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Review article

Collum-caput (COL-CAP) concept for conceptual anterocollis, anterocaput, and forward sagittal shift

Josef Finsterer [a,*], Concha Maeztu [b], Gonzalo J. Revuelta [c], Gerhard Reichel [d], Daniel Truong [e]

[a] Krankenanstalt Rudolfstiftung, Vienna, Austria
[b] Division of Neurology, General Hospital, Murcia, Spain
[c] Department of Neurosciences, Division of Neurology, Movement Disorder Program, Medical University of South Carolina, Charleston, USA
[d] Kompetenzzentrum für Bewegungsstörungen Paracelsusklinik Zwickau, Zwickau, Germany
[e] The Parkinson and Movement Disorder Institute, 9940 Talbert Ave, Fountain Valley, CA 92708, USA

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ABSTRACT

Objectives: Anterocollis as a rare subtype of cervical dystonia is difficult to treat and thus less appreciated than other subtypes of cervical dystonia. This review aimed at summarising and discussing recent advances in the management of anterocollis.
Methods: Literature review.
Results: Pure anterocollis is a rare condition but 1–24% of the cases of complex cervical dystonia present with an anterocollis component. Applying the collum-caput concept, anterocollis may be subdivided into conceptual anterocollis, anterocaput, and forward sagittal shift, which is useful to direct selection of dystonic muscles for treatment. Additionally, identification of dystonic muscles in conceptual anterocollis, anterocaput, or forward sagittal shift is achieved by electromyography, computed tomography, magnetic resonance imaging, or FDG-positron emission tomography. Treatment of choice is botulinum toxin A. In case of treatment failure, more rarely affected muscles need to be identified and injected. Deep muscles, as are frequently involved in conceptual anterocollis, anterocaput, and forward sagittal shift, should be injected only under guidance of electromyography, endoscopy, or imaging. The more accurately affected muscles are identified, the better the outcome.
Conclusions: Anterocollis as a subtype of cervical dystonia, responds poorly to botulinum toxin but management of this condition can be improved by application of identifying and guiding technologies.

Contents

* Corresponding author at: Postfach 20, 1180 Vienna, Austria, Europe.
E-mail address: fifigs1@yahoo.de (J. Finsterer).

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1. Introduction

Dystonia is defined as involuntary, centrally-driven muscle hypercontraction, which may be sustained (spasmodic), resulting in abnormal posture, or may be repetitive, resulting in rhythmic dystonic tremor or phasic cloniform dystonia, or both [1,2]. Dystonia may be painless or painful and may slow voluntary movements and reduce range of motion [1,2]. Concerning the distribution, dystonia may be focal, multifocal, segmental, or generalised. With disease progression focal or segmental dystonia may spread and generalise and the degree of dystonia may increase [3]. Generalisation may be accompanied by motor overflow (overflow dystonia) or contralateral mirror dystonia [4]. Dystonia may occur spontaneously or may be triggered by certain voluntary movements, head positions, walking, standing, or sitting [5]. The most frequent of the focal dystonias is cervical dystonia (CD) [6]. It occurs with a prevalence of 1:10,000. A rare subtype of CD is anterocollis [7]. The following review focuses on the management of anterocollis since this subtype of CD is rare and difficult to treat and thus less appreciated than other subtypes of CD. Literature for this review was collected by a systematic search of PubMed applying appropriate search terms.

2. Taxonomy

The term anterocollis is usually applied in a double sense. First, anterocollis is used as a phenomenologic term (phenomenologic anterocollis (PACOL)) describing phenomenologically an abnormally forward flexed head position also termed head drop, head ptosis, dropped head, or head tilt. Second, the term anterocollis is used in a causal sense for describing a subtype of CD. PACOL is due to weak head extensor muscles and not to dystonic, overactive head flexor muscles. This is also the case for camptocormia where the entire spine is bent forward due to weak trunk extensor muscles.

