Heterotopic Salivary Gland Located in the Middle Neck

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A rare case of a heterotopic salivary gland located in and draining to the middle neck is presented, and the literature is reviewed. Youngs and Scofield (1967) suggested that such glands are located mainly in the lower neck and developed by heteroplasia within a remnant of the precervical sinus of His. Clinically, doctors should be attentive of the differential diagnosis of draining sinuses such as a second branchial cleft sinus in the neck. The present case is of interest because: 1) such a lesion in the middle neck is extremely rare (this is, to our knowledge, the third case reported in the literature), and 2) a sinogram through the opening showed an arborizing pattern suggesting the possibility of a heterotopic salivary gland rather than of a second branchial cleft sinus before surgery. This lesion can be easily cured if diagnosed correctly and especially when a sinogram is made before surgery.

Key Words: Heterotopic salivary gland, middle neck, sinogram

The presence of a draining sinus along the anterior border of the sternocleidomastoid muscle in the neck is usually suggestive of a second branchial cleft sinus. But a heterotopic salivary gland, a rare lesion, may also be present at the same site with somewhat different clinical features.

This lesion is one which the surgeon should remember when treating draining sinuses and cysts of the neck. Klimko and Horanyi (1958) reported the first histologically proven case of a heterotopic salivary gland in the lower neck, but reported that cases of such lesions in the neck are rare. After reviewing 11 cases, Youngs and Scofield (1967) suggested that such lesions arise most probably by heteroplasia within a remnant of the precervical sinus of His. Clinically this lesion is somewhat different from the second branchial cleft sinus: 1) episodes of drainage are usually associated with a sense of fullness in the affected area and 2) the amount of drainage usually increases during eating or mastication. Possibly this lesion can be diagnosed with the aid of a sinogram before surgery. Our present case exhibited some of the typical clinical features of this entity, and a preoperative sinogram also suggested it. The objective of this report is to draw attention to this rare entity which is very similar to a second branchial cleft sinus.

CASE REPORT

A healthy 18-year-old Korean boy visited Severance Hospital with a draining sinus in the middle of his right neck (Fig. 1). Since birth, his parents had noticed an intermittent clear mucoid discharge from an opening located on the side of his middle neck. He complained of frequent mucoid discharge proceed by a sense of fullness of the affected lateral neck especially during eating or mastication. There was no history suggestive of infection at any time. On physical examination there was a small opening at the anterior border of the sternocleidomastoid muscle about 6 cm above the clavicle but there was no definite mass or cord-like structure around the opening. A sinogram using water-soluble contrast medium was obtained to delineate the sinus and its course. The tract was about 5 cm in length and directed cephalad. The tract was ectatic and at the end of the tract an arborizing
pattern was noted (Figs. 2 and 3). After injecting a gentian violet solution to sharply demarcate the tract, an elliptical incision was made around the opening. The dissection was carried 5 cm posterosuperiorly at which point a fat-like globular stained structure of about 1 cm² embedded within the muscle was found and removed without difficulty.

Grossly the specimen consisted of a cylindrical, tubular structure measuring 5 cm in length, and 0.5 cm in its average diameter but its midportion was severely dilated. Microscopically, a mixed seromucinous salivary gland with irregularly dilated excretory ducts was observed. The duct was surrounded by dense fibrous connective tissue. In the center of the ductal mucosa, squamous metaplasia was clearly seen (Figs. 4 and 5). After pathological confirmation of the salivary gland tissue, we tried to identify the normally located submandibular glands. The right duct of Wharton was cannulated and a sialogram and upper neck CT scan were performed which showed normal configuration and the location of both sub-

Fig. 1. Anterior view shows an external opening (arrows) in the middle neck, along the anterior border of the right sternocleidomastoid muscle.

Fig. 2. Sinogram through the opening shows a well defined smooth margined tubular structure passing upward 5 cm in length, 1.5 cm in maximum diameter. (arrows; opening site)

Fig. 3. Close-up view shows several arborizing, branching tubules originating from the end of the main duct.
Fig. 4. An embedded salivary gland in the skeletal muscle is shown. Histologically the glands (mixed sero-mucinous) are essentially normal. (H & E, ×100)

Fig. 5. The excretory duct is irregularly dilated and surrounded by dense fibrous connective tissue. In the center of the mucosa, squamous metaplasia is evident. (H & E, ×400)
mandibular glands (Figs. 6 and 7).

DISCUSSION

A heterotopic salivary gland of the neck is a rare lesion usually located in the lower neck. Klimko and Horanyi (1958, 1960) reviewed the literature and found three previously reported cases (not proven histologically) and added one case which occurred in a ten-month-old male baby bilaterally. That case was the first one proven histologically (a serous salivary gland) in the lower neck. Jemstrom and Prietto (1962) reported one case arising at the base of neck. Youngs and Scofield (1967) reported 11 cases culled from the files of the Armed Forces Institute of Pathology, Sabini et al. (1970), Parsons (1972), Rothner (1973), Stingle and Priebe (1974), Soucy (1985) separately reported one case each located in the lower neck. Extremely rare cases located in the upper neck (benign mixed tumor from a heterotopic mucinous salivary gland) by Pesavento and Ferlito (1976), and in the middle neck by Youngs and Scofield (1967) and Singer and Applebaum (1979) were reported. This present case in the middle neck is, to our knowledge, the third one reported in the literature. In attempting to explain the occurrence of a salivary gland in the neck, Youngs and Scofield (1967) concluded that on the basis of loca-
tion, bilateral occurrence, presence at birth and with histological findings, the salivary gland tissue had developed within a remnant of the precervical sinus of His by heteroplasia. Heteroplasia means the anomalous differentiation of ectodermal cells within a remnant of the precervical sinus to salivary tissue. According to a collective review by Rothner (1973) this lesion occurred bilaterally in one third of the cases with the right side being more common (4:1). In 65% of the reported cases the glands were of the mixed sero-mucinous type, the remainder being either serous or mucinous.

Clinically, a heterotopic salivary gland in the neck possesses several important features which may be similar to those of a second branchial cleft sinus: 1) the same location of the opening along the anterior border of the sternocleidomastoid muscle in the neck; 2) both commonly present at birth; 3) intermittent fluid discharge. But there are also distinguishing features: 1) absence of history of infection; 2) increased amount of discharge associated with eating, mastication, and/or menstruation (Rothner 1973; Stringle and Priebe 1974) in heterotopic salivary gland lesions.

A sinogram may be necessary for accurate diagnosis and to give helpful information to the surgeon. According to Parsons (1972), a branching (arborizing) pattern resembling a sialogram may be obtained. A sinogram is relatively easy and uncomplicated to obtain. With the sinogram the surgeon can identify the course of the tract and the distal end-structure (cyst or communication with the pharynx in the second branchial cleft sinus, arborizing pattern in the heterotopic salivary gland). We think that the above mentioned clinical features plus a sinogram may aid in the differential diagnosis of various sinuses occurring in the neck without difficul-

In our case precise pathogenesis cannot be understood due to unusual location, but we think that this case may arise from heteroplasia of ectodermal cells within a remnant of precervical sinus of His. Treatment should be total excision, which can be easily accomplished.

REFERENCES

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