Musculoskeletal Science and Practice 52 (2021) 102321

Contents lists available at ScienceDirect

Musculoskeletal Science and Practice

journal homepage: www.elsevier.com/locate/msksp

Original article

The association between specific temporomandibular disorders and cervicogenic headache

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ARTICLE INFO

Keywords:
Temporomandibular joint
Cervicogenic headache
Neck pain
Cervical pain
Orofacial pain

ABSTRACT

Background and aims: Upper neck signs, symptoms and hypomobility have been shown to present with a higher prevalence in patients with temporomandibular disorders (TMDs). However, there is currently no evidence of an association between specific TMDs and cervicogenic headache (CGH). Therefore, the aim of this study was to evaluate the odds ratio and the relative risk of CGH in patients with specific TMDs.
Method: 116 participants, including 74 patients with TMD (pain-related/intraarticular/mixed TMD) and 42 healthy controls took part in this study. The TMD diagnosis was made by senior faculty members of the Dental School according to the Diagnostic Criteria for TMD, while the cervical diagnosis was made by a qualified senior physical therapist. The analysis comprised the evaluation of the odds ratio of CGH among patients with TMD and the relative risk (RR) for CGH during 14–24 months of follow-up.
Results: Significantly higher odds ratios of cervicogenic headache were found among pain-related and mixed TMD (12.17 and 10.76, respectively) versus healthy controls. During the 14–24 months of follow-up, there was no significant difference of relative risk for CGH among patients with TMD versus healthy controls.
Summary and conclusions: The results support a clear clinical association between painful TMD (pain-related and mixed TMD) and cervicogenic headache.

1. Introduction

Temporomandibular disorders (TMDs) consist of a group of conditions that cause pain and dysfunction in the masticatory muscles, the temporomandibular joint (TMJ), and their associated structures (Ghurye and McMillan, n. d.; Sonia and Ohrbach, 2018). TMDs are very common among the general population, and they comprise the most ubiquitous musculoskeletal conditions in the USA after chronic low back pain (Sonia and Ohrbach, 2018). The diagnostic system for these disorders is the so-called Diagnostic Criteria for TMD (DC/TMD) which provide valid and reliable characteristics of common TMDs for clinical and research purposes (Schiffman et al., 2014). According to epidemiological studies, approximately 10% of the adult population has TMDs, and the majority of them are women aged 20–40 years (Sonia and Ohrbach, 2018).

After 80–90 years of debating the notion of headache originating from the cervical region among different study groups, Sjasstad et al. established diagnostic criteria for cervicogenic headache (CGH) (Sjaastad et al., 1998). The International Headache Society recognizes CGH as a specific secondary headache (Olesen and Steiner, 2004), although the diagnostic criteria it supports differ from those of the CGH International Study Group which emphasize reduced neck range of motion, mechanical provocation, unilateral headache dominancy with ipsilateral neck/shoulder/arm and relief by local anesthetic block as the definitive criteria for CGH (Leone et al., 1998; Sjaastad, 1999; Sjaastad et al., 1998). Based on these valid and reliable criteria (Leone et al., 1998; Sjaastad, 1999; Van Suijlekom et al., 1999), the prevalence of CGH is 0.4–2.5% in the general population and 15–20% in patients with chronic headache (Antonaci and Sjaastad, 2011; Haldeman and Dagenais, 2001; Knackstedt et al., 2010; Sjaastad and Bakketeig, 2008). The cervical flexion-rotation test is a special physical test which assesses the rotatory mobility of the upper cervical spine, and it is considered as being highly valid and reliable in the diagnosis of CGH (Hall and Robinson, 2004; Hall et al., 2010a; Ogince et al., 2007; Satpute et al., 2019; Takasaki

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https://doi.org/10.1016/j.msksp.2021.102321
Received 28 April 2020; Received in revised form 19 December 2020; Accepted 8 January 2021
Available online 12 January 2021

et al., 2011).