3. Collum-caput (COL-CAP) concept

One approach to assess CD is by application of the COL-CAP concept, introduced by Reichel et al. in 2014 [8]. The COL-CAP concept relies on the distinct differentiation between dystonic muscles affecting exclusively the position of the cervical spine (collum type, conceptual anterocollis CACOL), exclusively affecting the position of the head (caput-type, ACAP), or affecting both (shifts). According to the COL-CAP concept, 11 subtypes of CD can be differentiated (torticollis, torticaput, laterocollis, laterocaput, retrocollis, retrocaput, anterocollis, anterocaput, lateral shift, posterior sagittal shift (combination of retrocollis and anterocaput ("double chin")), and forward sagittal shift (FSS)) [8]. In the majority of the cases, CD manifests as a combination of these 11 subtypes with one subtype usually predominating. Pure subtypes of CD are rare. Maximally 23 paired muscles are affected in CD but some muscles are preferentially affected compared to others in each of the subtypes and in each individual. Among the 11 subtypes of CD, three present with anterocollis, CACOL (Fig. 1), ACAP (Fig. 2), and FSS (Fig. 3), CACOL is characterised by a forward flexion of the cervical spine with reduced angle between the thoracic spine and the cervical spine but preserved angle between skull base and vertebrum C1 in the sagittal plane (Fig. 1). ACAP is characterised by a normal angle between cervical spine and thoracic spine but decreased angle between C1 and the skull base in the sagittal plane (Fig. 2). FSS is characterised by a decreased angle between the cervical and thoracic spine but an increased angle between C1 and the skull base (retrocaput) (Fig. 3). Differentiation between CACOL, ACAP, and FSS is essential for determining which muscles are the ones most likely affected and most likely to respond to treatment.

4. Aetiology and pathogenesis

The cause of PACOL is muscle weakness or hypotonia of the neck extensors. The cause of CACOL, ACAP, and FSS is dystonic over-activity of the neck-/head-flexors or neck extensors. Only in mitochondrial disorders, which frequently manifest with myopathy but occasionally also with dystonia, can both types of anterocollis coexist. The most frequent causes of PACOL are myopathy (myasthenia, scapuloperoneal muscular dystrophy, nemaline myopathy, myotonic dystrophy, polymyositis, dermatomyositis, inclusion body myositis, necrotising myopathy, steroid myopathy, hypokalemic myopathy, radiation-induced myopathy), neuro-/neurono-pathy (amyotrophic lateral sclerosis, post-polio syndrome, Guillain–Barre syndrome, chronic inflammatory demyelinating polyneuropathy, Bannwarth syndrome), movement disorders (Parkinson syndrome, multisystem atrophy), neurodegenerative disease (Lewy-body disease), metabolic disease (mitochondrial disorder, carnitine-deficiency, acid-maltase deficiency, hypothyroidism,

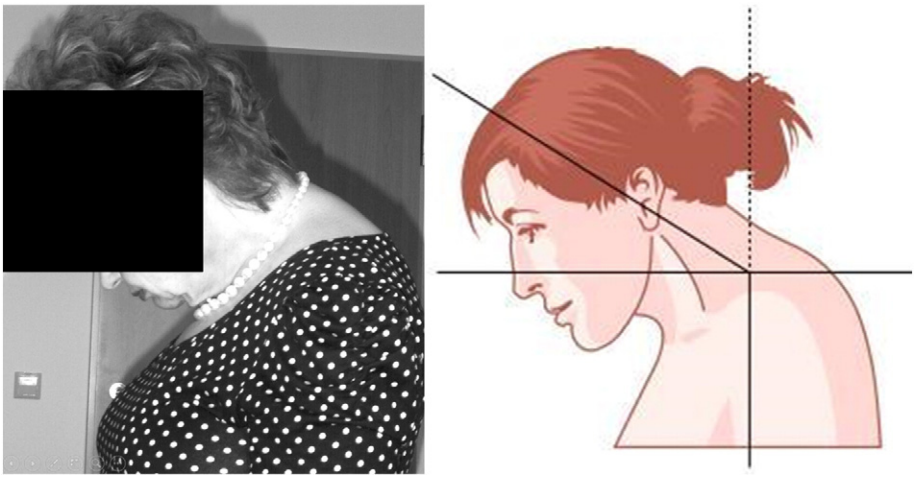

Fig. 1. Example of a patient with conceptual anterocollis.