

# Quantitative examination of isometric tongue protrusion forces in children with oro-facial dysfunctions or myofunctional disorders

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## Summary

Oro-facial dysfunctions (OFD) or oro-facial myofunctional disorders in children lead to severe problems in teeth and jaw position, articulation, chewing and swallowing. The forces of the tongue, the central muscle for articulation, chewing and swallowing are focused on in several studies. In this examination, isometric tongue protrusion forces (TPF) of children with OFD and controls were compared. Thirty participants with OFD and 30 controls were presented a target force level as a straight line on a monitor that they were supposed to match by generating an isometric tongue force for different target levels (0.25 N and 0.5 N). Correlations of the severity of OFD (symptom score) with the capacities of the TPF 0.25 N and 0.5 N were calculated. Statistical differences were obvious in TPF variability and the accuracy, depending on the weight. Tongue contact time, expressed as per cent (TCT, total contact: 100%), was significantly lower in children with OFD ( $P = .005$ ). Mean and median TPF was not different between groups. The predictive value of TPF for OFD revealed a level of 58.6% for TPF 0.25 N and 74.5% for TPF 0.5 N. Correlations of the severity of OFD were seen for some parameters. Subjects with OFD show significantly lower competencies in accuracy and endurance of tongue protrusion forces. This may have a high impact on phenotyping children with OFD and influence therapeutical approaches.

## KEYWORDS

accuracy, childhood apraxia of speech, myofunctional disorder, oro-facial dysfunction, tongue protrusion force

## 1 | BACKGROUND

Oro-facial dysfunctions (OFD) or myofunctional disorders include changes of the appearance, position and motility of lips, tongue, cheeks, jaw, throat and neck with a negative impact on oral posture and function.<sup>1</sup> Tongue thrusting and persistent infantile swallowing, mouth breathing, oro-facial muscular imbalance and malposition of the teeth and the jaw influence the integrity of the oro-facial system. This often leads to severe articulatory problems (phonetic disorders) or speech sound disorders (SSD). These difficulties may persist

throughout life and are detrimental to scholar, social and vocational development<sup>2,3</sup> with a long and intensive need for therapy and a high burden for the public health system.<sup>4</sup> The prevalence of SSD in children is estimated 2-15%, mostly 6-8%.<sup>5,6</sup> The prevalence of OFD is not known, but was estimated up to 87.6% for distinct oro-facial functions in healthy Brazilian schoolchildren, obtained by a screening which was based on an interview and a brief examination (NOT-S)<sup>7</sup> and for various diseases such as ectodermal dysplasia, adenotonsillar hypertrophy, Parkinson's disease, Prader-Willi syndrome, oromandibular dystonia and Treacher Collins syndrome (for review, see Bergendal

**TABLE 1** Patients' demographics

	OFD	Controls
n	30	30
Female	7 (23.3%)	12 (40%)
Male	23 (76.7%)	18 (60%)
Age (years) (mean)	9.0 ± 2.3 SD	9.6 ± 2.2 SD
Right-handed	23 (76.7%)	22 (73.3%)
Left-handed	6 (19.4%)	7 (23.3%)
Non-determined	1 (3.3%)	1 (3.3%)

et al).<sup>8</sup> Some children with oro-facial problems have childhood apraxia of speech (CAS). They often have extraordinary pronounced symptoms with distinct articulatory problems and the need of alternative communication devices (eg "talker").<sup>9</sup>

The pathogenesis for OFD or myofunctional disorders is likely multicausative. Pathognomonic symptoms that identify children with OFD or myofunctional disorders are not available. A high variability in symptoms presents challenges to establish a reliable classification. The current standard in OFD or myofunctional disorders diagnosis relies on clinical expert opinion, as there are no standardised and validated assessments available.<sup>9-11</sup> The oro-facial system is mainly evaluated by health professionals, among them speech-language pathologists, phoniatricians, ENT doctors, dentists, orthodontists, oral surgeons, paediatricians, neurologists and physiotherapists, all of whom are focused on different domains. The routine clinical examination assesses different movements, of which some are seen as particularly important, especially the motor coordination of the tongue, the core muscle involved in articulation.<sup>12,13</sup> However, the quality of clinical examination is limited by insufficient standardisation and lack of inter- and intra-rater reliability.<sup>14</sup> Complexity of clinical features and need of special materials are limitations same as the lack of objective approaches.<sup>9,10</sup> These limitations prevent reliable comparisons of study results and lead to inconsistent conclusions concerning diagnostic criteria and therapeutic recommendations.<sup>14</sup> Thus, the need for exploring and establishing standardised assessments has been emphasised.<sup>15-17</sup>