Numerous reports in the literature have assessed specific measurable impairments of the cervical spine in patients with TMD (Armijo-Olivo et al., 2012; 2010; Armijo Olivo et al., 2006; Ballenberger et al., 2018; Greenbaum et al., 2017; Grondin et al., 2015; Olivo et al., 2010; 2006; von Piekartz et al., 2016). Two of those impairments, which are mainly related to the upper neck, were consistently present in patients with TMD: one was reduced mobility, as mainly expressed in the cervical flexion-rotation test (Ferreira et al., 2019; Greenbaum et al., 2017; Grondin et al., 2015; von Piekartz and Hall, 2013), and the other was poor muscular performance of the deep craniocervical flexors (Armijo-Olivo et al., 2012; 2010; Ferreira et al., 2019; von Piekartz et al., 2016). Despite this large volume of evidence to support the association of TMDs and cervical spine disorders, to the best of our knowledge, no study to date has assessed the association between specific TMDs and CGH. Therefore, the aim of this study is to evaluate such an association.

2. Materials and methods

Data for this study were collected from consecutive patients referred to the Orofacial Pain & TMD Clinic between May 2016 and June 2018. The objectives of the study were described to all the participants, and they all signed an informed consent form. This study was approved by the Institutional Review Board of the University on October 20, 2015 (Approval No.: 20,141,217 _09,280,561).

2.1. Inclusion/exclusion criteria

Based on the valid and reliable DC/TMD guidelines (List and Jensen, 2017; Ohrbach and Dworkin, 2016; Schiffman et al., 2014; Schiffman and Ohrbach, 2016), only consecutive patients who were registered at the university's Orofacial Pain & TMD Clinic during the data collection period, older than 18 years and younger than 75 years, diagnosed with pain-related TMD and/or intraarticular TMD and consented to take part were included in the study. This wide range in age well represents the overall population of TMD patients according to epidemiological data (Guarda-Nardini et al., 2017; Sonia and Ohrbach, 2018). Patients with degenerative joint disorders or any other form of TMD were excluded for better homogeneity of the cohort. The control group consisted of students and staff members from the schools of dental medicine or health professions and patients attending the Clinic who did not complain of dental pain or any TMD and did not meet the DC/TMD criteria (Schiffman et al., 2014). The members of the control group were free of any functional disorder of the masticatory system or complaints that could indicate its presence (such as pain or clicks during chewing) for the past year. The TMD assessment and classification as well as the screening of the healthy controls were performed independently of the cervical spine examination by four senior faculty members at the Orofacial Pain & TMD Clinic of The School of Dental Medicine, who had completed the DC/TMD Training and Calibration Course at the Department of Orofacial Pain and Jaw function, the Faculty of Odontology, Malmö University, Sweden (EW, SR, AEP, and PFR). The training process of all four examiners was carried out prior to this study according the DC-TMD protocol which had been shown to achieve high levels of inter-observer reliability (Schiffman et al., 2014; Schiffman and Ohrbach, 2016).

2.2. The diagnosis procedure of patients with TMD

Based on the updated DC-TMD (Schiffman et al., 2014), the main criteria for pain-related TMDs were: 1) Pain while chewing, pain modified by jaw movement, function, or parafunction; 2) Confirmation of pain in masticatory muscle(s) or in the TMJ as verified by the examiner, and 3) pain in the masticatory muscle(s) or TMJ with either muscle palpation or maximum opening as verified by the examiner. The main criteria for intraarticular TMDs were TMJ noises that were objectively audible or reported by the patient in the past or during the examination and click(s) that were objectively audible with mouth opening and closing and/or lateral movement.

