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The clinical phenomenology and associations of trick maneuvers in cervical dystonia

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Abstract Sensory trick is an unusual clinical feature in cervical dystonia that attenuates disease symptoms by slight touch to a specific area of the face or head. Using a semi-quantitative questionnaire-based study of 197 patients with idiopathic cervical dystonia, we sought to determine probable pathophysiologic correlates, with the wider aim of examining its eventual clinical significance. The typical sensory trick, i.e., light touch, not necessitating the use of force leading to simple overpowering of dystonic activity, was present in 83 (42.1 %) patients. The vast majority of the patients required a specific sequence of sensorimotor inputs, including touch sensation on the face or different areas of the head, and also sensory and motor input of the hand itself. Deviations often led to a significant decrease in effectiveness and lack of expected benefit. Moreover, patients able to perform the maneuver reported compellingly higher subjective effect of botulinum toxin treatment (median 7 vs. 5 on a scale of 0–10; $p < 0.0001$) and lower depression score (median 10 vs. 14 on the Montgomery Åsberg Depression Rating scale; $p < 0.001$). Overall, the results point to marked disruption of sensorimotor networks in cervical dystonia. The mechanism of the sensory trick action may be associated with balancing the abnormal activation patterns by specific sensorimotor inputs. Its presence may be considered a positive predictive factor for responsiveness to botulinum toxin treatment.

Keywords Sensory trick · Cervical dystonia · Botulinum toxin · Treatment effectiveness

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Introduction

Cervical dystonia (CD), which is the most common form of focal dystonia, is a syndrome characterized by involuntary repetitive twisting movements of the head, leading to intermittent or constant abnormal postures interfering with voluntary movements (Fahn 1988; Fahn et al. 1998). Frequently, CD patients adopt maneuvers to ameliorate the dystonic posture and reduce the abnormal movements that can cause significant discomfort (Poisson et al. 2012; Patel et al. 2014a, b; Ramos et al. 2014). This classic hallmark, referred to as sensory tricks or geste antagoniste, has also been confirmed in many other focal dystonias, including writer's cramp (Kaji et al. 1995), blepharospasm (Gómez-Wong et al. 1998), and lingual dystonia (Tan and Chan 2005). Typical stimuli are light touches in particular areas. For example for cervical dystonia, light touching usually involves the face or neck, which can result in complete cessation of abnormal head movements (Schramm et al. 2004). This occurs even though the applied force is weak and not sufficient to counteract dystonic contraction by simply overpowering it (Ramos et al. 2014). Moreover, complex trick maneuvers have been described as dancing (Molho et al. 1996), walking backward and hopping in truncal dystonia (Fahn 1984) and vocalization in facial dystonia (Wiener and Nora 1984). Owing to its heterogeneity (sensory trick varies from person to person and within the same individual) and bizarre character, the

sensory trick used to be interpreted as an indicator of a possible psychogenic etiology of dystonia (Munhoz and Lang 2004). Despite numerous studies attempting to elucidate the pathophysiology of dystonia (Filip et al. 2013; Abbruzzese and Berardelli 2011; Vidailhet et al. 2009), which propose various neural node affections in this syndrome (Marsden et al. 1985; Inoue et al. 2004; Filip et al. 2013, 2014), the mechanism underlying the sensory trick phenomenon remains elusive. The most common hypothesis suggests that sensory tricks influence proprioceptive input; therefore balancing the ratio of inhibition to facilitation (Ramos et al. 2014).

Interest has not only grown in the phenomenology of sensory trick (Jahanshahi 2000; Ochudło et al. 2007; Martino et al. 2010; Müller et al. 2001), but also in its neurophysiological (Wissel et al. 1999; Tang et al. 2007) and imaging correlates (Naumann et al. 2000). However, these descriptions are often based on small cohorts and do not provide information on eventual clinical relevance of this feature that would delineate a specific patient group with slightly different problems or needs.

The primary aim of our study was to define the characteristics of sensory trick, and the conditions necessary to perform the maneuver, to see if specific motor and sensory patterns are required for complete or partial induction of this response. Secondarily, we focused on different aspects of the disease, both clinically and as perceived by the patient, including information on satisfaction with the treatment, social integration, depressiveness, and the influence of dystonia on daily activities of the patients. We wanted to determine the specific characteristics of the group of patients able to use sensory trick.

