CASE REPORT

Anomaly of digastric muscle: an alarm bell for head and neck surgeon

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Abstract. Objectives: Here follows the discussion of a case of hypoplasia of both bellies of digastric muscle and the difficult during neck dissection, because of his surgical importance like a pivotal landmark. Methods: We reported a case report concerning digastric muscle abnormalities, accidentally discovered during neck dissection due to surgical excision of a glottic squamous cell carcinoma staged as cT3N0. A brief literature review was done to compare and research similar cases. **Results:** Literature counts several reports of digastric muscle abnormalities, namely involving the anterior belly. Little has been written about hypoplasia of digastric muscle. **Conclusion:** Digastric muscle abnormalities are rare, which can be absent or, more frequently, duplicated. Albeit its anomalies are anecdotal, it is advisable to give due consideration to the hypoplasia of both bellies of the digastric muscle during the analysis of radiological imaging, in order to prevent the risk of operative complications. According to our knowledge, this is the first and singular case of digastric muscle's hypoplasia. (www.actabiomedica.it)

Keywords: Digastric muscle, Absence of digastric muscle, Neck dissection, Anatomical variants, Surgical landmarks

Introduction

The digastric muscle is a fundamental landmark in neck surgery. It is called digastric because it is formed by two bellies, the anterior and posterior one, connected on the hyoid bone by an intermediate tendon. This muscle acts to pull downwards the jaw and to rise the hyoid bone during swallowing; it also contributes during chewing and speaking. (1)

The anterior belly goes from the digastric fossa on the internal side of the midline mandible to the body of the hyoid bone. (2) It derives from the first pharyngeal arch, together with other muscular structures and the trigeminal nerve (V C.N.), which innervates it through the inferior alveolar nerve, branch of the mandibular one. (1) Its arterial supply comes from a branch of the facial artery: the submental artery.

The posterior belly goes from the medial side of the mastoid tip to the greater cornu of the hyoid bone. (3) It derives from the second pharyngeal arch, together with several other structures, among which the facial nerve (VII C.N.), responsible for its motor innervation. Its arterial supply comes from two branches of the external carotid artery: the posterior auricular and the occipital arteries. (1)

The digastric muscle plays an important role during neck dissection. It helps dividing the suprahyoid region into two different triangles:

 Sub-mandibular triangles: one per each side, delimitated posteriorly by the posterior belly of the digas2

tric muscle, anteriorly by the anterior one and superiorly by the inferior margin of the mandible. (1) At this level, the digastric muscle plays an important role as a landmark for the lingual artery, which can be detected with the identification of two different triangle: Béclard's triangle and Pirogoff's triangle. The former is delimitated by the posterior belly of the digastric muscle; the greater cornu of the hyoid bone and the hyoglossus muscle. The latter is delimitated by the intermediate tendon of the digastric muscle; the mylohyoid muscle and the hypoglossal nerve (XII C.N.). (4)

• Sub-mental triangle: on the midline, delimitated on each side from the anterior bellies of the digastric muscle and inferiorly from the hyoid bone body.

The digastric muscle is also of pivotal importance as a landmark for the research of the carotid artery, and hypoglossal nerve which lies in a deeper plane than the muscle. Infact it constitutes the upper limit of the Farabeuf triangle, while it is delimitated laterally from the inner jugular vein and medially from the tiro-linguofacial branch. (5) The anterior belly represents a fundamental landmark in the sublevel classification described by Robbins et al. dividing sublevel Ia from Ib. (2,6) Moreover, during parotidectomy, it is an important indicator of the emergence depth of the facial nerve. (7)

More specifically, the posterior belly of the digastric muscle is also known as "resident's friend" because its identification during neck dissection helps protecting essential anatomical structures, such as the internal jugular vein, the accessory cranial nerve, the hypoglossal nerve, the marginal mandibular nerve, the submandibular gland and the retromandibular vein.

Given its anatomical importance, the hypoplasia of the digastric muscle, albeit rare, must be an alarm bell for the head and neck surgeon during neck dissection, since there is an increased risk of surgical complications due to the lack of landmarks aimed to identify and preserve the aforementioned structures.

Case Report

Here follows the case of a 54-year-old man, referred to our attention due to self-reported dysphonia and dysphagia, arisen during the previous months, in absence of dyspnea. An endoscopic examination followed by a neck-thorax contrast-enhanced CT scan was hence performed. They showed the presence of an expanding lesion in the glottic-supraglottic region, which involved the false and true vocal cords, including the anterior commissure with invasion of anterior left paraglottis space. The lesion was then biopsied, revealing a squamous cell carcinoma of Grading 3 originating from the glottic larynx. According to 8th Edition of AJCC TNM a cT3 N0 lesion was described.

