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DIAGNOSTIC METHODS: Original Research

Is there a difference in response to manual cranial bone tissue assessment techniques between participants with cervical and/or temporomandibular complaints versus a control group?

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A B S T R A C T

Aim: Manual cranial bone tissue techniques (CBTT) are used by physiotherapists as a tool for assessment and treatment of complaints in the craniocervical, face, and head regions. The goal of this study was to determine whether CBTT were able to discriminate between subjects with cervical and/or temporomandibular disorders (TMD) and a healthy group. In addition, the inter-rater reliability when applying CBTT was also investigated.
Methods: A cross-sectional study was conducted and included 60 participants. Six standardized passive techniques were applied and judged for resistance, compliance, and sensory answer. In order to evaluate the inter-rater reliability of these techniques a cohort of participants was measured twice (by two evaluators) prospectively. A logistic regression model and Receiver Operating Curve (ROC) analyses were used to determining the discriminative validity of these techniques.
Results: Logistic regression identified a significant difference for five techniques for resistance and/or compliance and/or the sensory answer between the groups. Based on the Area Under the Curve (AUC) analysis, the discriminative ability of the temporal rotation to distinguish between the groups was fair to good (for resistance AUC = 0.7775 and for compliance AUC = 0.8065). The highest agreement between the two assessors was for the resistance with occipital compression (73%) technique.
Conclusion: This study highlights that some of the CBTT could be potentially useful in distinguishing subjects with cervical and/or TMD from healthy subjects. Inter-rater reliability was moderate. CBTT could be potentially integrated in the examination of participants with complaints in the craniofacial region.

1. Introduction

Cranial bone tissue techniques (CBTT) can be used to examine and treat patients with complaints in the craniocervical and head regions (Chaitow, 2005; Krützkamp et al., 2014; Von Piekartz, 2015). Each of the CBTT consists of a manual external pressure given to the cranial bones in a standardized direction. These manual techniques may be appropriate for treating different kinds of headache, tinnitus, craniofacial pain, vestibular disturbances, and other craniocervical related-conditions. However, until now, research looking at the effectiveness and assessment provided by CBTT has been limited (Jäkel and von Hauenschild, 2012; Krützkamp et al., 2014). Most of the evidence about influencing the cranium and its behaviour is described in growth studies performed in dental orthopaedics, plastic surgery and neurosurgery areas (Krützkamp et al., 2014; Opperman, 2000; Oudhof et al., 2001; Proffit et al., 2014; Robling et al., 2006). This evidence is useful but cannot be directly transferred to the techniques we use as physiotherapists/manual therapists. To the best of the authors' knowledge, there are no studies looking at CBTT effectiveness or at determining whether these techniques are useful when used as assessment techniques. Therefore, it is important that clinical research is performed to evaluate these techniques.

The effectiveness of these techniques lies in the complex interconnections between the cervical and craniofacial regions at different levels (e.g. neurological, biomechanical, and morphological). It is known that nociception of the head structures such as the

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cranial dura mater and the large cerebral arteries, and extracranial structures, such as the head, face, and neck muscles, may strongly influence the trigeminal system. (Schueler et al., 2013; Chichorro et al., 2017). Recently, it has been shown that noxious stimulation of the extracranial structures could increase meningeal blood flow and release of neuropeptides increasing meningeal nociception. This evidence may advocate for the effect of cranial manual therapy on complaints like headaches (Meßlinger et al., 2016; Schueler et al., 2013).

Evidence has shown that subjects with pain in the jaw and neck present with alterations in the craniofacial region (Olivo et al., 2010). Likewise the relation between the temporomandibular joint (TMJ) and headaches has been described (Conti et al., 2016). Other studies have described the relationship between the neck and the craniofacial region (Olivo et al., 2006) or the biomechanical relationship between the TMJ in relation to the head and/or cervical region (Matheus et al., 2009; Olivo et al., 2006; Von Piekartz and Hall, 2013; Von Piekartz et al., 2016). No study has been conducted concerning cranial bone tissue techniques and their relationship with the cervical region and/or temporomandibular region. To the best of our knowledge, this is the first study that examines CBTT in a clinical research study. The assessment of dysfunctions with manual CBTT aims at the reproduction of the patient's symptoms by observing the resistance, compliance, and the sensory answer which will be described in detail in the methods section.

Therefore, the main purpose of this explorative study was to provide validity evidence for these techniques when used as assessment techniques. Specifically, we wanted to determine if CBTTs or a cluster of CBTT were able to discriminate between subjects with cervical pain/temporomandibular disorders-TMD and a healthy group. The secondary goals of this study were to determine the intra- and inter-rater reliability when applying manual CBTT at detecting clinical parameters such as resistance, and compliance between a control and a TMD/cervical pain group.

2. Methods

2.1. Design and general overview

This study was a cross sectional and a reliability study. Subjects with TMD and/or cervical dysfunction were compared with healthy subjects while applying manual CBTT as an assessment. Sample size was determined by the rule of thumb stated by Stevens (2002) and Harrell and colleagues (Harrell et al., 1996) using logistic regression for the analysis. According to this rule, each covariate used in the model (that is: each type of technique applied, age, and gender of participants) needed to have between 15 and 20 participants. Therefore 60 participants were targeted for this study.

Two investigators, both physical therapists with recognized education in musculoskeletal therapy (International Federation of Orthopaedic Manual Therapy –IFOMPT), a certificate in management of cranial facial-mandibular pain (Cranial Facial Therapy Academy-CRAFTA), and with more than eight years of experience with CBTT, examined the two groups (TMD and/or cervical dysfunction versus a healthy group with no complaints) with six standardized passive techniques on the cranium. Each of the manual cranial bone tissue techniques was judged with three parameters: the resistance, the compliance and the sensory answer (described below). Two of them were determined by the investigators (resistance and compliance) and were dichotomized as normal or abnormal, the third one was scored by the participant (sensory answer).

