Focal dystonia in ENT

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Part 1 - Laryngeal dystonia
Part 2 - Oro-mandibular dystonia

Laryngeal dystonia

Introduction

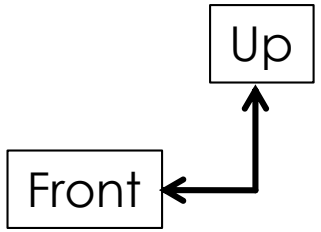
- Synonyms:
  - Spastic dysphonia
    - Should be avoided because it seems to be related to the pyramid route, which is not the case.
  - Spasmodic dysphonia
  - Laryngeal dystonia

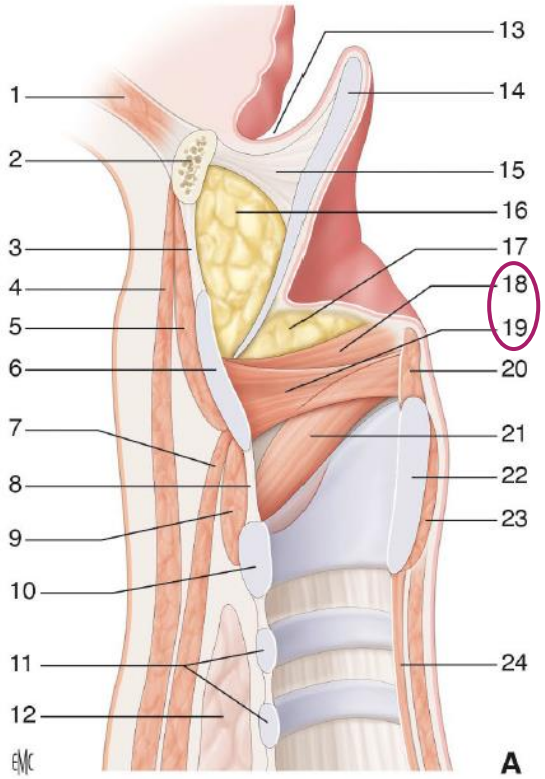
Spasmodic dysphonia-Definition

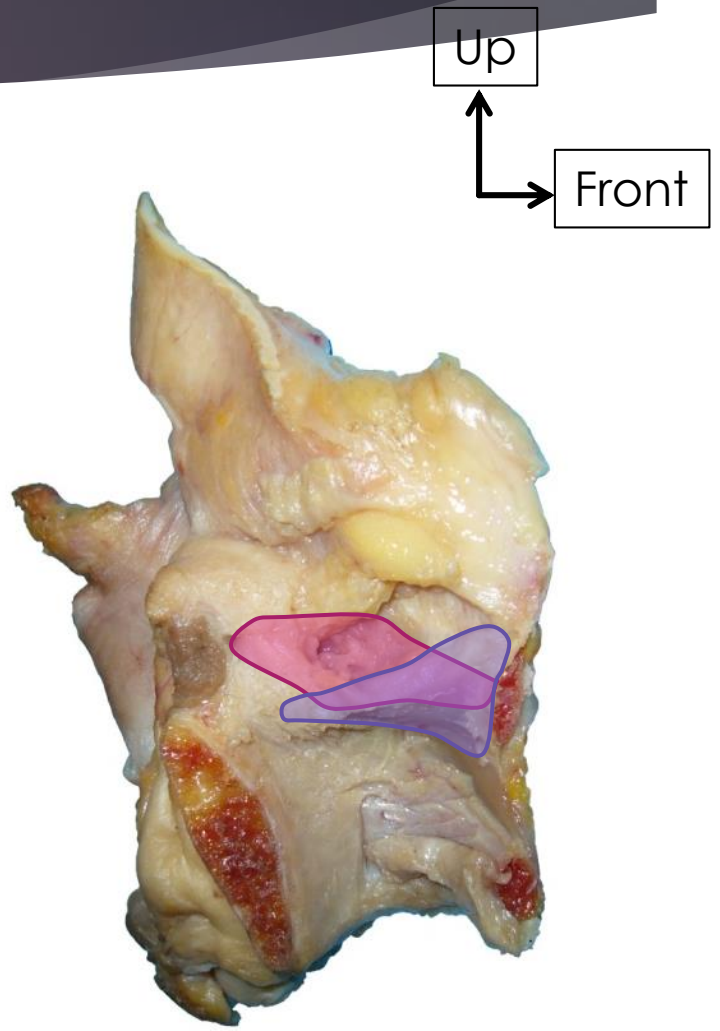
- Spasmodic dysphonia or laryngeal dystonia
  - Action-induced laryngeal movement disorder
  - May affect the adductor and/or abductor muscles
- Rare vocal disorder
- Profile:
  - begins around the age of 40,
  - Predominantly female

