

Heterotopic salivary gland presenting as a discharging sinus in the base of the neck

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Abstract

We report a case of congenital heterotopic salivary gland with draining sinus in the lower neck on the right side of a 10-year-old female, which we initially thought to be a branchial fistula. Heterotopic salivary glands are rare lesions in the neck and when present appear very similar to branchial cleft sinus or fistula. This congenital lesion is rare. This is probably the first report from India. It is important to report this case to raise the awareness of this condition.

Introduction

The presence of a draining sinus along the anterior border of the sternocleidomastoid muscle in the neck usually indicates a second or third branchial cleft sinus or fistula. A heterotopic salivary gland is a rare lesion, which may present at the same site as a cutaneous discharging sinus. There may be only minor clinical differences in their presentation.¹ The rarity of this congenital lesion, together with the fact that there are no available reports of this lesion from India, stimulated us to report this case to raise awareness of this condition.

Case Report

A 10-year old female presented to the otolaryngology department of our hospital with a draining sinus in her right lower neck. Since birth her parents had noticed an intermittent clear mucoid discharge from an opening located in her lower neck. This was preceded by mild swelling or fullness in the lower neck, which was not associated with her having eaten. There was no history suggestive of any infection at any time in the form of pain, redness or pus discharge. On examination, there was a pin-point opening at the anterior border of the sternocleidomastoid muscle approxi-

mately 1 cm above the right sternoclavicular joint, but there was no swelling or thickening around the opening. A sinugram using iohexol, a non-ionic contrast dye, was obtained to delineate the sinus/fistula tract (Figure 1). The tract was approximately 5 cm in length and in a cephalad direction. It was dilated in its central portion. The patient was taken for excision of the tract. After injecting methylene blue solution to delineate the tract, an elliptical incision was made. The dissection revealed a descending branch and a larger posterosuperior tract. In the middle of the tract, a dilated structure of approximately 1 cm² containing mucoid collection and embedded within the muscle was found. The tract ascended further for another 1 cm and ended blindly in a superficial plane. The dissection was easy to perform and without problems.

The specimen was made up of a tubular structure 5 cm in length and 0.5 cm in its average diameter, dilated in its middle portion surrounded by muscle. Microscopic examination revealed mucinous salivary gland tissue embedded in striated muscle and inflammatory cells were seen (Figure 2). There was no lining of branchial cleft fistula.

Discussion

Heterotopic salivary gland tissue (HSGT) consists of salivary tissue outside of the major and minor salivary glands. Heterotopic salivary gland tissue is found most commonly in the periparotid and intra-parotid lymph nodes. The neck, at the anterior border of the sternocleidomastoid muscle near the sternoclavicular joint is also an area of salivary gland heterotopia.² Klimko and Horanyi reported the first histologically documented case of HSGT in the lower neck in 1958.¹ The other rare locations in the head and neck region include the mandible, maxilla, middle neck, larynx, hypopharynx, middle ear and thyroglossal duct. They are usually asymptomatic incidental findings but can present as a mass or a fistula. Benign and malignant neoplasms can arise in such heterotopic salivary tissue, which may sometimes be the initial presenting feature.

Recognition of salivary gland as a heterotopia is based on the anatomic location of the tissue, and microscopically by the association and relationship to surrounding tissues. For instance, a salivary gland surrounded by skeletal muscle or in a (extraparotid) lymph node is heterotopic.³

The origin of salivary gland tissue in the lower neck is obscure. As salivary glands do not migrate during development, like parathyroids or the thymus, it is presumed that most heterotopias of salivary glands in neck arise de novo from the endoderm of the pharynx.³ In the

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lower neck, it is believed that the tissue originates from a pre-cervical sinus of His or a cervical vesicle, a distinct embryologic structure that forms in the lower part of the neck between the second branchial arch and the upper thoracic wall, which normally disappears before birth.⁴ This is thought to occur by heteroplasia or anomalous differentiation of ectodermal cells within the remnant of the pre-cervical sinus to salivary tissue. There are rare reports of occurrence of heterotopic salivary tissue in association with branchial cleft fistula which support the belief that the heterotopic glands arise through errors of development in the branchial apparatus.^{5,6} So HSGT should be looked for in every cervical anomaly related to any branchial apparatus derived lesion.

