient presenting with a port-wine stain (PWS) along the dermatome distribution of the left ophthalmic (V1) and maxillary (V2) branches of the trigeminal nerve, accompanied by a lesion on the tip of the nose



Figure 2

Postoperative frontal and left lateral views of the patient, taken two weeks after the procedure.

Discussion

Basal cell carcinoma (BCC) is the most prevalent form of skin cancer, with ultraviolet (UV) radiation from sunlight identified as the primary risk factor. The most effective method of prevention is to minimize exposure to sunlight. However, a multitude of factors have been posited as potential triggers for tumor formation, including arsenic, burns, and chronic irritation in areas not exposed to sunlight (8). Radiation exposure is also associated with this type of cancer.

PWS is the most common vascular malformation seen in humans (8). PWS is seen with enlargement of papillary dermal capillaries and venules on histological examination.

In our literature review, we found 79 cases of BCC arising from PWS in 36 patients, although we did not find data showing that PWS directly increases the risk of this type of cancer (9).

When we re-examined the causes of BCC, we found that topical radiotherapy (thorium X) used in the treatment of PWS was among these causes. This treatment method was among the possible causative factors for the cases encountered during the literature review. However, this treatment method has already been abandoned in the treatment of PWS, and current treatment methods, argon laser and pulsed dye laser, are defined as the gold standard (10). However, BCC

cases developing from PWS also occur de novo. In cases of BCC that developed de novo on PWS, there was an increase in growth factors. The hypothesis was that increased vascularization and vascular ectasia were the causative factors. The increased growth hormones made the epidermis more vulnerable to sunlight, and this is considered to be a de novo developmental mechanism (8). It is also thought that the blood flow irregularity in PWS may contribute to the formation of BCC.

When the skin structure of port wine stain cases is examined histologically, epidermal atypia and structural changes in basal keratinocytes are encountered. This makes this skin vulnerable to cancerization. Again, the reflection of the same histological area in the clinic is seen in multiple foci. Again, the unclear reflection of the stain boundary lines makes the diagnosis difficult. Especially cases that have received the topical radiotherapies mentioned in the past or who have broadly limited port wine stains, as in our case, should be followed up dermatologically. It is thought that these structural changes in the background of port wine stains will play a triggering role in tumor development (10).

The most reasonable method to consider de novo tumor development is the exclusion of possible causes. Our patient has not received any PWS treatment (thorium X or laser) before. Hence, in our patient, the fact that the patient has not received

topical radiotherapy brings us closer to a de novo mechanism, while the sun exposure of the facial area for years distances us from the possibility of de novo. The increased growth hormone level due to increased vascularization and vascular ectasia in port wine stain confuses whether the developing cancer is caused by sunlight or whether sunlight triggers de novo mechanisms in sensitized skin.

Since the normal skin appearance is disrupted in areas affected by PWS, the risk of this tumor increases in patients who have previously received radiotherapy for PWS, and the clinician should pay attention to the risk of skin cancer development in the patients they follow up with. GNAQ mutation has also been found in non-syndromic port wine stain patients (11). However, its association with basal cell carcinoma has not been demonstrated (12).

BCC is a tumor known for its low recurrence rate and low risk of metastasis in general. Treatment is planned according to the risk ratio determined according to the National Comprehensive Cancer Network guidelines. Following a risk assessment of the patients, non-surgical treatment options are employed in low-risk cases, and surgical excision is used in high-risk cases. It can thus be concluded that the most effective treatment method is Mosh surgery (5).

Consequently, BCC is a common skin cancer. Surgical excision is the gold standard treatment modality in high-risk cases. In this case, we present our approach to a high-risk case arising from PWS in an elderly male patient. Our clinical recommendation for reconstruction in large-sized defects is the paramedian forehead flap.

Conflict of Interest Statement

The authors declare no conflict of interest.

Ethical Approval

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Consent to Participate and Publish

Informed consent was obtained from patients for being included in the study. Presented in additional page

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Availability of Data and Materials

Data available on request from the authors.

Artificial Intelligence Statements

The authors declare that they have not used any generative artificial intelligence for the writing of this manuscript, nor for the creation of images, graphics, tables, or their corresponding captions.

Authors Contributions

YB: Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Validation; Visualization; Writing-original draft.

BCI: Investigation; Validation; Writing-original draft.

EA: Conceptualization; Formal analysis; Funding acquisition; Investigation; Methodology; Project administration; Resources; Supervision; Validation; Writing-review & editing.

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