A task that has proven to be sensitive to examine motor dysfunctions in neurological disorder with motor impairments is the objective and quantitative assessment of tongue protrusion forces (glossomotography),<sup>18</sup> part of the Q-Motor battery established particularly for use in neurodegenerative diseases.<sup>19,20</sup> Application in multicentre clinical trials in Chorea Huntington allowed placebo-effect-free (mavoglurant, a selective metabotropic glutamate receptor 5 antagonist versus placebo) and sensitive detection of motor performance.<sup>19</sup>

We therefore hypothesised that glossomotography might open new approaches towards establishing criteria for diagnosing and assessing disability in patients with OFD or myofunctional disorders and possibly assist in evaluating novel therapeutical approaches in future trials. To explore the applicability of glossomotography in OFD or myofunctional disorders, this study was conducted to evaluate whether deficits in tongue protrusion forces in children with OFD or

myofunctional disorders can be detected compared to controls and whether they are correlated to clinical deficits.

## 2 | METHODS

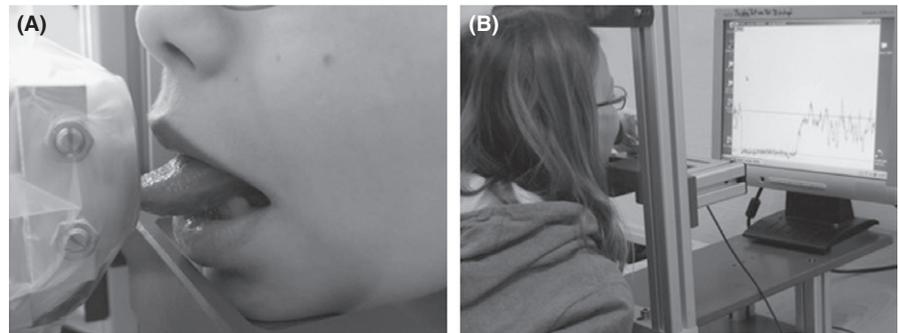
### 2.1 | Participants

Sixty children between 6 and 15 years were included in the study. Thirty children, 7 girls and 23 boys, mean age 9.0 (±2.3) years (SD) (median 8.6 years, range 5.1-14.9 years) with OFD or myofunctional disorders and 30 healthy controls, 12 girls and 18 boys, mean age 9.6 (±2.2) years (SD) (median 9.3 years, range 5.8-14.2 years) participated in the study after they and their parents gave their informed written consent to research and to publication in accordance with the Declaration of Helsinki. They were recruited from the outpatient clinic of Audiology and Phoniatrics and the Center for Chronic Sick Children, Charité University, Berlin, Germany. Inclusion criteria were a normal non-verbal intelligence (IQ) in the coloured progressive matrices (CMP), a normal hearing threshold on both sides (better than 20 dB HL), not more than one otitis in the last year, transitory evoked otoacoustic emissions reproducible over 60% on both ears, no arthritis, normal visus (anamnestically and tested before start of school), no co-existing neurological diseases, no cerebral palsy or epilepsy, no psychiatric diseases, no known sociological or emotional problems, capability of following simple verbal instructions. The diagnosis of OFD or myofunctional disorders was defined after carrying out the Berlin oro-facial Screening (BoS), a comprehensive examination of oro-facial motoric and sensible competences as described before.<sup>21</sup> Briefly, it considers 4 structured parts: the personal history, the observation, the examination of well-defined oro-facial movements of the lips and the tongue and the results/recommendations. It has been developed and optimised to offer a structured assessment that considers all necessary parts of diagnostic, which can be carried out in a reasonable time and offers an encompassing conclusion for therapy, therapeutical progress and interprofessional communication.

