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Absence of the Sternocleidomastoid Muscle in a Patient That Underwent Neck Dissection for Squamous Cell Carcinoma of the Tongue

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The congenital absence of a skeletal muscle is a rare cause of congenital muscular torticollis, and the condition is associated with various unusual anatomical structures. We describe a rare case of congenital absence of the sternocleidomastoid muscle associated with squamous cell carcinoma of the tongue in a patient who underwent neck dissection. In this case, both the external jugular vein and the spinal accessory nerve were absent. However, we found that branches of the C3 nerve extended from the cervical plexus to the trapezius muscle and seemed to be acting as trapezius muscle motor nerves in place of the accessory nerve. After the operation, the patient was able to lift and abduct his right arm, and his shoulder did not droop.

INTRODUCTION

Lymph node metastasis is one of the most important prognostic factors for patients with oral squamous cell carcinoma. However, surgical treatment for lymph node metastasis from squamous cell carcinoma sometimes causes shoulder problems due to the complete interruption of the motor nerves innervating the trapezius muscle (12,19).

There have been various reported cases of congenital muscular torticollis (CMT) with agenesis of sternocleidomastoid (SCM) and trapezius muscles (1,5,6,11,17). However congenital absence of SCM and spinal accessory nerve (SAN) has been only a few case. Canby and Cookman (3) described left-sided absence of SCM and trapezius accompanied by the loss of innervation by SAN in two cadavers. This is the first reported case of a patient with CMT involving the absence of SCM and SAN to undergo modified radical neck dissection for lymph node metastasis from squamous cell carcinoma. It is worth noting that the patient achieved good postoperative trapezius muscle function.

CLINICAL CASE

A 40-year-old Japanese male presented with a month-long history of pain and a stomatitis on the right side of his tongue. Clinical examinations revealed a 22 x 18 x 8 mm ulcerated tumorous lesion, which exhibited spontaneous and contact pain, on the right lateral margin of his tongue (Fig. 1). The patient's face was symmetric, but the right side of his neck was a little thinner than the left side. He had no clinical symptoms which suggest the nerve anomaly.



Fig.1. Intraoral findings showing an extensive ulcer with induration on the right tongue

Computed tomography (CT) examinations demonstrated an enlarged lymph node in the patient's neck (level III), but the SCM and external jugular vein could not be found (Fig. 2). Magnetic resonance imaging (MRI) detected a thin region of fibrous tissue where the SCM should have been on the right side of the patient's neck (Fig. 3).



Fig.2. CT image showing the mid internal jugular node

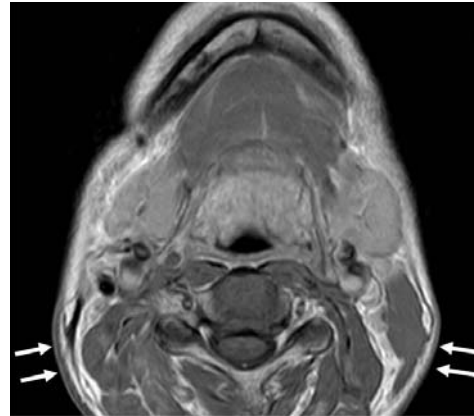


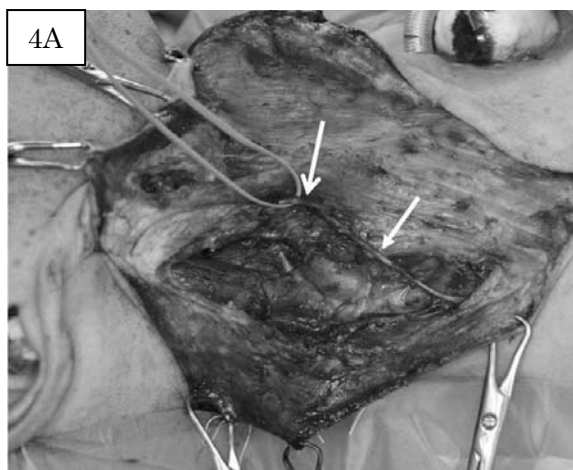
Fig.3. MRI image showing a thin region of fibrous tissue on the right side of the neck

The patient underwent an incisional biopsy of the lesion and obtained a diagnosis of well-differentiated squamous cell carcinoma.

Under a diagnosis of tongue cancer (T1N1M0), the patient underwent a partial glossectomy, modified radical neck dissection, and concomitant reconstructive surgery using a radial forearm flap. A neck tissue specimen that was contiguous with the primary tumor was removed via the pull-through approach.

A Y incision was made, and flaps from the subplatysmal plane were elevated, but the external jugular vein was not present. We left the greater auricular nerve intact and tried to find the SAN at the point (Erb's point)(15) where the greater auricular nerve crossed the posterior border of the abovementioned fibrous tissue, which was considered to be degraded SCM tissue. However, we only found a bunch of sensory cervical nerves, which did not react to nerve stimulation (Fig. 4A).

Even after we had divided the posterior belly of the digastric muscle, the SAN could not be found around the jugular vein (Fig. 4B). A level 5 dissection was performed, and we found C3 nerve branches that extended from the cervical plexus to the trapezius muscle, which was confirmed by electrical nerve stimulation using. We preserved these nerves, as they seemed to act as motor nerves for the trapezius muscle in place of the SAN (Fig. 4C).



