Case Report · Description de cas · Fallbericht

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Congenital Submandibular Salivary Gland Fistula

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Key Words
Salivary gland · Fistula, congenital

Abstract
In this study, we present a patient with a congenital cervical submandibular gland fistula who was admitted to the hospital for nodular thyroid hyperplasia. After bilateral subtotal thyroidectomy, the fistula tract and the submandibular salivary gland on the right side of the neck and the obliterated sinus on the left side of the neck were excised. The pathological examination led to the diagnosis of adenomatous hyperplasia of the thyroid gland with focal lymphocytic thyroiditis and mixed-type main salivary gland (submandibular gland) with cystic dilatation in one duct. Congenital salivary gland fistulae are rare and may be associated with abnormalities of the branchial apparatus or arise from aberrant or accessory salivary tissue. Excision of the fistula and the remaining submandibular gland is considered the treatment of choice in congenital fistulae, but traumatic fistulae generally resolve with conservative treatment within 3 weeks.

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Kongenitale Fistel der Unterkieferspeicheldrüse
Dieser Bericht präsentiert den Fall einer Patientin mit einer kongenitalen Halsfistel der Unterkieferspeicheldrüse, die wegen einer Struma nodosa das Krankenhaus aufsuchte. Nach bilateraler subtotaler Thyreoidektomie wurden auch der Fistelgang und die Unterkieferspeicheldrüse auf der rechten sowie die obliterierte Fisteltasche auf der linken Halsseite reseziert. Der pathologische Befund ergab die Diagnose adenomatöse Hyperplasie der Schilddrüse mit fokaler lymphozytärer Thyreoiditis und eine Hauptspeicheldrüse (des Unterkiefers) vom gemischten Typ mit zystischer Erweiterung des Ganges. Kongenitale Fisteln der Speicheldrüsen sind selten und können mit Anomalien der embryonalen Kiemengänge einhergehen oder aus verlagertem oder zusätzlichem Speicheldrüsengewebe entstehen. Die Resektion der Fistel und der verbleibenden Unterkieferspeicheldrüse wird als Therapie der Wahl angesehen, aber traumatisch bedingte Halsfisteln heilen im Allgemeinen unter konservativer Behandlung innerhalb von 3 Wochen.

Fistule congénitale des glandes salivaires sous-maxillaires
Ce travail rapporte le cas d'une patiente adressée pour une hyperplasie thyroïdienne nodulaire et qui présentait, en plus, une fistule congénitale des glandes sous-maxillaires. Le traitement a été chirurgical. Au cours de la thyroïdectomie bilatérale subtotale pour laquelle elle avait été adressée, la fistule et la glande sous-maxillaire droite ont été enlevées. A gauche, le trajet fistulaire, oblitéré, a été réséqué et la glande préservée. L'examen histologique a montré l'existence d'un adénome pléomorphe dans la glande droite, associé à une dilatation cystique. Le traitement des fistules salivaires congénitales est chirurgical; par contre, la littérature mentionne que les fistules salivaires post-traumatiques guérissent généralement spontanément dans les 3 semaines.

Case Report

A 60-year-old woman was referred to our clinic with the diagnosis of a nodular goiter for operation. She was under antithyroid treatment and 'euthyroid' at the time of admission. Physical examination revealed a nodular thyroid gland and fistula orifices of approximately 2–3 mm in diameter over the lower end of the anterior borders of the sternocleidomastoid muscle bilaterally. She complained of a draining serous discharge from these orifices since childhood, especially when crying. There was neither surgery nor trauma in the history so we suggested that the fistula was congenital. Other physical findings were normal. We decided to perform bilateral subtotal thyroidectomy for nodular goiter. We did not need to make any fistulography or other further studies such as ultrasonography (US) because the orifices were in the surgical exploration area (fig. 1). We planned to make intraoperative US or fistulography if needed. The patient was taken to the operation room after the routine preparations. After bilateral subtotal thyroidectomy, exploration was extended to the site of the orifices, and the fistula tract was exposed; then, the submandibular salivary gland on the right side and the obliterated sinus on the left side of the neck were excised. Pathological diagnosis was adenomatous hyperplasia of the thyroid gland with focal lymphocytic thyroiditis and a mixed-type main salivary gland (submandibular gland) with cystic dilatation in one duct (fig. 2).

