Case Report

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Orifices at the Lower Neck: Heterotopic Salivary Glands

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Key Words

Heterotopic salivary-gland tissue

Fistula in the lower neck

Congenital abnormality

Abstract

Heterotopic islands of salivary-gland tissue are rare and have been described in a number of sites in the head and neck. We describe a patient with salivary-gland fistulas in the lower third of the neck, without evidence of any other congenital abnormalities. The appearance of sinuses in association with heterotopic salivary-gland tissue suggests that its embryologic origin may be from the branchial clefts. Surgical treatment is recommended.

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Introduction

The embryologic development of head and neck structures is complex and anatomical abnormalities often occur. Heterotopic islands of salivary-gland tissue have been described in a number of sites in the head and neck. In this report, we describe the presence of normal salivary-gland tissue in the lower part of the neck; the clinical characteristics, management and embryologic significance of this unusual anomaly are discussed.

Case Report

An 11-year-old girl complaining of two small openings at the base of the neck was admitted to our department. Her mother had first noticed the appearance of the two small orifices when she was 1 year old. The girl complained of the formation of two small nodules (0.5-1 cm in diameter) which increased in size intermittently, mainly in relation to the ingestion of food. On palpation of the nodules, a clear mucoid fluid was discharged. The patient was healthy without evidence of any other congenital abnormalities, the family history also being negative in this respect.

On physical examination there were two symmetrical orifices 2-3 cm above the sternoclavicular joint. The openings measured 1 mm in diameter and were located at the anterior border of the

sternocleidomastoid (SCM) muscle (fig. 1). At the left orifice, cannulation for radio-opaque contrast injection revealed a 3-cm-long sinus. Cannulation of the right side was unsuccessful. The lesions were excised and their histological examination showed a sinus tract lined by pseudostratified cylindric epithelium. Normal acini of serous and mucous types (mixed type) of salivary glands were identified in the adjacent connective tissue (fig. 2).

Discussion

Heterotopic salivary glands have been described in several areas within the oral cavity and other more distant sites: the pituitary gland, the middle and external ear, the mastoid bone, the thyroglossal duct, the capsules of the thyroid and parathyroid glands, the mandible, the lymph nodes and the sterno-clavicular joint [1,2].

Most of the reports in which ectopic salivary-gland tissue was found in the middle and lower parts of the neck emphasize the close relationship to the anterior border of the SCM muscle [2], as in our case. The appearance of sinuses in association with heterotopic salivary-gland tissue suggests that its embryologic origin may be from the branchial clefts; the lower neck position strongly supports an association with the pre-cervical sinus. The overlapping of the third and fourth branchial clefts forms the precer-

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Fig. 1. Fistulas in the lower neck. On palpation, discharge of a clear mucoid fluid.

BUST Fig. 2. A sinus tract and normal salivary tissue. $\times 280$.

vical sinus at the sixth week of gestation. The sinus normally disappears leaving two small ectodermal cysts, called cervical vesicles, along the lower and anterior border of the SCM muscle. It seems reasonable to suppose that salivary-gland tissue may also arise from the heteroplastic ectodermal lining of the precervical sinuses or their cervical vesicles [3,4]. Cutaneous orifices are infrequently noted at birth, but this may be due to minimal symptomatology [2]. Commonly, a draining sinus opening on the anterior neck is seen with a saliva-like secretion and a tendency to increase in size with ingestion of food [4]. Usually there is no family history. Local complications are very infrequent, but neoplasms may develop in ectopic

salivary glands [5,6], so surgical treatment is recommended. Associated developmental anomalies have been described in very exceptional cases [7, 8].

It is surprising that this pathology is hardly referred to in the dermatological literature [9]; heterotopic salivary tissue is yet another anomaly which must be considered in the differential diagnosis of lesions in this region.

In conclusion, a fistula not connected with the alimentary tract situated in the lower neck and secreting a mucoid fluid (exacerbated by food ingestion) should arouse the suspicion of a heterotopic salivary gland.

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