Anatomy

- Thyro-arytenoid muscle

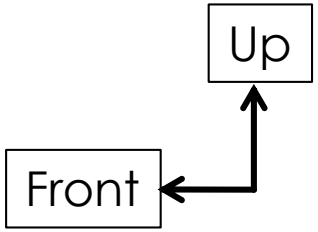


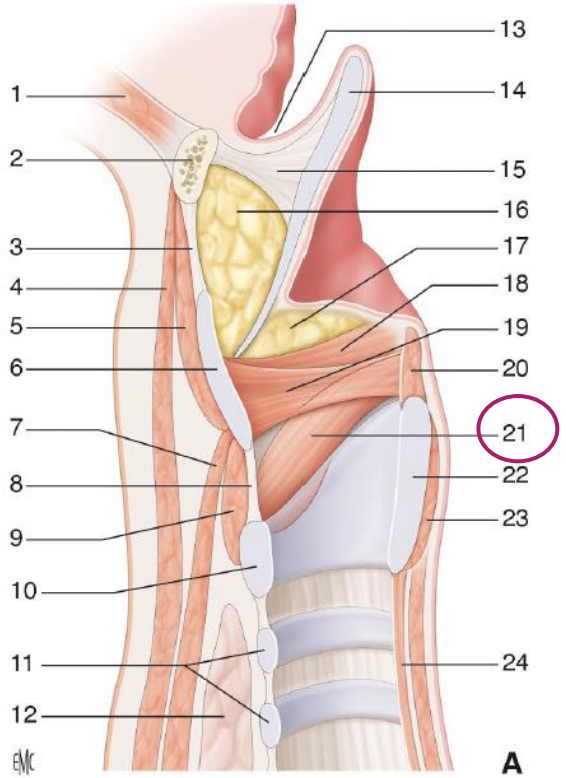


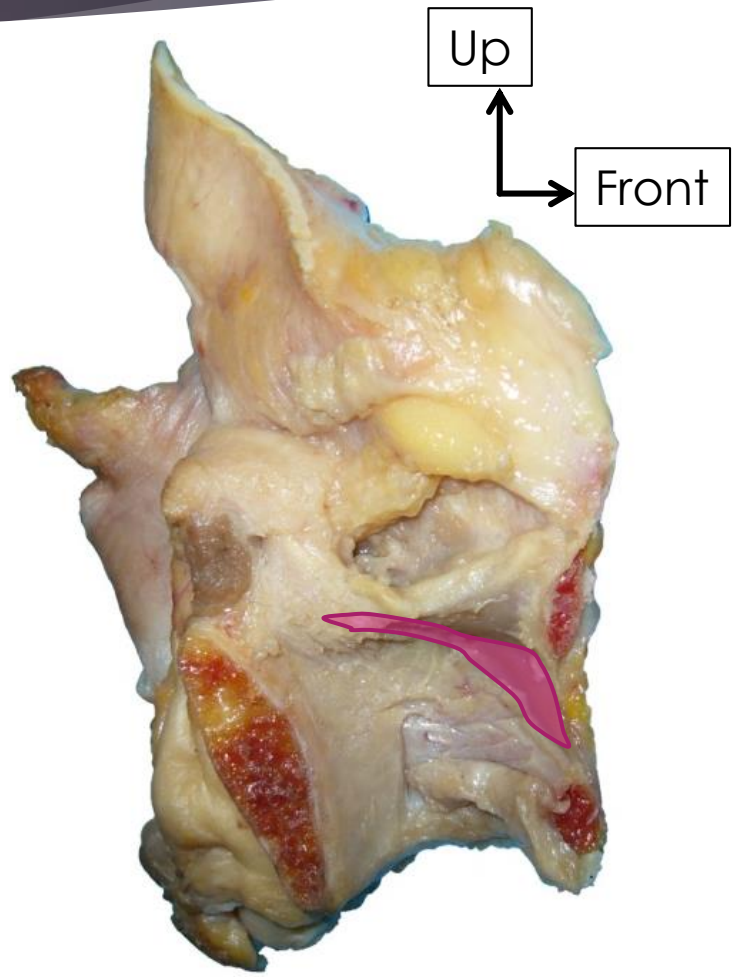


Anatomy

- Lateral crico-arytenoid muscle

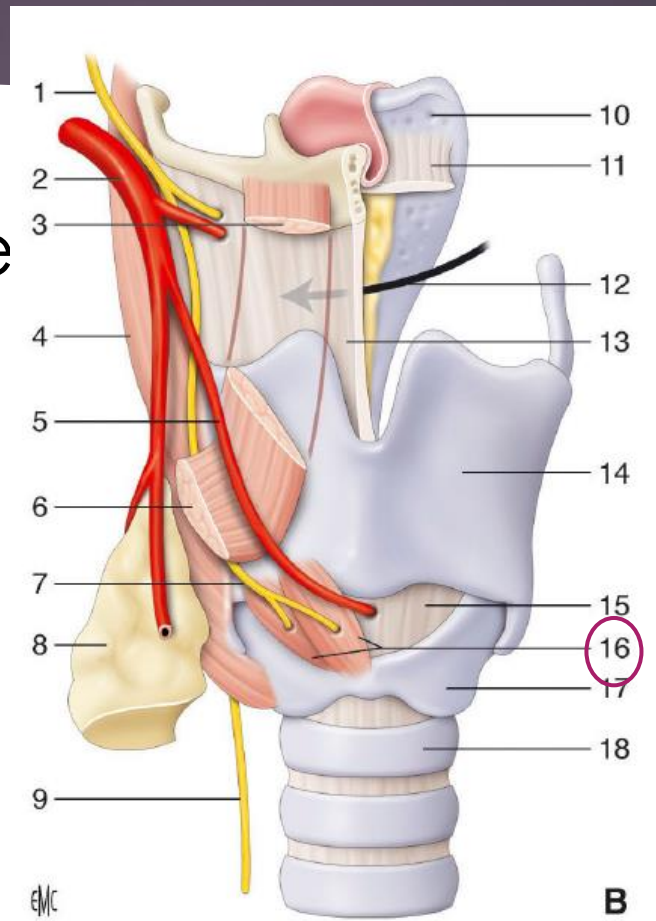






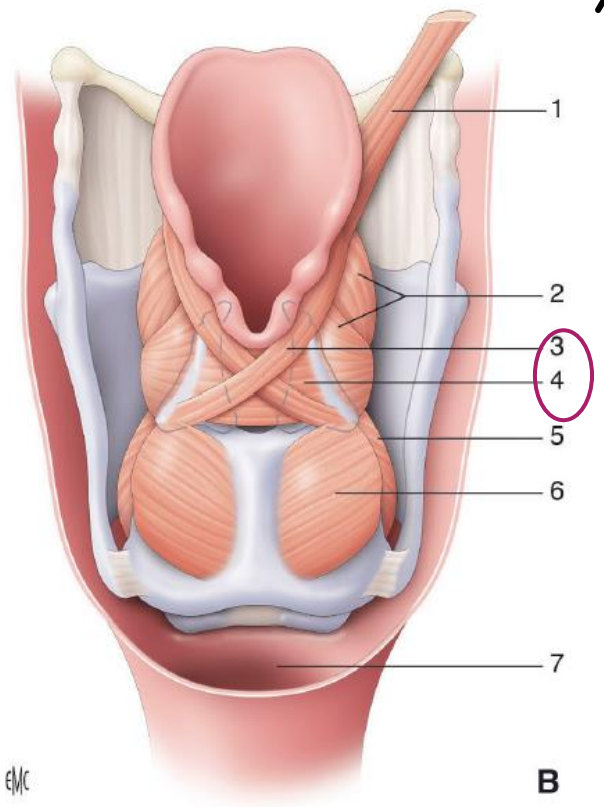
Anatomy

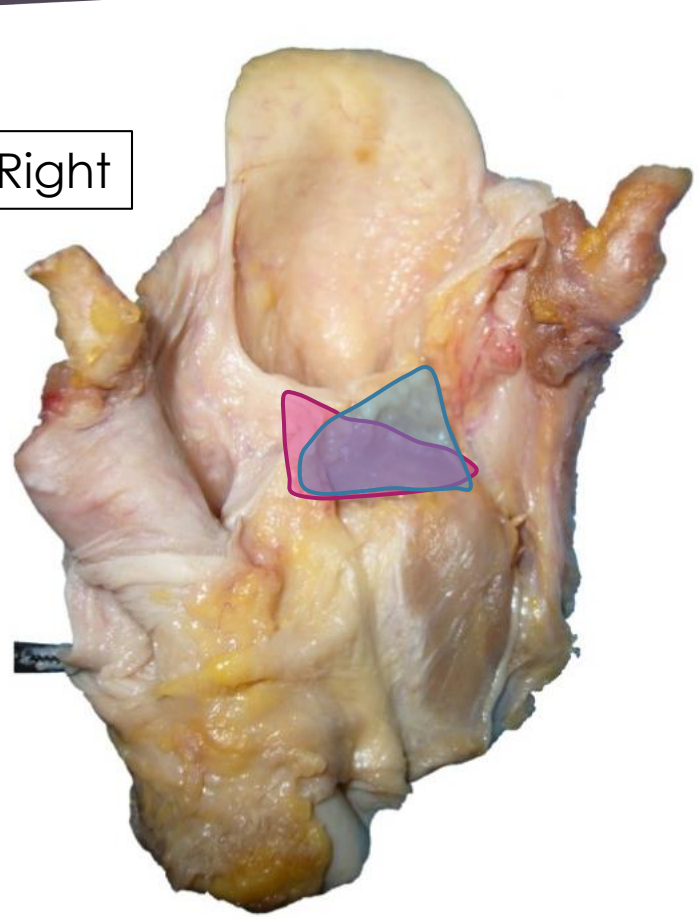
- Front view
- Crico-thyroid muscle



Anatomy

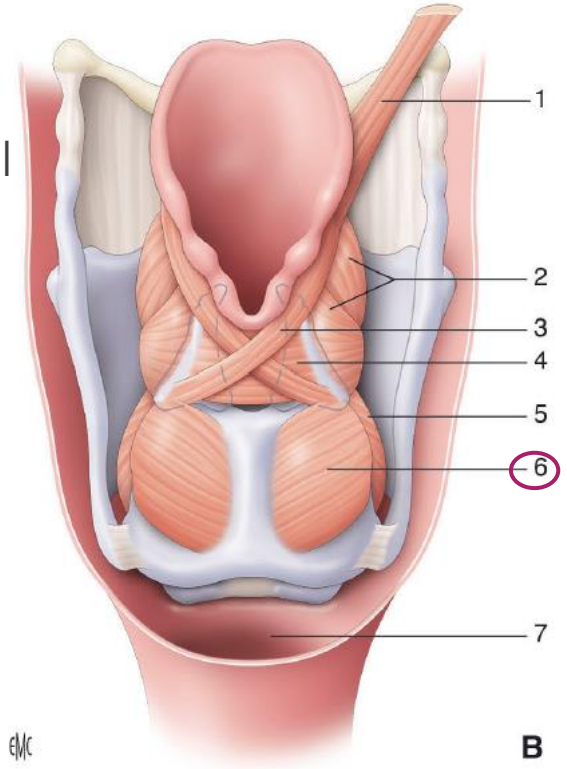
- Posterior view
- Arytenoid muscles

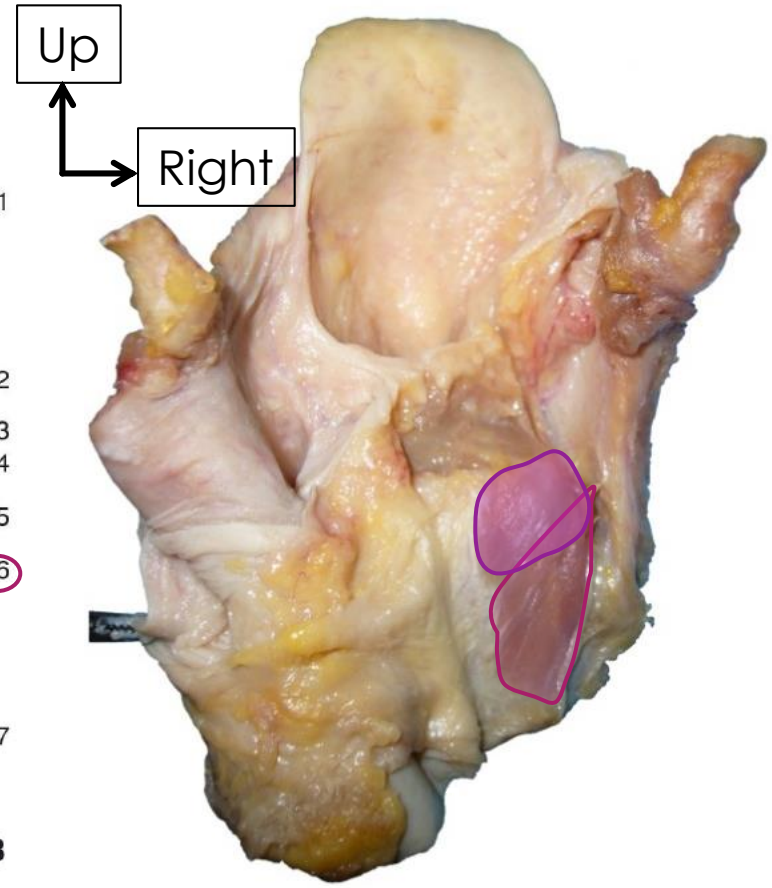




Anatomy

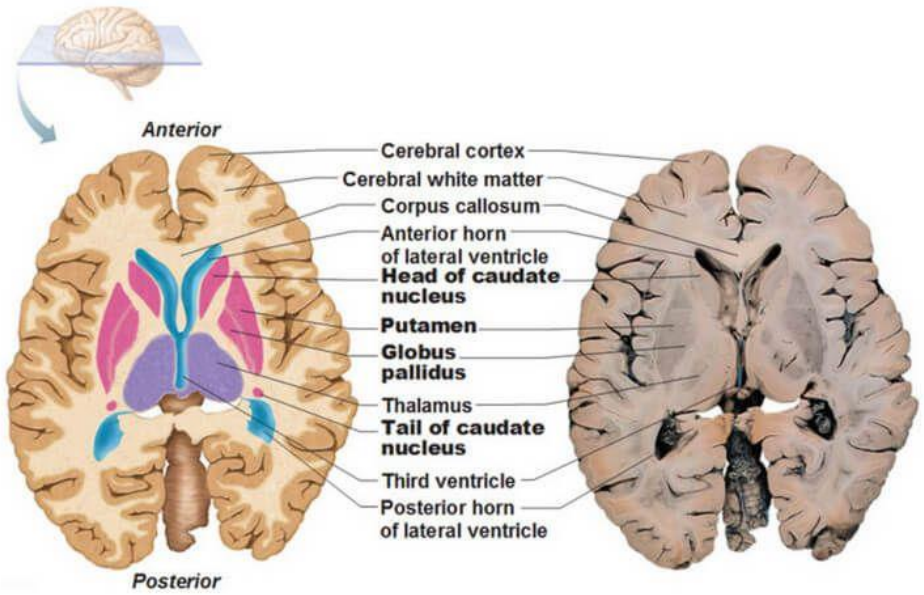
- Rear view
- Posterior crico-arytenoid muscle
- Only abductor muscle of the vocal folds





Etiology

- Origin of dystonia in general
  - Dysfunction of the basal ganglia
    - Idiopathic
    - Linked to genetic/metabolic/etc. pathologies that disrupt this network (e.g. Wilson's disease, DYT 1, vascular lesions, etc.).



Etiology

- The origin of spasmodic dysphonia: controversial
  - Psychogenic origin?
    - More than 50% of patients describe an emotional shock before the onset of the disease.
      - However, this is also true in dystonias of organic origin and other disorders of the basal ganglia, typically revealed by physiological or psychological stress (e.g. DYT12, etc.).
    - Sudden onset, after bereavement or a sudden break in the patient's life
    - Hyperactive, overworked, often authoritarian people in positions of responsibility
    - Could reflect internal conflict
    - Improvement of symptoms by alcohol, sedatives and other psychotropic drugs
      - Also true for organic abnormal movements (DYT 11, essential tremor, certain myoclonic disorders, etc.).
    - Aggravation by stress

Etiology

- Controversial... neurological?
  - No difference in personality test results between patients and the general population
  - Similar observations in true dystonia
    - Spasmodic dysphonia as part of more complex dystonic pathologies (e.g. Wilson's disease, DYT 6, etc.)
  - Motor disorder authenticated on electromyography
  - Association with other focal or segmental dystonias
    - Importance of the neurological examination and collaboration between the neurologist and ENT specialist

Clinical presentation

- Spasmodic adduction/adductor dysphonia
  - Tight, strangled, choppy voice, which can make it impossible to emit any sound voluntarily
  - Sudden start and end of voicing
  - Weak, monotone voice
  - Frequent vocal tremor, perceived with a slowed speech rate
  - Reduced intelligibility
  - Vocal projection sometimes difficult
  - Possible improvement in laughter, whispering or non-communicative voice
  - Severe forms: permanent dyspnoea

Clinical presentation

- Spasmodic adduction/adductor dysphonia

Clinical presentation

- Voice strain
  - Uncomfortable for the patient and for the contact person
- Unattractive voice
- Withdrawal of oral communication
  - The telephone is particularly difficult
- Sometimes improved in singing and loud voices

Clinical presentation

- Spasmodic abduction/abductor dysphonia
  - Whispered voice, barely audible, out of breath
  - Forced voice, with sudden stops, leading to long pauses

Clinical presentation

- Mixed spasmodic dysphonia
  - Adductor and abductor damage

Examination of the larynx

- Diagnosis by clinician's ear
- Laryngoscopy
  - may be normal at rest
  - Phonation
    - Jerky movements of the vocal folds (adducted dysphonia)
    - Abduction dysphonia (abduction dysphonia)
  - "Dance of the arytenoids »
    - During phonation
    - Sometimes at rest

Examination of the larynx

Examination of the larynx: adduction dysphonia

Electromyography

- Principle:
  - Locating muscles with a needle
    - Thyro-arytenoid for adducted dysphonia
    - Posterior crico-arytenoid for dysphonia in abduction
  - Collecting the muscle's electrical signal
  - Observation of electrical hyperactivity
    - Either continuously or in puffs,
    - At rest or during phonation
  - Help with therapeutic procedures: guiding injections

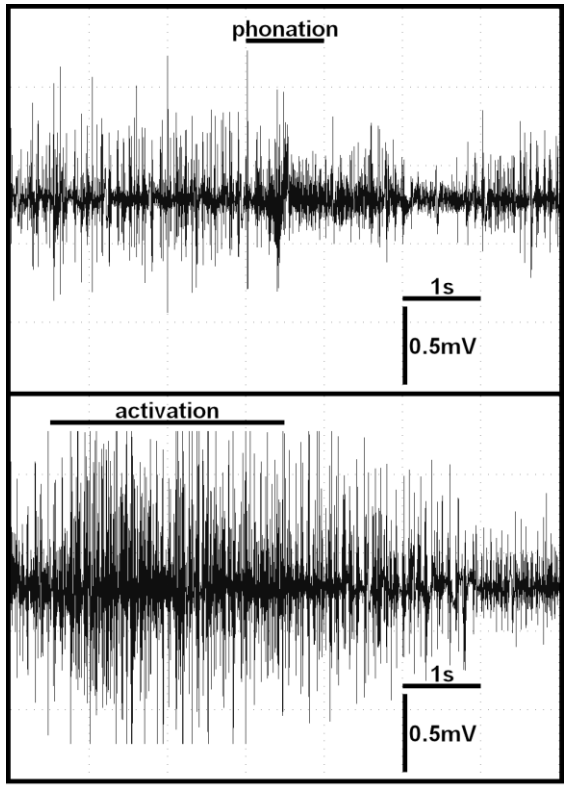


Fig. 1. Needle EMG of the LCM at rest, phonation, and activation

J. Michelis, F. Schuler, I. Debove, J. Müllner, L. Lachenmayer, E. Seifert. Combined EMG and endoscopy guided botulinum toxin injection of the longus capitis muscle in cervical dystonia with anterocaput [abstract]. *Mov Disord.* 2018; 33 (suppl 2).

Electromyography

Treatment

- Psychiatric care
  - In cases of confirmed psychogenic origin
  - Often disappointing results
  - Emphasise the potential of full recovery given absence of organic damage (Stone et al. 2016)
  - Explain the nature of the problem to the patient and discuss the possibility of a functional origin
- Speech therapy
  - Relaxation methods

Treatment

- Immobilisation of one or both vocal cords
  - Temporary effectiveness
    - Injection of botulinum toxin the muscle responsible
      - Current reference technique
      - Less invasive
      - Injection under electromyography control
      - Effective in 2 to 3 days
      - Dysphonia (hypophonia), transient aspiration
      - Average duration of efficacy: 2 to 6 months
      - Iterative injections

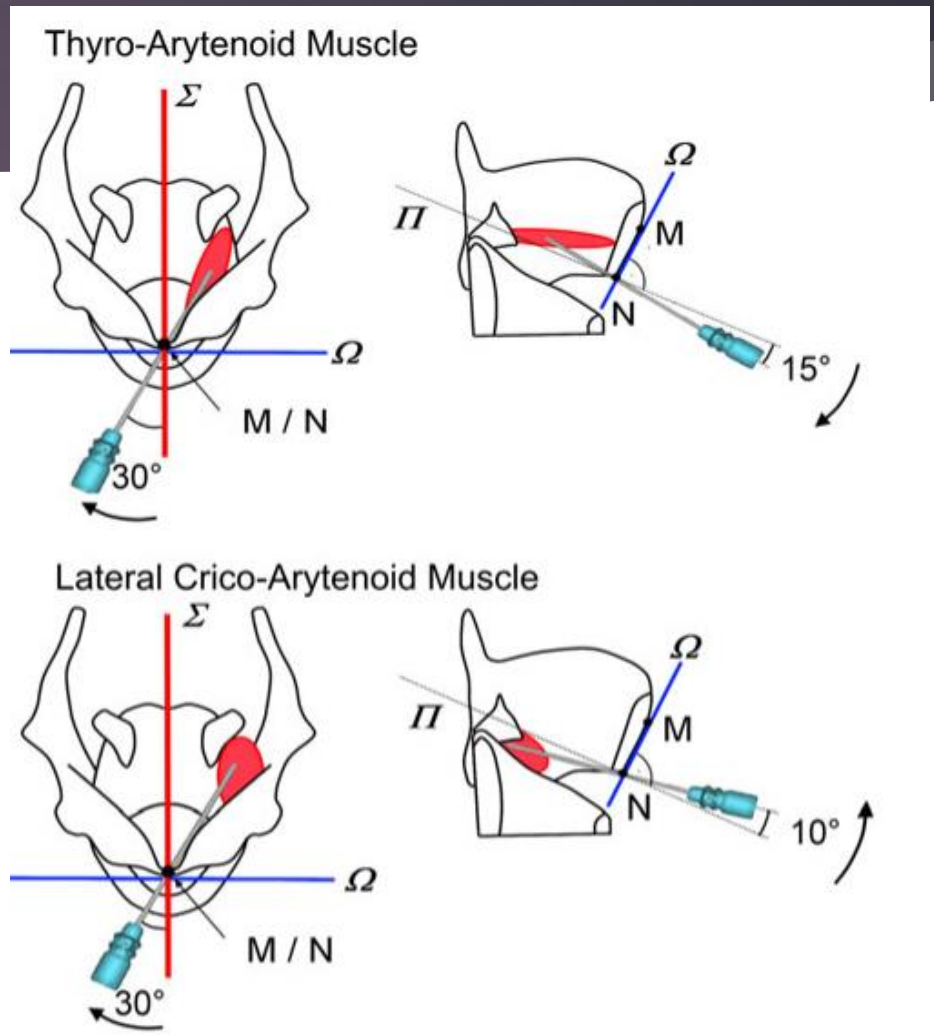
Treatment

- Immobilisation of one or both vocal cords
  - Temporary effectiveness
    - Sectioning/crushing of one laryngeal recurrent nerve
    - Endoscopic coagulation of the terminal branches of the recurrent nerve
  - Long-term effectiveness
    - Selective laryngeal denervation/reinnervation of the adductors