Resistance. Resistance is described according to the principles of Maitland in 1986 (Maitland, 2013). During manual pressure on healthy tissue in a body structure, a first resistance (R1) occurs. This resistance exponentially increases until the limit (R2) (Hengeveld and Banks, 2013). During this study, the investigators needed to determine whether there was an abnormal resistance of the cranial tissues while performing passive movements on the cranium. For example, they could assess whether R1 started earlier during the range, or that R1-R2 distance was smaller than normal, or the limit (R2) was reduced (Von Piekartz, 2015).

Compliance. Compliance is the capability of adaptation of structures to external forces by absorbing these forces or leading to shape and form changes (McCarthy and Frassica, 2014). Compliance seems to play an important role in the growth of cranial tissue which is based on the stress-transducer system mechanisms between bone tissues (Oudhof et al., 2001; Gabutti and Draper-Rodi, 2014; Proffit et al., 2014; Robling et al., 2006). Cranial compliance changes may be due to the adaptation of the cranial tissue itself, but also to the tissues inside the cranium, such as intracranial fluid blood, cerebrospinal fluid, brain, connective, and neuromeningeal tissue (Heisey and Adams, 1993; Von Piekartz, 2015). The investigators in this study had to judge whether there was a counter reaction of (bone) tissue after a passive movement of the cranium. This is also called “rebounce“.

Sensory answer. Every subjective experience during the cranial manual intervention, such as pain, vertigo, tinnitus, sensory awareness of an extremity among others could be defined as a sensory answer (Von Piekartz, 2015). The investigators had to ask the volunteers if they experienced any sensory answer. The participants scored their sensory answer using a Coloured Analogue Scale (CAS). This tool was developed for determining the intensity of headache in people of all ages (McGrath et al., 1996). At one side of the scale the participant can register their current level of complaints on a coloured line which differs in color, area and length. This side corresponds directly with the other side where there is a numerical rating scale from 1 to 10 cm (Bulloch et al., 2009; McGrath et al., 1996). This tool has demonstrated good reliability (McGrath et al., 1996).

The six CBTTs will be summarised later in the procedure section of this manuscript, and are described in detail in Appendix 1. The CBTTs used in this study are part of the education program of the Cranial Facial Therapy Academy (CRAFTA). They are based on challenging cranial bone tension and stress; and are also implemented in the fields of orthodontics, plastic and neuro surgery and follow functional/biological/and pragmatic models (Chaitow, 2005). CBTTs do not pretend to mobilize the sutures of the skull or influence the craniosacral rhythm (von Piekartz, 2007; 2015).

2.1.1. Reliability design

To determine which data from which investigator would be used for the rest of the analyses for this study, an intra-rater reliability study was done. In order to evaluate the inter-rater reliability of the six cranial techniques a cohort of participants was measured twice (by two evaluators) prospectively.

The protocol of this study followed the Declaration of Helsinki and was approved by the ethics committee of the University of Applied Sciences Osnabrück, Germany (WiSO MS-MP-WS 1617–05).

2.2. Subjects

Participants were recruited in a general private physical therapy practise of the principal investigator (PI) in the east of Netherlands. Prior to participation, participants gave written informed consent. To be included in this study, the following inclusion criteria applied: the participant would 1) need to read and understand the informed consent; 2) be over 18 years of age, and; 3) be able to lie down for 15 min. Participants were excluded from the study for the following reasons: 1) if they had had surgery, head trauma, or pathology of the central nervous system which occurred in the last year, or; 2) had central sensitization as evaluated by the Dutch-CSI questionnaire.

To make a clear distinction between cases (i.e. subjects with pain) and control subjects, subjects classified as "healthy subjects" should not have had chronic complaints and should have been pain-free in the head and neck region for at least a year before starting of the study. Short, insignificant periods of headache, neck pain, or toothache (for dental reasons, for a maximum of one week) were allowed. Participants complaining of headache episodes whose frequency was more than once per month were excluded from the healthy group. Participants with neck pain were excluded from the control group if it had required treatment in the last year (Hall and Robinson, 2004).

2.3. Measurements

Prior to the study, three questionnaires were used to include or exclude participants and to classify them into a cervical/TMD group or a healthy group. In addition, a subjective assessment and clinical tests of the cervical spine and the TMD were done.

2.3.1. Questionnaires

1) **The Dutch Central Sensitization Inventory.** The cut-off score for determining central sensitization is determined to be 40 out of 100. This questionnaire is a reliable tool and has a good construct valididity for determining central sensitization (Kregel et al., 2015; Mayer et al., 2012; Neblett et al., 2016).
2) **The Neck Disability Index-Dutch Language Version.** This questionnaire is the most commonly used questionnaire to determine the influence of neck complaints on activities of daily life. It is validated and has a high reliability (Vernon, 2008; Vos et al., 2006).
3) **The TMD pain screener.** This tool is recommended by the Diagnostic Criteria/Temporomandibular Dysfunctions (DC/TMD) (Ohrbach et al., 2016; Schiffman et al., 2014). The TMD pain screener is a reliable and valid instrument for any pain-related TMD (Schiffman et al., 2014).

Subjective Assessment: A brief subjective assessment was done to determine complaints in the last year in addition to duration, quality, and location of the complaints. Subjects were asked about trauma in the craniofacial region, and were assessed for limited movements of the jaw and the cervical spine, and for joint sounds as described in the study of Visscher et al. (2000). Participants drew their complaints on a body chart (Hengeveld and Banks, 2013). Intensity of complaints was measured with a Coloured Analogue Scale (CAS). This scale has a high reproducibility (Bulloch et al., 2009; McGrath et al., 1996).