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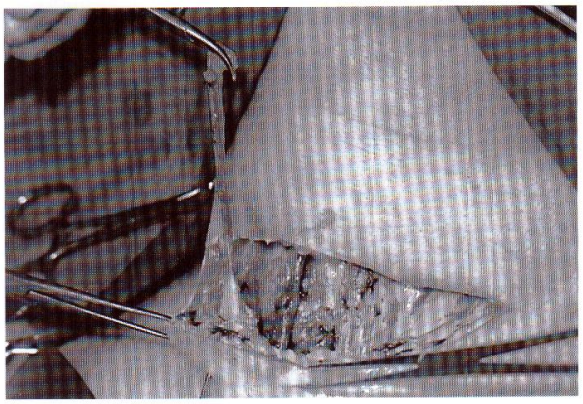


Fig. 1. Excision of the fistula tract on the right side of the neck.

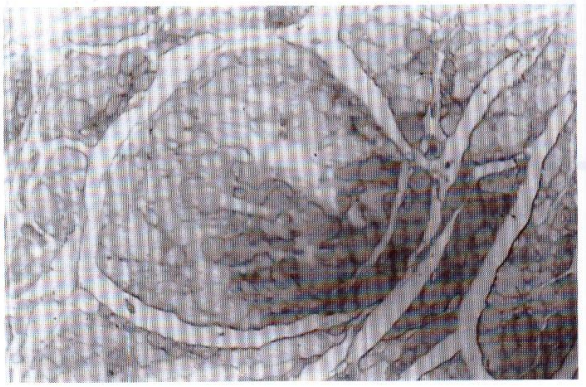


Fig. 2. Histological appearance of mixed-type main salivary gland (submandibular gland) with cystic dilatation in one duct. HE. × 40.

The patient was discharged from the hospital on the 3rd postoperative day. The follow-up period was uneventful with no complication since then.

Discussion

The submandibular salivary gland is one of the paired major salivary glands consisting of a large superficial part and a smaller deeper part, continuous with each other around the posterior border of the mylohyoid muscle. The submandibular duct is about 5 cm long and has a thinner wall than the parotid duct. It starts from the gland's lowest part, below the mandible in lateral views, ascends vertically above the mandible's lower border and turns sharply forwards gradually ascending to its opening [1].

External salivary fistulae are uncommon entities which are generally traumatic in origin as unpleasant complications of upper aerodigestive tract surgery, but also foreign bodies and calculi may be causative factors [2–5]. Congenital fistulae have been reported especially from the parotid gland [6] and from the ectopic deposits of salivary tissue [7] and almost all have been external fistulae. The presence of a congenital sinus opening along the anterior border of the sternocleidomastoid muscle is generally thought to be due to a second-cleft branchial fistula [7]. Congenital salivary fistulae may be associated wih abnormalities of the branchial apparatus or arise from aberrant or accessory salivary tissue. Most authors support the theory of heteroplasia as the origin of this fistula. Histologic reviews of branchial arch specimens show that in half of the cases seromucous glandular tissue surrounds the fistula [8]. Most of the fistulae are reported to be cutaneous, but rare cases of fistula between the submandibular gland and the oropharynx have been reported [9].

Histologic examination reveals squamous or columnar ciliated epithelium with abundant lymphoid tissue which is not a feature of normal salivary gland ducts. Mucous glands are rare, drainage is usually mucoid rather than serous, but there are exceptions as seen in our case. Drainage may increase with any stimulus to salivation as the patient in our case complained of increased discharge when crying. Incomplete tracts are less common than complete fistulae [7].

Excision of the fistula and the remaining submandibular gland is considered the treatment of choice in congenital fistulae [3], but traumatic parotid fistulae generally resolve with conservative treatment within 3 weeks [6].

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