Clinically, they are located along the anterior border of sternocleidomastoid muscle with a cutaneous opening draining mucoid or serous discharge, which resembles branchial cleft fistulas or sinuses.⁷ The opening may be anywhere, in mid or lower neck. In our case, it was near the sternoclavicular joint. This made us doubt it was a branchial cleft fistula where the opening is usually at the junction of upper 2/3rd and lower 1/3rd. Other differentiating features are the absence of a history suggestive of infection and an increase in the discharge at the time of meals, mastication or menstruation in case of heterotopic salivary gland sinuses.¹ Our patient reported no discharge while eating. Most reported cases of cervical HSGT have a right-sided predilection. Cases generally present at birth or by early childhood, but they are rarely diagnosed in adulthood.⁸

Congenital abnormalities of the branchial apparatus can manifest as different abnormal conditions in the neck, including a cyst (a small, partial malformation with no external

opening), a sinus (a malformation with one superficial opening), or a fistula (a complete malformation with both internal and external communications).

Fistulas of the second branchial cleft are the most common, accounting for as many as 90% of all branchial cleft fistulas.⁹ They are present along the anterior border of the sternocleidomastoid muscle. The tract crosses superiorly-lateral to the common carotid artery, the glos-

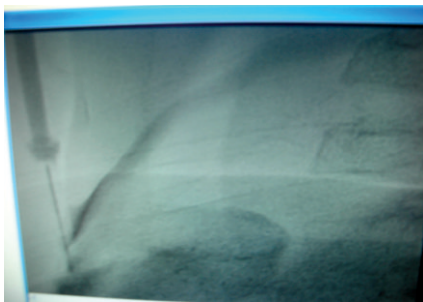


Figure 1. Sinugram showing the tract running cephalad with dilatation at its end.

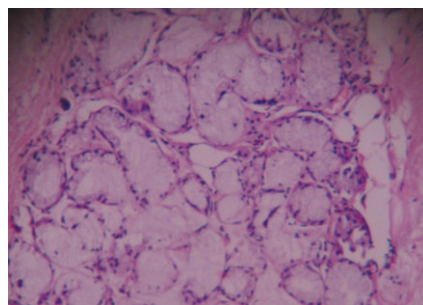


Figure 2. Heterotopic salivary gland tissue embedded in the skeletal muscle (H&E $\times 40$).

sopharyngeal nerve, and the hypoglossal nerve, and lies between the internal and external carotid arteries. The sinus often ends close to the middle constrictor muscle. In other cases, the sinus opens into the region of the tonsillar fossa. Fistulas of the third branchial cleft, which are rare, also appear at the anterior border of the sternocleidomastoid muscle. Here, the tract terminates by piercing the lateral thyrohyoid membrane at the piriform sinus.

Surgery for heterotopic salivary gland fistula is limited in extent compared to surgical removal of a branchial fistula.¹⁰ In our patient, the tract was always in a superficial plane. Another horizontal incision was given superiorly to facilitate the dissection but the tract soon ended and appeared as if some fiber of strap muscle had been removed.

A sinugram/fistulogram is helpful in accurate pre-operative diagnosis and differentiating HSGT from branchial cleft fistulas. With the fistulogram, the surgeon can delineate the course of the tract, which may be superficial in a heterotopic salivary gland with an arborizing pattern resembling a sialogram or deep between carotid arteries in a branchial fistula.¹ It also shows the distal end structure, which may be a cyst, an internal communication with the pharynx in case of a branchial cleft fistula, or a heterotopic salivary gland. The fistulogram or a sinugram can give information regarding the width of the tract and any ectasia or dilatation, a feature of HSGT, as was present in our case.

Therefore, the above mentioned clinical features and a pre-operative fistulogram may help to differentiate between various draining fistulas of neck present at the anterior border of sternocleidomastoid. HSGT should be included in the list of congenital fistulas of the lateral neck.

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