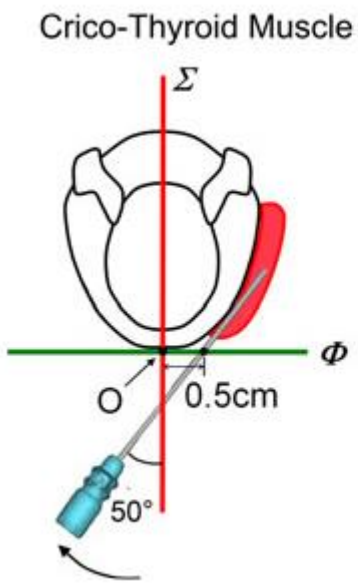
Injection of toxin: injection technique

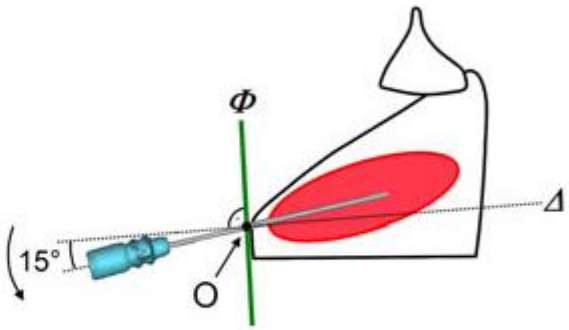
- Adductors
  - Uni- or bi-lateral
    - Greater efficiency in bilateral relations
    - More side effects
  - Doses:
    - 1-3U if bilateral,
    - 5-10 U if unilateral

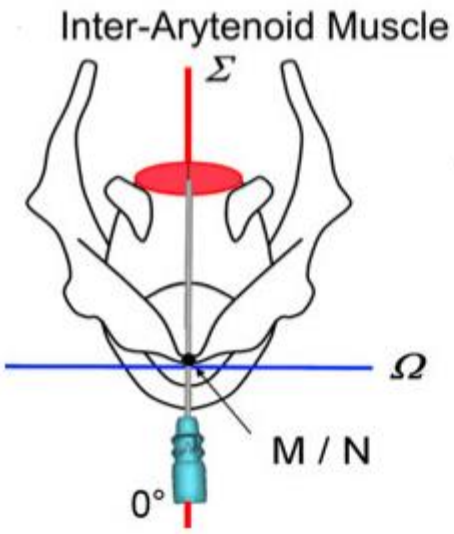
Injection technique

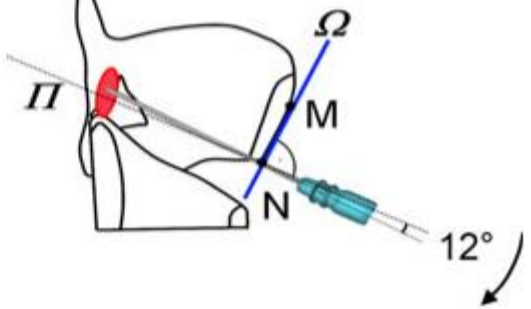


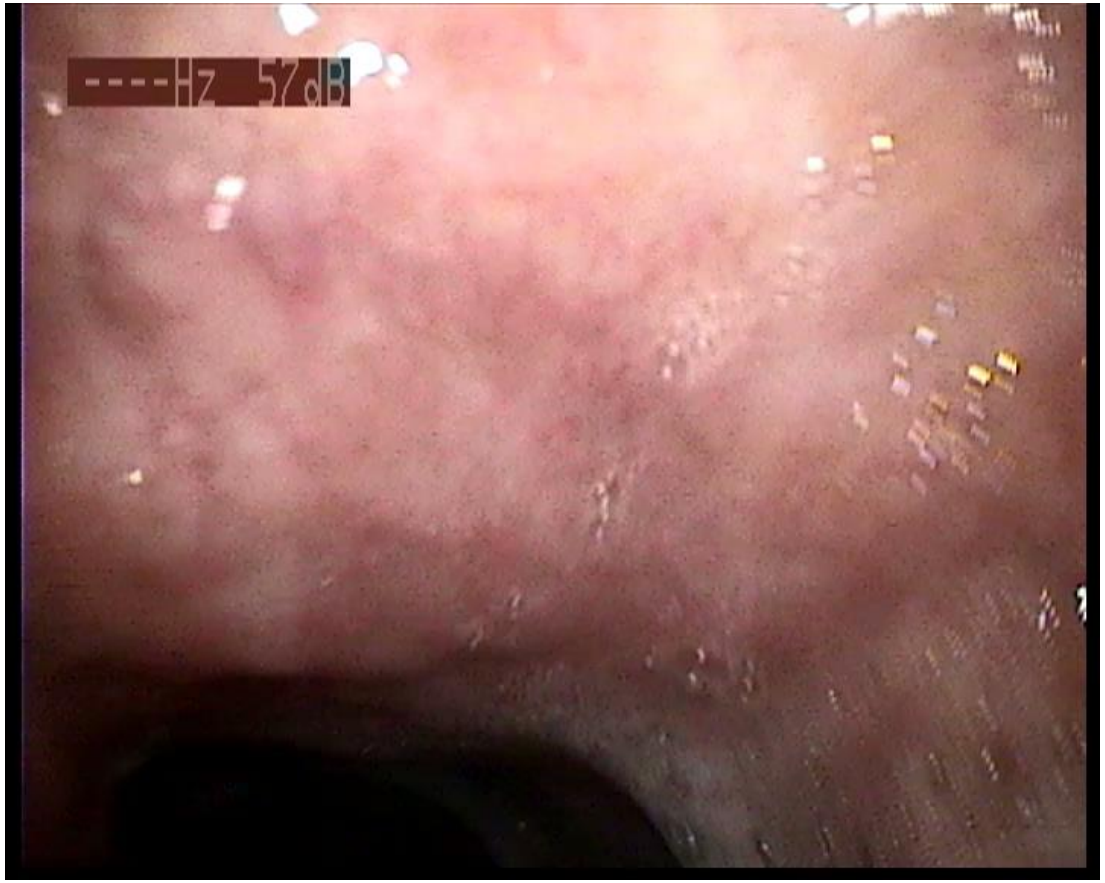
Injection technique







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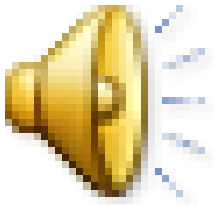
- Before injection

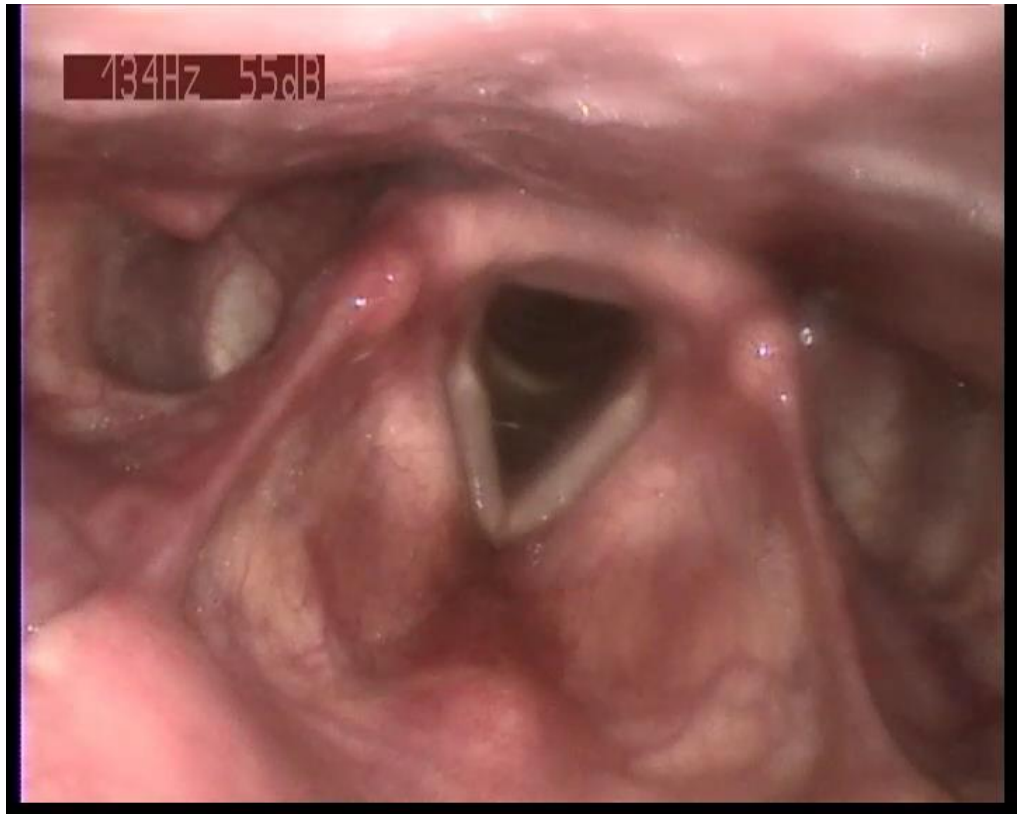


- After injection

Side effects

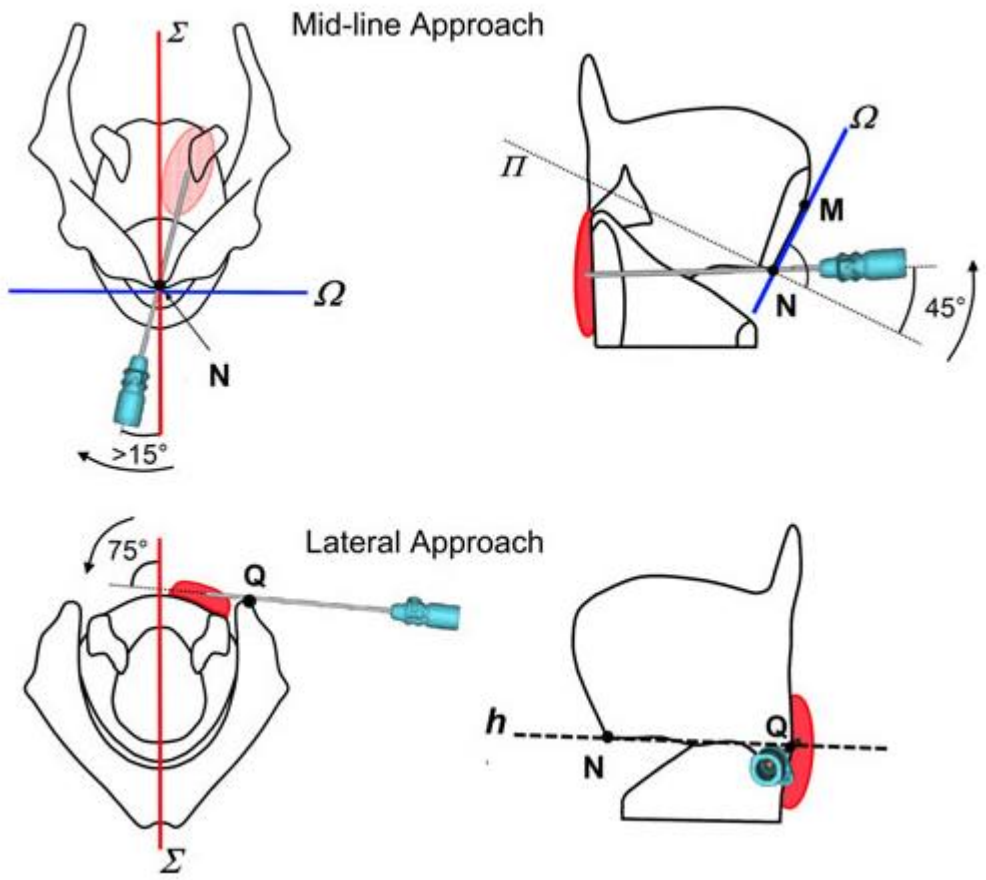
- Our experience based on 19 patients, within a unilateral TA injection cycle:
  - Transient aphonia/hypophonia: 15 to 20 days in 4 patients
  - Dysphagia with liquids : 5 patients
    - Up to 2 months for 1 patient
  - Dysphagia with solids: 2 patients
    - Up to 15 days

Examination of the larynx: dysphonia in abduction



Injection of toxin: injection technique

- Posterior crico-arytenoid muscle
- Abductor:
  - Always one-sided
  - 5U
- Side effects
  - Dyspnoea
  - dysphagia



Respiratory laryngeal dystonia

- Borderline "respiratory" forms
  - Paradoxical vocal cord movement or vocal cord dysfunction syndrome
    - Brief episodes of vocal cord adduction
      - In asthmatic patients
      - At sports
      - During exercise or at rest
      - Rather young, performance anxiety

Respiratory laryngeal dystonia

- Differential diagnosis:
  - Bilateral laryngeal paralysis (laryngeal diplegia)
    - Permanent dyspnoea, increased during sleep
    - Room neighbour test
- Treatment
  - Rehabilitation, relaxation, psychotherapy
  - Indication of botulinum toxin in case of failure

Respiratory laryngeal dystonia

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Oro-mandibular dystonia

Definition

- A form of dystonia involving
  - The lower part of the face, particularly the lips (85%),
  - The masticatory muscles (61%),
  - Language (17%)
  - The pharynx (?)