Controls were examined the same way and had to be inconspicuous in all oro-facial motoric and sensible items. Most children of both groups were right-handed (see Table 1).

### 2.2 | Experimental set-up and task: tongue protrusion force (glossomotography)

Isometric tongue protrusion force (TPF) was measured as described before.<sup>18</sup> Briefly, subjects were seated upright on a chair in front of a table with their chin resting on a height-adjustable base. A pre-calibrated and temperature-controlled force transducer (Mini-40; ATI Industrial Automation, Apex, NC, USA) was mounted 2 cm in front of their lips (Figure 1A). The force transducer had a circular plane contact surface measuring 40 mm in diameter, which was coated with a disposable plastic cover. A monitor was placed 30 cm in front of the subject's eyes presenting feedback on the tongue force exerted (Figure 1B). Subjects were instructed to open their mouth widely and, following a cueing tone, protrude their tongue to



**FIGURE 1** (A) In glossomotography, tongue protrusion force (TPF) is measured with a force transducer. (B) A monitor displays the target forces. Target force level (straight line) and individual TPF in a subject with mild deficits in tongue force variability and tongue contact time (TCT)

establish contact with the force transducer. They were presented a target force level as a straight line on a monitor that they were supposed to match by generating an isometric force with their tongue. After 35 seconds, a second cueing tone marked the end of each trial, and subjects were asked to retract their tongue. After test trials, four trials were recorded for each of the two target force levels 0.25 N and 0.5 N. Adherence of subjects to the instructions was closely monitored by the examiner. If subjects supported their tongue with teeth or lips to stabilise tongue protrusion, trials were excluded from evaluation and repeated. Data were sampled at 400 Hz, stored and analysed on a laboratory computer system (WinSCP, open-source software, Prague). Mean and median TPF (defined as mean and median of TPF from the 4 trials for each force per group), TPF variability expressed as coefficient of variation (TPF-CV, defined as standard deviation TPF divided by the mean TPF, shows how precise the subjects are able to coordinate their tongue protrusion forces), tongue contact time expressed as per cent (TCT, sign of endurance, defined as time in which the participant contacted the sensor with the tongue; total contact: 100%), and, as a sign of accuracy, the percentage of time the TPF remained (i) within a range of  $\pm 10\%$ ,  $\pm 20\%$  and  $\pm 50\%$  (TPF  $\pm 10\%$ , TPF  $\pm 20\%$  and TPF  $\pm 50\%$ ) of the target force level (the percentage of time in which the TPF stayed between the target of  $\pm 10\%$ : 0.225 N-0.275 N (0.25 N) and 0.45 N-0.55 N (0.5 N);  $\pm 20\%$ : 0.2 N-0.3 N (0.25 N) and 0.4 N-0.6 N (0.5 N);  $\pm 50\%$ : 0.125 N-0.375 N (0.25 N) and 0.25 N-0.75 N (0.5 N) and (ii) below 10%, 20% and 50% of the target force level (TPF < 10%, TPF < 20%, TPF < 50%; <10%: <0.225 N (0.25 N) and <0.45 N (0.5 N); <20%: <0.2 N (0.25 N) and <0.04 N (0.5 N); <50%: <0.125 N (0.25 N) and <0.25 N (0.5 N)) were calculated during a 20-second period, starting at 10 seconds after the first cueing tone and ending 5 seconds before the second cueing tone.

Oro-facial dysfunctions symptom score included the results (0 = not realisable, 3 = totally correct; Likert scale) of the movements of eight clue functions of tongue and lips (lips pursed, lips broad, change of lips pursed and broad, forming a mouth of a fish, tongue to the right side, tongue to the left side, tongue to the sides-consecutive movement, moving the tongue around the lips) (mean).

