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Tremor and Other Hyperkinetic Movements

Case reports

Torticaput versus Torticollis: Clinical Effects with Modified Classification and Muscle Selection

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Abstract

Background: Several different subtypes are distinguished in cervical dystonia, depending on their different levels of movement. In simple rotation, classified as torticollis spasmodicus, we now differentiate between torticollis and torticaput dependent on whether only the head or the neck is turned. The new classification system permits for different injection schemes.

Case reports: In a retrospective study of 22 patients, we examined whether modifying the injected muscles leads to improvement in the results as evaluated in the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS). The results showed that both injection schemes do in fact lead to improvements while differentiating between caput and collum has significantly better effects.

Discussion: Due to our results we recommend the classification differentiating between torticollis and torticaput type.

Keywords: Torticaput, torticollis, cervical dystonia, botulinum toxin, TWSTRS, Obliquus capitis inferior, sonography

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Introduction

In the last few years, a distinction has been made in cases of cervical dystonia between the so-called head (caput) and neck (collum) types, for example, between torticaput and torticollis.[1,2] Relying on this classification, different muscles have been injected from what was previously the case. The obliquus capitis inferior muscle (Figure 1) in particular has gained attention, which had earlier been but rarely treated and which for understandable reasons called for injection under ultrasound control.[3] However, to date no data have been published on controlled studies supporting the claim that the new classification involving injections in different muscles is in fact superior.

That is why we asked if there is any advantage of the classification differentiating torticaput and torticollis (Figure 2), the consequent change in injection scheme, and the selection of different muscles.

Patients and methods

From 2013 to 2018, we consecutively examined patients at two centers (Freiburg and Wolfach) with a torticaput who had up to then been treated for torticollis and in whom the ipsilateral splenius capitis muscle and the contralateral sternocleidomastoideus muscle had been injected.

Patients were excluded if they had any other subtype of cervical dystonia or a dystonic tremor. After this change in classification, the previous dosage was adhered to, while the choice of target muscles was altered so as to inject the contralateral sternocleidomastoideus, the contralateral trapezius, and the ipsilateral obliquus capitis inferior muscles (Video 1). All injections were performed under ultrasound control.

Before injecting, all patients were given the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS)[4] according to the previous treatment

scheme, and again 4 weeks post-injection according to the same scheme and according to the modified one.

Results

During the study period, a total of 22 patients (13 women and 9 men, mean age 58.7 ± 11.85 years) with a typical torticaput (without any further subtype) were examined and treated (Table 1). Almost all subjects benefited from the injection, according to both the standard treatment scheme (−2.68 points) and the modified one (−4.77; $p < 0.0001$ each). Relevant undesired side effects were not reported in any one of the two groups. The injection under the modified injection scheme demonstrated a superiority to the standard scheme (−2.09; $p < 0.0001$).

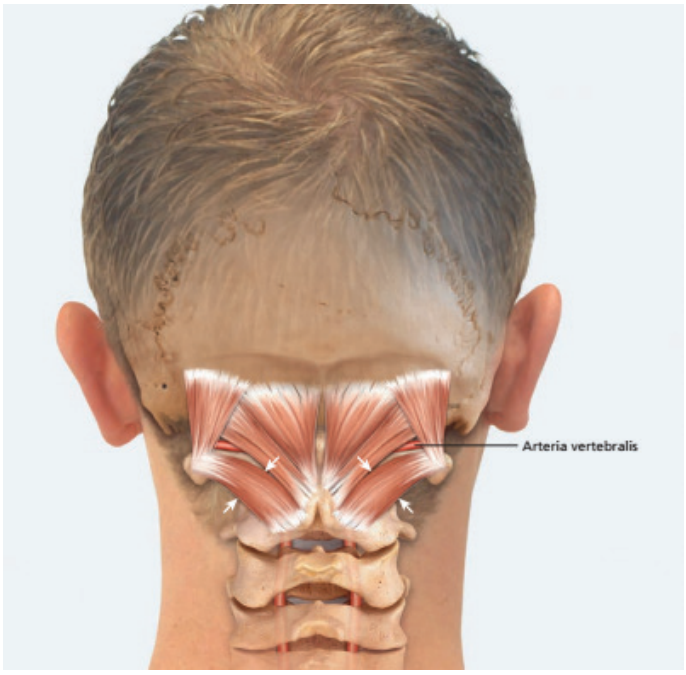


Figure 1. **Anatomy of short neck muscles.** Short Neck Muscles with Marked Obliquus Capitis Inferior Muscle[9] (with Permission of the Author and Publisher).