Cervical spine tests: For the cervical spine the following tests were included: 1) active range of motion (AROM) of the neck in all directions (Hengeveld and Banks, 2013; Visscher et al., 2000). Degrees were measured with a single inclinometer. This device is reliable for measuring AROM (Hole et al., 1995; Williams et al., 2010); 2) the flexion-rotation test (for experienced examiners: sensitivity = 0.90, specificity = 0.88) (Hall et al., 2004; 2008); 3) Passive Accessory Intervertebral Movements (Hengeveld and Banks, 2013). Hall and colleagues found a good reliability (PABAK$\geq$ 0.70) for the upper three segments in headache patients; 4) Palpation of the neck structures. Pain was determined verbally on a scale from zero to ten. It is known that even participants without complaints may have a high intensity of pain on palpatio;n therefore, a cut-off value of 2 was chosen to distinguish between pain and no pain (Visscher et al., 2000). For the maximum verbal-scores as a dependant variable, a sensitivity of 52.3 and a specificity of 76.3 have been reported (Visscher et al., 2000).

TMJ Tests: In order to determine subjects with TMD, the criteria of the DC/TMD (Axis I) were used as described by Schiffman et al. (2014). The examination involved movements of the jaw and palpation of the chewing musculature and the TMJ.

3. Study procedures

3.1. Therapist training

Before the actual data collection, two experienced manual and craniofacial therapists were trained in the application of the manual CBTT. They received an instruction film with the correct cranial techniques. The techniques that were used have been developed by the Cranio Facial Therapy Academy (CRAFTA®). This is an eclectic, pragmatic approach of examination and treatment with CBTT by searching for functions/dysfunctions which can be related to the clinical pattern of the craniocervical and craniofacial region (Von Piekartz, 2015). Each of the CBTTs consists of a manual external pressure applied to the cranial bones. The pressure can vary from a force applied in the direction of compression or distraction, in transversal lateral-medial, in longitudinal caudal-cranial or in a rotational direction. Pressure can be applied to one cranial bone or to two related cranial bones. There are six standard techniques for examination described: a. Compression of occipital/frontal bone; b. Compression of occipital/frontal bone diagonally (right occiput/left frontal); c. Compression of occipital/frontal bone diagonally (left occiput/right frontal); d. Compression of occiput; e. Anterior rotation of both temporal bones; and f. Transversal movement of the sphenoid against the occipital bone.

A detailed explanation of the techniques is described in Appendix 1. Each of these techniques can be varied in direction, rhythm and force or the sequence of bone pressure can be changed, similar to the variations in techniques as described in the Maitland approach of manual therapy (Hengeveld and Banks, 2013). Techniques were practised and calibrated for two hours with an experienced clinician. To standardize the techniques, the applied force was measured with a dynamometer (MicroFET2 from Biometrics Motion bv.). To make sure of feeling the quality of the compliance, the force magnitude was set at seven kilograms. It is known that a large force is needed to have an influence on the compliance of the cranial bones (Heifetz and Weiss, 1981; Seimetz et al., 2012). At the start of each examination day, the force applied by investigators 1 and 2 was calibrated again on the dynamometer and with the experienced clinician. Techniques were practiced until a good agreement ($\geq$80%) was achieved between therapists and the clinician.

3.2. Application of the techniques

The participants were told to arrive at a predefined day and time. During the examination, the participant was asked to lie down on a table in a standardized position (legs extended next to each other, arms in front of the belly, no pillow was allowed). Both assessors were blinded to the group the participant belonged to. Investigator 1 carried out the sequence of cranium techniques which were: compression occiput/frontal, compression occiput right side/frontal left side diagonally, compression occiput left side/frontal right side diagonally, compression occiput, rotation temporal, and transversal lateral sphenoid/occiput (Appendix 1). Each technique was repeated three times in this fixed sequence. Starting with the occipital/frontal technique gave the assessors a good impression of the compliance of the cranium of that specific participant. The order of techniques was chosen to minimize changes in hand positions for convenience of the examiner and to prevent giving an extra stimulus to the participant.

The participant had to score on a CAS the amount of sensory answer experienced for each technique. At the same time, investigator 1 recorded the quality of resistance and compliance (normal/abnormal) felt when applying each one of these techniques on a score card. After all of the six techniques had been performed, the participant waited in the clinic for half an hour. Investigator 1 then examined the next participant with the same procedure.

After half an hour of rest, the first participant lay down again, and investigator 2 examined them with the same sequence of techniques as investigator 1, and the same procedures were repeated. After being evaluated by both assessors, the participant was thanked and informed about the possibility of contacting the principal investigator in case of any questions. Investigator 3 did the logistics, welcomed the participants and led them to the rooms. After each session, the score cards were deposited in a blind box. The score cards were collected and a fourth investigator anonymized the data. Investigator 3 did the analysis. (Fig. 1). After four weeks, a cohort of 20 participants (ten for each evaluator) was measured again to evaluate the intra-rater reliability of the six cranial techniques.