1. Based upon the above principles, the following diagnoses were made according to the Axis I of the DC/TMD index:
   a. Local myalgia: Pain of muscle origin that is affected by jaw movement, function or parafunction (e.g., chewing gum or biting nails), and occurrence of the index pain only at the site of calibrated palpation of the temporal or masseter muscles. This diagnosis has very high sensitivity (0.9) and specificity (0.99) (Schiffman et al., 2014).
   b. Myofascial pain with referral (MFP): Same as for myalgia with referral of pain beyond the boundary of the muscle being palpated. This diagnosis has high sensitivity (0.86) and specificity (0.98) (Schiffman et al., 2014).
   c. Headache attributed to TMDs (HATMDs): Headache in the temple area that is affected by jaw movement, function or parafunction, and occurrence of the index pain only at the site of calibrated palpation on the temporal muscle. This diagnosis has very high sensitivity (0.89) and specificity (0.87) (Schiffman et al., 2014).
   d. Arthralgia: Pain of joint origin that is affected by jaw movement, function or parafunction, and occurrence of the index pain only at the site of calibrated palpation of the TMJs. This diagnosis has very high sensitivity (0.89) and specificity (0.98) (Schiffman et al., 2014).
   e. Disc displacement with reduction (DDWR): An intra-articular biomechanical disorder involving the condyle-disc complex. In the closed position, the disc is in an anterior position in relation to the condyle and becomes reduced during condylar translation (opening or eccentric condylar translation). The reduction is accompanied by a clicking, popping or snapping noise. The diagnosis is positive if the joint noise is detected with palpation during at least one of three repetitions of condylar translation. Without imaging (MRI), This diagnosis has low sensitivity (0.34) but high specificity (0.92) (Schiffman et al., 2014).
   f. Disc displacement without reduction (DDWOR): Same as for DDWR, but the disc is not reduced during condylar translation. DDWOR is associated with persistent limited range of mouth opening (<40 mm). Without imaging (MRI), this diagnosis has relatively moderate sensitivity (0.80) but very high specificity (0.97) (Schiffman et al., 2014).

Based on the DC-TMD manual (Schiffman et al., 2014), imaging is not required for diagnosis in population studies. However, the low sensitivity of the disc-related disorders in our study bears a high risk for false negative and is therefore a study limitation.

Based on these criteria, each TMD patient was categorized into one of the following three specific TMD groups: pain-related (at least one of the following: myalgia, MFP, HATMD, or arthralgia), intraarticular (at least one of the following: DDWR or DDWOR), or mixed (combined pain-related and intraarticular).

2.3. The diagnosis procedure of patients with CGH

Examination of the cervical spine for the diagnosis of CGH was carried out by a qualified senior physical therapist with a master's degree in musculoskeletal and sports physiotherapy and 15 years of clinical practice in manual therapy of the cervical spine. The examiner was blinded to the group assignment (i.e., study or control) and to the specific TMD diagnosis throughout the study.

To be diagnosed with CGH participants had to fulfill the following criteria:

1) Meet all of the diagnostic criteria suggested by the CGH International Study Group with the exception of diagnostic anesthetic blocks (Sjaastad et al., 1998)
2) Found as being positive in the cervical flexion-rotation test, which is specific for the mobility of the upper cervical spine. This test is considered positive when the range of motion is less than 32° to at least one of the sides due to either pain or stiffness (Hall and Robinson, 2004; Ogince et al., 2007).

2.3.1. Diagnostic criteria for CGH

Patients were diagnosed as having CGH according to the criteria proposed by Sjaastad (Sjaastad et al., 1998) and further modified by Antonaci and Sjaastad (2011) with the exception of diagnostic anesthetic blocks:

(1) Unilaterality of the head pain without side shift, starting in the upper posterior neck or occipital region, eventually spreading to the oculofrontotemporal area on the symptomatic side. This criterion was screened by specifically questioning the participants.

(2) Restricted neck range of motion. This criterion was screened by applying the Cervical Flexion-Rotation Test (described in detail in 2.3.2).

(3) Headache triggered by neck movement and/or sustained awkward positions. This criterion was screened by specifically questioning the participants.
(4) Headache elicited by external pressure over at least one of the upper cervical joints (C0-3). This criterion was screened by applying posteroanterior (PA) pressure to each of the upper three cervical motion segments (Hall et al., 2008). A positive response was determined by the eliciting of a relevant pain by at least one of the segments.
(5) Moderate-to-severe, non-throbbing and non-lancinating headache. This criterion was screened by questioning the participants.
(6) Headache frequency of at least 1 per week for a minimum of 3 months. This criterion was screened by specifically questioning the participants.

2.3.2. The cervical flexion-rotation test

Upper neck mobility was evaluated by the cervical flexion-rotation test with a cervical range of motion device (Performance Attainment Associates, Roseville, Minnesota, USA) (Fig. 1), one of the most reliable and sensitive tools for measuring cervical spine mobility (Prushansky et al., 2010). The cervical flexion-rotation test is considered the best clinical means of assessing relative isolated mobility of the upper cervical spine (Satpute et al., 2019). It has well-established high sensitivity and specificity in the diagnosis of CGH (Hall and Robinson, 2004; Hall et al, 2008, 2010a, 2010b; Takasaki et al., 2011).