### 2.3 | Statistical analysis

Statistical analysis was performed using SPSS 22.0<sup>®</sup> (IBM Corp., Armonk, NY, USA). Groups were compared using nonparametric

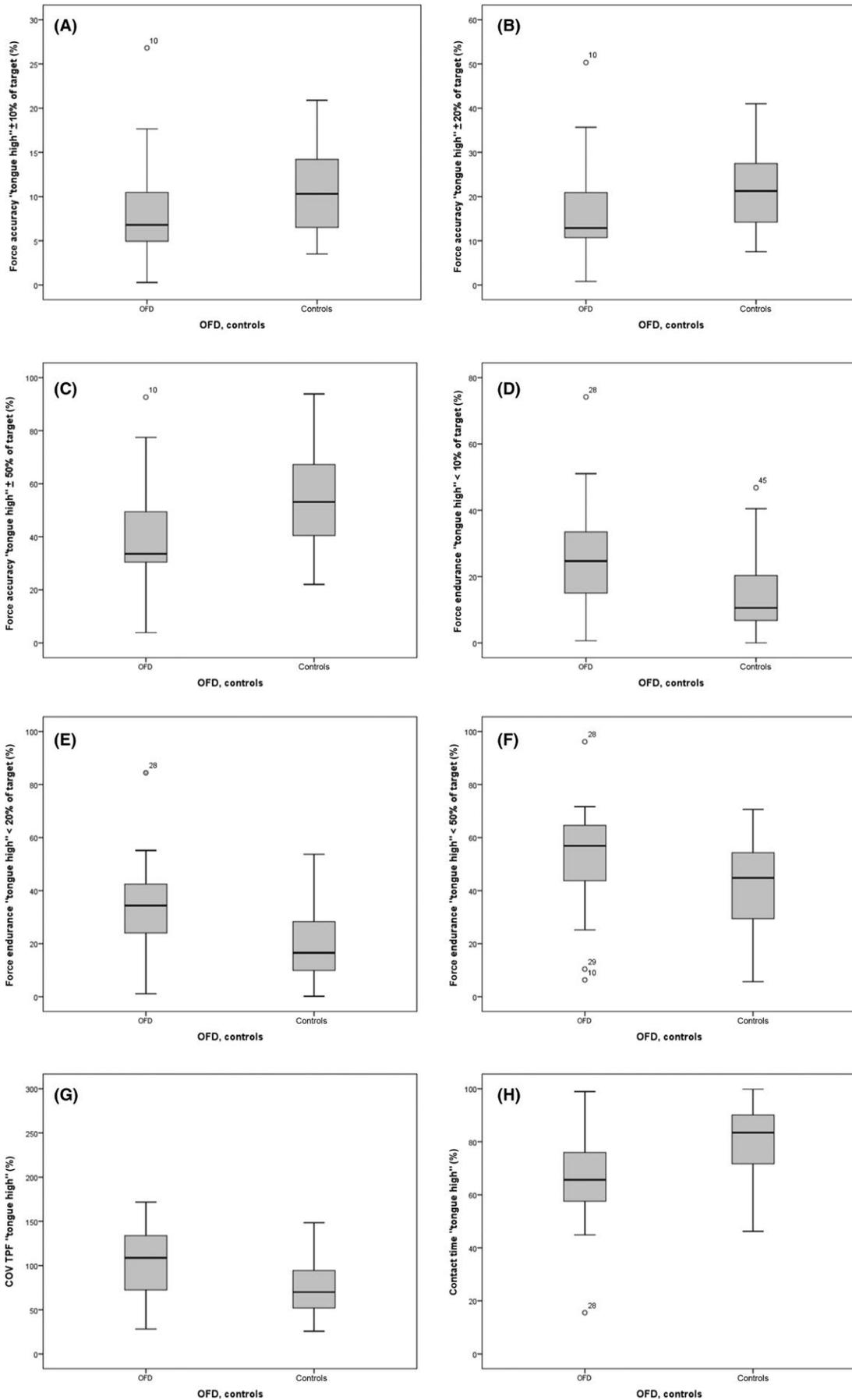
Mann-Whitney *U* tests; nonparametric Spearman's rank tests were calculated to analyse correlations of measures with the OFD score. Statistical significance was accepted at the  $P \leq .05$  level. No multiple-testing correction was applied, as the results of the study can be defined as hypothesis generating. Results of the study were expressed in box-whisker plots of the median and quartiles.