4. Statistical analysis

Data were analysed descriptively. Means and SD were used to describe continuous data such as age and sensory answer (CAS). Frequencies and percentages were used for variables that were dichotomous or categorical such as the description of resistance and compliance between groups. Significance of age differences between the cervical/TMD group and the healthy group was calculated with an independent sample *t*-test. To determine whether there were differences in the proportion of subjects with pain (i.e. neck pain and TMD) and healthy subjects in the response to each of the cranial techniques on resistance, several Chi square analyses were conducted. After that, a purposeful modeling strategy using a logistic regression model was followed. First, a single logistic regression was done separately to examine the association between each independent variable (e.g. response to each type of the technique applied, age and gender of participant) and groups involved (TMD/cervical pain and healthy subjects) (dependent variable). The dependent variable was whether the subject had TMD/cervical pain or not (yes/no). The independent variables that were significant at *p-value* ≤ 0.20 were entered into a multiple logistic regression model. Variables were entered in a forward stepwise fashion. After the addition of each one of the independent variables, a Receiver Operating Characteristic (ROC) curve analysis was run to determine the discriminative ability of the model for distinguishing between subjects with pain and those who were healthy. Therefore, a series of receiver operating characteristic (ROC) curve analyses were performed. These ROC analyses were compared and the best model with the highest Area Under the Curve (AUC) was chosen as the final model. The ROC analysis provides the AUC which is an overall measure of discrimination; that is, it identifies how accurately the model can correctly classify subjects as healthy or belonging to the pain groups. The most useful model can then be identified by comparing the AUC, where models with higher AUC values are better at discriminating between groups (Cleves and Rock, 2002; Fritz and Wainner, 2001). The following guidelines are recommended for interpreting the discrimination performance of a model from the AUC: excellent discrimination (AUC = 0.90 to 1.0); good discrimination (AUC = 0.80 to 0.90); fair discrimination (AUC = 0.70 to 0.80); poor discrimination (AUC = 0.60 to 0.70); and discrimination no better than chance (AUC $\leq$0.50) (Hanley & McNeil, 1982). After running the multiple logistic regression model, variables that were not statistically significant at a *p-value*<0.05 were removed from the model. The final model included all variables that were statistically significant at $p < 0.05$. All data analyses were performed using STATA software and guided by a statistical expert.

Initially we planned to use Kappa (κ) statistics for assessing the agreement between the 2 investigators to determine the outcomes of interest for each one of the techniques (resistanceand compliance). However, it was not possible to get adequate results with the use of kappa. It is well known that there is the so-called “kappa paradox” where high agreement can accompany low kappa scores, and this might have happened in our study (Feinstein and Cicchetti, 1990). Therefore, we decided to report the percentage of agreement between assessors. Kappa (κ) was also intended for usein calculating the intra-rater reliability while performing the cranial techniques, but this was also not possible for the aforementioned reasons. Instead, percentage of agreement was calculated, as well as a measure of intra-rater reliability. The highest intra-rater agreement would reveal the assessor who had better reliability in his judgments, and thus his data would be used for further analyses.

5. Results

5.1. Participants

The cervical/TMD group consisted of 30 participants (33% male (n = 10); 67% female (n = 20)) with a mean age of 45.13 (SD ± 14.41). The healthy group involved 30 participants (57% male (n = 17), 43% female (n = 13)) with a mean age of 41.97 (SD ± 18.98). No significant age difference was found between the cervical/TMD group and the healthy group (p = 0.470). Subjects included in the cervical/TMD group were subdivided as follows: pure cervical complaints (n = 18), pure TMD complaints (n = 6) and a mixed group with both complaints (n = 6). No significant age difference was found among the three cervical/TMD subgroups was found (p = 0.112).

5.2. Intra-rater reliability of manual CBTT

The intra-rater agreement was acceptable for most of the CBTTs. The occipital/frontal compression and the occipital compression alone techniques had an agreement of 70% or higher for both examiners and for both resistance and compliance outcomes. The rest of the techniques had variable intra-rater agreement for examiner one and two on resistance or compliance. For example, examiner 1 had a good intra-rater agreement for occipital/frontal diagonally from right occiput to left frontal (for both resistance and compliance) and good intra-rater agreement for temporal rotation on resistance and occipital/sphenoid transversal lateral on compliance. On the other hand, examiner 2 had good intra-rater agreement for occipital/frontal diagonally from left occiput to right frontal and the occipital/sphenoid transversal lateral (for both resistance and compliance). See Table 1 for details of intra-rater agreement.

5.3. Inter-rater reliability

Agreement between examiners was the best for resistance with occipital compression (73%), followed by occipital/frontal compression (straight and diagonal from occiput right to frontal left) (67%) techniques. For compliance, only compression of the occipital bone had an acceptable agreement between examiners (70%). Rotation of both temporal bones to anterior and the transversal lateral movement of the occiput versus sphenoid had the lowest agreement between examiners (Table 2).

Table 1
Intra-rater agreement (for examiner 1 and 2) on resistance and compliance for all the 6 techniques.

Technique	Evaluator 1		Evaluator 2	
	Resistance (Agreement in %)	Compliance (Agreement in %)	Resistance (Agreement in %)	Compliance (Agreement in %)
O/F	80%	70%	80%	80%
O/F left occiput	60%	30%	70%	70%
O/F right occiput	100%	70%	50%	50%
O	80%	80%	70%	80%
Te/Te	80%	50%	50%	50%
O/S	40%	70%	70%	70%

Abbreviations: O/F= Occipital/Frontal compression; O/F right and left occiput = Occipital/Frontal diagonally from right/left occiput to left/right frontal; Compression occipital/frontal bone diagonally (left occiput/right frontal) Te/Te= Temporal rotation; O/S= Occipital/Sphenoid transversal lateral; O=Occiput compression.

Table 2
Percentage of inter-rater agreement (between examiners) for all of the 6 CBTT techniques.

Technique	Resistance (Agreement in %)	Compliance (Agreement in %)
O/F	66,7%	60,0%
O/F left occiput	58,3%	53,3%
O/F right occiput	66,7%	60,0%
O	73,3%	70,0%
Te/Te	55,0%	53,3%
O/S	53,3%	51,7%

Abbreviations: O/F= Occipital/Frontal compression; O/F right and left occiput = Occipital/Frontal diagonally from right occiput to left frontal; Te/Te= Temporal rotation; O/S= Occipital/Sphenoid transversal lateral; O=Occiput compression.

