



CASE REPORT

# Large esophageal schwannoma mimicking thyroid tumor with egg-shell calcification on preoperative ultrasonography



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**Summary** Schwannoma tumors in esophagus are extremely rare and clinically present as dysphagia in most reported cases. Because of their rarity and need for histopathological confirmation using immunohistochemistry, an erroneous diagnostic and therapeutic approach can be adopted. A 36-year-old woman presented at the hospital with complaints of an anterior neck mass. On ultrasonography, a large left thyroid mass with egg-shell calcification was suspected. However, the thyroid surgeon found that it was not a thyroid tumor. An incision biopsy was performed for histopathological analysis, which revealed a schwannoma. Then, salivary leakage occurred through the cervical incision site, suggesting that the incisional biopsy had caused esophageal perforation. She was transferred to our department and underwent emergency surgery. We successfully resected the tumor and controlled the infection without any further injury to the esophagus, although it was a revision surgery and the wound was greatly infected. We believe that it is important to always keep in mind that an atypical presentation of esophageal schwannoma may lead to the development of, for example, a large nodule in the left thyroid gland involving the esophagus.

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## 1. Introduction

Esophageal schwannoma is extremely rare. Since it was first reported by Chaterlin and Fissore<sup>1</sup> in 1967, only 30 articles on esophageal schwannoma have been published in English.<sup>2–7</sup> Because of its rarity and difficulty in diagnosis until histopathological examination of a surgical specimen, esophageal schwannoma is hardly ever considered in patients with a large mass located at the lower anterior neck. Other tumors originating from the thyroid gland, the thymus, or the lymph node are considered.

Herein, we report the case of a large esophageal schwannoma mimicking a thyroid tumor on preoperative ultrasonography (US), resulting in iatrogenic esophageal perforation.

## 2. Case Report

The patient was a 36-year-old woman who initially presented with a history of progressive anterior neck mass that had lasted several months. The patient had mild dysphagia, but was able to consume a regular diet. The patient visited our hospital (but not our department) and underwent neck US. On the US, a large mass with egg-shell calcification was identified in the left thyroid gland, presenting posterior acoustic shadowing (Fig. 1). There was no abnormal lymph node in the bilateral cervical region. The patient was evaluated according to the guidelines issued by the American Thyroid Association under the clinical impression of a large thyroid nodule, and underwent fine needle aspiration and thyroid function test. The result of fine needle aspiration was reported as atypia with undetermined significance, and thyroid-stimulating hormone level was within normal range. Thyroidectomy was planned to confirm the thyroid malignancy and manage her discomfort. However, after thyroidectomy, the thyroid surgeon found that the tumor was not originating from the thyroid gland. It was

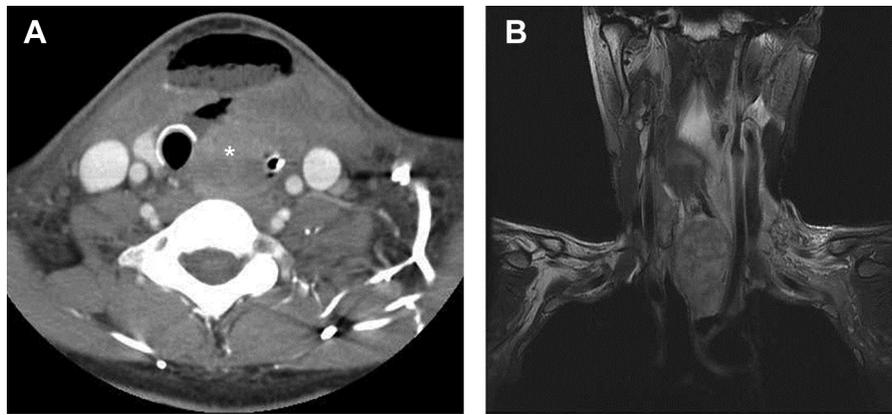


**Figure 1** On initial ultrasonography, a large mass with egg-shell calcification was identified in the left thyroid gland, presenting posterior acoustic shadowing.

located posterior to the thyroid gland, compressing the thyroid gland anteriorly. On the basis of this finding, the thyroid surgeon did not perform complete excision of the mass; instead, an incisional biopsy was performed for histopathological diagnosis analysis. Histopathology revealed schwannoma. Seven days after initial surgery, the patient was referred to our department in a poor clinical condition. The patient was unable to eat or drink. Salivary leakage through the cervical incision site was occurring since Postoperative Day 2, suggesting that incisional biopsy had caused esophageal perforation. The wound was greatly infected and foul smelling. To determine the tumor size and extent of infection, computed tomography (CT) and magnetic resonance imaging (MRI) were performed. On CT scan, the tumor was identified as located in the cervical esophagus, displacing the trachea to the right side and obstructing the esophageal lumen (Fig. 2A). Collection of saliva was found, and severe inflammation was noted. On coronal MRI, a fusiform tumor of approximately 6.5 cm in size, which extended to the substernal area, was identified (Fig. 2B). We planned surgical exploration to control the infection causing the abscess and to excise the tumor completely as soon as possible because more severe adhesion was anticipated if further delay was introduced. During surgery, we first identified the recurrent laryngeal nerve, which ran along the esophagus, and the perforation site at the esophagus. A large tumor was palpated at the posteromedial aspect of the cervical esophagus. With best effort to preserve mucosal integrity of the esophagus, we attempted to expose the tumor, which was located within the submucosal plane of the esophagus. After peeling the tumor off the esophageal wall laterally and superiorly, we could completely dissect the tumor, including its substernal portion, by pulling it superiorly without sternotomy (Fig. 3). The esophageal perforation site—the result of the previous incisional biopsy—was closed primarily and reinforced with an inferior-based sternocleidomastoid muscle rotation flap. On gross inspection, the tumor was well capsulated and was 6.5 cm × 4.5 cm in size. The cut surface of the tumor appeared homogenous yellow (similar to the appearance of a typical schwannoma), but there was no calcification (Fig. 4). Pathologic examination confirmed a benign schwannoma. A barium swallow study on Postoperative Day 4 revealed that there was no leakage. The patient started oral intake of a soft diet. The patient was allowed to resume a regular diet on Postoperative Day 7. At the 1-year follow-up, the patient was well without any complications.