## 3 | RESULTS

At the *high target force level* (0.5 N, see Figure 2), statistical differences between the groups of symptomatic subjects and controls were seen in the measures of TPF variability expressed as coefficient of variation (TPF-CV;  $P = .009$ ) and the endurance expressed as the percentage of time. As a sign for accuracy, the tongue remained (i) within a range of  $\pm 10\%$ ,  $\pm 20\%$  and  $\pm 50\%$  (TPF  $\pm 10\%$ ,  $P = .043$ ; TPF  $\pm 20\%$ ,  $P = .015$ ; and TPF  $\pm 50\%$ ,  $P = .005$ ) of the target force level and (ii) below 10%, 20% and 50% of the target force level (TPF < 10%,  $P = .006$ ; TPF < 20%,  $P = .005$ ; and TPF < 50%,  $P = .037$ ). Tongue contact time, expressed as per cent (TCT, total contact: 100%), was significantly lower in children with OFD or myofunctional disorders ( $P = .005$ ). Mean and median TPF was not different between groups, see Table 2, indicating a resilient distribution for statistical computation. The most significant differences were seen in TCT, TPF  $\pm 50\%$  and TPF < 10% and < 20%, respectively.

At the *low target force level* (0.25 N), TPF  $\pm 50\%$  was significantly lower in children with OFD or myofunctional disorders compared to controls ( $P = .039$ ), and TPF  $\pm 10\%$  ( $P = .059$ ) and TPF  $\pm 20\%$  ( $P = .07$ ) did not differentiate well between the two groups. TPF-CV, mean TPF, TCT and precision of TPF  $\pm 10\%$  and  $\pm 20\%$  as well as TPF < 10%, 20% and 50% did not show significant differences between children with and without OFD or myofunctional disorders. The predictive value of TPF for OFD or myofunctional disorders revealed a level of 74.5% for TPF 0.5 N and 58.6% for TPF 0.25 N.

Correlations of the measures of the severity of OFD or myofunctional disorders, expressed as the OFD symptom score with the capacities of the TPF 0.5 N, revealed a weak but significant positive correlation to the parameter TPF  $\pm 50\%$  ( $r = .28$ ,  $P = .039$ ) and positive and negative correlations to the TPF-CV ( $r = -.237$ ), TPF  $\pm 10$ ,  $\pm 20$  ( $r = -.237$ ,  $r = .235$ ) TPF < 10%, < 20% and < 50% ( $r = -.236$ ,  $r = -.248$ ,  $r = -.228$ ) as well as TCT ( $r = .278$ ). Correlations between OFD symptom score and TPF 0.25 N showed a significant positive correlation



**FIGURE 2** Motor abilities in children with OFD (OFD) and controls (C) for the target forces 0.5 N, as shown in the box plots (A-H) and statistical results. Groups are best distinguished at 0.5 N in TCT in %, TPF  $\pm$  50% (C) and TPF < 10% (D) and 20% (E) (all shown in box-whisker plots of the median and quartiles)

**TABLE 2** Tongue protrusion forces. Between-group comparison and group medians

	Target force (N)	OFD vs C (P)	C, Median (quartiles)	OFD, Median (quartiles)	Correlation coefficient r sum score/P
TPF-CV	0.25	.210	117.24 (74.51-180.60)	151.00 (106.00-188.00)	-.166/.213
	0.5	.009*	70.00 (50.70-95.21)	108.78 (72.4-136.68)	-.237/.082***
Mean TPF (N)	0.25	.216	0.14 (0.10-0.21)	0.20 (0.12-0.23)	-.164/.219
	0.5	.346	0.31 (0.24-0.37)	0.29 (0.21-0.31)	.022/.871
Median TPF (N)	0.25	.088	0.13 (0.10-0.20)	0.20 (0.13-0.24)	-.228/.088***
	0.5	.346	0.30 (0.24-0.35)	0.28 (0.23-0.31)	-.014/.917
TCT (%)	0.25	.657	51.66 (43.74-73.18)	52.19 (42.25-65.14)	.059/.661
	0.5	.005**	83.44 (71.10-90.21)	52.19 (42.25-65.14)	.248/.068***
TPF $\pm$ 10% (%)	0.25	.059	7.07 (5.15-11.03)	5.90 (4.39-7.12)	.25/.058
	0.5	.043*	10.30 (6.43-14.49)	6.79 (4.9-10.56)	-.237/.082***
TPF $\pm$ 20% (%)	0.25	.07	13.55 (10.96-21.26)	11.66 (9.01-14.66)	.245/.063
	0.5	.015*	21.27 (13.97-27.97)	12.88 (10.57-21.31)	.235/.084***
TPF $\pm$ 50% (%)	0.25	.039*	35.51 (14.51-37.74)	28.72 (23.48-39.36)	.273/.038*
	0.5	.005**	53.15 (40.18-67.98)	33.59 (30.00-49.71)	.28/.039*
TPF < 10% (%)	0.25	.141	25.57 (14.61-37.74)	31.94 (22.14-41.69)	-.195/.143
	0.5	.006*	10.54 (6.63-20.34)	24.69 (15.00-34.00)	-.236/.083***
TPF < 20% (%)	0.25	.246	55.11 (32.06-61.98)	52.99 (39.11-63.73)	-.154/.250
	0.5	.005**	16.56 (9.37-28.91)	34.36 (22.50-43.33)	-.248/.068***
TPF < 50% (%)	0.25	.895	55.11 (32.06-61.98)	52.99 (39.11-63.73)	-.018/.896
	0.5	.037*	44.77 (29.37-54.77)	56.91 (42.00-64.67)	-.228/.094***