  - Concept of the "stomatognathic" system

- Variable terminologies
  - Oro-mandibular dystonia
  - Oro-bucco-lingual dystonia (or even "OBL dyskinesias" in the broad sense)
  - Cranial dystonia

Epidemiology

- 2 Women/1 Man
- Onset between the ages of 50 and 60
- 62% open, 20% closed, 18% mixed
- Associated lingual dystonia in 27%,
  - More often with open or mixed dystonias
- Rare, but probably underestimated:
  - Prevalence: 0.1-6.9/100,000
  - Problem of the medical referent (often a dentist or maxillo-facial surgeon)
  - Could in fact be as common as cervical dystonia or blepharospasm

Screening questionnaire

Table 1 Ten-item questionnaire for oromandibular dystonia. The self-administered questionnaire includes questions concerning the clinical features of oromandibular dystonia, such as stereotypy, task-specificity, sensory tricks, and morning benefit

Have you experienced the following problems? Please mark one answer per question.	Not at all	A little	Moderately	Quite a bit	Extremely
1. Do you experience involuntary contractions or movements in the mouth, jaw, tongue, or lips?	□	□	□	□	□
2. Do you have muscle contractions or movements that you are unable to control?	□	□	□	□	□
3. Is the region of the muscle contractions or movements (jaw, cheek, temple, tongue, or lips) always the same?	□	□	□	□	□
4. Is the direction of the muscle contractions or movements (mouth closing, opening, or tongue protrusion) always the same?	□	□	□	□	□
5. Do your symptoms appear only during a specific task (speaking, eating, opening mouth, etc.)? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
6. When something is in your mouth (chewing gum, candy, or a mouth piece), or you touch your mouth or chin with hands or fingers, do the symptoms become milder? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
7. Are you able to sleep without symptoms?	□	□	□	□	□
8. Are your symptoms less severe or absent in the morning and gradually worsen during the day? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
9. Is the severity of your symptoms affected by tension or relaxation? Or, was it so in the early phase when you noticed the symptoms?	□	□	□	□	□
10. Is it difficult for you to control the symptoms when you are nervous or under stress?	□	□	□	□	□

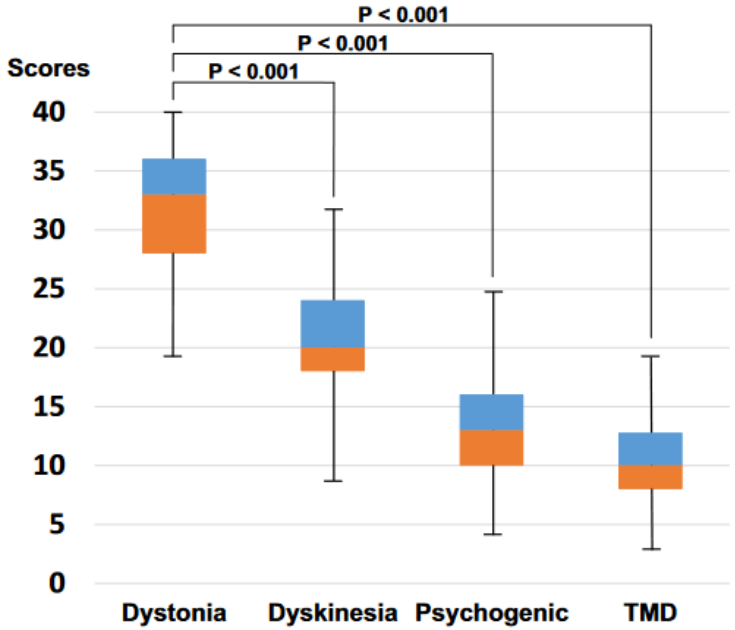


Fig. 1 Comparison of the results of total scores of the questionnaire among four groups. Total scores of the questionnaire significantly differed among oromandibular dystonia, temporomandibular disorders ($p < 0.001$), oral dyskinesia ($p < 0.05$), and psychogenic movement disorder ($p < 0.01$). Horizontal lines: median values, boxes 25th to 75th percentile; TMD temporomandibular disorders

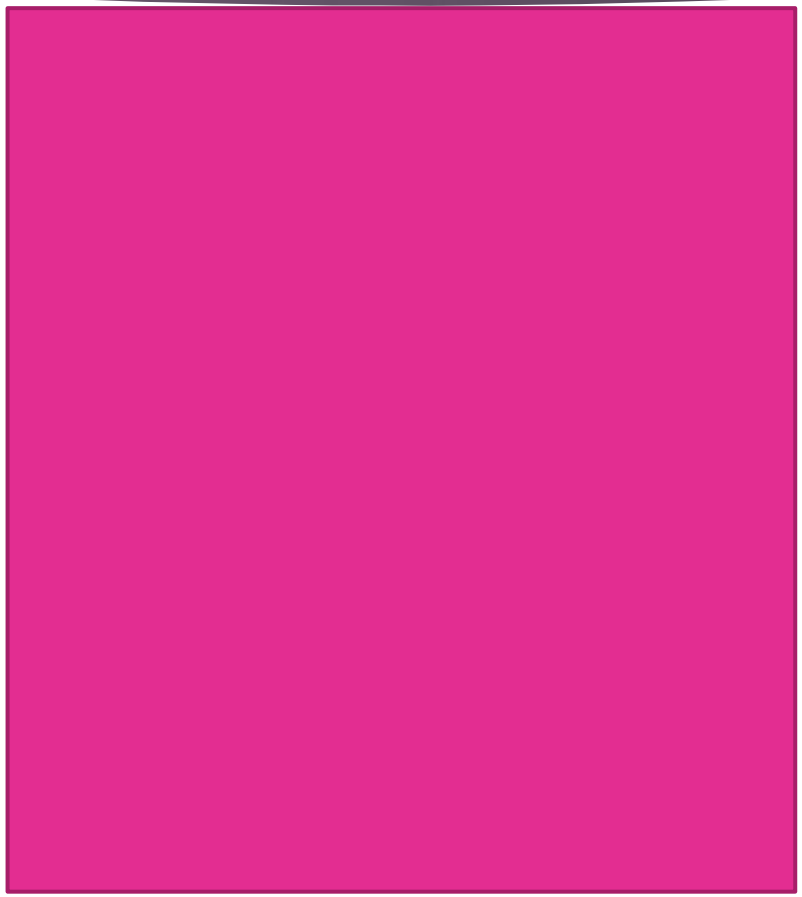
Nosological entities

- DOM as part of segmental dystonia (43%)
  - Classic case= Meige's Sd (or "craniofacial" dystonia): Blepharospasm + DOM
  - Other frequent associations: cervical dystonia, writer's cramp, spasmodic dysphonia

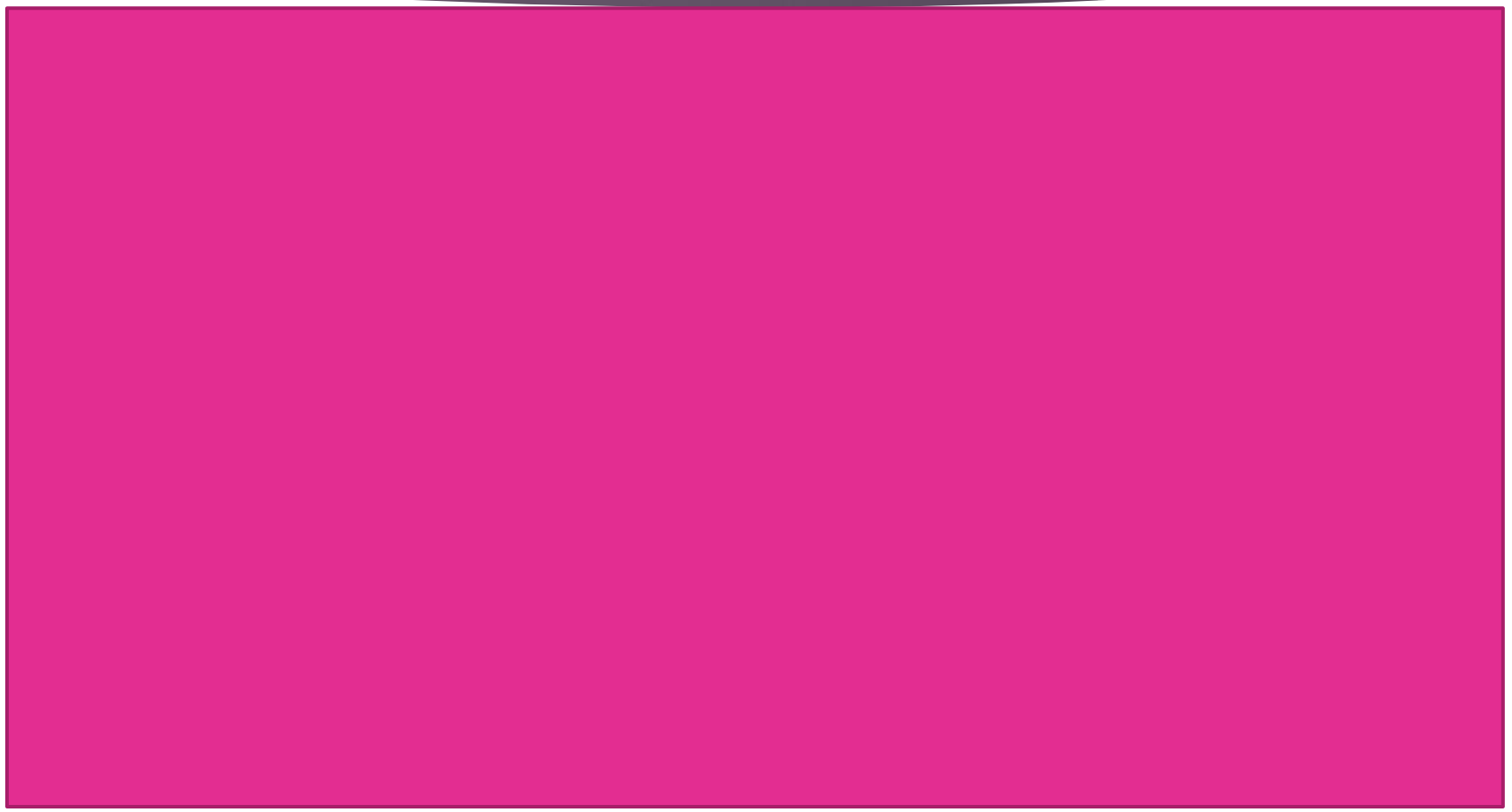
- DOM in the form of isolated focal dystonia (39%)
  - Iatrogenic
    - Late onset (neuroleptics)
    - Isolated oral (labial) dyskinesia with tonic component: "rabbit syndrome" associated with certain SSRIs and/or neuroleptics
  - Idiopathic (rare)
  - Sometimes triggered by local trauma

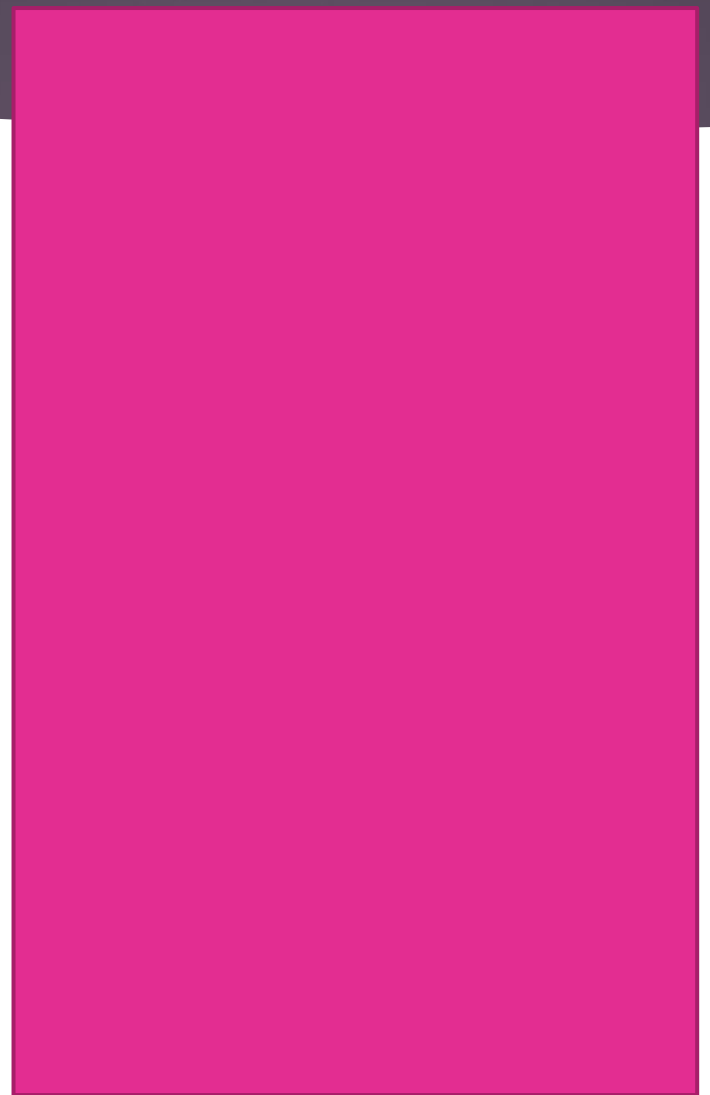
- DOM as part of generalised dystonia (10%)
  - But which can be the only/principal mode of expression
    - Pure" genetic dystonia - DYT 6 , DYT 25, DYT 24/ANO 3
    - "Dystonia +": Wilson's disease, Neuroacanthocytosis (to be eliminated even if rare pathology, particularly if associated with chorea, epilepsy, amyotrophy, dementia), post-anoxic, PKAN, Parkinson's disease, Lesch-Nyhan syndrome.