Methods

Participants

In total, three-hundred and ten questionnaires were sent to active patients at the Movement Disorders Centre of St. Anne's Teaching Hospital, Brno, Czech Republic. The patients were chosen according to the diagnosis stated in their medical records and careful review of the available patient documentation. All the patients showed only symptoms of cervical dystonia. Diagnosis was made by an experienced neurologist in accordance with the current diagnostic criteria (Albanese et al. 2011). The accuracy of the diagnosis was supported by previous long-term clinical follow-up. Exclusion criteria included: presence of additional neurological conditions, psychiatric illness, and extensive drug and alcohol history.

All the participants provided written informed consent. The study was approved by the institutional review board of the St. Anne's Teaching Hospital, Brno, Czech Republic.

Materials and study design

The study is based on two data sets: clinical data using the severity score of the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) (Jost et al. 2013), and a complex self-administered questionnaire set. The questionnaires were sent to the patients meeting the study criteria by post, the patients filled the forms at home.

Clinical data of interest (age, sex, history of dystonia, treatment, and TWSTRS score collected at patient visits, i.e., at the time of maximal clinically and subjectively observed disease severity) acquired from patient medical documentation, were subject to further analyses. No objective data were systematically collected between regular patient visits assessing the situation during the period of peak treatment effect. The TWSTRS score was available only for 81 patients; the medical records for the remaining subjects included either the Tsui score or did not use any standardized scoring system.

The first part of the questionnaire focused on the sensory trick itself. The questions and possible answer options to be included in this instrument were devised based on literature review and previous clinical experience. The information of interest included the presence of the sensory trick, preconditions required for successful induction of the sensory trick, and its history and effectiveness. Additionally, to assess the subjective effect of botulinum toxin treatment after a usual, average administration, the most common method of symptom management (Ramirez-Castaneda and Jankovic 2013; Wissel et al. 2001), we added an 11-point scale [0–10; with 0 corresponding to no effect, 10 corresponding to complete suppression of symptoms and clearly delineated cut-offs for no or negligible (0–2), partial (3–4), good (5–6) very good (7–8) and complete effect (9–10)]. The pilot version of the instrument was assessed for clarity during clinical examination in 20 patients. The second part consisted of the Cervical Dystonia Impact Profile (CDIP-58) (Cano et al. 2004) and of the Montgomery Åsberg Depression Rating Scale (MADRS) (Montgomery and Asberg 1979). The CDIP-58 scale measures the impact of cervical dystonia on the life of the patient in eight subscales grouped into three conceptual domains: the symptoms (comprising subscales for the head and neck, pain and discomfort, and sleep), daily activities (the subscales for upper limb activities and walking), and psychosocial sequelae (the subscales for annoyance, mood, and psychosocial functioning). After transformation, the individual scores and the total score of CDIP-58 ranged from 0 (no effect) to 100 (most impact). The data were collected by a commissioned postal survey of patients with questionnaires mailed to the home addresses of the patients.

Data analysis

The analyses were performed using the Statistica 12 software (Statsoft Inc., Tulsa, OK, USA). First, questionnaires with missing responses were excluded from the analysis to ensure the reliability of the scores and avoid biases, leaving in total 197 valid subject data sets. Second, continuous variables were checked for normal distribution by graphing the normal probability plot and using Kolmogorov–Smirnov test, revealing global significant departures from normality. After that, demographic variables and the main parameter of interest (presence of sensory trick) were compared between patients with available TWSTRS score, and without it using the Mann–Whitney *U* test. The aim of this process was to ensure that these patient groups did not show any significant clinical differences. Because no significant group differences were obtained ($p > 0.05$ for all clinical indexes), the subjects were treated as a single cohort in all the subsequent analyses. The Mann–Whitney *U* test was also used to compare the variables of interest with the presence of the sensory trick as the independent variable. Analysis of categorical variables was performed using Pearson's Chi-squared test. Correlation analyses were based on the Spearman Rank test. The significance value of $p < 0.01$ was used for all the comparisons.