Medians, upper and lower quartiles (in parentheses) of tongue force measures.

C, controls; OFD, children with oro-facial dysfunction or myofunctional disorders; N, Newton; TPF-CV, tongue protrusion force coefficient of variation; TPF  $\pm$  x%, % forces stay in a range of x% of the target force; TPF < x%, % forces stay below x% of the target force. The correlation coefficient r describes the correlation between the OFD symptom score and the distinct tongue movement.

\* $P \leq .05$ , \*\* $P \leq .005$ , \*\*\*non-significant, but comprehensible correlation.

to the precision  $\pm$ 50% ( $r = .273$ ,  $P = .038$ ) and positive and negative correlations to the median TPF ( $r = -.228$ ) and TPF  $\pm$ 10,  $\pm$ 20 ( $r = .25$ ,  $r = .245$ ).

No significant difference could be detected between the groups within the variables gender, age and handedness.

## 4 | CONCLUSION

This study demonstrates that the Q-Motor glossomotography test detects motor variability in children with OFD or myofunctional disorders compared to control subjects during a tongue protrusion force-matching task. We observed that the *absolute* forces of isometric TPF did not differ significantly between groups (Table 2), which may explain contradictory results seen in previous studies.<sup>22,23</sup> However, all measures of *variability* of isometric TPF were able to discriminate between children with OFD or myofunctional disorders and controls in the 0.5 N target force condition; variability in

the 0.25 N target force only reached statistical significance for the parameter TPF  $\pm$ 50%.

Skilled movements presume to be associated with improvement of accuracy and a lower variability compared with novice performance.<sup>24</sup> Accuracy in a scientific context is a qualitative term that indicates whether there is an agreement between a measurement made on an object and its target value. The dynamic of accuracy of the task performance in the present study was described by calculating the percentage deviation of the forces from both target forces (0.25 N and 0.5 N). The results for accuracy of the high task force revealed higher, statistic significant dynamic changes compared to the lower task force. For the high force results, TCT and TPF  $\pm$ 50% revealed the highest statistic significances, indicating that children with OFD or myofunctional disorders have lower power of endurance and higher variation in isometric forces. This corroborates results from other studies in which high hold forces of an oro-facial task were more likely to show a significant dynamic compared to low hold forces.<sup>25</sup> It has been observed before that force control tasks at higher force levels are easier to perform

than lower force levels in finger force control tasks.<sup>25,26</sup> Motor units firing rate variability and motor recruitment are discussed in this context, as motor units are recruited per size. The smallest motor units are recruited first, while larger units fire successively to increase force output. This suggests that accuracy, with a lack of variability of motor output for movements of lower forces, may be the major driver of pathology in OFD or myofunctional disorders. Deficits in “extraction” of motor commands from the inherent “noise” in the nervous system are hypothesised.<sup>27,28</sup> As fine coordination of tongue movements is essential for articulation, chewing or swallowing, this observation may explain the clinical deficits seen and expected to impact on motor programming and planning in OFD or myofunctional disorders.<sup>15,29</sup>

