A 54-year-old female with complex regional pain syndrome (CRPS) type 1 secondary to a motor vehicle accident presented to the hospital with a 2- to 3-week history of worsening generalized body pains seeking intravenous immunoglobulin (IVIG) infusion. At presentation, the patient also noted mouth dryness and pain radiating to her head, symptoms typical for her CRPS. Additionally, the patient later reported several months of increased multifocal pain with a particular focus in the region of the left upper palate. As the patient’s primary team searched for an appropriate immunomodulatory regimen to reduce the patient’s reliance on IVIG for analgesia, an oral maxillofacial surgery consult was obtained, and a contrast-enhanced maxillofacial computed tomography (CT) was subsequently ordered.

Multiplanar contrast-enhanced maxillofacial CT was performed and demonstrated facial soft tissues within normal limits, along with no evidence of a facial bone fracture or malalignment. A defect in the left posterior maxilla was noted secondary to prior dental extractions, which was unchanged from imaging 3 years prior. Also unchanged from prior imaging was complete absence of the anterior belly of digastic muscle (ABDM) on the right side that was compatible with agenesis (Figure 1).

Incidentally, an abnormal intracranial vascular malformation was noted on the maxillofacial CT, which prompted magnetic resonance imaging of the head, which also demonstrated agenesis of the right ABDM (Figure 2).

Our patient was nonsyndromic, had no cosmetic abnormalities, and reported no difficulty in swallowing or phonation. She denied prior neck surgeries and was completely asymptomatic.

Figure 1. Axial computed tomography (CT) utilizing soft-tissue window (width 400, level 50) demonstrates a fat-filled right submental fossa at the presumed location of the anterior belly of the right digastic muscle (red arrow). A normal anterior belly of the left digastic muscle is seen (yellow arrow).

Figure 2. Coronal T2-weighted (left) and postcontrast T1-weighted (right) magnetic resonance imaging (MRI) brain obtained at the same hospitalization to further evaluate the intracranial vascular anomaly confirmed aplasia of the anterior belly of the right digastic muscle (red arrows). A normal anterior belly of the left digastic muscle is seen (yellow arrows).
Complete absence of the ABDM is a rare anatomic anomaly that has not been frequently reported in the literature, noting that it is most commonly seen in syndromic patients having hemifacial microsomia. MacQuillan et al found that in patients who had features matching hemifacial microsomia such as orbital deformity, mandibular hypoplasia, ear deformity, and cranial nerve (CN) VII deficits, agenesis of the anterior belly of digastric was found to be 40%. In syndromic patients with CN VII deficits, the ABDM can be used for reanimation of the lower lip by using an ABDM transfer. Due to such a high rate ABDM agenesis in syndromic patients, MacQuillan et al recommended obtaining preoperative imaging to confirm the presence of the ABDM.

Gibson et al attempted to establish the incidence of ABDM agenesis or atrophy in patients who are nonsyndromic and who have not had previous surgeries on the neck. In their study using ultrasound on 1484 patients, unilateral atrophy or agenesis was found in 3 patients with an overall incidence of 0.2%. All 3 of the patients with agenesis of the ABDM were cosmetically normal without decreased functionality, similar to our patient.

The ABDM is uniquely valuable to the otolaryngologist as a prominent surgical landmark. Neck dissections are generally performed by locating the superficial borders and exposing the anatomic structures in a superficial to deep fashion. Level Ia and Ib selective neck dissections are often performed for removing submental and submandibular lymph nodes in oral cancer by using the ABDM as the borders. Injury to the marginal mandibular nerve during a level I neck dissection can occur resulting in a defect in depressing the lower lip and an asymmetric smile. Therefore, it is important for the surgeon to be familiar with the anatomy of level Ia and Ib neck dissections. Accordingly, agenesis of the ABDM may result in intraoperative confusion leading to an increased risk of iatrogenic injury to the nerve.

Accurately identifying the anomalous absent ABDM by the radiologist and/or surgeon is crucial as it may reduce the risk of intraoperative iatrogenic nerve injury, alter the plans to reanimate a lower lip deficit, and prevent misdiagnosis of other submental pathology.

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