Results

Demographics and basic data on dystonia history

Of 320 questionnaires sent to the patients meeting the study criteria, 212 questionnaires were returned, yielding a return rate of 66.3 %. After excluding the questionnaires with missing responses, the data set contained 197 respondents, 148 (75.1 %) women and 49 (24.9 %) men. The median age was 58 years (range 26–83 years). The median disease duration was 12 years (range 1–49 years). The most common presenting symptom of the disease was rotation of the head, 135 (68.5 %) patients; tremor in 97 (49.2 %) patients; pain in 44 (22.3 %) patients; with 131 (66.5 %) patients stating only one main symptom at the onset of the disease, 50 (25.4 %) patients stating two main symptoms, and 16 (8.1 %) patients reporting all three listed symptoms already at the beginning of the clinical manifestation of the disease. Twenty (10.2 %) patients responded that the disease was also present in another member of their family.

Treatment in this group of CD patients was mainly based on botulinum toxin injections; specifically 187 (94.9 %) patients. Median treatment length was 7 years (range 0–23 years). The median dose per treatment session was 500 units of Dysport (range 0–800 units) and 125 units of Botox (range 0–200 units). The median interval between two treatment sessions was 105 days (range 62–155 days). The median general subjective effect of the botulinum toxin treatment, assessed using the above described scale of 0–10, was 5. Further oral medication for dystonia treatment (including analgesics due to dystonia associated neck pain) was used in 57 (28.9 %) patients.

Sensory trick

For the question focused on sensory trick presence, we clearly delineated the difference between typical sensory trick (light touch) and "forcible trick" (a maneuver similar to sensory trick, but necessitating the use of force antagonistic to the direction of dystonia) because we did not consider simple overpowering of dystonic muscle activity to be associated with more subtle processes underlying geste antagoniste.

Typical sensory trick was present in 83 (42.1 %; based on the number of 197 patients with valid questionnaires) patients, all treated with botulinum toxin injections. The patients used the following areas of touch: neck 51 (61.4 %) patients, cheek 45 (54.2 %) patients, chin 34 (41.0 %) patients, occiput 16 (19.3 %) patients, temples 10 (12.0 %) patients, forehead 8 (9.6 %) patients (see Fig. 1). Six (7.2 %) patients stated a different area, including atypical tricks, for example, biting the shirt collar, reported by one patient. Usually, only one (36 patients; 43.4 %) or two (28 patients, 33.73 %) possible areas were reported of touch that enabled successful performance of the maneuver. However, a minority of patients could perform the trick using multiple areas: three areas in 11 (13.3 %) patients, four areas in 1 (1.2 %) patient, five areas in 1 (1.2 %) patient and all the six listed areas in 6 (6.2 %) patients. The patients usually discovered the trick almost immediately after they started to perceive the symptoms of the disease; 56 (67.5 %) of patients. The effect of the sensory trick usually remained stable over the years with

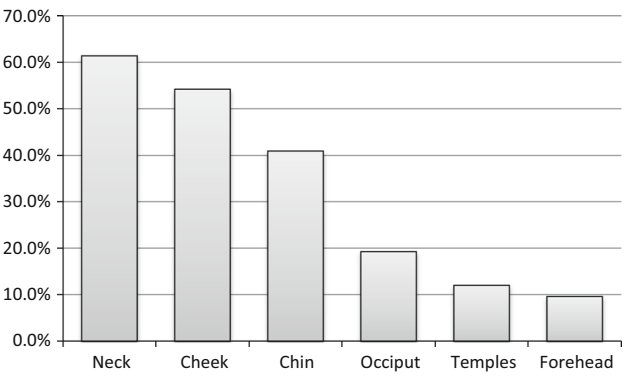

Fig. 1 Percentage of patients using the listed areas of touch in sensory trick; including patients with multiple possible areas (based on 83 patients who reported sensory trick)

57 (68.7 %) patients reporting no change. Only 7 (8.4 %) patients claimed that its effect had increased in comparison with the original level and 19 (22.9 %) patients stated that there was a slow decrease of its effects. Maximal effect of the maneuver was mostly reached without problems in every attempt with 36 (43.4 %) patients stating that the trick always functioned and 20 (24.1 %) patients reporting a success rate of about three out of four attempts. Twenty (24.1 %) patients benefited from a sensory trick in 1/2 attempts and 7 (8.4 %) patients stated even lower frequency. Subjectively perceived effect on symptom severity during the sensory trick was measured using a simple scale ranging from 1 to 10 (10 corresponding to complete disappearance of symptoms). The median stated effect was 7 (Fig. 2). The effect of the sensory trick was usually present for only a few seconds after removing the stimulating hand; 50 (60.2 %) patients. Seventeen (20.5 %) patients stated that the effect disappeared immediately and 16 (19.3 %) patients perceived at least partial relief lasting for a minute or longer.