5.4. Discriminative ability of the manual CBTT to distinguish between subjects with cervical/TMD group or a healthy group

5.4.1. Univariate analyses

The univariate analysis (Chi-square) indicated statistically significant differences (≤ 0.2) between subjects with pain and healthy subjects for seven variables. It involved two techniques for the resistance (i.e. temporal rotation and occiput/sphenoid transversal lateral), two for the compliance (i.e. occiput compression and temporal rotation) and three for the sensory answer (i.e. occiput/frontal straight compression and diagonally from the right occiput to left frontal and temporal rotation) (see Table 3 for results from the univariate analyses). All abnormal values for the sensory answer were scored in the pain group. The results of the single logistic regression analysis supported the Chi-square results obtaining similar results. The results of the logistic regression for resistance and compliance responses are listed in Table 3. All results from the temporal rotation technique (i.e. resistance, compliance, sensory answer) were demonstrated to significantly discriminate between groups ($p \leq 0.05$) in the univariate analyses.

5.5. Multivariable analyses

To determine the most optimal model to distinguish between subjects with cervical/TMD pain and the healthy group, all seven significant variables from the univariate analyses (i.e. for resistance temporal rotation and transversal lateral of occiput/sphenoid; for compliance occiput compression and temporal rotation; for the sensory answer compression of occiput/frontal straight and diagonally (from the right occiput to left frontal), and temporal rotation), in addition to age and gender of participants, were added to a multiple logistic regression analysis using a forward stepwise method. After the addition of each one of the variables, a ROC analysis was performed for each one of the models. Each model was graphically represented by a ROC curve, so they could be compared. The AUC was used to determine which one of the models and variables included in the model distinguished between subjects with cervical/TMD pain and the healthy group and the most optimal model could be determined. The AUC values ranged from 0.69 to 0.80. The highest scores for the AUC were found when the responses from the resistance and compliance of the temporal technique were included in the model; these scored fair (AUC = 0.7775 for resistance and 0.8065 compliance respectively) (Fig. 2a). Afterwards, combinations of the obtained ROC curves were made in order to find the combination which led to the highest AUC. None of the combinations led to a higher value of the AUC when compared to only including the separate responses of the temporal technique, except when combining all temporal outcomes (resistance, compliance and sensory answer). This model increased to a value of 0.8160 (Fig. 2b, highlighted in orange), which means that the combination of these techniques discriminated well between a group with pain and a group without complaints.

6. Discussion

This study explored whether CBTT were able to distinguish between subjects with cervical/TMD and healthy subjects. Also, an intra-rater and inter-rater reliability study was performed to examine the agreement within and between two experienced clinical examiners using these techniques.

6.1. Discriminative ability of the manual CBTT

The results of this study highlighted that the temporal rotation was able to discriminate well between the cervical/TMD group and the healthy group. The discriminative ability of this technique could be explained by the fact that the temporomandibular joint (TMJ) is closely related to the temporal bone.

It was expected that other techniques would discriminate as well between a group with and without complaints as we do see in clinical practice. However, we could not observe this discrimination in our study. Although several studies have reported a relation between the cervical spine and the TMD (Conti et al., 2016; Matheus et al., 2009; Olivo et al., 2006 & 2010; von Piekartz and Hall., 2013; Von Piekartz et al., 2016), to the best of our knowledge, no study has been performed to test the ability of the manual CBTT to distinguish between a group of subjects with cervical/TMD group and a healthy group. Therefore it is difficult to compare the results obtained in the present study.

6.2. Intra-rater and inter-rater reliability of the manual CBTT

The intra- and inter-rater reliability were found to be moderate. The agreement between reviewers for the compression of the occiput technique met the criterion of 70% percent agreement, and two occipital/frontal techniques were near our cut-off (67%). The poor results for the occipital/sphenoidal technique might be

Table 3
Single logistic regression and chi-square results for each one of the CBTT techniques for the three parameters of interest (resistance, compliance, and sensory answer).

Technique	Resistance				Compliance				Sensory answer			
	p-value (chi- square)	Odds Ratio	CI 95%	p-value (single logistic regres.)	p-value (chi-square)	Odds Ratio	CI 95%	p-value (single logistic regres)	p-value (chi-square)	Odds Ratio	CI 95%	p-value (single logistic regres)
O/F	1.000	1.00	0.31, 3.14	1.000	0.426	0.65	0.22, 1.86	0.427	0.150*	2.35	1.06, 5.14	0.034**
O/F right occiput	0.448	1.80	0.38, 8.32	0.452	0.519	1.52	0.42, 5.47	0.520	0.076*	2.44	0.90, 6.58	0.078*
O/F left occiput	0.273	0.54	0.18, 1.62	0.276	0.432	0.66	0.23, 1.85	0.433	0.313	2.86	1.10, 7.36	0.030**
O	0.640	0.64	0.09, 4.15	0.643	0.095*	0.30	0.07, 1.29	0.107*	0.313	2.49	0.95, 6.56	0.064*
Te/Te	0.001**	7.42	2.07, 26.55	0.002 **	0.000**	8.49	2.37, 30.46	0.001 **	0.036**	2.23	1.31, 3.80	0.003**
O/S	0.152	2.31	0.72, 7.40	0.157*	0.243	2.00	0.61, 6.46	0.247	0.688	1.49	1.0, 2.20	0.049

Abbreviations: O/F= Occipital/Frontal compression; O/F right and left occiput = Occipital/Frontal diagonally from right occiput to left frontal; Te/Te= Temporal rotation; O/S= Occipital/Sphenoid transversal lateral; O=Occiput compression R = Resistance; C=Compliance.

explained by the fact that it is hard to prevent the head from moving during transversal lateral movement. This might occur at the expense of the sensitivity of the palpating hand.

