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Unilateral Syringoma of the Face Associated with Hyperthyroidism

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Abstract

Syringomas are benign tumors derived from the intraepidermal portion of eccrine sweat ducts. They usually occur on the periobital area, but have also been found on the scalp, forehead, cheeks, axillae, abdomen, extremities, genitalia, and buttocks. We describe a patient with an unusual presentation of unilateral syringoma of the face associated with hyperthyroidism.

Key words: unilateral syringoma; face; hyperthyroidism

Introduction

Syringomas are common tumors of the intraepidermal eccrine sweat gland ducts. Their occurrence is twice as common in women as in men, and they most commonly appear in adolescence. However, further lesions can develop later in life, and they can occur between the first and sixth decades of life. Clinically, syringomas appear as small, multiple, firm, skin-colored-to-yellowish papules, 1 to 3 mm in diameter, localized most commonly on the lower eyelids and malar areas, but also on the axillae, neck, chest, upper arms, and abdomen. The lesions usually are bilateral and symmetrically distributed (1). A unilateral facial syringoma case has not previously been reported in the literature.

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Fig. 1. Multiple papules seen on the right side of the face

Case Report

A 34-year-old female presented with a 2-year history of unilateral erythematous papular eruption on the right cheek. The patient's notable past medical history was hyperthyroidism. She has been on antithyroid drug therapy (propylthiouracil 50 mg/day) for seven years. In her previous history, the patient was euthyroid at the onset of the syringoma lesions. The family history was unremarkable. Physical examination revealed multiple, reddish brown, flat-topped papules 1–2 mm in diameter on the right cheek and right zygomatic areas. In addition, she has

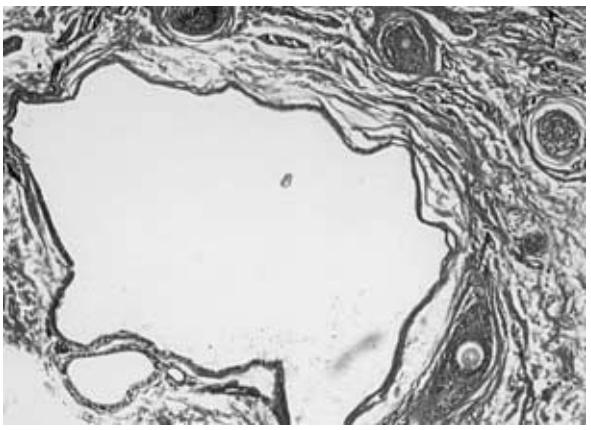


Fig. 2. A cystic cavity lined with a double row of epithelium in the dermis (HE ×4)

exophthalmus (Fig. 1).

Histopathological examination of the skin lesions showed numerous small ductal structures within the dermis (within fibrous stroma). In addition, there was a cystic structure, lined by a double row of epithelial cells (Fig. 2). A complete blood count, serum biochemistry, and thyroid hormone levels were within their normal ranges. The cranial magnetic resonance imaging (MRI) was normal.

Discussion

Unilateral cases of syringoma are quite uncommon in the literature. Unilateral syringomas have been reported to be localized on the extensor aspect of the left upper arm, the left antecubital fossa, the left side of the chest (2), on the nipples (3), on the right side of the chest and right arm (4), and over the right shoulder with anterior extension down the upper arm (5). To our knowledge, unilateral facial syringomas have not been previously reported.

A few familial cases have also been reported (5). In addition, there are reports of unilateral or segmental syringomas with no family history (6). Our case is a 34-year-old woman with no familial history.

It has been suggested that syringomas are hormonally responsive, based on observations that they increase in size during pregnancy, the premenstrual period and the use of oral contraceptives, and that they are occur most commonly in women and during puberty. A study of nine non-vulvar syringomas from both males and females demonstrated an estrogen receptor in one case and progesterone receptors in the remaining eight syringomas by immunohistochemical staining (1). In recent case reports, immunohitochemistry of vulvar (1) and eruptive syringoma (7) revealed progesterone positivity. Syringoma is also seen with diabetes (7). Our patient has been on an antithyroid drug for seven years because of hyperthyroidism. At the time of admission, she was euthyroid. Syringomas associated with hyperthyroidism have not been previously reported in the literature.

Despite the hormonal interactions mentioned above, we have not found any information suggesting that hyperthyroidism has a role in the development of syringoma. Therefore, at the moment, we can not speculate whether hyperthyroidism and syringoma coincidentally occur or hyperthyroidism may have a role in the pathogenesis of syringoma.

Syringomas typically present in a bilateral and symmetrical distribution, but there have been reports of unilateral, unilateral linear nevoid, bathing trunk, and generalized distributions. Clinically, syringomas may be mistaken for acne vulgaris, sebaceous hyperplasia, milia, lichen planus and eruptive xanthoma (8). However, syringomas demonstrate distinctive histopathologic features. Examination of the dermis demonstrates numerous small ducts lined by a double row of flattened epithelial cells. Often, the outer layer extends into the surrounding stroma, forming a comma-like projection. The ductal lumina are filled with an amorphous periodic acid-Schiff-positive material (1, 5, 6, 9).

Syringomas are benign and usually asymptomatic, so treatment is offered primarily for cosmetic reasons (9). The multiple treatment options include carbon-dioxide laser (9, 10), trichloracetic acid, and carbon-dioxide laser combination (11), der-

mabrasion, excision, electrodesication with curettage, and cryosurgical destruction (9). Additionally, oral and topical retinoids (8) and topical atropine (12) are also used in treatment. However, chemical and surgical destruction may produce scarring and do not prevent recurrence of the syringoma (9). The present case received a topical retinoid treatment.

In conclusion, unilateral facial syringoma associated with hyperthyroidism is unusual. To our knowledge, no such a case has not been previously reported in the literature.

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