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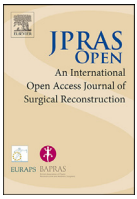

Original Article

Multiple Foci of Basal Cell Carcinoma Arising in Rhinophyma: A Case Report and Literature Review

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ABSTRACT

Background: Rhinophyma is a benign condition caused by the excessive growth of sebaceous glands in the nasal tissue, presenting with symptoms such as nasal hypertrophy, erythema, and papules. Cases of basal cell carcinoma in rhinophyma have been reported in literature, but its etiological role remains unclear. It is uncertain whether rhinophyma is predisposed to neoplasm development or if their coexistence is coincidental.
Material and Method: We conducted a literature survey to identify such cases reported over the years.
Results: We identified 22 studies reporting a total of 47 cases in the literature, all involving male patients. The most common pattern of occurrence was the rapid growth of a nodular formation within the context of rhinophyma.

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Discussions and Conclusion: The elucidation of the association between basal cell carcinoma and rhinophyma remains challenging. The presence of multiple foci supports the theory that rhinophyma may play a role in their development, but larger studies are needed to establish a causal relationship.

Introduction

Rhinophyma is a benign condition characterized by the excessive growth of sebaceous glands in the nasal tissue.[1] It primarily affects Caucasians[2] between the fifth and seventh decade of their life.[3] Rhinophyma is more commonly observed in male patients than in female patients (with a ratio ranging from 12:1 to 30:1).[4] This gender disparity has led some researchers to hypothesize the potential role of androgen hormones in its etiology.[2] Rhinophyma is commonly associated with a history of rosacea,[5] alcohol abuse, nutritional deficiencies,[6] and prolonged steroid therapy.[7] Clinical presentation of rhinophyma includes hypertrophy of nasal soft tissues, erythema, telangiectasia, and papules.[8] Various treatment modalities, ranging from medical therapy to laser or mechanical ablation and surgical intervention[9] are available.

Typically, neoplasms such as basal cell carcinoma (present in approximately 3% of cases[10]), squamous cell carcinoma,[10] schwannoma,[11] cutaneous angiosarcoma,[12] and B-cell neoplasm[13] coexist with rhinophyma. However, it has not been clearly established whether rhinophyma itself acts as a precancerous condition, especially since these neoplasms are typically identified in a single location and in a clinically significant stage. Herein, we present a case (written in line with the Surgical Case Report (SCARE) criteria[14]) and describe the uniqueness of multiple preclinical stage basal cell carcinoma foci in a patient treated for rhinophyma.

Case Report

A 65-year-old male patient presented to our department with a rhinophyma that had been present for approximately 6 years and previously untreated. Upon initial examination, there was evidence of hypertrophic, sebaceous skin without any suspicious nodular formations or sebaceous drainage (Figure 1). The patient complained of respiratory problems due to nasal ostium compression. No other comorbidities or history of other facial skin neoplasms were reported. Tangential excision of the skin was performed using an electrosurgical knife. The excised tissue was sent to the pathology department, which reported the presence of “rare superficial nests of basal cell carcinoma within the excision margins” (Figure 2a-c). Subsequently, the patient was followed up in our clinic and achieved complete healing of the nasal skin in the following 3 weeks (Figure 3a-b).

Materials and Methods

We conducted a literature search on PubMed and Scopus using the keywords “rhinophyma,” “basal cell carcinoma,” “basal cell,” and “basal cell neoplasms”. We excluded studies that were not case reports, case series, cohort studies, or case-control studies.[15] The language of the articles was not a criterion for exclusion. Additionally, we manually scanned the citations within the identified papers. We extracted only the data pertaining to the incidence of this specific neoplasm from studies that also examined the occurrence of neoplasms other than basal cell carcinoma.

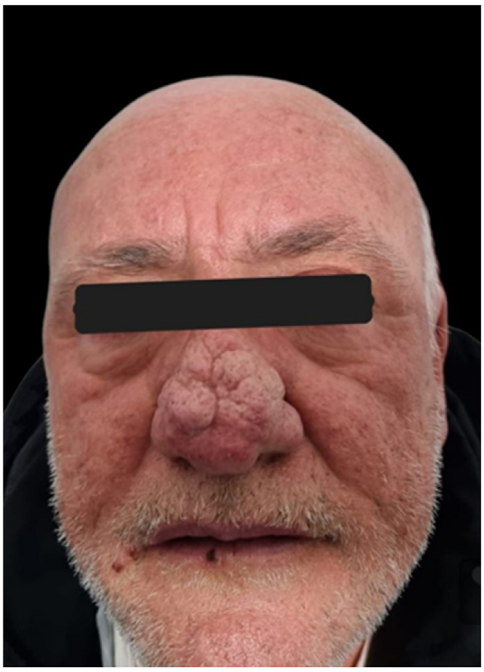

Figure 1. Preoperative appearance.