Also, the temporal technique had poor agreement. The disadvantage of this technique is the early start and the inclination of the resistance. This makes it hard to determine the behaviour of the resistance. Another prominent finding was the difference in the diagonal techniques of occipital/frontal. A possible explanation for the difference in scores might be dominant hand of the investigators. One investigator was left-handed, and the other right-handed. Better sensory perception can be developed with the dominant hand; this might have influenced the results. In clinical practice this will mean that it is important not only to search for a change in outcomes, but also for a change in symptoms, and to combine these findings when determining the response to these techniques.

Although the inter-rater agreement was low, as expected, in some of the techniques, the intra-rater agreement was appropriate for most of the techniques. This is important for clinical practice since evaluation and treatment are generally performed by the same clinician. Therefore, these results add to the validity evidence of these techniques in the clinical context.

For future studies it will be important that assessors standardize and practise these techniques before they are applied to patients, to improve consistency in the results.

6.3. Limitations

It is important to acknowledge that this project has some limitations that we discuss below:

***Application without a trial*:** Most participants were not familiar with cranial techniques. If a participant was stressed, felt afraid of the treatment, or was simply not able to relax, this could be expressed in motor control changes in, for example, the neck and/or masticatory musculature. This could have changed the tension in the musculature which might have influenced all outcomes. In future studies this can be avoided by giving a trial treatment to get the participant accustomed to the techniques and the examination situation, or by including questionnaires about fear and beliefs to get insights into this factor.

The blinding of the assessors: both *assessors* were experienced craniofacial physical therapists and were familiar with these types of patients. Therefore they may have recognized orofacial-cervical dysfunction during the examination, although they were blinded to group status.

Diversity in groups of participants: Participants with large variability of age and gender (without a significant difference) were included in this study. This makes the groups less homogenous.

Complaints of the participants: In this study we examined a group with TMD/cervical pain. If other groups with different complaints would have included, perhaps other results could have emerged.

6.4. Clinical implications

In clinical practice the examination of a patient is far more comprehensive and the hypothesis of the patients' problem will be made and adapted step-wise by the clinician. This means that the interpretation of the results of all six CBTTs in relation to the patients complaints may lead to a pattern recognition and a specific clinical diagnosis (Higgs and Jones, 2000; Hengeveld and Banks, 2013). This implies that outcomes of these techniques such as resistance, compliance, and sensory answer are only one factor, and therefore it might be helpful to cluster these techniques with other diagnostic/ examination tools (for example: CBTT with cervical ROM, main

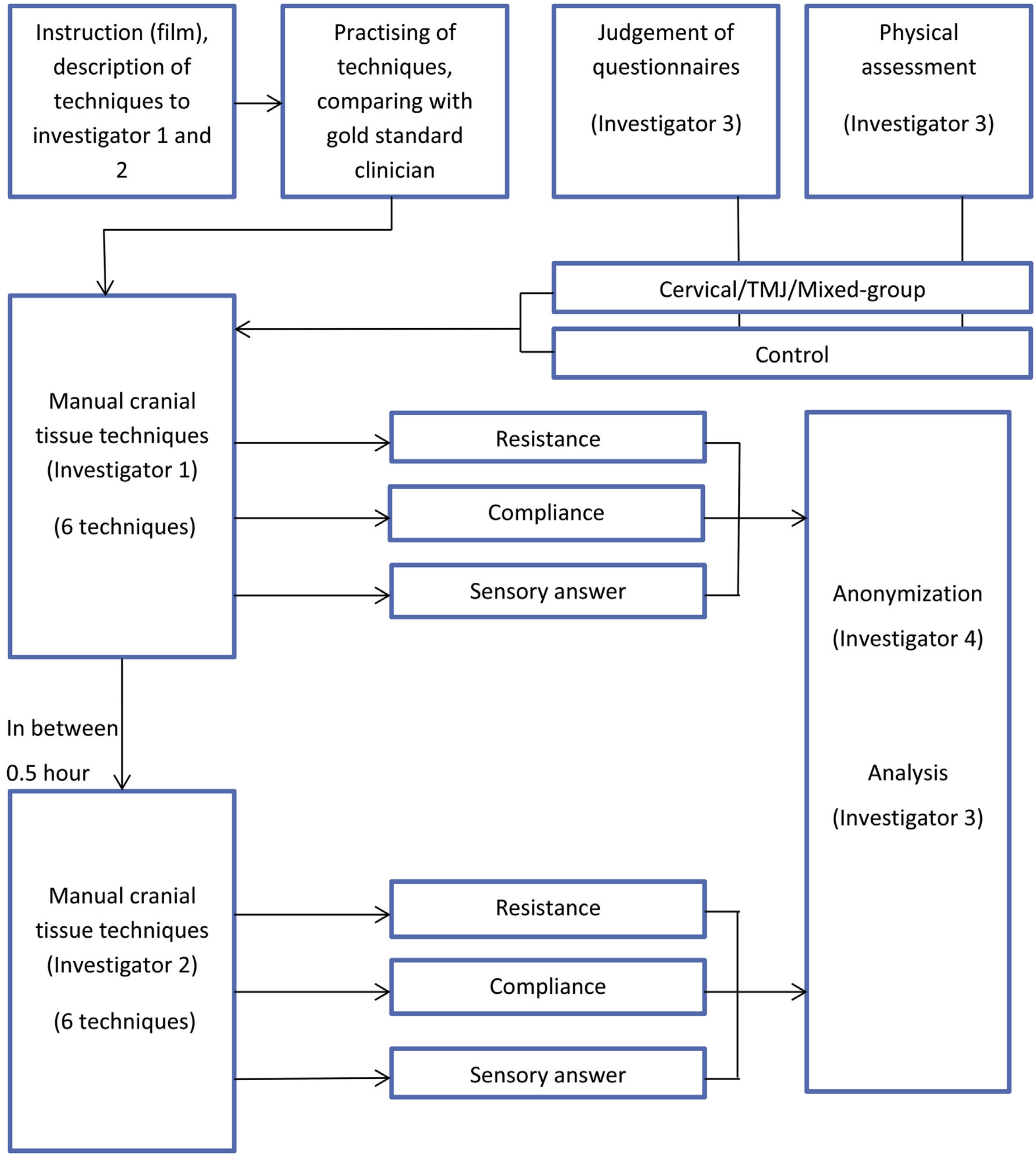
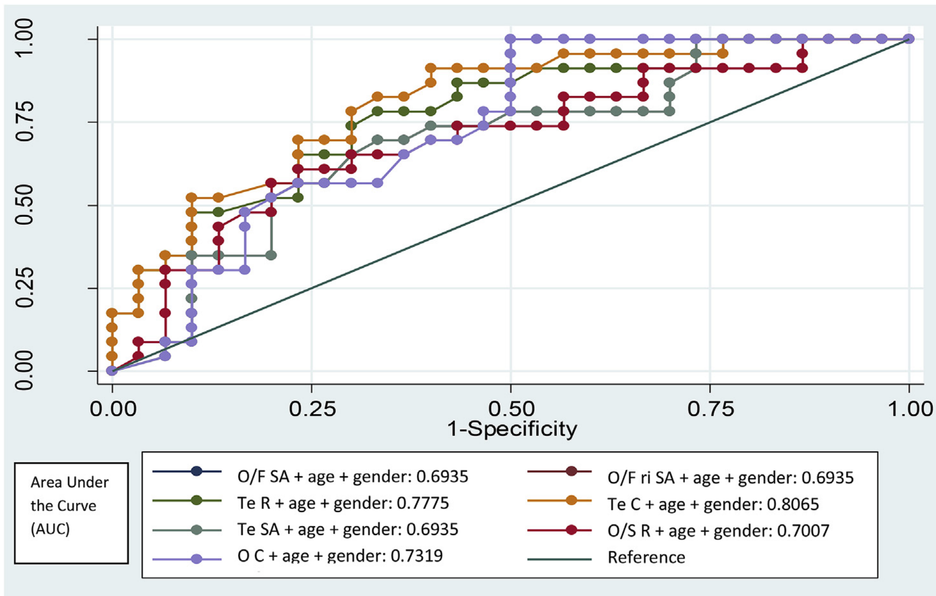