Late neuroleptic syndrome



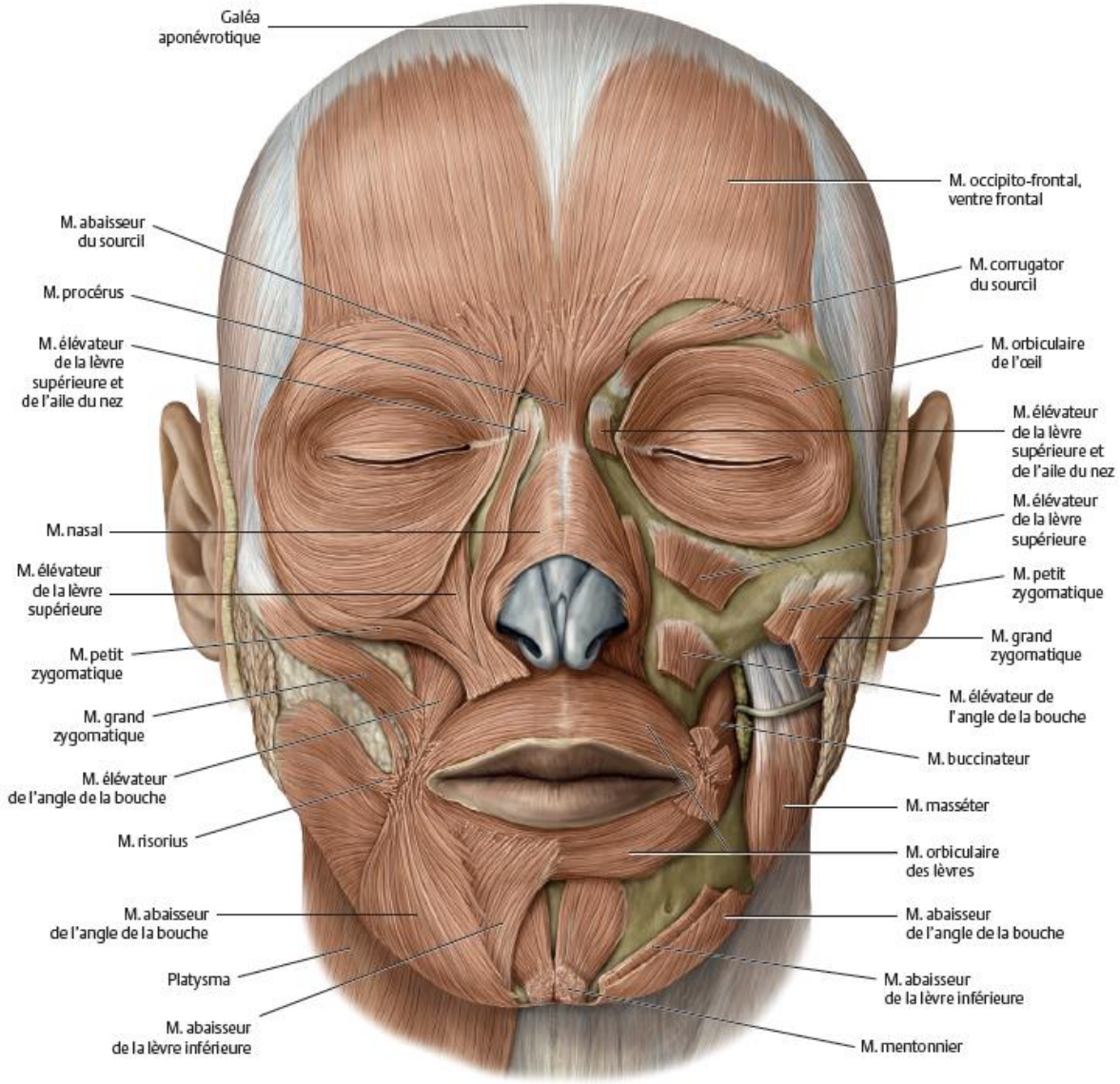
Meige syndrome





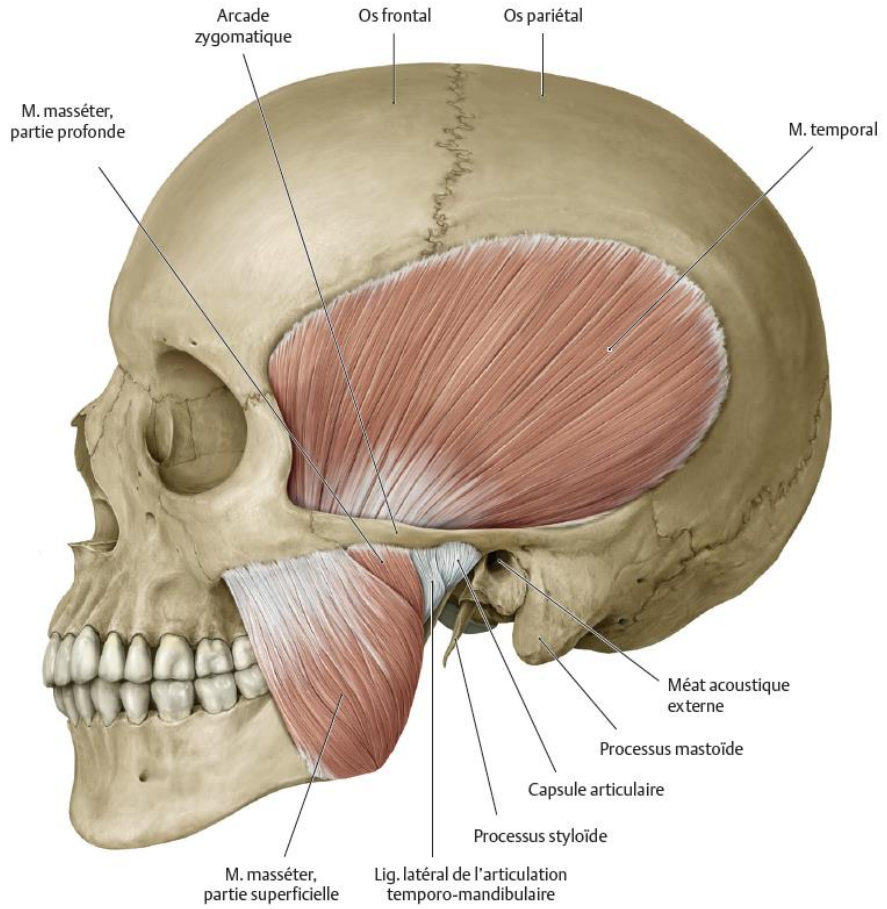
Anatomy

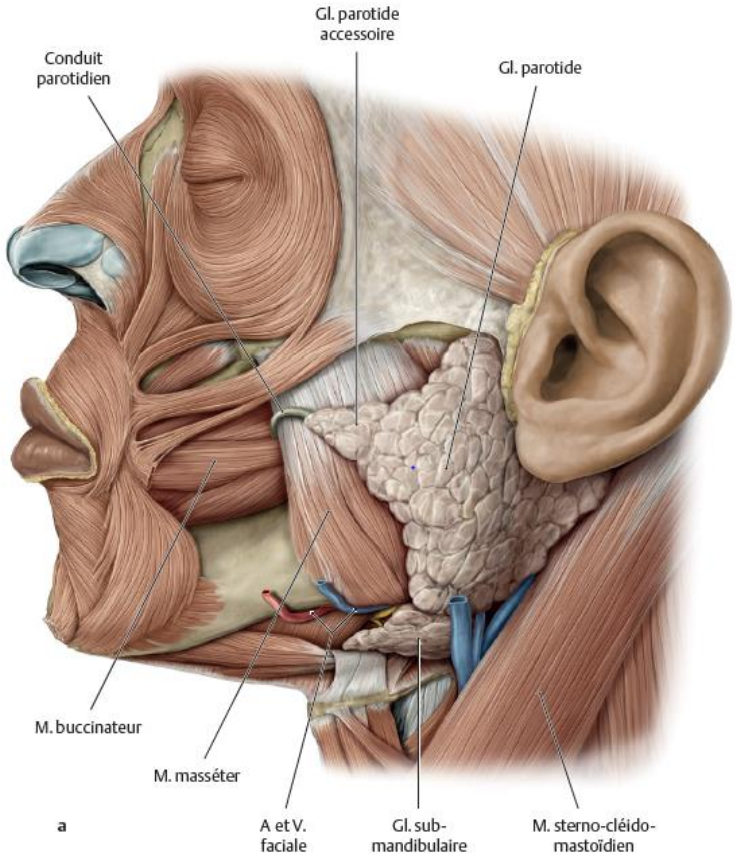
- Labial dystonia



Muscles de la mimique, vue antérieure.
Partie droite de la face, couche superficielle, partie gauche de la face, couche profonde.