Therefore, during multidisciplinary consultation, the patient was selected for open partial horizontal laryngectomy preserving the right crico-arythenoid unit (OPHL type IIA + Left Ary) and bilateral selective neck dissection (levels II-IV).

The surgery began with the bimastoid P. Andrè incision. A sub-platysma flap was set up. After the right selective neck dissection (SND levels II-IV), the next step was to detach the investing layer of the deep cervical fascia from the left sternocleidomastoid muscle. Later on, the accessory nerve was identified at its insertion on the sternocleidomastoid muscle, hence exposed in its course across level II. It was then followed superiorly, identifying the internal jugular vein. For this step is mandatory to search the posterior belly of digastric muscle, needed to delimitate the upper boundary of the neck dissection.

During the research of this anatomical landmark the operators became aware of an anomaly that was an important hypoplasia of posterior belly of the left digastric muscle; the dissection continued with the inspection of the anterior belly of the muscle, that, similarly, appeared hypoplastic (Fig. 1). The dissection hence continued, even if it was more difficult to identify and preserve anatomical structures in absence of the digastric muscle.

This anatomical anomaly only affected the left side. On the right side there was no evidence of alteration of the various surgical landmarks.

The surgical operation then continued with the supracricoid laryngectomy and subsequent temporary tracheostomy. The post-operative period was unevent-ful, and the patient was discharged from the hospital 19 days after surgery. Nasopharyngeal tube was removed on 12th day with an optimal swallowing and a satisfactory laryngeal function.



Figure 1 (A, B): Intraoperative view: the white arrow indicates the hyopoplasia of digastric muscle. OM: Omohyoid muscle; SMG: Submandibular Gland; PG: Parotid Gland, SCM: Sternocleidomastoid Muscle; IJV: Internal Jugular vein.

Discussion

The digastric muscle is a pivotal landmark for head and neck surgeons. It is called "resident's friend" since its identification during surgery can help the operator to recognize and spare vital structures like the internal jugular vein, the accessory cranial nerve, the hypoglossal nerve, the marginal mandibular nerve, the facial nerve.

Given its particular embryogenetic development, with the anterior belly deriving from the first pharyngeal arch and the posterior belly from the second one, several abnormalities have been described. Most frequently the anterior belly is involved and generally they represent an accidental discovery, since these alterations are asymptomatic. In 2019 Kim and Loukas (1) analyzed literature and classified the most common abnormalities of the anterior belly of the digastric muscle which varied from the presence of a bilateral or monolateral anterior accessory bellies with normal insertions on the hyoid bone (8,9); in some cases, the accessory belly crossed the midline, ending on the contralateral intermediate tendon or on the middle part of the hyoid bone. (10-12) Yüksel in 2001 described an accessory anterior belly originating from the mylohyoid raphe and inserting on the ipsilateral intermediate tendon, mono or bilaterally. (13) Sargon in 2007 described the presence of multiple bilateral accessory bellies. (14) Various combination of these abnormalities involving the anterior bellies have been reported through time. (1-10, 14)

Concerning the abnormalities of the posterior belly of the digastric muscle, there are only few reports in current literature: in 2006 Ozgursoy at al. described a case of accessory posterior belly with normal proximal insertion on the mastoid bone and distal one on the strip muscle of the neck. (3) Later in 2015, Zhao at al. described a similar case of accessory posterior belly inserted distally on the infrahyoid muscles, associated with the absence of ipsilateral omohyoid muscle. (15, 16) Recently, Yuen at al. reported a case of complete absence of the posterior belly of the digastric muscle with normal anatomy of the remnant structures of the neck, the contralateral side wasn't examined.² Similarly to this last report, our patient presented a complete hypoplasia of the left digastric muscle, which was discovered during neck dissection during a horizontal laryngectomy for larynx squamous cell carcinoma. In all reported cases, authors highlighted the difficult to complete a safe surgery in absence of the "resident's friend", a meaningful landmark for head and neck surgery.

In our case, the neck dissection was performed with some difficulties for the absence of this landmark and with an elongation of usual surgical times. However, all anatomical structures were preserved.

After the operation, a more accurate study of preoperative contrast-enhanced neck CT scan confirmed



Figure 2. (A, B, C): CT scan imaging. In the three projections (Sagittal, Axial and Coronal projection). The anatomical defect was highlighted with a circle and compared with the contralateral side.

the absence of the posterior belly of the left digastric muscle, which had gone unbeknownst (Fig. 2).

This case reminds the importance for both surgeons and radiologists to analyze more thoroughly pre-operatory imaging since anatomical variants, even if rare, can seriously complicate surgery, especially in case of patients suffering from head and neck cancer, in which the anatomical subversion due to tumoral growth can further puzzle the whole picture. Even more attention should be paid from less expert surgeons, who could suddenly find themselves in trouble without the "resident's friend".