A CASE OF CMT UNDERGOING NECK DISSECTION

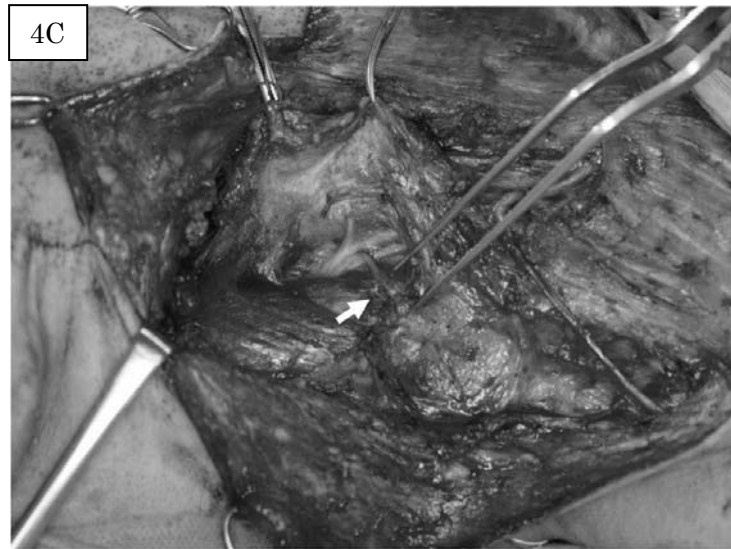


Fig.4. Intra operative findings. 4A: Only a bunch of sensory cervical nerves was found at Erb's point. 4B: Posterior belly of the digastric muscle was resected, and the internal jugular vein became visible (arrow), but the SAN could not be found around this. 4C: C3 nerve branches was confirmed using a nerve stimulator that extended from the cervical plexus to the trapezius muscle

Soon after the operation, the patient was able to lift and abduct his right arm, and his shoulder did not exhibit any drooping (Fig. 5). Histological examinations of the resected material demonstrated five metastatic lymph nodes, and postoperative radiation therapy was administered. The patient is alive without any clinical symptoms of recurrence or shoulder problems at 50 months after surgery.



Fig.5. Soon after operation, the patient could lift and abduct his right arm

DISCUSSION

The congenital absence of skeletal muscles is rare and displays an incidence of between 0.3% and 1.9% births (14). The congenital absence of the SCM has been described as a cause of torticollis and asymmetry of the neck in early infancy (17). Its pathogenesis remains unknown, but inflammatory, vascular, neuropathic, or myopathic insults during early embryogenesis and genetic causes have been implicated (1,11).

CMT is treated with infant stimulation and passive stretching, which result in improvements in more than 90% of infants. Torticollis that persists beyond the first year of life despite conservative therapy is treated with corrective surgery, e.g., the release of the fibrotic band (4,5,6,20). The patient in the present case achieved improvements in his CMT after stretching alone. In fact, he did not exhibit any clinical symptoms at the time of

his first visit to our hospital; however, CT and MR images showed that his right SCM was absent. He had no hoarseness after operation, so it seems to be no anomaly in cranial root of the accessory nerve.

The most important prognostic factor for patients with oral squamous cell carcinoma is the presence of lymph node metastasis. Radical neck dissection (RND) has become the standard treatment for neck metastasis from oral cancer. However, this surgical procedure causes functional morbidities in the shoulder because it sacrifices the SCM and SAN. So, various modified RND (mRND) procedures have been developed to reduce the severity of such postoperative functional disadvantages, which include pain and limited shoulder mobility (19). On the other hand, resection of the SAN during RND does not always result in shoulder function problems, while some patients who undergo mRND suffer shoulder problems despite the SAN being preserved (16).

In previous studies, it was reported that the SAN exited from the posterior border of the SCM (in the posterior triangle of the neck) at a mean distance of 1–1.5 cm from the great auricular nerve in the deep layer above the investing deep fascia (2,13). Salquarelli *et al.* (15) reported that the distance between the SAN and Erb's point (where the great auricular nerve emerges from the posterior border of the SCM) ranged from 0 to 3.8 cm (mean: 1.53 cm). In addition, the distance between the location where the SAN enters the trapezius muscle and the clavicle ranged from 2.5 to 7.3 cm (mean: 4.8 cm). We also use Erb's point to identify the SAN (19). Within the posterior cervical triangle, the SAN lies between the superficial and prevertebral layers of the cervical fascia and is just lateral to the levator scapulae muscle. The fibers of the SAN enter the trapezius muscle anteriorly, approximately 5 cm medial to the attachment of the muscle to the lateral third of the clavicle (18). In most cases, a very small additional branch arises from the SAN about 2 cm medial to the trapezius muscle. This branch enters the muscle approximately 2 to 3 cm cranial to the main nerve (9).

The SAN leaves the jugular foramen between the internal carotid artery and the internal jugular vein. It then passes ventrally (56%) or dorsally (44%) across the jugular vein (9) or in rare cases through it (3%) (18). The SAN always passes medial to the styloid process and the posterior belly of the digastric muscle. It then crosses the floor of the apex of the carotid triangle before entering deeper tissue. Khaki *et al.* present a variant of the spinal accessory nerve plexus that contributed to the formation of the ansa cervicalis. (8) Jones and Stell (7) reported that they found branches of the C3 or C4 nerves that passed across the posterior triangle and innervated the trapezius muscle directly or joined the SAN just before its entry into the trapezius muscle. It was reported that one to three (two in most cases) trapezius branches of the cervical plexus exist and contribute to the innervation of the trapezius muscle. These branches pass through the posterior triangle of the neck independently of the SAN, very rarely intermingling with it (9).

Kusakabe *et al.* (10) performed the double free muscle technique using the trapezius branches of the cervical plexus and reported that these branches appeared to be equivalent to the SAN with regard to the time to re-innervation. The present patient achieved good postoperative trapezius muscle function; i.e., he exhibited a range of motion of 180° during lateral abduction of his shoulder, and branches from the cervical plexus (C3) appeared to contribute to the motor innervation of his trapezius muscle.

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A CASE OF CMT UNDERGOING NECK DISSECTION

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