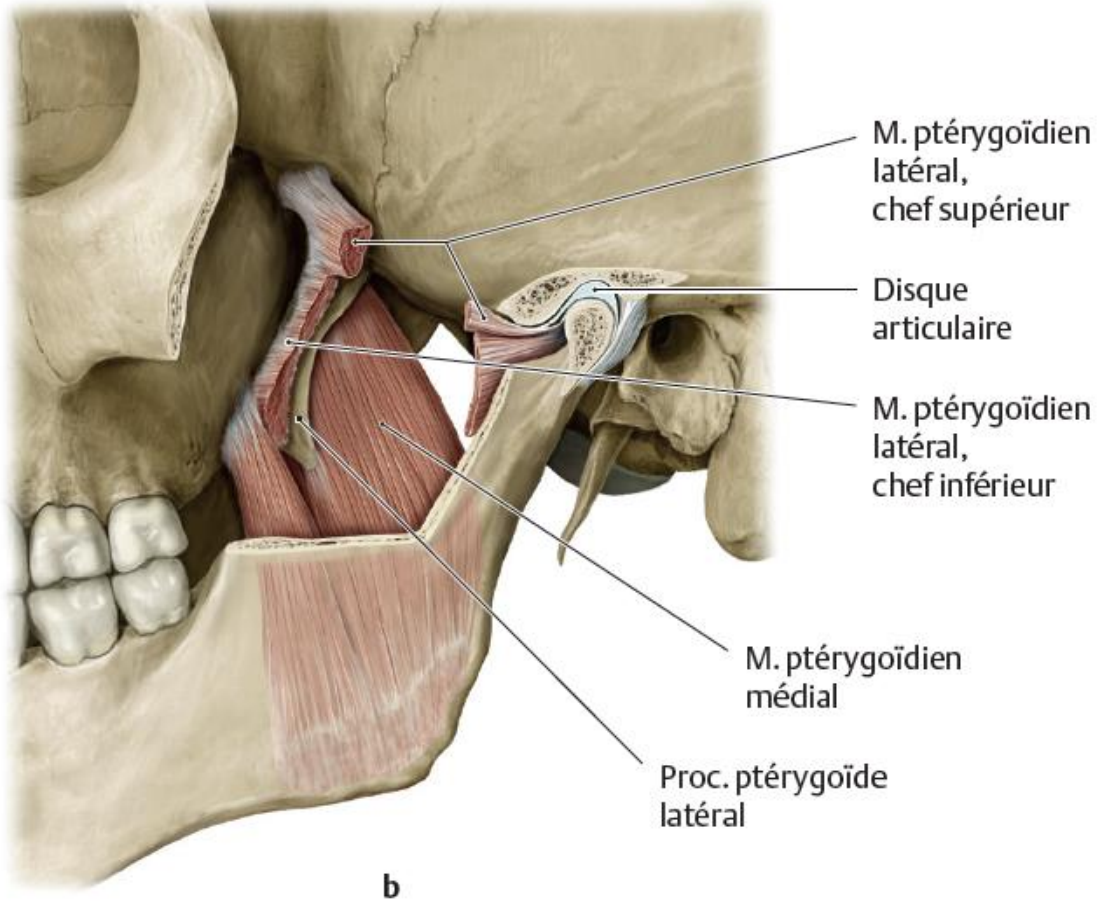
Anatomy





Muscles de la mimique, vue latérale

Anatomy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Disque articulaire
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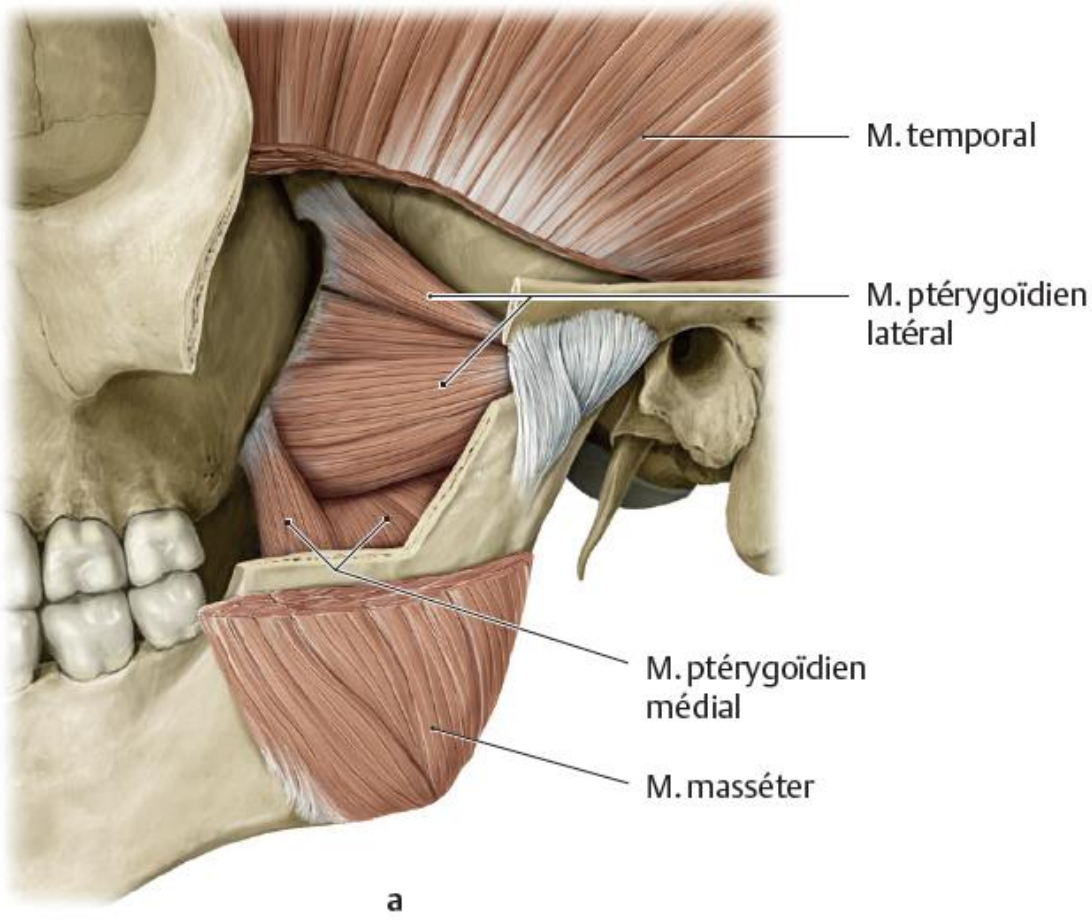
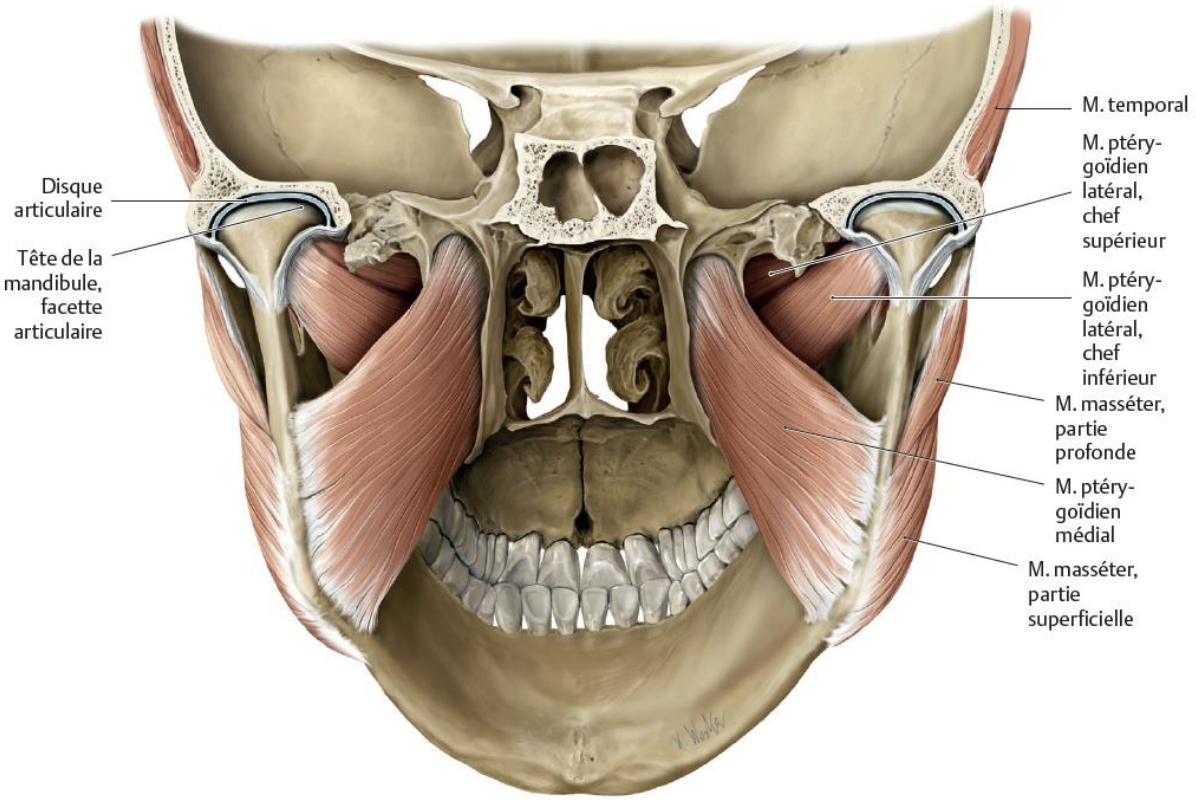
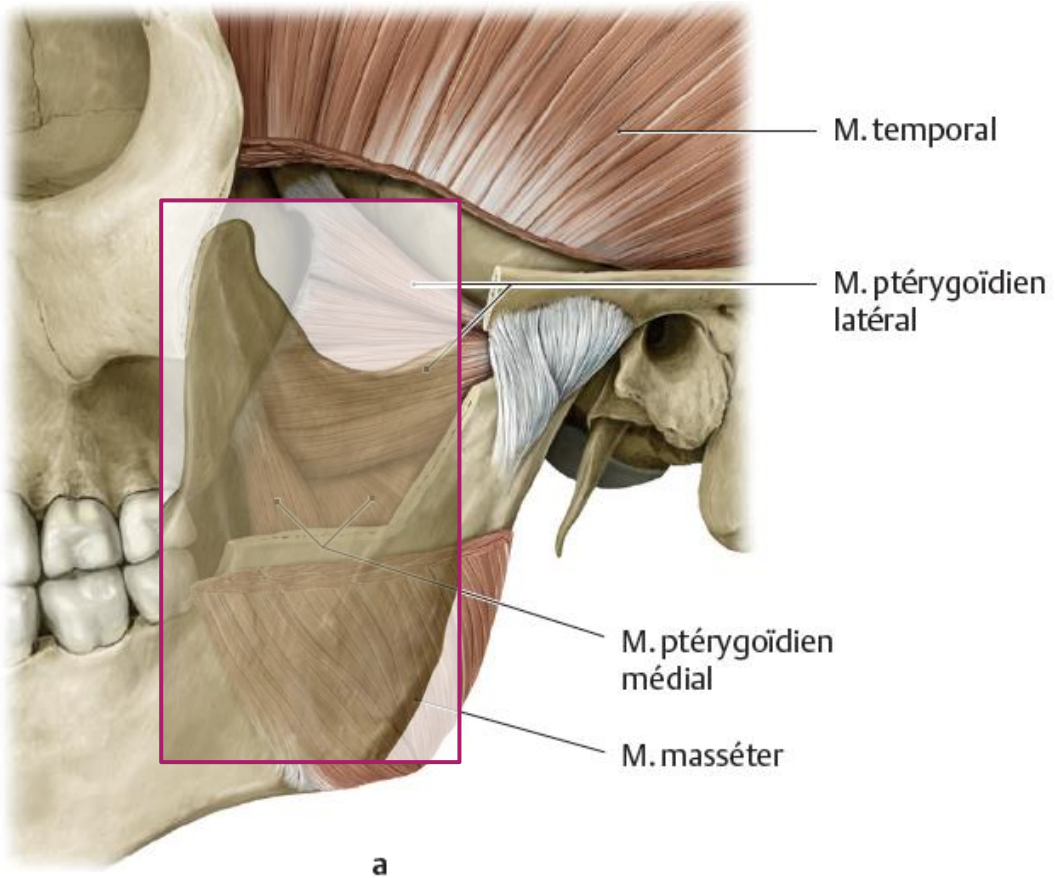
M. ptérygoïdien médial
Proc. ptérygoïde latéral
b


M. temporal
M. ptérygoïdien latéral
M. ptérygoïdien médial
M. masséter
a

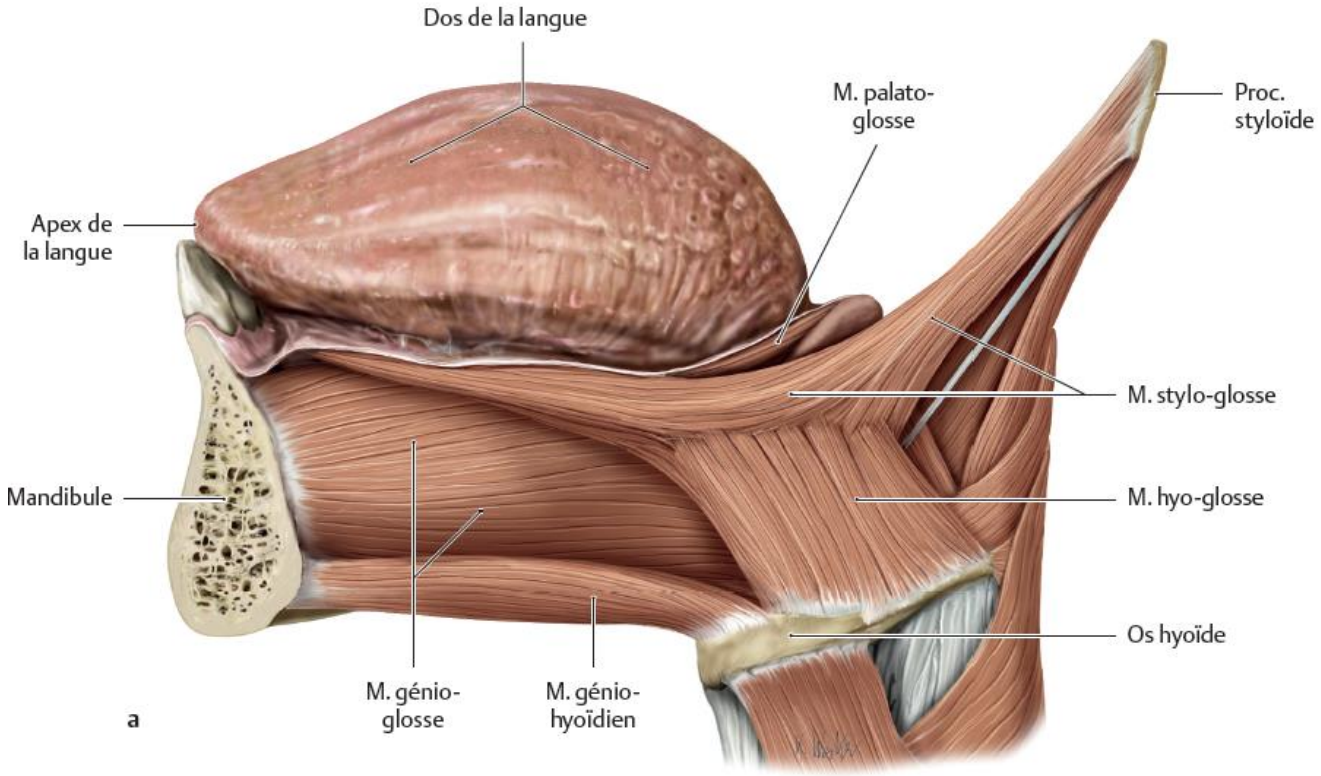
Anatomy

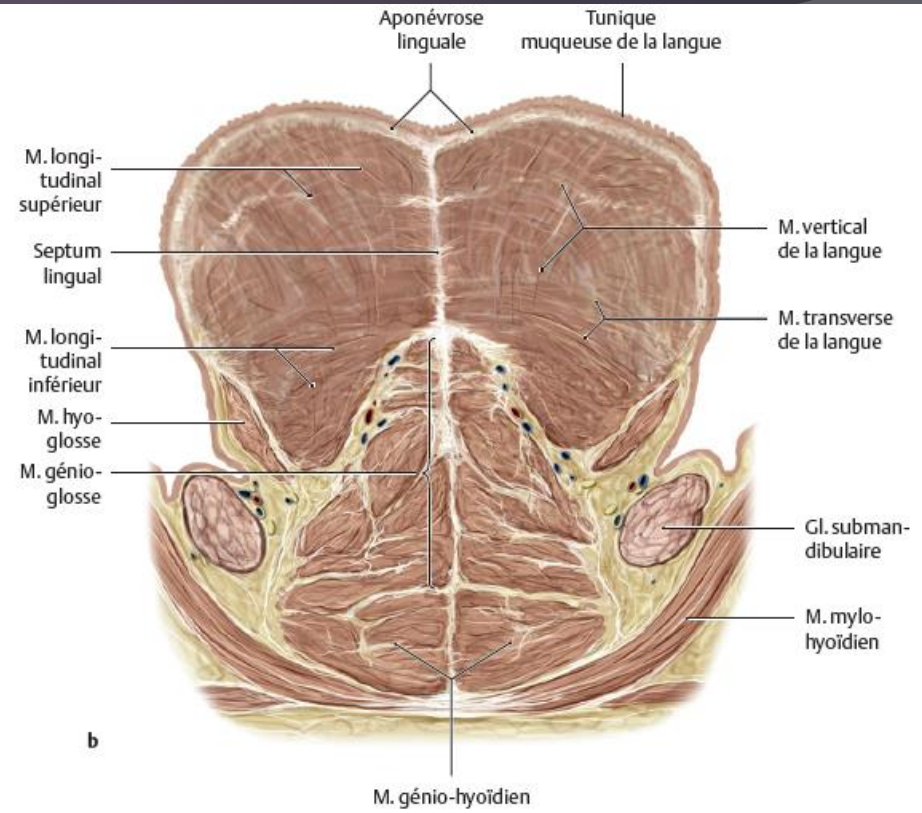

Disque articulaire
Tête de la mandibule, facette articulaire
M. temporal
M. ptéry-goïdien latéral, chef supérieur
M. ptéry-goïdien latéral, chef inférieur
M. masséter, partie profonde
M. ptéry-goïdien médial
M. masséter, partie superficielle

Anatomy


M. temporal
M. ptérygoïdien latéral
M. ptérygoïdien médial
M. masséter
a

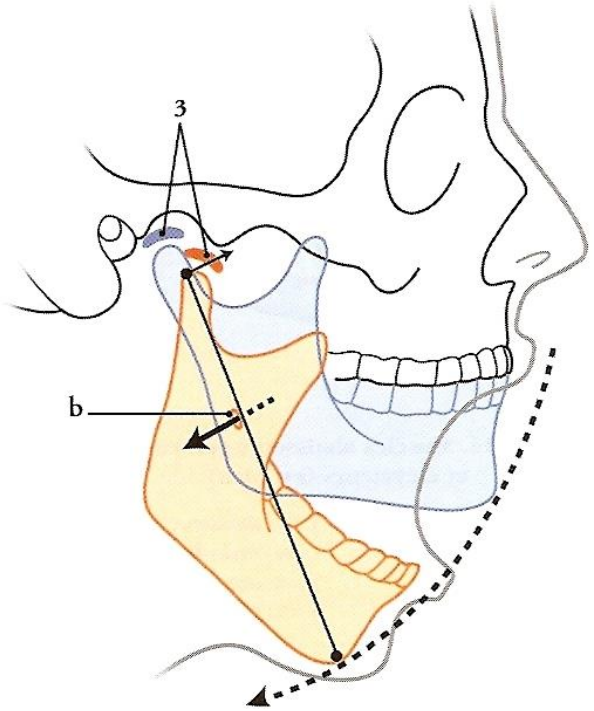
Anatomy





Muscles de la langue.
Vue gauche (a), vue antérieure d'une coupe frontale (b)

Functional anatomy-Masticators



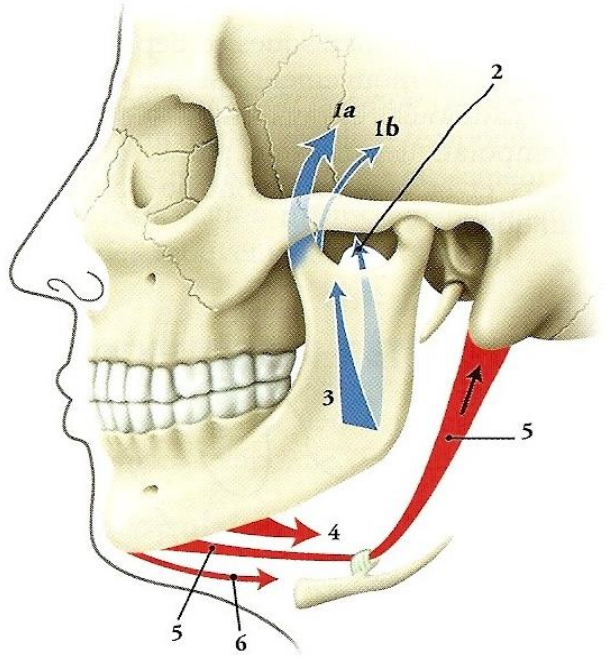


FIG. 6.14. Muscles abaisseurs (en rouge) et élévateurs (en bleu)

1. m. temporal (fibres antérieures a, fibres postérieures b)
2. m. ptérygoïdien médial
3. m. masséter
4. m. mylo-hyoïdien
5. m. digastrique
6. m. génio-hyoïdien