To determine specific motor and sensory patterns necessary for induction of the feature, we added an additional part asking the patients to perform the maneuver under non-standard conditions and to evaluate the effect of the trick using a simple scale: the same effect as when the maneuver is performed under standard conditions, partial effect and no effect (Fig. 3). Of the subjects with a present sensory trick, 55 (66.3 %) patients were able to use both hands and 38 (45.8 %) patients had specific touch areas on both sides of the head to reach at least partial alleviation of the symptoms. Interestingly, 31 (37.3 %) patients could use the contralateral hand to perform the trick with an identical effect as on the preferential side. Furthermore, when patients touched the side of the face contralateral to the usually used side with the hand that did not typically perform the maneuver, 34 (41.0 %) patients felt at least partial

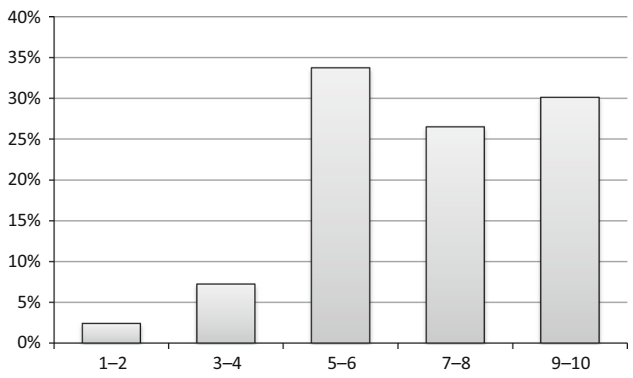

Fig. 2 Histrogram of subjective effectiveness of sensory trick, scale 1–10 with 1 corresponding to minimal effect and 10 to complete suppression of symptoms (based on 83 patients who reported sensory trick)

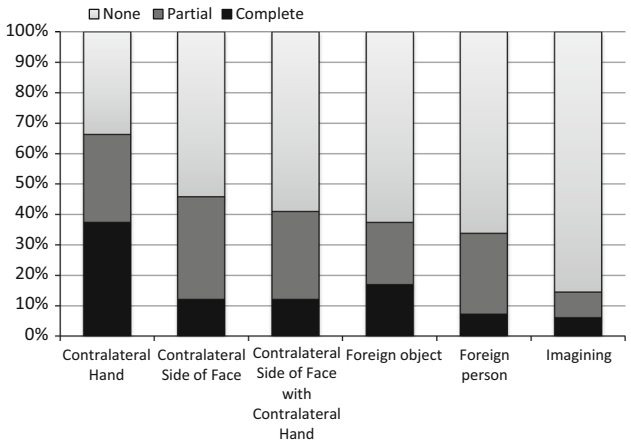

Fig. 3 Sensory trick under non-standard conditions; percentage of patients feeling complete/partial/no benefit after changing the maneuver in specified ways (based on 83 patients who reported sensory trick)

improvement. However, only 32 (37.3 %) patients stated at least partial improvement of the head position when using a foreign object to touch the preferential area with their preferential hand (a seemingly negligible change) and maintaining the movement pattern and tactile stimulus on the face. Touch from another person resulted in an even lower effect, bringing at least partial relief in only 28 (33.7 %) patients. Surprisingly, 12 (14.5 %) patients stated some improvement even when they were imagining that they were performing the maneuver.

Questionnaires

In the symptoms domain of the CDIP-58 scale, the median score was 33 (range 0–94); for daily activities it was 15 (range 0–100) and for psychosocial sequelae it was 24 (range 0–93), resulting in a total median score of 25 (range 0–93). The median MADRS score reached the level of mild depression; 12 (range 0–41).

