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Congenital Fistula from Ectopic Accessory Parotid Gland: Diagnosis with CT Sialography and CT Fistulography

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Summary: We report a case of congenital fistula from ectopic accessory parotid gland to the cheek demonstrated by CT sialography and CT fistulography. The right parotid gland was abnormally located lateral to masseter muscle. The fistula was arising from an ectopic accessory parotid gland with ectopic duct positioned anterior to masseter muscle. CT sialography and CT fistulography were very helpful in the diagnosis and surgical planning.

Index terms: Salivary glands, abnormalities and anomalies; Salivary glands, computed tomography; Fistula

Most parotid fistulas are acquired and arise secondary to trauma, surgery, malignant tumors, and inflammation (1). Rarely do they occur congenitally as malformation during fetal development (2). Although accessory parotid gland is a common variation (3), parotid fistula arising from an ectopic accessory parotid gland is extremely rare (2, 4).

We report a case of congenital parotid fistula arising from an ectopic accessory parotid gland demonstrated by computed tomography (CT) sialography and CT fistulography.

Case Report

A 5-year-old girl presented with a fistula at the facial skin of the right cheek from birth. She had a history of excision of right periaural appendix at 3 years of age. The outflow from the fistulous opening was especially abundant during food ingestion. The fluid was clear and serous. Clinical examination demonstrated a pinpoint-size opening located at the facial skin surface 20 mm posterior to the angle of the right commissure of the lips (Fig 1A). No additional abnormalities were noted.

A CT scan done before injection of contrast showed soft tissue along the lateral aspect of the masseter muscle on the right side. The parotid bed contained fat with no evidence of glandular tissue (Fig 1B). A section taken more caudad showed a small irregularly marginated soft-tissue density anterior to the right masseter muscle (Fig 1C). A CT sialogram obtained after injection of water-soluble contrast medium (Ultravist, Schering AG, Germany) into the normal orifice of the parotid duct in the oral cavity revealed opacification of the soft tissues lateral to the masseter muscle (Fig 1D). The separate soft-tissue nodule in the right buccal area was not opacified. The fistulous opening in the cheek was cannulated with a 22-gauge blunted needle, and 1.5 mL water-soluble contrast medium (Ultravist, Schering AG, Germany) was injected. A CT scan showed the soft-tissue nodule anterior to the masseter muscle in a pattern suggestive of salivary gland tissue (Fig 1E). The tissue lateral to the masseter muscle was not opacified. There was no communication between the fistula and Stenson's duct system.

The soft tissue lateral to the masseter muscle was considered to represent an ectopic parotid gland with a duct opening in the normal position in the oral cavity. The soft-tissue structure anterior to the masseter muscle was thought to represent an ectopic accessory parotid gland with an ectopic drainage pattern to the skin. The left parotid gland was considered to be normal.

Under general anesthesia, a fistulectomy (excision of the right accessory parotid gland) was performed. Pathologic examination revealed tissue typical of the parotid gland.

Discussion

Congenital salivary fistulas can originate from the parotid gland, submandibular gland, ectopic salivary gland, and, rarely, accessory parotid gland (2, 4). The sites of opening of the fistulas have been the retroauricular region, the facial skin of the cheek, the oral mucosa, and skin of the cervical region. Congenital fistulas from accessory parotid gland have typical

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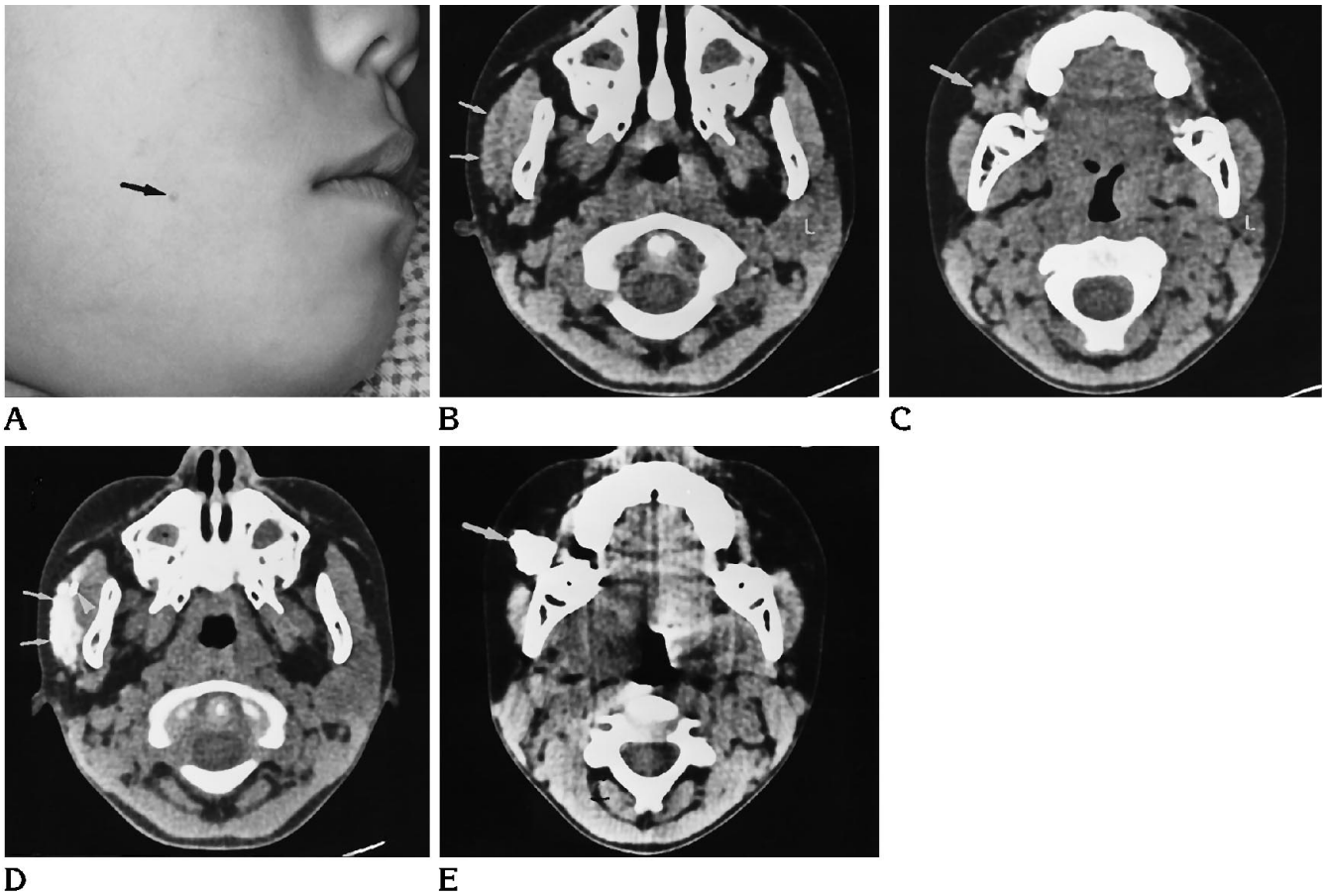