Functional anatomy-Masticators

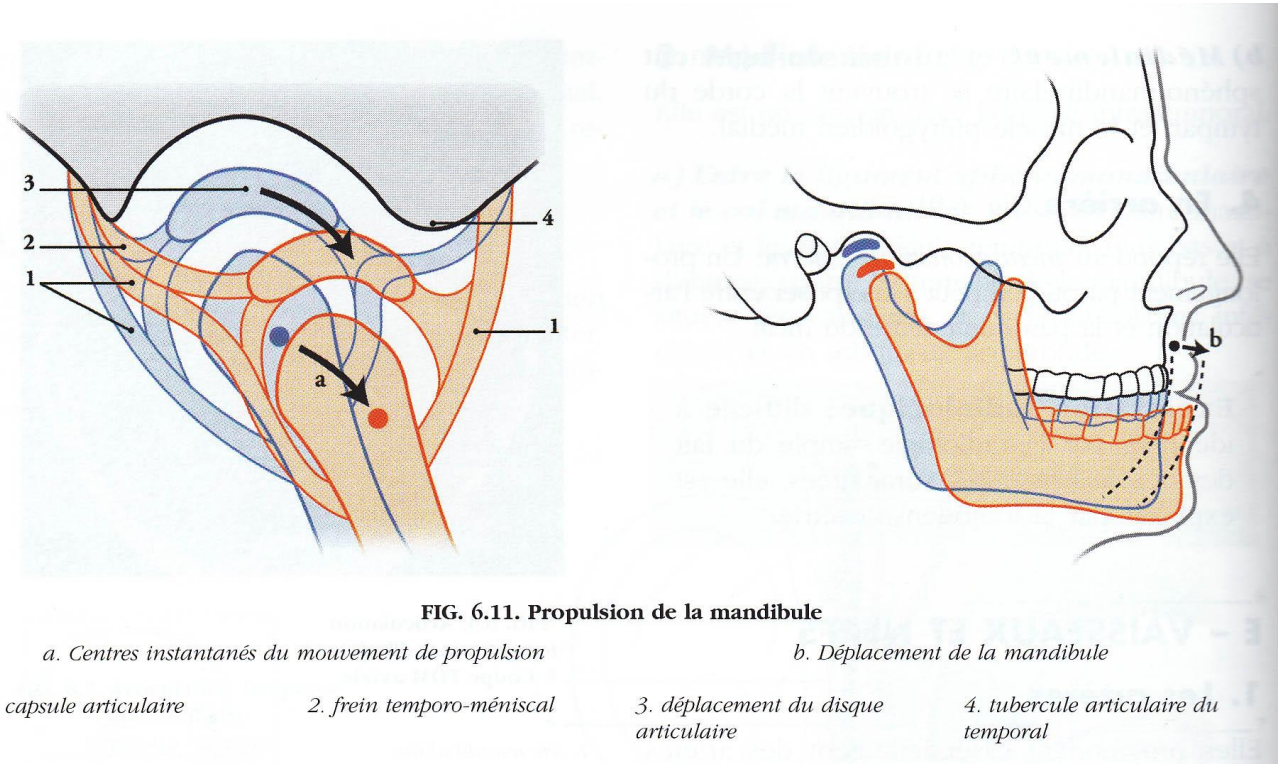


FIG. 6.11. Propulsion de la mandibule

a. Centres instantanés du mouvement de propulsion

b. Déplacement de la mandibule

1. capsule articulaire

2. frein temporo-méniscal

3. déplacement du disque articulaire

4. tubercule articulaire du temporal

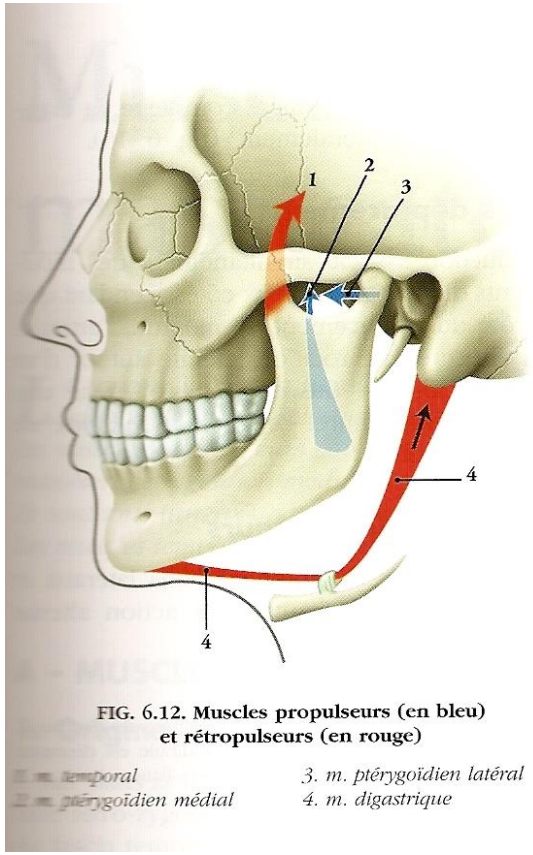


FIG. 6.12. Muscles propulseurs (en bleu) et rétropulseurs (en rouge)

1. m. temporal

2. m. ptérygoïdien médial

3. m. ptérygoïdien latéral

4. m. digastrique

Functional anatomy-Masticators

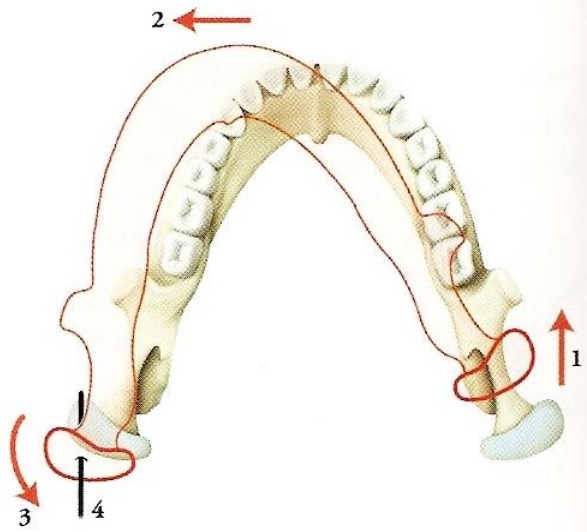


FIG. 6.15. Diduction

1. translation antérieure
2. déplacement latéral du menton
3. rotation
4. axe de la rotation

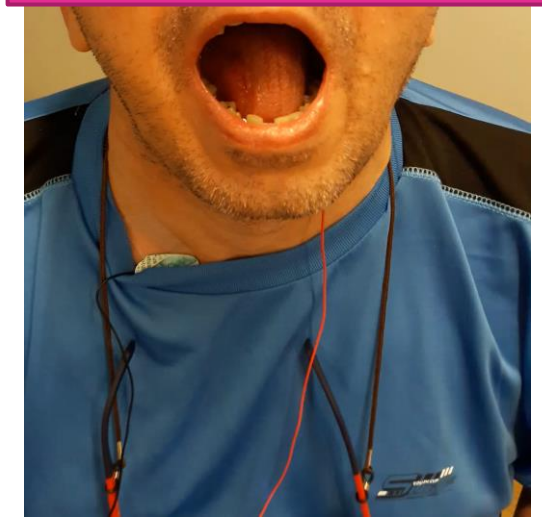
Phenomenology

- Tonic contractions or repetitive, sequential movements:
  - Trismus, bruxism
  - Involuntary opening or closing of the jaw,
  - Mandibular diduction
  - Involuntary tongue movements
  - Combinations of these movements
- Consequences
  - Biting of the tongue and/or cheeks
  - Dental problems (17%)
  - Articulation difficulties (64%)
  - Pain: headaches, SADAM (32%)
  - Chewing difficulties (49%)
  - Dysphagia (27%)

FIGURE 2 | Subtypes of oromandibular dystonia. Oromandibular dystonia includes jaw closing dystonia **(A)**, tongue dystonia **(B)**, jaw opening dystonia **(C)**, jaw deviation dystonia **(D)**, jaw protrusion dystonia **(E)**, and lip dystonia **(F)**.

Opening mouth dystonia

Lingual dystonia



Assessment of oromandibular dystonia

- Oromandibular dystonia rating scale

TABLE 1 | Video examination protocol for oromandibular dystonia.

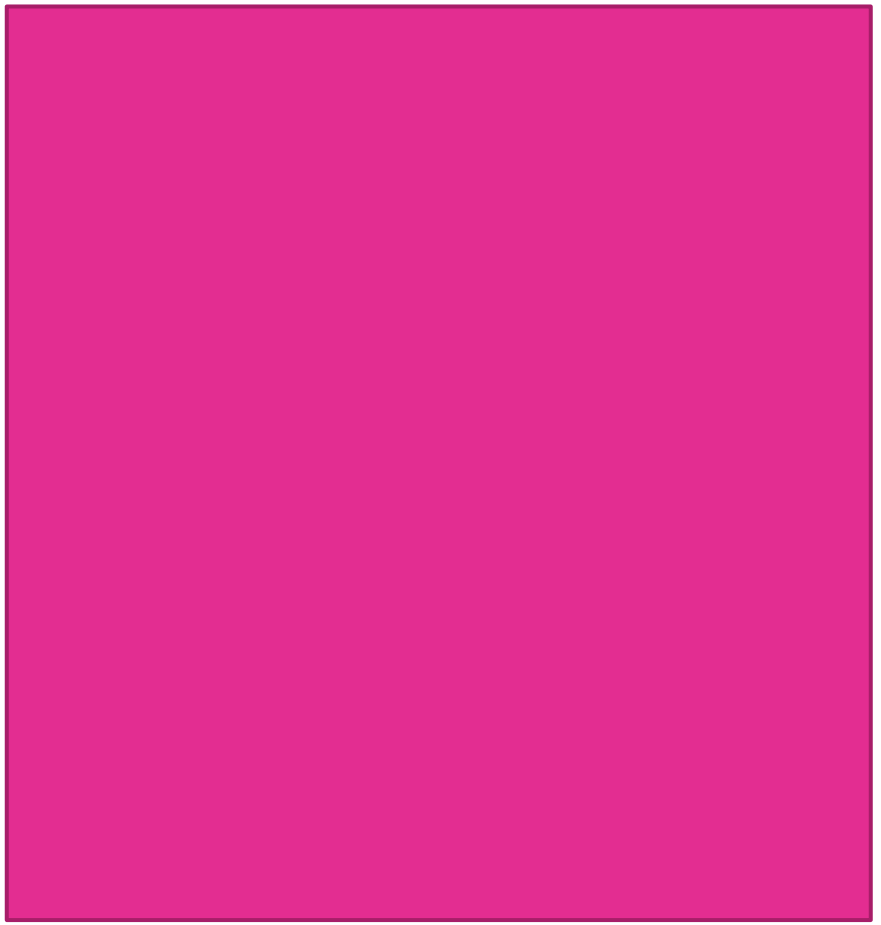
1.	At rest (10 s)
2.	Count from 1 to 10 aloud
3.	Open/close mouth (5 times)
4.	Lateral movements (5 times)
5.	Jaw protrusion (5 times)
6.	Tongue protrusion (hold for 5 s)
7.	Hold long vowel: "Ahh.." for 5–10 s
8.	Read: sentences
9.	Gum chewing (30 s)

Yoshida K (2020) Development and Validation of a Disease-Specific Oromandibular Dystonia Rating Scale (OMDRS). Front. Neurol. 11:583177. doi: 10.3389/fneur.2020.583177

DOM and dysphagia

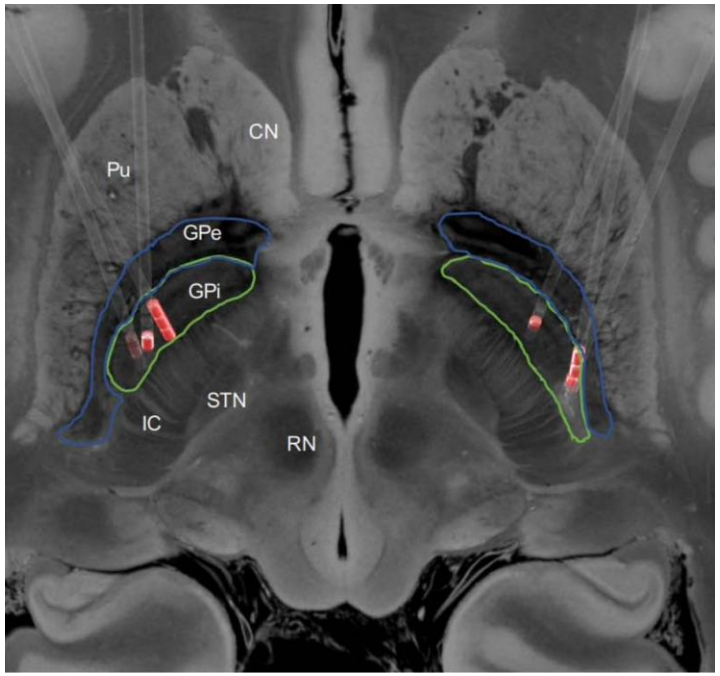
- Dysphagia present in 15.6% of patients
  - Weight loss in half of them, 5-6 kg
- Mechanisms
  - Difficulties getting into the mouth
  - Oral incontinence
  - Food expulsion
  - Poor chewing
  - Lingual propulsion defect
  - Slightly altered pharyngeal phase
  - Traumatic lesions of the mucosa (bites)
- Improvement in dysphagia with treatment
  - Tetrabenazine and/or botulinum toxin

DOM and dysphagia



Treatments

- No high level of evidence because rare and difficult to assess/no severity scale
- Dental rehabilitation, stabilising splints
- Distracting elements / conjuring gestures:
  - matches, sweets, chewing gum
- Tetrabenazine
  - 26% efficiency only
- Clonazepam
  - Would be effective on isolated Meiges>DOM
- DBS
  - GPi stimulation (generally bilateral)
  - Mainly for refractory materials
- (Pallidotomy)
- Coronoidotomy, masseter myotomy:
  - as a second-line treatment for refractory dystonia in closure