Fig. 1. Study procedure.
Investigator 1 and 2: Experienced craniofacial therapists who were blinded for the groups; Investigator 3: Principal investigator; Investigator 4: Anonymized the data.

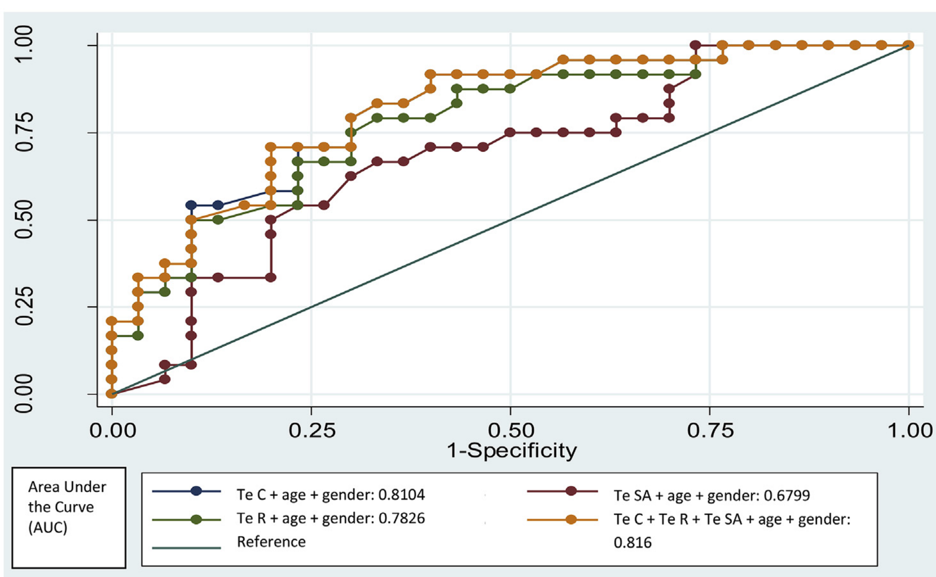
physiological TMJ movements or neurodynamic testing) as it is known that clustering can increase the diagnostic value and can help defining diagnoses for the clinical use (De Hertogh et al., 2007).

6.5. For clinical practice is useful to know that:

- The intra-rater reliability was acceptable for both assessors in most of the CBTTs. This means that in practical situations CBTTs have an acceptable value during reassessment and treatment performed by the same clinician.
- The temporal bone technique had a high discriminative ability to distinguish between the investigated groups during this study; all other techniques were less discriminative. Therefore it is highly recommendable in clinical practice to cluster tests and combine them with a hypothetically driven clinical reasoning approach. Manual CBTT should not stand alone in decision making about diagnosis and treatment possibilities in these types of patients.
- The sensory answers of three techniques (i.e. Occipital/Frontal straight and diagonally from the right occiput to the left frontal bone and temporal rotation) were able to discriminate between groups. Therefore involvement of the cranium has to be tested by trial treatments and reassessment of all found impairments and disabilities, including the sensory answer of the patient.
- Techniques of the temporal bone were significant for participants with TMD. These techniques can be used for screening



(a)



(b)