Therefore, a pre-operative imaging (MRI imaging better than CT scan) can help the surgeon to highlight details, which can be investigated and studied to exclude surprise during the dissection. The absence of the usual surgical landmarks, previously well pointed up, allows the surgeon to evaluate and make use of adjacent anatomical structures, able to guide him during the different steps.

Conclusion

The digastric muscle is an important landmark in head and neck surgery, but sometimes it can present anatomic abnormalities that can trick surgeons during neck dissection. Anatomical variants of this muscle namely involve the anterior belly, with its absence of duplication. There have been described some cases of posterior belly abnormality, with accessory posterior bellies or its complete absence. Our case, a hypoplasia of both bellies, was a singular anatomic anomaly not yet discussed in literature. The anomalies involving the digastric muscle can lead to more complicated neck dissection, being it fundamental to recognize and preserve neuro-vascular structures. Therefore, an accurate study of pre-operative imaging must always be the starting point for surgical planning, in order to avoid unwanted surprises.

Conflicts of interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

References

- 1. Kim SD, Loukas M. Anatomy and variations of digastric muscle. Anat Cell Biol. 2019;52(1):1-11.
- Yuen SN, Gengler I, Badhey AK, Tang AL, Zender C. Absent Posterior Belly of Digastric Muscle. The Laryngoscope. Published online December 10, 2020. doi:10.1002/ lary.29317
- Ozgursoy OB, Kucuk B. Unique variation of digastric muscle: a confusing landmark for head and neck surgeons. Acta Otolaryngol (Stockh). 2006;126(8):881-883.
- 4. Tubbs RS, Rasmussen M, Loukas M, Shoja MM, Cohen-Gadol AA. Three nearly forgotten anatomical triangles of the neck: triangles of Beclard, Lesser and Pirogoff and their potential applications in surgical dissection of the neck. Surg Radiol Anat SRA. 2011;33(1):53-57.
- 5. Singh M, Vashistha A, Chaudhary M, Kaur G. Forgotten triangles of neck. Ann Maxillofac Surg. 2016;6(1):91-93.
- 6. Robbins KT, Medina JE, Wolfe GT, Levine PA, Sessions RB, Pruet CW. Standardizing neck dissection terminology.

Official report of the Academy's Committee for Head and Neck Surgery and Oncology. Arch Otolaryngol Head Neck Surg. 1991 Jun;117(6):601-5.

- Mercante G, Marchese C, Giannarelli D, Pellini R, Cristalli G, Manciocco V, Ruscito P, Pichi B, Marchesi P, Spriano G. Oncological outcome and prognostic factors in malignant parotid tumours. J Craniomaxillofac Surg. 2014 Jan;42(1):59-65. Epub 2013 Mar 29. PMID: 23541937
- Larsson SG, Lufkin RB. Anomalies of digastric muscles: CT and MR demonstration. J Comput Assist Tomogr. 1987;11(3):422-425.
- Mori M. Statistics on the musculature of the japanese. Okajimas Folia Anat Jpn. 1964;40:195-300.
- Sakamoto Y, Akita K. Supernumerary muscle bundles in the submental triangle: their positional relationships according to innervation. Surg Radiol Anat SRA. 2004;26(3):245-253.
- Liquidate BM, Barros MD, Alves AL, Pereira CSB. Anatomical Study of the Digastric Muscle: Variations in the Anterior Belly. Int J Morphol. 2007;25(4):797-800.
- Ricciardiello F, Bocchetti M, Pellini R, Palladino R, Caraglia M, Boscaino A, Petruzzi G, Romano FJ, Chiurazzi B, Addeo R, Mazzone S, Mesolella M, Oliva F, Iorio B. Sarcomatoid larynx carcinoma differential clinical evolution, on field statistical considerations. Am J Otolaryngol. 2021 May-Jun;42(3):102934. doi: 10.1016/j.amjoto.2021.102934. PMID: 33526270.
- 13. Yüksel M, Yüksel E. A case with accessory digastric muscles

and its clinical importance. Ann Plast Surg. 2001;46(3):351-352.

- Sargon MF, Önderoglu S, Sürücü HS, Bayramoglu A, Demiryürek D, Özturk H. Anatomic Study of Complex Anomalies of the Digastric Muscle and Review of the Literature. Okajimas Folia Anat Jpn. 1999;75(6):305-313.
- Zhao W, Liu J, Xu J, Wang H. Duplicated posterior belly of digastric muscle and absence of omohyoid muscle: a case report and review of literature. Surg Radiol Anat. 2015;37(5):547-550.
- 16. Di Maio, P., Iocca, O., De Virgilio, A., Giudice, M., Pellini, R., D'Ascanio, L., Golusiński, P., Ricci, G. and Spriano, G. (2020), Narrow band imaging in head and neck unknown primary carcinoma: A systematic review and meta-analysis. The Laryngoscope, 130: 1692-1700.

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