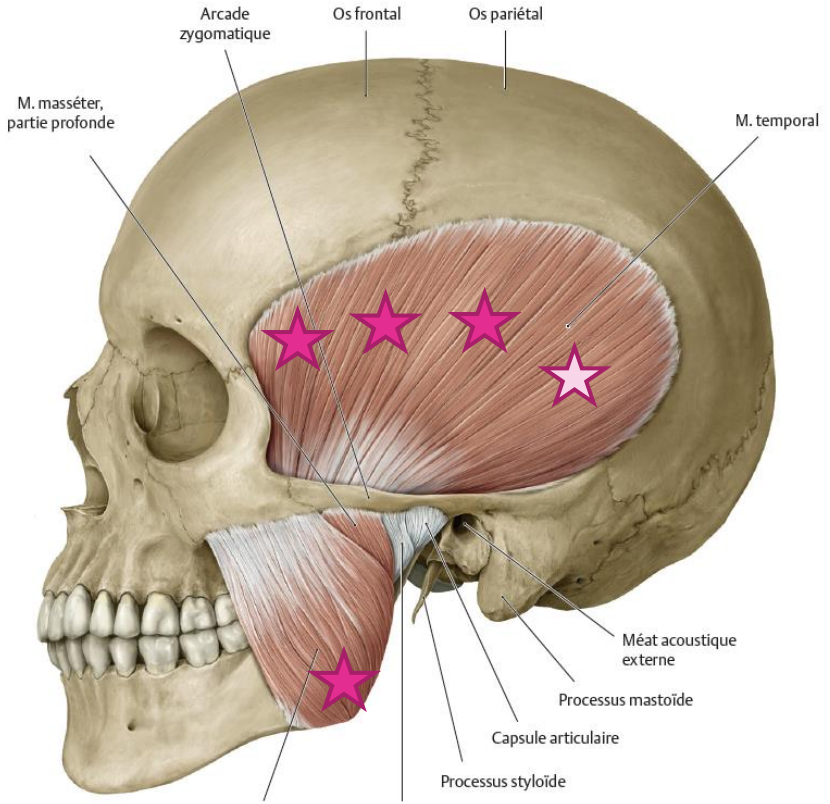


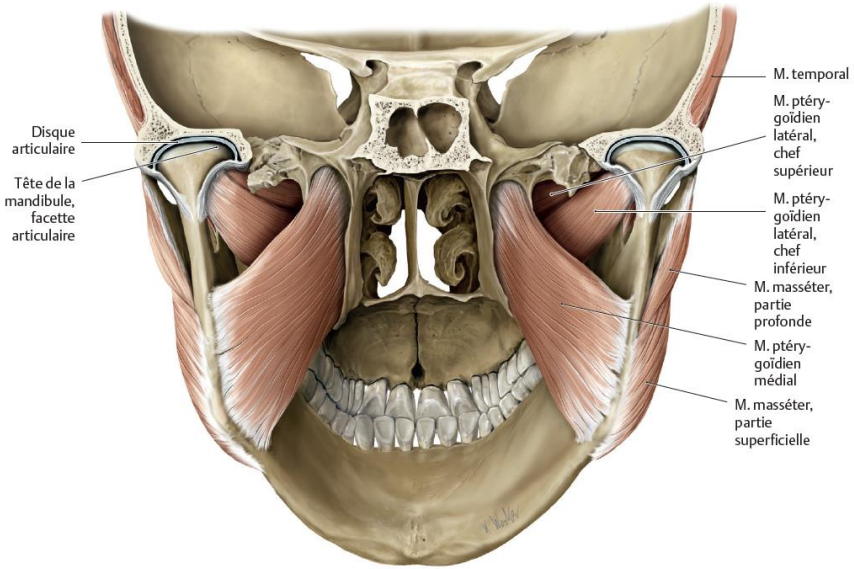
Botulinum toxin

- First line of treatment
  - Lack of randomised controlled trials,
  - But many open trials, on many patients
- Target suitable for semiological analysis
  - Best answers for dystonia in closure
- Lower efficiency than other sites
  - When combined, particularly blepharospasm

Injection techniques

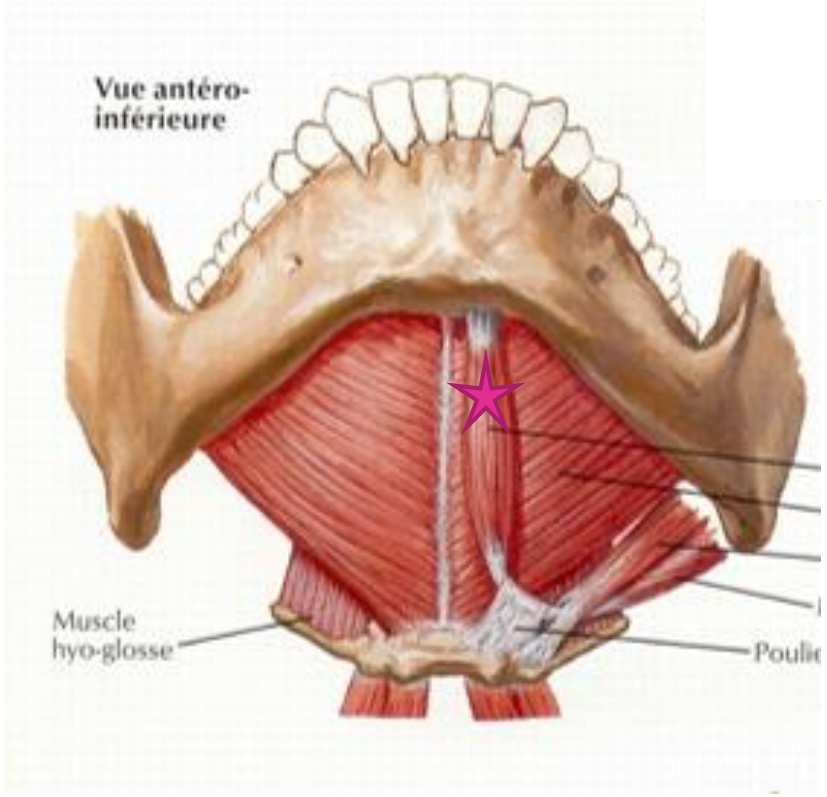
- Dystonia in closure
  - 1-Masseter
    - Not necessarily with EMG
    - 40-50 U to start with
  - 2-Temporal
    - 3-4 points high up in the temporal fossa
    - 20-40 U to start with
  - 3-Medial pterygoid
    - EMG
    - Inferior approach 5-10mm anterior to the mandibular angle, medial to the mandible, pointing upwards.
      - Facial artery on the way
    - or endo-buccal: inter-maxillary commissure
    - 20U to start with



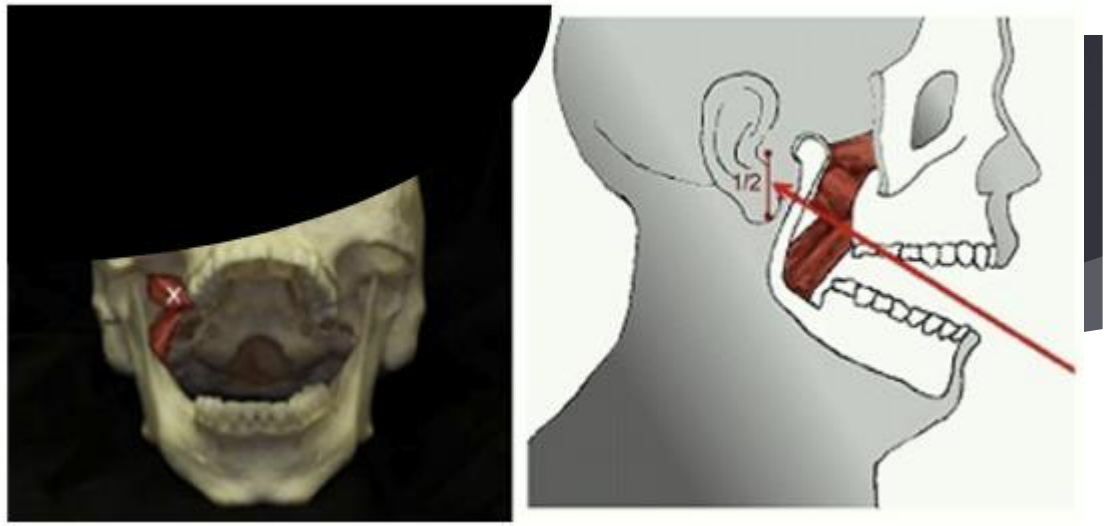


Injection techniques

- Opening dystonia
  - Submental complex
    - =Hyoid muscles
    - EMG
    - 1 cm from the anterior edge of the mandible
    - Slightly lateral to the median line
    - Literature: 30U to 200 U
      - Our experience: 5U per side
    - Risk of severe dysphagia
  - Lateral pterygoid

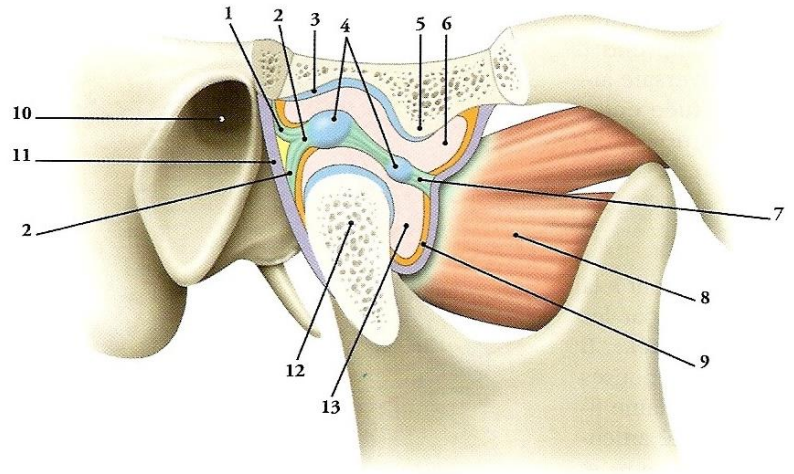


Injection techniques



Moscovich, 2015

- Deviated dystonia
  - Lateral pterygoid
    - Contralateral to the deviation
    - Intra-oral approach
    - External approach:
      - Slightly open mouth,
      - Approximately 35 mm from the ACE and 10 mm from the zygomatic ar
      - 15 degree upward angle
      - Risk: maxillary artery
      - Recommended doses: 20-40U



Injection techniques

- Tongue movements
  - Semiotic analysis +++
  - Risk of dysphagia +++
  - Small doses: 10U to start with
  - EMG location
  - Hogweed:
    - Posterior injections for lingual protraction
  - Hyoglossa:
    - for ipsilateral rotations
- Lip movements
  - Semiotic analysis
  - Risk of drooling/asymmetry of the face
  - Very low doses: 2.5-5U to start with

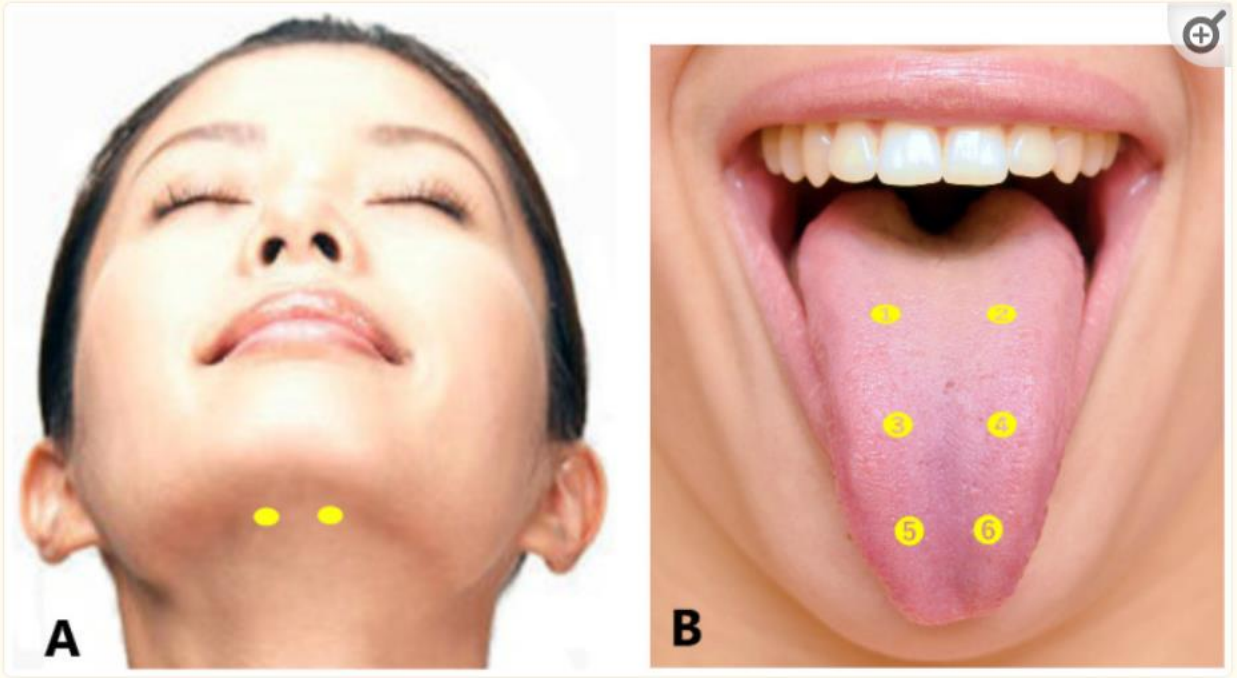


Figure 4

Submandibular sites of BoNT injection for lingual dystonia (**A**). Intraoral sites for BoNT injection (**B**) for protrusion type (❶–❹), laterotrusion type (right deviation (❶, ❸), left deviation (❷, ❹)), and curling type (❶–❻) [4,25,173].

Yoshida, 2022

Side effects of toxin injections

- 27.1% according to a 2021 meta-analysis (Dadgardoust et al.)
- Regional weakness
  - (excessive in relation to the desired effect, or nearby)
- Haematoma
- Painful tension at the injection site
- Depending on the injection site:
  - Minor discomfort when chewing
  - Asymmetric smile or absence of smile
    - Ptosis of the labial commissure and subsequent lip incontinence
  - Xerostomia
  - Changes in articulation (rhinophonia, lingual paresis)
  - Dysphagia

Side effects of toxin injections

- Dysphagia:
  - Possible impairment of 2 of the three swallowing phases
  - Oral phase
    - Reduced efficiency of bolus preparation
      - Mastication, insalivation, kneading with the tongue
    - Decreased efficiency of lingual propulsion
  - Pharyngeal phase
    - Velar elevation defect
      - Pharyngo-nasal reflux
    - Failure of the base of the tongue to retract/base-lingual propulsion
    - Pharyngeal propulsion defect

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