Motor symptoms in OFD or myofunctional disorders are usually assessed with semiquantitative tests which are limited by subjectivity and categorical design, and thus often exhibit low sensitivity.<sup>15-17</sup> To date, studies that examined tongue forces with objective methods revealed contradictory results or were not applied to children with OFD or myofunctional disorders.<sup>12,22,23</sup> In children with OFD or myofunctional disorders exhibiting verbal dyspraxia, reduced tongue forces and tongue strength endurance were reported before.<sup>30</sup> Variability of motor output in children with OFD or myofunctional disorders with an isometric tongue protrusion force exercise has never been formally assessed to the best of our knowledge. However, children with OFD or myofunctional disorders or CAS showed deficits in other motor functions, in coordination and temporal sequencing,<sup>30</sup> indicating that an overall, non-task-specific problem may exist that influences fine motor coordination.

With respect to clinical developments, our findings suggest that therapeutical interventions that focus on isolated tongue-muscle strengthening may not be warranted, but support therapeutical approaches aiming to improve accuracy of movements.

In other neurological diseases, for example Huntington's disease, the disease burden correlates with the isometric TPF variability.<sup>19</sup> In multiple sclerosis, TPF variability correlated with the individual disease burden (Expanded Disability Status Scale, EDSS) and with changes in diffusion tensor imaging of the brain.<sup>20</sup> In the present study, especially the variability of the TPF showed some significant correlations with clinical OFD scores. Our results showed that OFD score was positively correlated to TPF  $\pm$  50% for both forces (0.25 N and 0.5 N), some measures of precision ( $\pm$ 10,  $\pm$ 20; 0.25 N and 0.5 N) were positively and some measures of endurance (<10%, <20%, <50%; 0.5 N) were negatively correlated to the OFD symptom severity, confirming that a reduced accuracy of oro-facial function is accompanied by lower capacity in precise TPF output.

We acknowledge that our study has several limitations. Motivation of the children could have impacted the TPF-outcome. We minimised this by explaining the study conditions and tasks in detail to all participants. In addition, test trials were carried out before each task. Adherence of subjects to the instructions was closely monitored. If subjects supported their tongue with teeth or lips to stabilise tongue protrusion, trials were excluded from evaluation and repeated. We recruited 3 times more boys than girls in the OFD group. The distribution between the sexes of OFD or myofunctional disorders is not exactly known.<sup>7,8</sup> The prevalence of speech sound disorders (SSD)

is estimated twice to threefold as high in boys,<sup>31</sup> so that the tested study group represents the prevalence of speech sound disorders. In the OFD group, patients were selected within the age in which OFD or myofunctional disorders show a peak and can be diagnosed most reliably.<sup>21</sup> Other studies showed an age dependency of tongue force measures,<sup>23</sup> so that assessing the impact of age seems to be important for future studies.

Interestingly, the handedness, often discussed in the context with development of speech, was not associated with the results of TPF in the between-group comparison. This underlines result from others,<sup>32</sup> who did not find a higher number of left-handed in a group of children with SSD.

The development of objective and quantitative measures for functional deficits is a key goal for improving assessment and the understanding of OFD or myofunctional disorders in children and beyond. With respect to the variability in OFD or myofunctional disorders, we conclude that TPF-tasks performed with the Q-Motor glossomotography assessment may offer an examiner-independent method to quantify deficits in children with OFD or myofunctional disorders and to grade the severity of the disease. Future studies should further examine the correlation between clinical measures and the Q-Motor changes, the longitudinal behaviour of Q-Motor measures and the impact of therapeutic interventions on the measures proposed.

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## CONFLICT OF INTEREST

Dr. Reilmann is founding director and owner of the George-Huntington-Institute and QuantiMedis GmbH, which provides quantitative motor assessment in multicentre clinical trials to different sponsors. Glossomotography is protected by patent DE10309540A12004.09.16. The other authors have stated explicitly that there are no conflicts of interest in connection with this article.

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## SUPPORTING INFORMATION

Additional Supporting Information may be found online in the supporting information tab for this article.

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