Discussion

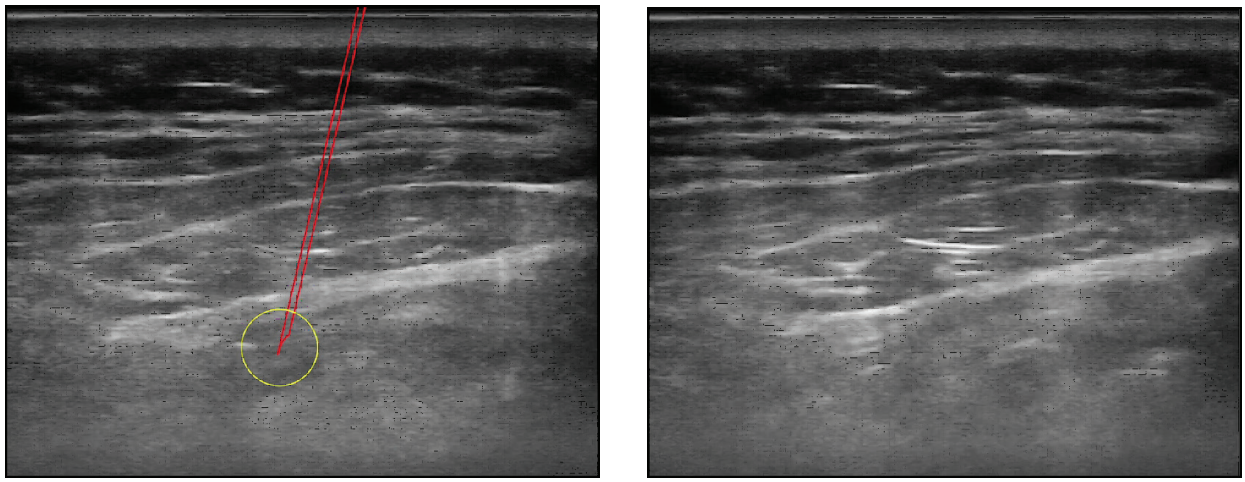
Since Reichel's publication in 2011 we have been discussing different subtypes of cervical dystonia.[2] This makes sense because different muscles are involved. We should take the two levels of movement into account: the upper one between the skull and C2 and the lower one between C2 and C7. The C2 vertebra can be regarded as a fixed point. When muscles that induce a rotation of the head are dystonic, the head rotates and the neck remains in the position, leading to a torticaput. If action takes place caudal to C2, a rotation of the neck occurs in relation to the trunk, termed torticollis. The position of the larynx offers help here in clinical orientation: in cases of torticaput, the larynx remains rather more in a medial position, and in torticollis it rotates laterally. Of course, in cases of torticollis, the head is also rotated due to the rotation of the neck (Figure 2).

The muscles traditionally injected in cases of torticollis are the contralateral sternocleidomastoideus and the ipsilateral splenius capitis[5] whereby it is important to note that the contralateral sternocleidomastoideus has an effect on head rotation and not that of the neck.[1,2]

Recently, an observational study was published, which compared torti- or laterocollis or –caput.[6] In the present analysis, we have concentrated on the clinical findings before and after the introduction of the new concept, especially for patients with torticaput.

The results of the underlying study show, first, that the traditional way of treating torticollis does work, which is not surprising in as much as it confirms our clinical experience.[5] They show, second, that interestingly the modification induces an improved effect, thus supporting a differentiated, flexible approach. It must be admitted that pure cases of torticaput are rare[1] and typically involve combinations.

Figure 2. **Torticaput vs. Toricollis.** In Cases of Torticaput (on the Left), the Larynx Remains Rather More in a Medial Position, and in Torticollis (on the Right) it Rotates Laterally.



Video 1. **Injection into the OCI under sonography control.** Once with graphic element (red marked needle), once without additional orientation.

Table 1. **Examined Patients with Torticaput**

Pat. No	Sex	Age	TWSTRS – 0	TWSTRS – 1	TWSTRS – 2	TWSTRS – 3
1	F	52	17	15	18	12
2	F	48	16	14	16	11
3	F	59	20	16	21	14
4	M	62	11	9	11	7
5	F	69	22	20	21	17
6	M	38	16	12	16	9
7	F	42	19	16	18	14
8	M	55	10	8	11	7
9	F	72	14	9	14	9
10	M	69	4	3	4	2
11	M	51	13	13	14	11
12	F	47	14	12	14	10
13	M	58	17	13	17	12
14	F	51	18	16	17	13
15	M	78	16	13	16	12
16	F	47	6	5	6	4
17	F	66	16	11	15	9
18	F	73	17	15	17	11
19	M	81	9	7	9	6
20	F	56	12	9	11	7
21	F	62	21	15	21	11
22	M	67	19	17	19	13
Mean		58.7	14.86	12.18	14.82	10.05
SD		11.85	4.68	4.19	4.60	3.53

Abbreviations: SD = standard deviation; TWSTRS – 0 = prior to conventional therapy; TWSTRS – 1 = 4 weeks after conventional therapy; TWSTRS – 2 = prior to reinjection; TWSTRS – 3 = 4 weeks after modified therapy.

Significance: 0 vs. 1: t(21) = 8.61; p = 0.25 *10^−7 (p < 0.0001); 1 vs. 3: t(21) = 8.9; p = 0.14 *10^−7 (p < 0.0001); 2 vs. 3: t(21) = 12.3; p = 0.48 *10^−10 (p < 0.0001).

Baseline TWSTRS values were lower than in studies with cervical dystonia, likely because pure torticaput generally has a lower overall score, because of the structure of TWSTRS. The improvement in the TWSTRS was also slightly lower than, for example, in the comparative study on ona- versus incobotulinumtoxin A, probably for the same reason.[7] This could be because it is mainly the rotation that is affected (not other parameters of the TWSTRS), and not the improvement of the symptoms is detected.

Furthermore, our study is retrospective and thus only considered therapy responders. The interval between the injections was 12–16 weeks so that even with a residual effect can be expected. Also, the use of ultrasound can influence the results, on the one hand, a placebo effect and, on the other hand, a more precise injection.

The next step should necessarily be conducted as a controlled, blinded, prospective study incorporating further subtypes, and de-novo patients. A cautionary caveat, however, must note that to date we still do not have a scale that takes the new classification into account according to head and neck type.[8] In addition, the subjective improvements and the quality of life should be recorded.

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