For this examination, the cervical range of motion device was firmly attached to the head of the participant who lay supine on a treatment couch. The participant was asked to relax while the neck was moved by the examiner to the end of the cervical flexion range. In this flexed position, the head and neck were passively rotated as far as possible within comfortable limits of pain or physiological stiffness. End of range was determined either by firm resistance encountered by the therapist or the subject reporting the onset of pain, whichever came first. The intention was to measure range of motion irrespective of cause of limitation and in the least provocative manner in order to prevent potential exacerbation of symptoms. The range was recorded and repeated two more times to both sides, with 30s of rest in-between the tests. The average of the results of the three tests to each side represented the final score. The result was considered positive when the range of motion was less than 32° to at least one of the sides.

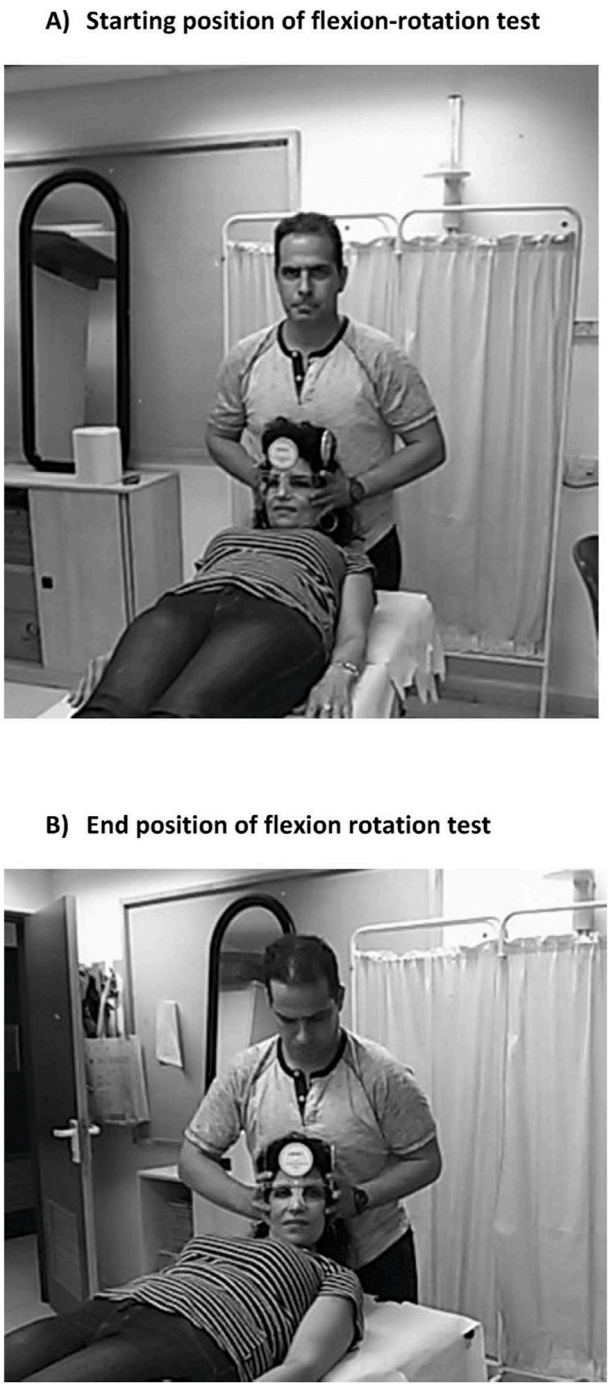

Fig. 1. The cervical flexion-rotation test.

2.4. Assessing the relative risk for CGH: the cohort study

Patients with TMD (pain related, intraarticular or mixed) but without CGH were assigned to the TMD group. They were also followed up for 14–24 months. These patients were monitored by a telephone call every 6 months for the development of CGH according to the above-mentioned diagnostic criteria. Any patient who reported any kind of new headache was invited to undergo a full clinical assessment as described earlier. The incidence of CGH was analyzed and compared to a healthy control group.