Analysis of further impact of sensory trick

Using the Mann–Whitney *U* test with the presence of the sensory trick as the independent variable and factors associated with the general condition (as perceived by the patient; CDIP-58, MADRS, botulinum toxin treatment effect) used as dependent variables, we found significantly higher subjective effect of botulinum toxin treatment in patients with the sensory trick (median 7 vs. 5; $p < 0.0001$; Fig. 4) and significantly lower MADRS score in this group (median 10 vs. 14; $p < 0.001$). According to the Spearman rank test, there was a perceivable trend in the interaction between the MADRS score and the subjective effect of botulinum toxin treatment. However, it did not reach

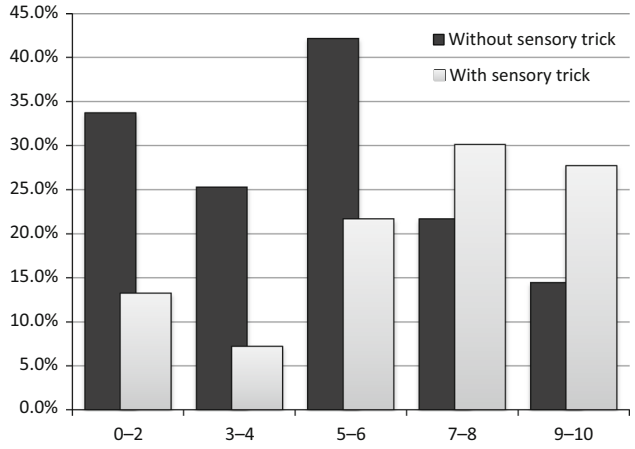

Fig. 4 Histogram of subjective effectiveness of botulinum toxin in patients with sensory trick and patients without it, scale 0–10 with 0 corresponding to no effect and 10 to complete suppression of symptoms (based on 83 patients who reported sensory trick)

statistical significance at the determined level (Spearman $R = -0.181$, $p = 0.011$). Furthermore, there were no significant differences found in the CDIP-58 scale or its domains. In an analysis of the demographic characteristics and disease severity of the patients with the sensory trick, we did not find any association with age, disease length, or TWSTRS score of the individual. Pearson's Chi-squared test did not reveal any association between sensory trick and sex or family history.

Discussion

The results of our study are in agreement with previous research defining the sensory trick as an episodic specific maneuver that ameliorates dystonia in a manner that may not be perceived as necessary to counteract involuntary movement (Ramos et al. 2014). Moreover, our data extend further and shows that there is a distinct motor and sensory pattern that needs to be followed to induce the sensory trick. Sensory input from the touch area itself, even maintaining the typical motor pattern of the individual, does not usually reproduce the desired effect, as seen in the maneuver amendment where the patient touched a specific area with a foreign object. Omitting the motor input (in the variation where the touch was provided by another person) led to an even lower effect of the trick, thus allowing us to hypothesize that the motor dimension of the feature is of considerable importance as well. Furthermore, the abundance of trick patterns in some individuals, such as the distinct degree of freedom in choosing the hand to perform the maneuver, and to some extent, even choosing the side of the face, may point to a more complex motor and sensory network underlying the mechanism of action of this response. Several studies have suggested the existence of impaired integration of proprioceptive input (Tinazzi et al. 2009) and abnormal kinesthetic perception (Putzki et al. 2006) in focal dystonia pathophysiology, thus proposing a possible hypothesis of the sensory trick as an adjustment of an abnormal link between sensory input and motor output. A compelling positron emission tomography study that demonstrated reduced activation of supplementary motor and primary sensorimotor cortex after sensory trick use (Naumann et al. 2000) may be considered a confirmation of this theory. When coupled with the sizeable group of patients that felt an improvement even when imagining doing the trick (Ramos et al. 2014; Greene and Bressman 1998), our data seem to indicate the presence of an even more elaborate system hidden behind this seemingly simple trick. However, as imagination of movement leads to similar brain activation patterns as the movement itself (Hanakawa et al. 2008), the influence of premotor structures may well explain the partial effect described by some subjects. Hence, with dystonia pathogenesis involving abnormal inhibition at multiple levels of the central nervous system (Kanovsky et al. 2003), the mechanism of the sensory trick as a complex sensorimotor input may lie in balancing the ratio of inhibition and facilitation (Gómez-Wong et al. 1998), thus temporarily ameliorating dystonia symptoms.