Fig. 2. a ROC of all individual cranial bone tissue techniques (CBTT).
Each curve displays the discriminative ability of each of the individual cranial bone tissue techniques (CBTT). A higher AUC represents better discriminative ability to distinguish between healthy and subjects with neck and jaw complaints ($p \leq 0.20$) Abbreviations: O/F= Occipital/Frontal compression; O/F ri = Occipital/Frontal diagonally from right occiput to left frontal; Te= Temporal rotation; O/S= Occipital/Sphenoid transversal lateral; O=Occiput compression R = Resistance; C=Compliance; SA= Sensory Answer. **b**: **ROC of combined CBTT of temporal resistance, compliance and sensory answer.**
Each curve displays the area under the curve when all significant techniques ($p \leq 0.05$) plus age and gender are combined in one model. Abbreviations: Te= Temporal rotation; C = compliance; R = Resistance; SA= Sensory answer.

patients with TMD but also as treatment techniques. Cranial techniques of the temporal bone might have an influence on patients with TMD and therefore they are of importance to take into consideration in screening and in (trial) treatments.

7. Conclusion

The results of this cross sectional study highlight that some of the CBTT could be potentially useful in distinguishing subjects with cervical pain and/or TMD from healthy subjects. This indicates that cranial bone tissues may behave differently in subjects with pain. Inter-rater reliability was acceptable for some of the techniques and intra-rater agreement was acceptable for most of them. CBTT could be potentially integrated in the examination of participants with complaints in the cranio-facial region. This study offers only preliminary evidence and more research investigating these techniques is warranted.

Conflicts of interest

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors. No conflict of interest is declared. The study was approved by the Research Ethics Commission of the University of Applied Science Osnabrück (WiSO MS-MP-WS 1617—05).

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Appendix. The six Cranial Bone Tissue Techniques (CBTT) which are used during the study

1a. Compression occipital/frontal bone: The left hand grasps the frontal bone with the lower arm perpendicular to the cranium on the frontal bone. The right hand grasps the occiput. Both hands move towards each other (approximation).

1b. The same technique as described in 1a, but now there is a diagonal compression from the occipital and frontal bones (right occiput/left frontal);

1c. The same technique as described in1a, but now the compression occurs diagonally in the other direction from the occipital and frontal bones (left occiput/right frontal)

1d. Compression of the occiput: the occiput rests on both the practitioner's hands; the hypothenars (edge of the hand) are moved in bilateral, transversal directions toward the midline; and in the meantime the ventral (between the thumb and palm) part of both hands push the central part of the occiput in the posterior-anterior direction

1e. Anterior rotation of the temporal bone: the index finger and thumb are located on the zygomatic arc; the middle finger contacts the external acoustic meatus, the ring finger and little finger are located on the mastoid, and both forearms are perpendicular to the temporal bone, executing an anterior rotation on both sides.

1f. Transversal movement of the sphenoid against the occipital bone: The right hand grasps the occiput, and the left thumb and left index finger are respectively placed on the right and left greater wing of the sphenoid bone. In this position, a transversal movement of the sphenoid from left towards the right and in reverse, is initiated.

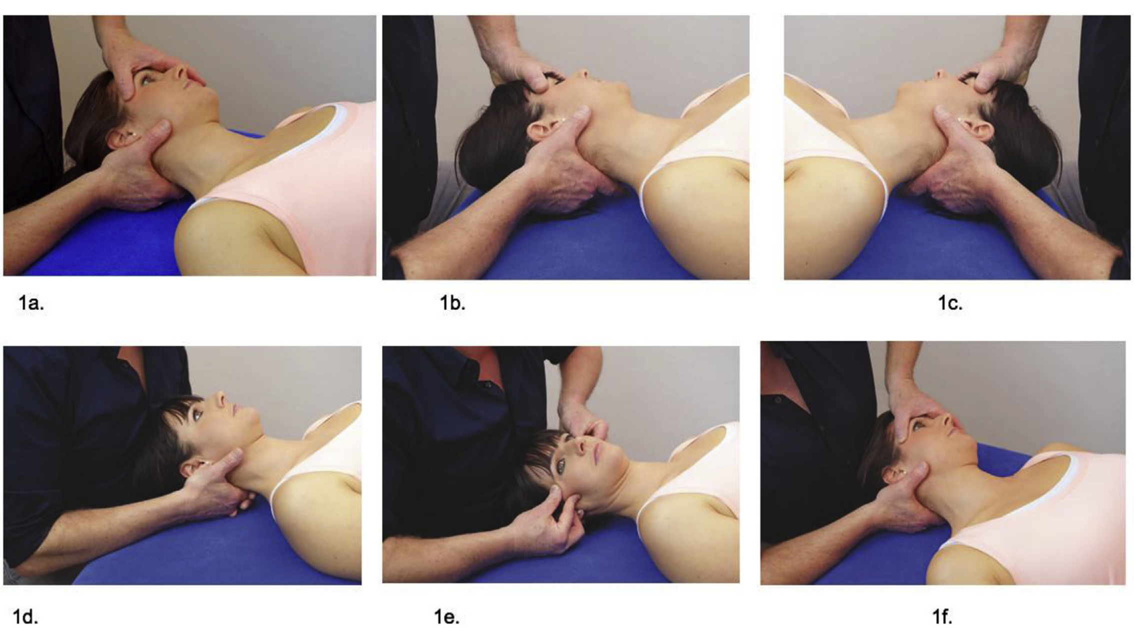

Fig. 1. : **The 6 cranial techniques which are performed during the study**.
a. Compression occipital/frontal bone; b. Compression occipital/frontal bone diagonally (right occiput/left frontal); c. Compression occipital/frontal bone diagonally (left occiput/right frontal); d. Compression occiput; e. Anterior rotation of both temporal bones; f. Transversal movement of the sphenoid against the occipital bone.

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