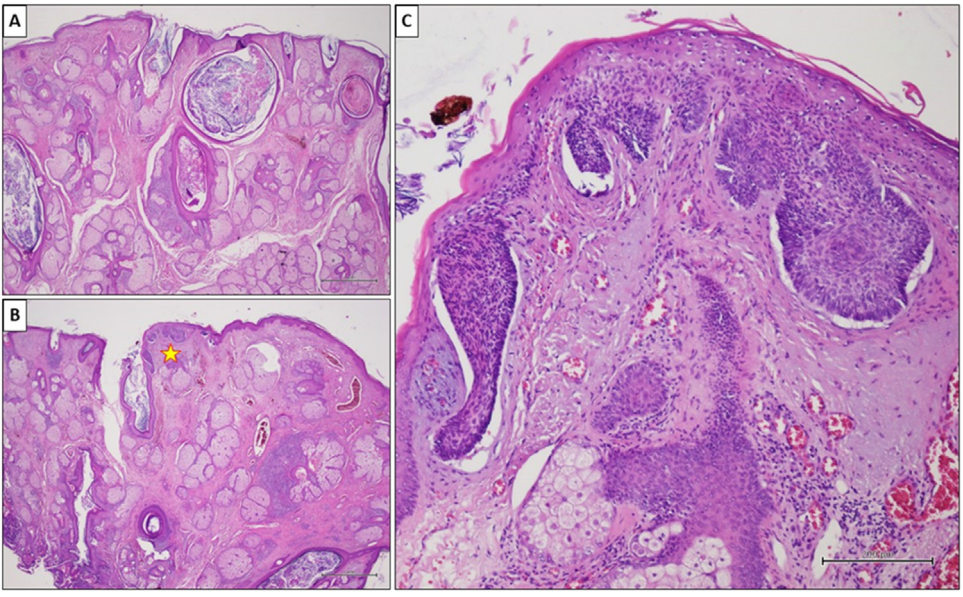

Figure 2a-c. Histological findings. Histological examination results showing sebaceous gland hypertrophy, follicular plugging, and peri-infundibular lymphocytic inflammation, consistent with the diagnosis of rhinophyma (A and B, hematoxylin and eosin stain, original magnification 20 ×). In this morphological setting, a superficial basal cell carcinoma is present (B, yellow star). The typical morphological findings of basal cell carcinoma are shown in (C), including epidermal attachment of neoplastic lobules, peripheral palisading, and cleft formation between tumor lobules and stroma (C, hematoxylin and eosin stain, original magnification 200 ×).

Figure 3a-b. Postoperative appearance.

Results

The occurrence of basal cell carcinomas arising from rhinophyma was described over a century ago,[16] with some authors reporting incidence of neoplasms between 15% and 30% in patients with rhinophyma.[10,17,18] In our literature analysis, we identified 22[10,16, 18-37](Tables 1, 2) studies reporting a total of 47 documented cases of neoplasm. In most cases (40), the neoplasm was clinically detectable. Several reported cases involved a single basocellular neoplasm, although in some instances, it was accompanied by other types of neoplasms such as squamous cell carcinoma and sebaceous adenoma. Surgical excision was the predominant treatment modality reported.

The first reported case in the literature described a 76-year-old man with a 10-year history of rhinophyma.[16] Subsequently, there have been anecdotal case reports.[18–20] The first fairly structured analysis was conducted in 1967[10] in a study involving 47 veterans (45 men and 2 women) with a history of rhinophyma lasting over an average duration of approximately 8 years. Histological analysis of surgical specimens revealed 5 cases of basal cell carcinoma, 1 case of adenoacanthoma, and 1 case of sebaceous adenoma.

Table 1
Reports of basal cell carcinoma arising on Rhinophyma.

Year	Author	Number of patients	Type of tumor	Clinical features
1904	Wende et al.[16]	1	Basal cell carcinoma	Not specified
1931	Eisenklam et al.[19]	1	Basal cell carcinoma	Not specified
1955	Rees[18]	2	Basal cell carcinoma	In situ
1967	Acker et al.[10]	5	Basal cell carcinoma	Nodular
1970	Freeman[20]	1	Basal cell carcinoma	In situ
1970	Fisher[3]	1	Basal cell carcinoma	In situ
1973	Kornblut et al.[22]	1	Basal cell carcinoma	Infiltrated
1977	Brubaker et al.[23]	1	Basal cell carcinoma	Nodular
1980	Preaux[24]	1	Basal cell carcinoma	Not specified
1984	Barhelemy et col.[25]	14	Basal cell carcinoma	Not specified
1988	Keefe et al.[26]	1	Basal cell carcinoma	Nodular
1990	Silvis et al.[27]	1	Basal cell carcinoma	Infiltrated
1998	Baruchin et al.[28]	4	Basal cell carcinoma	Nodular, ulcerated, nodular, in situ
1999	Tamir et al.[29]	1	Basal cell carcinoma	Infiltrated
2001	Lutz et al.[30]	1	Basal cell carcinoma	Infiltrated
2005	McKenna et al.[31]	1	Basal cell carcinoma	Nodular
2008	Nambi et al.[32]	1	Basal cell carcinoma	Ulcerated
2008	Leyngold et al.[33]	1	Basal cell carcinoma	Infiltrated
2011	Qassemyar et al.[34]	3	Basal cell carcinoma	Nodular, ulcerated, in situ
2011	Lazzeri et al.[35]	2	Basal cell carcinoma	Nodular, ulcerated,
2013	De Seta et al.[36]	1	Basal cell carcinoma	Nodular
2021	Chlebicka et al.[37]	2	Basal cell carcinoma	Infiltrated, ulcerated

