INTRODUCTION

Sebaceous neoplasms are rare adnexal tumors that can present a challenge to clinicians. Only four cases of sebaceous carcinoma with sebaceoma have been reported in the literature. Herein, we describe the case of a sebaceous carcinoma originating from a sebaceoma in a solitary nodule of the posterior neck. Immunohistochemically, the tumor cells were strongly positive for epithelial membrane antigen and p53. It is possible that adnexal carcinomas may arise from malignant transformation of their benign counterparts as well as de novo. Malignant transformation was likely in this case because the lesion was composed of distinct benign and malignant components, and the benign component showed the typical histopathological features of sebaceoma. This case underscores the fact that partial and superficial biopsies sometimes may not provide the correct diagnosis. If a surgeon suspects malignancy based on a clinical examination, then it is mandatory to perform a deep biopsy.

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brown-to-reddish skin nodule about 10 mm in diameter with an erosive dimple on the upper portion (Fig. 1). There was no other abnormality of other organs and systems. The patient also had no significant past medical history. Under local anesthesia, the nodule was completely excised with a 5-mm free margin. Histological examination of the biopsy taken from the lesion revealed a sebaceous carcinoma arising in a sebaceoma. The large nodule in the upper part of the lesion corresponded to sebaceous carcinoma with ulceration. The specimen consisted of multiple variably-sized, discrete nodules and was centered in the superficial dermis (Fig. 2). The sebaceoma was composed of an admixture of basaloid germinative sebaceous cells and mature sebocytes. There was no nuclear pleomorphism and the mitotic activity was sparse. The sebocytes appeared mature with clear to lightly eosinophilic bubbly cytoplasm. The sebaceous carcinoma displayed an infiltrating growth pattern. The tumor cells showed marked nuclear pleomorphism and frequent mitoses. In addition, the sebaceous carcinoma was immunohistochemically characterized by strong positivity for epithelial membrane antigen. Mutations and nuclear accumulation of p53 tumor suppressor gene expression were also noted as features that may be correlated with a poor outcome. The patient was discharged without any postoperative complications. There was no sign of recurrence or metastasis after excision during a 5-month follow-up period.

**DISCUSSION**

This case is interesting because the lesion was composed of distinct benign and malignant components, and the benign component showed the typical histopathological features of sebaceoma. Few cases of sebaceous carcinoma arising in a preexisting sebaceoma have been reported in the literature. Although sebaceous carcinoma is sometimes accompanied by sebaceoma, these two types of neoplasms (sebaceous carcinoma and sebaceoma) are usually sharply segregated [4]. Adnexal carcinomas, such as clear cell hidradenocarcinoma and microcystic adnexal carcinoma are generally thought to develop de novo [5,6]. However, some adnexal carcinomas arise from preexisting benign counterparts; for instance, 20% to 50% of porocarcinomas are associated with poromas. Most spiradenocarcinomas and cylindrocarcinomas arise from their benign counterparts. This evidence suggests that some adnexal carcinomas arise from malignant transformation of their benign counterparts as well as de novo [7,8].

Sebaceoma, a term proposed by Troy and Ackerman [9], is a benign skin adnexal tumor with sebaceous differentiation, which can occur as a primary lesion or as a secondary tumor to either nevus sebaceous or seborrhoeic keratosis. The architectural pattern is characterized by sharp circumscription, smooth borders, and symmetry. The lesions have aggregations of basaloid undifferentiated sebocytes admixed with single or small clusters of mature vacuolated sebocytes, often in association with sebaceous duct-like structures and dense eosinophilic sclerosis. Like sebaceous adenomas, these neoplasms can be associated with Muir-Torre syndrome [10]. The major difference between sebaceoma and sebaceous adenoma is that the former has randomly scattered sebaceous cells without nuclear atypia, whereas the latter displays more highly organized, irregularly shaped lobules of sebaceous cells with an outer rim of more than a single layer of small germinal cells. There are no known immunohistochemical differences in addition to the architec-
Sebaceous carcinoma is a rare neoplasm, of which 75% of cases occur on the periorbital region and approximately 25% occur in the extraocular region. Of those cases, 70% arise in the head and neck region, with the parotid gland being the most common location. The most frequent clinical presentation of sebaceous carcinoma is a painless subcutaneous nodule. Other presentations include diffuse thickening of the skin, pedunculated lesions, or an irregular mass. Its clinical manifestations are often mistaken for benign conditions, resulting in a delayed diagnosis and management [12]. Metastasis to regional lymph nodes and other sites is common in these situations. Pathologists must be aware of the varied clinical manifestations of this condition, its histopathological morphology, and its multiple biomarkers to ensure an accurate diagnosis [13,14]. The protean appearance of sebaceous carcinoma complicates the diagnosis. It often presents with an appearance similar to more common benign lesions and is frequently misdiagnosed. It is not unusual for a sebaceous carcinoma to be treated multiple times as a chalazion before the diagnosis is made. An incomplete biopsy specimen, either because of its proximity to the eye or because sebaceous carcinoma is not in the clinical differential diagnosis at the time, can both complicate matters and delay therapy. Treatment is surgical, with Mohs micrographic surgery having the best results; there is an 11% recurrence rate after Mohs micrographic surgery and a 30% rate after standard excision [15].

Sebaceous carcinoma arises from two possible cell types: pluripotent cells with the capacity for sebaceous differentiation or ectopic sebaceous cells that develop during embryogenesis. Other reported locations include the submandibular glands, extremities, toes, penis, chest wall, sole of the foot, external auditory canal, and anterior neck region. The varied origin of this neoplasm highlights the importance of considering sebaceous...
carcinoma regardless of the anatomical location [16]. Several papers have been published on the relationship between sebaceous cancer and human papillomavirus [17,18]. Overexpression of p53 is involved in the carcinogenesis of this tumor, and is a risk factor suggesting a poor prognosis [17]. The histogenesis of sebaceous carcinomas remains unclear. Intraepidermal or intraepithelial sebaceous carcinoma in the epidermis or conjunctiva, as well as sebaceous carcinoma in association with a Bowen’s disease-like lesion, may suggest de novo histogenesis of sebaceous carcinoma from abnormal sebaceous germinative cells within the epidermis or conjunctival epithelium [19].

In conclusion, partial and superficial biopsies may not provide the correct diagnosis in many cases. The need for deep biopsy in cases of skin tumors should be emphasized. In fact, if a surgeon suspects malignancy on clinical examination, then it is mandatory to perform a deep biopsy. Time is important in such cases because sebaceous carcinoma is an aggressive tumor that spreads to deeper structures and also metastasizes to other sites very rapidly. Pathological examinations are limited regarding microscopic features in certain cases of very small and superficial biopsies that do not show a clear-cut malignant tumor. Therefore, the authors emphasize the importance of awareness of the possibility of sebaceous carcinoma with combined sebaceaoma, which may lead to a better evaluation and treatment of this condition.

NOTES

Conflict of interest
No potential conflict of interest relevant to this article was reported.

Ethical approval
The study was approved by the Institutional Review Board of Soonchunhyang University Cheonan Hospital (IRB No. 2021-02-051) and performed in accordance with the principles of the Declaration of Helsinki. Written informed consent was obtained.

Patient consent
The patient provided written informed consent for the publication and the use of her images.

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