

## CASE REPORT

# Ectopic salivary gland fistula in the sternoclavicular joint region

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**Key Clinical Message**

Although rare, ectopic salivary gland fistula should be considered in the differential diagnosis when treating a fistulous lesion on the neck because its natural history and the level of difficulty of surgery are different from those of the branchial fistula.

**KEYWORDS**

congenital fistula, neck lesion, salivary gland

## 1 | INTRODUCTION

Among fistulous lesions occurring in the neck of infants, the branchial fistula is relatively well-known.<sup>1</sup> On the other hand, the ectopic salivary gland fistula (ESGF) is relatively rare. Usually, this congenital anomaly is differentially diagnosed based on its clinical course and/or imaging findings on ultrasonography, computed tomography (CT), or magnetic resonance imaging (MRI).<sup>2</sup> However, making a definitive diagnosis in every case of congenital fistulous lesion of the neck is difficult. In this report, we present a case of ESGF in the sternoclavicular joint region which was successfully treated by surgical excision.

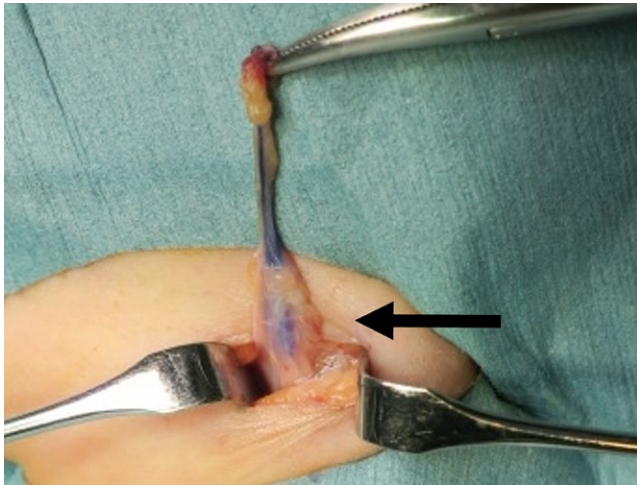
## 2 | CASE REPORT

A 7-month-old male presented at the outpatient clinic of Tokyo Metropolitan Children's Medical Center with the complaint of clear mucous drainage from a congenital pin-hole fistula in the right sternoclavicular joint region first detected at age 1 month. On physical examination, no subcutaneous mass was palpable (Figure 1). Though we suspected the

lesion to be some kind of congenital anomaly, we decided not to remove it until the patient reached an age at which imaging studies under sedation and general anesthesia could be safely performed. During the follow-up period, no signs of infection were observed, and the clinical presentation was stable. At age 1 year 11 months, an MRI was carried out, revealing a small, tubular structure continuous with the skin surface and



**FIGURE 1** Clinical presentation of the lesion. Drainage of clear mucous fluid was observed



**FIGURE 2** Resected specimen of the lesion. A soft mass suspected of being ectopic salivary gland tissue was observed after staining with crystal violet (arrow)

extending deep into the tissue in front of the sternal head of the right sternocleidomastoid muscle.

For prophylaxis and a definitive diagnosis, the lesion was surgically removed after staining of the fistula lumen with crystal violet when the patient was 2 years old. The deep end of the fistula was attached to the fascia of the sternocleidomastoid muscle (Figure 2). A soft mass was observed near the deep end of the fistula, which was stained with crystal violet and identified easily. Histopathological examination revealed lobules of salivary gland tissue around the fistula. Based on these findings, ESGF was definitively diagnosed.

### 3 | DISCUSSION

Ectopic salivary gland fistula is a rare, congenital anomaly of the head and neck region. Until now, most reports of ESGF described involvement of the anterior lower neck<sup>3-7</sup> as in the present case. Although lesions in other locations such as the cheek<sup>8</sup> and hyoid region<sup>9</sup> have been reported, such locations are considered to be quite rare. Based on this characteristic location, the origin of the anomaly is thought to lie in the branchial apparatus<sup>3-5,9</sup> or embryonic placodal duct.<sup>7</sup>

The differential diagnosis of ESGF includes branchial anomalies and congenital cutaneous fistula at the sternoclavicular joint. Although the former have been well-studied,<sup>1</sup> the congenital cutaneous fistula at the sternoclavicular joint was only recently recognized as a disease, which is considered to be a skin-side remnant of the 4th branchial cleft.<sup>10</sup> Both are more likely to involve infection,<sup>10,11</sup> which may enable them to be differentiated from ESGF, which is characterized by intermittent drainage of clear fluid, a fistula opening at the anterior border of the sternocleidomastoid muscle, and an absence of a history of infection.<sup>3,5,6</sup> There are few reports

of ESGF with infection, possibly due to the manner of its development or the shortness of the duct. A palpable mass (salivary gland tissue) close to the sinus opening may also be another distinguishing feature.<sup>7</sup> Although rare, ESGF is now recognized as an independent disease entity among congenital sinuses or fistulas.

As an example of preoperative imaging of a case of ESGF, Stingle et al<sup>6</sup> demonstrated a sinogram of a 15-year-old patient which aided in correct presurgical diagnosis. However, a sinogram is not always easy to perform in an infant. Radiation exposure may also be a concern. Although we did not perform a sinogram in the present case, we were able intraoperatively to visualize a small mass, later identified as the salivary gland, by crystal violet staining. Given the difficulty of the differential diagnosis, we believe that crystal violet staining may also be useful for avoiding an intraoperative misdiagnosis.

Surgeons who encounter a congenital lesion of the lower neck should be aware of ESGF as a possible diagnosis despite its rarity since this type of lesion is more readily removable than a brachial fistula.<sup>9</sup>

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All the authors have read the manuscript and have approved this submission. This case report was approved by the Institutional Review Board of Tokyo Metropolitan Children's Medical Center (TMCMC).

### CONFLICTS OF INTEREST

None declared.

### AUTHORS' CONTRIBUTIONS

Agata Kato designed the study and wrote the initial draft of the manuscript. Ikkei Tamada contributed to the analysis and interpretation of data, and assisted in the preparation of the manuscript. All other authors have contributed to data collection and interpretation, and critically reviewed the manuscript. All authors approved the final version of the manuscript, and agree to be accountable for all aspects of the study.

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