2.5. Statistical analysis

All the data were collected in the Orofacial Pain & TMD Clinic and downloaded directly into an Excel spreadsheet. The SPSS,22.0 version (SPSS Inc., Chicago IL, USA) was used for the statistical analyses. The odds ratio (OR), its standard error, and 95% confidence interval were calculated according to Altman (1991). The relative risk (RR), its standard error and 95% confidence interval were also calculated according to Altman (1991). The sample size was based on a power analysis aimed to reach enough participants for the follow-up cohort group of patients with TMD but without CGH. The calculation (Eng, 2003) was based on the estimated CGH prevalence of 4% in the general population (Antonaci and Sjaastad, 2011) and hypothesis of 30% in the TMD cohort group.

3. Results

Out of 198 consecutive patients who were seen in the Clinic for the first time during the data collection period, 74 met the inclusion criteria. Their data were retrieved, and the results were analyzed for the three specific TMD groups (pain-related $n = 37$, intraarticular, $n = 17$, and mixed $n = 20$) and compared to those of the healthy control group who met the inclusion criteria ($n = 42$ out of 47 volunteers). The mean age of all participants was 34.2 (±standard deviation 12.3) years, with no significant age difference between all the groups (Table 1). Most of the 116 participants were women ($n = 86$, 74%) and there was no significant sex difference between the groups (Table 1).

3.1. CGH odds ratios

Significant differences between the patients with TMD and the healthy controls were found for the odds ratio of CGH (OR = 7.92; 95% CI = 1.75-33.77; $p < 0.001$). Twenty-one of the 74 TMD patients were diagnosed with CGH (28%) compared to only 2 out of the 42 healthy controls (5%) (Table 2). There was a significant difference for the odds ratios of CGH between the TMD subgroups (Table 2). The pain-related (OR = 12.17; 95% CI = 2.53-58.39)) and the mixed (OR = 10.76; 95% CI = 1.98-58.45) TMD groups showed significantly higher rates of CGH compared to the healthy controls ($p < 0.001$). No significant difference was found for the CGH odds ratio between the patients with intra-articular TMD and the healthy controls (OR = 0.46; 95% CI = 0.02-10.14) (Table 2).

3.2. CGH relative risk

During the first 12 months of data collection, a total of 39 patients with TMD (32 females and 7 males) were diagnosed as not having CGH and therefore assigned to the TMD cohort group. They were followed-up for an average of 21.6 months, and none was lost to follow-up. Three TMD patients developed CGH (7.6%) (Table 3) during the follow-up period. Thirty healthy controls with no CGH were followed up within the same period and none had any new CGH diagnosis. No significant difference of relative risk for CGH was found between the groups (RR = 5.42; P = 0.25) (Table 3).

Table 1
Age and sex groups description.

Group	N	Age (SD)	Female (%)
Pain Related TMD	37	34.1 (11.8)	29 (78%)
Intra-Articular TMD	17	32.0 (12.6)	11 (64%)
Mixed TMD	20	39.0 (14.3)	17 (85%)
Healthy	42	32.0 (11.0)	29 (69%)
Total	116	34.2 (12.3)	86 (74%)

4. Discussion and conclusions

The main findings of this study demonstrate that the odds ratios of CGH were significantly higher only for the pain-related and mixed TMD groups (OR = 12.17 and 10.76, respectively), and not for the intra-articular group (OR = 0.46) compared the healthy control group. In contrast, there were no significant differences between the patients with TMD and the healthy controls in the incidence rates of CGH throughout the 14- to 24-month follow-up period.

The average age of the patients with TMD in this study was 34 (±12) years, and the female-to-male ratio was 3:1, with no significant differences between the study groups for age and sex. These age and sex data are in accordance with previous epidemiological studies of patients with TMD (Guarda-Nardini et al., 2012; Sonia and Ohrbach, 2018), and therefore support the external validity of our study findings.

A literature search failed to yield any previous material relating to the prevalence of CGH among patients with TMD, despite the recognized and evidence-based clinical correlation between the masticatory system and the cervical spine. According to several high-quality epidemiological studies, the prevalence rate of CGH in the healthy adult population is 0.5–2.5% (Knackstedt et al., 2010; Sjaastad and Bakketeig, 2008; Vincent and Luna, 1999). The current study found an overall CGH rate of 28% among patients with TMD (21 out of 74 patients; OR = 7.92), and 5% in the healthy control group (2 out of 40 participants) (Table 2).

