Unusual Cystic Variant of Rhinophyma

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Rhinophyma is a slowly progressive, disfiguring disorder of the nose seen almost exclusively in men. It consists of large nodular masses of hypertrophic and hyperemic connective tissue and sebaceous glands. We present an 89-year-old white man with an unusual cystic variant of rhinophyma. While rhinophyma has 2 different presentations, our case contains not only both spectrums of histologic findings but also multiple cysts. These cysts were present on gross pathologic and microscopic examination and contained both squamous and mucinous epithelium.

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Case Report

An 89-year-old white man presented with a 9-year history of hyperplasia of the soft tissue of the nose, cheeks, and forehead, with a preserved nose silhouette, consistent with the clinical presentation of a cystic variant of rhinophyma and long-standing rosacea. He was otherwise healthy, with no relevant medical history. The rhinophyma was partially excised by tangential shaving, allowing reepithelialization from the residual deep pilosebaceous appendages. At 6-month follow-up, the patient had substantial clinical improvement. Gross pathologic examination of the tissue showed multiple cystic cavities up to 10 mm in size (Figure 1). Microscopic examination of hematoxylin and eosin–stained paraffin-embedded tissue sections showed an epidermis of normal thickness with prominent sebaceous hyperplasia, perifollicular inflammation, and dermal telangiectases consistent with rhinophyma (Figure 2). Focal marked dermal thickening with absence of pilosebaceous structures and increased dermal mucin were present (Figure 3). Several dilated follicles with smaller cyst formation also were seen. In addition, multiple large cysts, corresponding to grossly visible cysts, were observed (Figure 1). These cysts had a mixed lining consisting of squamous epithelium and mucinous epithelium with gobletlike cells (Figure 4). Transition from the squamous to mucinous epithelium appeared gradual. Substantial lymphocytic infiltrate was present under the mucinous epithelium (Figure 5). None of the examined large cysts had connection with adnexal structures.

Comment

Phymas are slowly progressive, disfiguring disorders of the face and ears that represent the end stage of severe rosacea, a common inflammatory condition of facial skin.[1,2] Although rosacea has a higher incidence in females, rhinophyma, the most common of all phymas, is seen almost exclusively in men older than 40 years.[1,3,4] Rhinophyma consists of large nodular masses of hypertrophic and hyperemic connective tissue and sebaceous glands positioned on the distal half of the nose.[4,5] There are reports of other diseases mimicking rhinophyma, such as microcystic adnexal carcinoma, sebaceous carcinoma, metastatic carcinoma, angiosarcoma, sarcoidosis, and amyloidosis.[6-11] The development of skin cancer, both basal and squamous cell carcinomas, in rhinophyma appears to be coincidental; however, it should not be overlooked as a possible occurrence.[1,3,12]

The pathophysiology of rhinophyma still remains unclear. Reports on the association between alcohol consumption and rhinophyma are conflicting and are supported with little or no statistical evidence.[3] Some studies suggest chronic inflammation could lead to connective tissue hypertrophy and fibroplasia,[12,13] while others point out proliferation of the sebaceous glands to be the main mechanism.[14,15] Other evidence suggests that fibrosis and scarring may play a prominent role in the pathophysiology of rhinophyma because of elevated transforming growth factor (TGF)–β1 and transforming growth factor–β2 levels.[16]

Histologic findings in severe forms of rhinophyma rarely are reported in the literature.[15]

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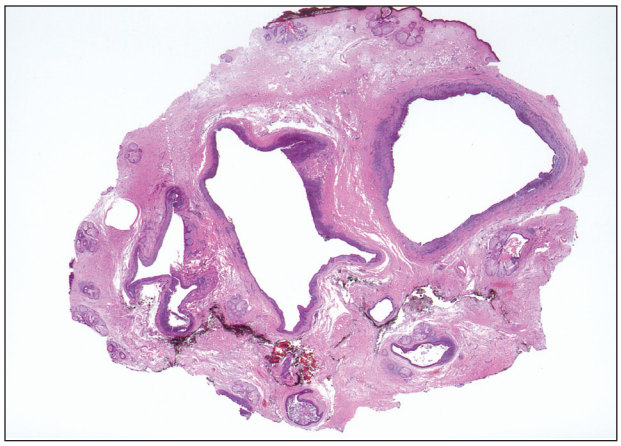

Figure 1. Multiple cystic cavities (H&E, original magnification ×40).

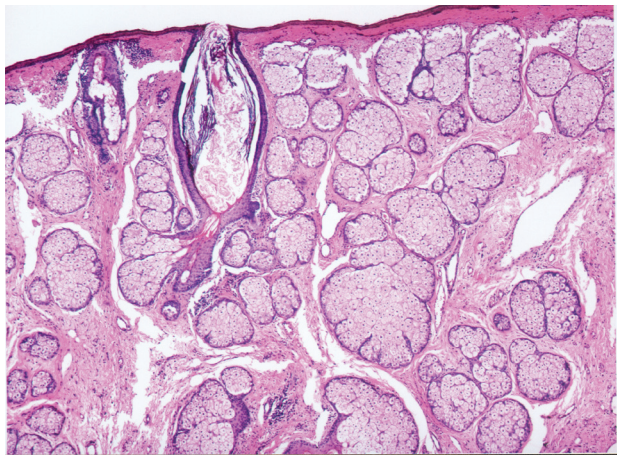

Figure 2. Prominent sebaceous hyperplasia, perifollicular inflammation, and dermal telangiectases (H&E, original magnification ×100).