## 3. Discussion

Unlike in previous reports of esophageal schwannoma, our patient's chief complaint was anterior neck mass rather than dysphagia. Of the approximately 30 case reports on esophageal schwannoma, in only one report the chief complaint of the esophageal schwannoma was palpable neck mass as well as dysphagia.<sup>7</sup> In our case, tolerable swallowing despite a large esophageal mass was possibly due to the tumor's characteristic of indolent growth and elasticity of the esophagus. Therefore, US was used as the first imaging modality for neck mass, rather than endoscopic examination for esophageal lesion. On US, the mass



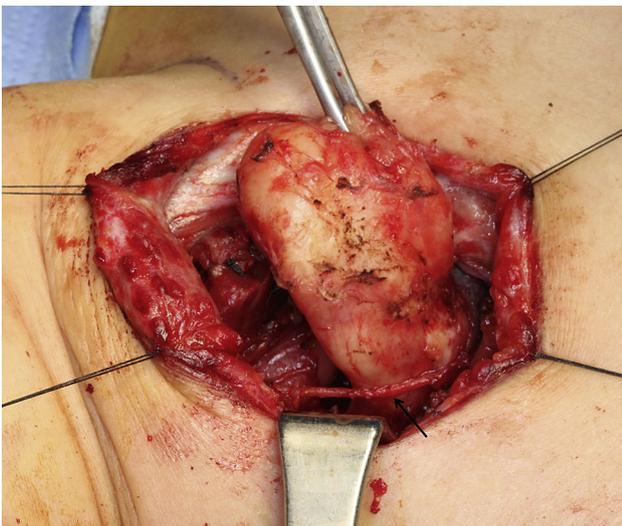
**Figure 2** (A) Neck computed tomography scan shows a large esophageal mass (asterisk) displacing the trachea to the right side and nasogastric tube to the left side. Collection of leaked saliva and severe inflammation are suggested. (B) Coronal T2 magnetic resonance imaging reveals a 6.5-cm fusiform tumor extending to the substernal area.

appeared to be a thyroid tumor with egg-shell calcification occupying almost the entire left thyroid gland. Interestingly, however, echogenicity of the esophagus was not found on that image. The radiologist might have considered that it was due to posterior acoustic shadow of egg-shell calcification or compression of esophagus by the tumor. Unfortunately, once thyroid nodule was suspected as the causative lesion of the neck mass, additional imaging modalities, such as CT or MRI, were not performed because US alone is widely accepted as a diagnostic imaging tool for thyroid nodule, and none of the guidelines issued by the American Thyroid Association and American Association of Clinical Endocrinologists/Associazione Medici Endocrinologi/European Thyroid Association recommend the routine use of CT, MRI, and positron emission tomography for evaluating thyroid nodules.<sup>8,9</sup> However, in review of the initial US image obtained, we suggest that the hyper-echogenic lining that appeared within the left thyroid gland

was not egg-shell calcification of the tumor, but mucosal lining of the esophagus, which was anteriorly displaced by the esophageal tumor. In fact, we did not find any calcified lesion within the tumor on macro- and microscopic examinations of the specimen.

Because benign schwannoma is a submucosal tumor, the tumor was excised completely without perforation of the esophagus or performing esophagectomy in almost all reported cases.<sup>2,3,5-7</sup> However, in the present case, tumor excision was very challenging because curative surgery for esophageal schwannoma was performed as a revision surgery, and the wound was greatly infected. Nevertheless, we successfully resected the tumor and controlled the infection without any further injury to the esophagus, after making the decision of immediate surgical exploration.

In a review of the literature, only one Czech report was found in which esophageal schwannoma was misdiagnosed as a thyroid tumor preoperatively, as in our case.<sup>10</sup> Although esophageal schwannoma is extremely rare, the surgeon can collect the necessary clinical information for differential diagnosis if sufficient imaging studies are performed. It is important to always keep in mind that an atypical presentation of esophageal schwannoma may lead



**Figure 3** After peeling the tumor off the esophageal wall laterally and superiorly, we could completely dissect the tumor, including its substernal portion, by pulling it superiorly, while identifying and preserving the recurrent laryngeal nerve (arrow).



**Figure 4** Cut surface of the tumor appeared homogeneously yellow, similar to a typical schwannoma, but no calcified tissue (suspected on the preoperative ultrasonography) is observed.

to the development of, for instance, a large nodule in the left thyroid gland involving the esophagus.

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