Table 2
Aggregated data of the cases reported in literature.

NUMBER OF CASES	47
Clinical presentation of the tumor	13 nodular 6 infiltrated 5 ulcerated 5 in situ 18 non specified
Associated symptoms	6 chronic drains 4 infections 1 bleeding
Treatment	7 tangential excision 20 surgical excision 1 radiotherapy 2 Mohs surgery 2 laser excision 14 not specified
Age	Range: 51-82 years Mean: 69 years Median:72
Sex	47 male
Years with rhinophyma	Range: 4-50 years Mean: 19 years Median: 15 years
Other associated neoplasm	Yes: 4 No: 43
Tobacco Consumption	4 Yes 4 No 39 Not reported
Alcohol Consumption	6 Yes 3 No 38 Not reported
Comorbidities	3 Type II Diabetes 2 Fatty liver cirrhosis 1 Polymyalgia rheumatica 1 Kidney failure 1 Tuberculosis

Subsequent studies continued to report anecdotal cases of basal cell carcinoma arising from rhinophyma until 1984 when a French group[25] analyzed 169 histological samples from patients with rhinophyma. They identified 14 cases of basal cell carcinoma and 8 cases of squamous cell carcinoma. In a 1998 Israeli study,[28] 4 cases of surgical excision of basal cell carcinoma were reported, all of which exhibited advanced symptoms.

A particular case was reported in a French study in 2011[34], where a patient aged 82 years with advanced rhinophyma and nodular and ulcerated lesions was found to have 5 tumors. Among these, 3 were Basal Cell Carcinoma (BCC) and 2 were Squamous Cell Carcinoma (SCC).

Discussion and Conclusions

Over time, attempts have been made to assess whether the presence of rhinophyma is a causal factor in the development of basal cell carcinomas or if the coexistence of these conditions is merely coincidental. Several hypotheses have been proposed. Acker et al.[10] hypothesized that the presence of lymphocytic infiltrates and pronounced inflammation associated with hyperplastic foci could be an etiological factor in the development of basal cell carcinoma. They concluded that the presence of rhinophyma increases the risk of developing these neoplasms. However, these conclusions were later criticized by Keefe et al.[26] indicating that the examined population was small and spread over a long period. Another explanation for the development of basal cell carcinomas was proposed by Brubaker et al.[23] and they identified the presence of papillary buds with high cellular proliferation

within the basal layer of dilated follicles in histological samples from patients with rhinophyma. These structures with high cellular proliferation rates are believed to be the origin of basal cell carcinomas.[38] Other authors hypothesized that trauma[27] and presence of scars[39] may be the triggering factors in the development of basal cell carcinomas, contributing to the occurrence of neoplasms in rhinophyma.

The presence of a correlation between basal cell carcinoma and rhinophyma was challenged by some authors[40] who questioned the findings of previous studies, citing the limited cumulative population and the incidence rate of detected basal cell carcinomas, which is not comparable to that of the general population. By analyzing the Scottish population (which has a basal cell carcinoma risk of 2.2%, likely underestimated), and comparing it to various incidence rates reported in the literature for patients with rhinophyma, they concluded that these findings were consistent with the overall risk of developing these neoplasms.

There is still no unanimous agreement regarding the correlation between basal cell carcinoma and rhinophyma. In this context, our case represents a novelty, as it is characterized by the presence of multiple non-localized foci of basal cell carcinoma that had not progressed to a clinically detectable stage. This demonstrates the presence of a "field of cancerization" throughout the affected surface of rhinophyma rather than being confined to a single area. This result indicates that the occurrence of these neoplasms is not likely to be coincidental with the presence of rhinophyma, but rather suggests that rhinophyma creates an environment that is predisposing to the development of these neoplasms. Furthermore, these associations indicate the need to perform histological examinations even for seemingly benign lesions, considering the potential presence of neoplastic lesions. Further investigations and studies are needed to confirm this hypothesis, but these findings can bring clarity to the research area where consensus has not yet been reached.

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The patient consented to the publication of the case.

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