Fig 1. A 5-year-old girl with congenital accessory gland fistula.

A, There is a pinpoint-size fistulous opening (*arrow*) at the right cheek near the mouth angle.

B, On precontrast CT scan, the left parotid gland (*L*) is normal in shape and size, but no gland tissue is found in right parotid bed. Note the abnormal soft tissue lateral to right masseter muscle (*arrows*).

C, Precontrast CT scan caudad to B shows a small irregular marginated soft tissue on the buccal fat pad at the anterior border of the right masseter muscle (*arrow*), which is thought to be an ectopic accessory parotid gland. *L* indicates lower pole of left parotid gland.

D, CT sialography with cannulation of right Stensen's duct reveals abnormal location of opacified right parotid gland, which is just lateral side of masseter muscle (*arrows*). Separate soft-tissue nodule in the right buccal fat pad shown in C was not opacified (not shown). Note the opacified Stensen's duct (*arrowhead*).

E, CT fistulography with cannulation of fistulous opening reveals opacification of right ectopic accessory parotid gland (*arrow*). Opening of cutaneous fistula was anteroinferior to this ectopic accessory parotid gland (not shown).

sites of fistulous opening at the facial skin of the cheek near the mouth angle and are almost always accompanied by aural appendage (5). In this case, the patient had a history of right periaural appendix excision at the age of 3 years.

Accessory parotid glands occur rather frequently. In one study (3), 21% of dissections revealed clearly detached accessory glands at variable distances from the main gland. The accessory gland was found in close relation to Stensen's duct, because the duct passed along the lateral aspect of the masseter muscle and was usually positioned on or above the duct. In our case, the abnormal tissue was below Stensen's duct and was more anterior than usual, lying at the anterior border of the masseter muscle.

The accessory gland usually has one major tributary emptying into Stensen's duct, and thus sialography can reveal the duct system of accessory parotid glands. But we failed to opacify the small soft tissue anterior to the masseter in CT sialography and could not find any connection between duct system of this abnormal tissue and Stensen's duct at surgery. We think that *ectopic accessory parotid gland* is a more appropriate term than *accessory parotid gland* for

this condition. These separate ductal systems were also noted in another case of congenital parotid gland fistula (2).

In our case, right parotid gland was not found in the normal parotid bed but had ectopic anterior position overlying the masseter muscle. Embryologically, the parotid gland develops as an outgrowth from the buccal cavity, spreading back towards the ear (6), and the arrest during this developmental process could result in the unusual anterior position of parotid gland.

CT sialography is obtained by CT scanning of the parotid gland during injection of water-soluble contrast medium after cannulation of Stensen's duct. It has been used in patients with parotid mass and may be helpful in distinguishing intrinsic from extrinsic lesions, differentiating benign from malignant tumors, and determining the relationship of these tumors to the facial nerve (7). CT sialography allowed mapping of the gland's parenchyma and revealed abnormal location of right parotid gland in our patient. But for most of the clinical settings, magnetic resonance imaging or contrast-enhanced CT has replaced CT sialography.

CT fistulography with cannulation of fistulous opening revealed opacification of ectopic accessory parotid gland, which was unopacified at CT sialography. It was a useful technique to demonstrate the connection between fistulous tract and ectopic accessory parotid gland. Exact location of gland tissue and identification of the ductal communication with glands have therapeutic implication.

CT sialography and CT fistulography were very helpful in the diagnosis of congenital parotid gland fistula originating from an ectopic accessory parotid gland and allowed proper surgery.

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