This notion provides a viable foundation for the intriguing difference in the subjective effectiveness of botulinum toxin treatment. Neuroimaging analyses have shown that botulinum toxin administration is associated with significant changes in functional sensorimotor maps (Opavský et al. 2011). These results complement the studies using various neurophysiological modalities (Currà et al. 2004) confirming pronounced cortical alterations associated with botulinum toxin treatment. Therefore, in patients capable of correcting abnormal activation patterns in these very areas using the sensory trick, botulinum toxin could indeed be affecting an environment displaying stronger tendencies for pathology normalization. This suggestion of a close association between the sensory trick and botulinum toxin treatment is also supported by reports of a restoration of sensory trick effectiveness after botulinum toxin administration (Ochudło et al. 2007; Müller et al. 2001). Furthermore, the group of patients with the sensory trick showed significantly lower MADRS scores than the remaining cervical dystonia patients; a result not attributable to differences in disease severity, as the analysis of TWSTRS scores failed to reveal any particularity. However, TWSTRS data were available only in a subsample of patients and even if no differences in demographic variables or the presence of sensory trick were found between the group with TWSTRS data available and the group without this

information, the concerns of possible lack of study power are tangible and further studies will be needed to elucidate this issue. The lower level of anxiety and depression could possibly play at least a partial role in the perceived higher effectiveness of botulinum toxin treatment; nevertheless, the interaction of the MADRS score and subjective treatment effect did not reach statistical significance. Nonetheless, this possible association must be considered as a factor that plays a seemingly minor role in the perception of treatment effect by the patients.

Nevertheless, some limitations need to be considered with regard to these results. First, the main weakness of this exploratory questionnaire-based study lies in the subjective characteristic of the information obtained regarding treatment effectiveness. Future studies incorporating clinical evaluation of the botulinum toxin effect will be helpful to parse the objective and subjective outcomes of the treatment, including various factors likely to influence patient satisfaction. Second, information based on a non-validated questionnaire without the option of providing further explanation to the subject and retrospective review of demographic and clinical data is a limitation. Furthermore, the usage of a self-administered questionnaire may be associated with a strong under-ascertainment bias. The discrepancy in the frequency of the sensory trick in the present study and most published literature, which relied on clinical examination, is partly supporting this view as well. In our study, 42.1 % of the patients reported the presence of the sensory trick, whereas sensory trick prevalence reported in literature is about 17–89 %, with the majority stating 70–80 % (Ramos et al. 2014). This variance could be partially explained by our strict differentiation between the "forcible trick" necessitating pressure to alleviate the disease symptoms, and the typical sensory trick, characterized exclusively by light touch, without any forcible movement induction; a distinction not expressly observed by other authors. However, this factor itself is unlikely to fully account for this difference, opening the relationship between the sensory trick presence and the botulinum toxin efficacy to bias and questioning the full validity of this finding. Moreover, the repeated performing of the maneuver, even if modified, could also be suspected of confounding to some extent, as little information is available on the effectiveness of the sensory trick after repetitive use. The ascertainment whether the reuse of sensory trick within a short time period leads to improvement or deterioration of its effect could be of much interest in further studies.

Owing to the retrospective characteristic of this study, the TWSTRS score was available only in about 40 % of the patients. This limitation is partly attenuated by statistically confirmed absence of significant differences between the patient groups (with and without available TWSTRS score), so the subjects could be treated as a single cohort in all the subsequent analyses.

And lastly, the pool of active cervical dystonia patients from an established botulinum toxin clinic used for this analysis excludes both milder patients not referred to specialized centers and many patients with insufficient or negligible botulinum toxin effect, including the resistant patients. This bias, even though difficult to address, could also be of interest in further studies.

Nevertheless, this study provides one of the most complex descriptions of the sensory trick, underscoring the pivotal role of the sensorimotor integration characteristic of this feature. Taken together, these findings highlight a clinical perspective of the sensory trick as a possible predictive factor of the effect of botulinum toxin treatment, and it agrees well with a growing body of evidence hypothesizing dystonia as a complex network disorder. Future studies incorporating further aspects of this feature and more complex clinical examinations will be helpful in expanding upon the currently provided results.

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Conflict of interest There are no potential conflict of interests regarding this paper and no financial or personal relationships that might bias this work.

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