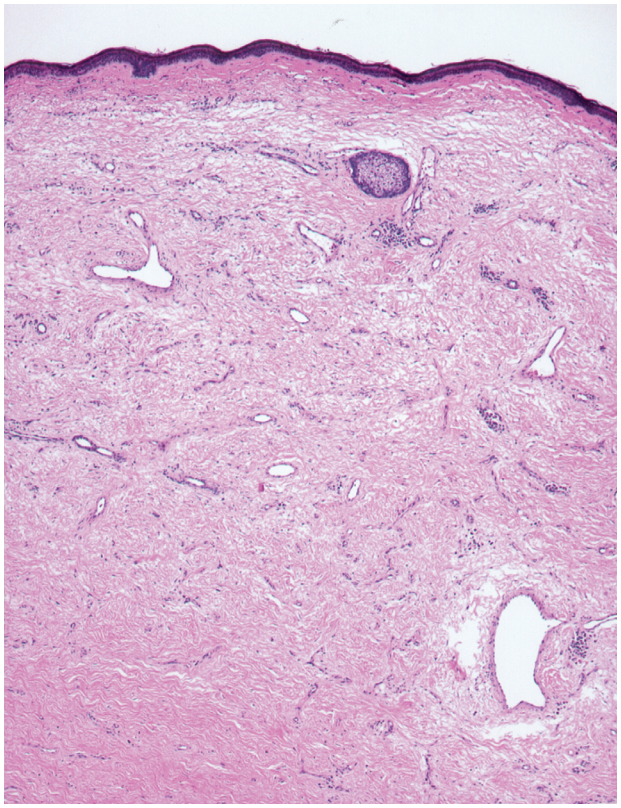

Figure 3. Focal marked dermal thickening with absence of pilosebaceous structures (H&E, original magnification ×100).

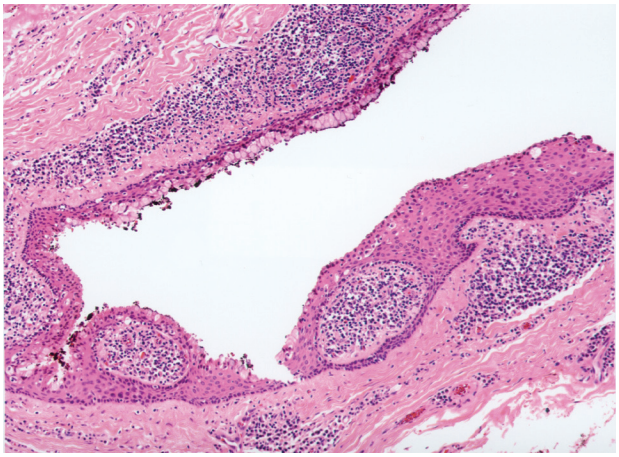

Figure 4. Lining of large cyst consisting of squamous epithelium with gradual transition to mucinous epithelium (H&E, original magnification ×200).

Rhinophyma has 2 different histologic presentations. The first is characterized histologically by sebaceous hyperplasia, perifollicular inflammation, dermal telangiectases, and occasional follicular cysts, while clinically it presents as rhinophyma with a preserved nose profile. The second presentation histologically shows dermal fibrosis, reduction or absence of pilosebaceous structures, presence of telangiectases, and increased amounts of dermal mucin, and clinically shows a markedly distorted proboscis.[15]

Our case is unusual because the histologic findings contain both spectrums described in rhinophyma. Also, to our knowledge, there is no previous report of large mixed squamous and mucinous cysts in conjunction with rhinophyma. We believe that they probably represent squamous and mucinous metaplasia in cysts derived from dilated follicular structures. The absence of a connection between these cysts and pilosebaceous structures is probably due to the chronic pericystic inflammation that destroyed the residual pilosebaceous structures. Another possibility that cannot be completely excluded is that the cysts may represent misplaced metaplastic sinonasal respiratory mucosa. The differential diagnosis also includes mucoepidermoid carcinoma, which consists of mucus-secreting and

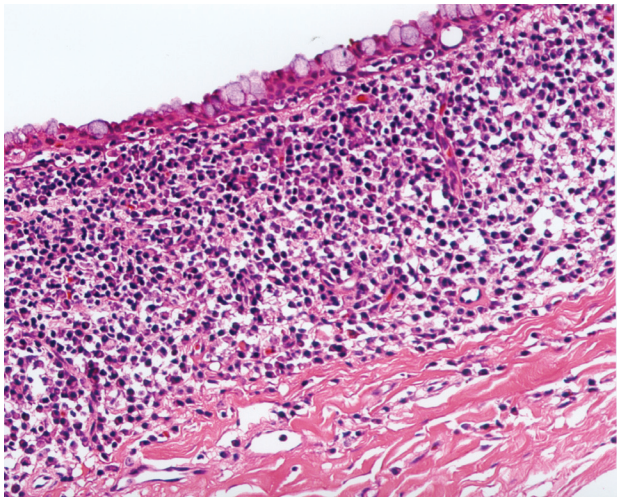

Figure 5. Lymphocytic infiltrate under the mucinous epithelium (H&E, original magnification ×400).

epidermoid cells. However, mucoepidermoid carcinoma would be poorly circumscribed with very small cysts and substantial cytologic atypia.

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