Analysis of the TMD subgroups revealed the highest CGH rate as being that for the pain-related TMD groups (14 out of 37 patients, 38%; OR = 12.17), followed by the mixed TMD group (7 out of 20 patients, 35%; OR = 10.77) (Table 2). No patient in the intra-articular TMD group was diagnosed with CGH. Therefore, patients with pain-related TMD, with or without intra-articular disorder, are significantly more likely to be diagnosed as also having CGH than patients with isolated intra-articular TMD and healthy controls. The relatively high rates of CGH among "painful" patients with TMD may also explain their significantly impaired cervical spine performance and high pain levels, given that CGH patients reportedly have similar neck impairments (Rubio-Ochoa et al., 2016). The mechanism that explains those high comorbidity rates is likely to be the neuro-anatomical convergence of nociceptive stimuli from the upper neck and the trigeminal nerve into the same trigeminocervical nociceptive neuron (TCN) (Bartsch and Goadsby, 2003; Goadsby and Bartch, 2010). The finding that no intra-articular TMD patients were diagnosed with CGH, along with the fact that it is not characterized as a pain disorder further supports the mechanism of somatically referred pain via TCNs as the main contact linking the upper neck and the masticatory system. If a mechanical dysfunction had been the underlying mechanism of a connection between the upper neck and the masticatory system, one would have expected to find several intra-articular TMD patients with CGH, but this was not the case. This study provides the first reference of CGH prevalence among patients with TMD, and its findings warrant further research on larger study groups.

This cohort study was the first to assess the relative risk for CGH in patients with TMD. Three of the 39 TMD patients (7.6%) who entered the study without CGH ultimately developed it during the 14–24 months of follow-up. Although the incidence rate of CGH in the TMD group in this study was higher than the rate of CGH in the general population (0.5–2.5%), an incidence comparison between the TMD cohort and the healthy control group yielded no significant difference (P = 0.25). The relatively small size of the study groups, however, precludes the ability to support a causal connection between having TMD and developing CGH, as suggested by the convergence theory (Bartsch and Goadsby, 2003; Goadsby and Bartch, 2010; Haldeman and Dagenais, 2001). One possible explanation could be the relatively short follow-up period. Presumably, the development of CGH in patients with TMD takes longer than 14–24 months on average, and therefore establishing a relationship would necessitate a longer follow-up. Another possible explanation could be that the causal connection has a one-way and not a two-way

Table 2
Odds ratio of cervicogenic headache.Red represents statistical significance (P⟨0.05).

GROUP	N	Cervicogenic Headache		CGH %	Odds ratio (compared to controls)	95% Confidence Interval	P value versus controls
		Yes	No				
Healthy	42	2	40	5%	NA	NA	NA
Pain related TMD	37	14	23	38%	12.17	2.53-58.39	0.0018
Intra-Articular TMD	17	0	17	0	0.46	0.02-10.14	0.6249
Combined TMD	20	7	13	35%	10.76	1.98-58.45	0.0059
TMD total	74	21	53	28%	7.92	1.75-35.77	0.0071

Table 3
Relative risk for CGH in patients with TMD.

GROUP	N	Cervicogenic Headache		CGH %	Relative risk (compared to controls)	95% Confidence Interval	P value versus controls
		Yes	No				
Healthy	30	0	30	0	NA	NA	NA
TMD	39	3	36	8.3%	5.42	0.29–101.18	0.2572

design, whereupon patients with CGH would be more prone to develop TMD (Mingels et al., 2019) than patients with TMD would be prone to develop CGH. Further research on CGH patients is needed to confirm the latter notion.

The main strength of this study is that it is the first to assess odds ratios of CGH in specific TMDs as well as to evaluate the relative risk of those patients to develop CGH during a period of 14–24 months. Its main limitation is the relatively small groups of patients with specific TMDs as well as the lack of a specific data regarding the characteristic of pain (time and severity).

In view of the strong association between the diagnosis of painful TMD and CGH, we suggest that these patients should be routinely screened for CGH by a trained musculoskeletal clinician.

Funding sources

This work was financially supported by the American Academy of Orofacial Pain, New Jersey, USA (2000$ grant).

A statement “significance”

To the best of our knowledge this work is the first ever to assess the association between specific Temporomandibular disorders and Cervicogenic Headache.

Declaration of competing interest

None.

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