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ORIGINAL ARTICLE

Spinal deformity and malocclusion association is not supported by high-quality studies: results from a systematic review of the literature

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Abstract

Background Several reports in the literature have suggested a causative association between oral occlusion and spinal deformity such as scoliosis and Scheuermann's disease or kyphosis. Based on these findings, a growing number of adolescents with spinal deformity receive orthodontic treatment, supposing a beneficial effect on the spine.

Objective The aim of this study was to verify the association between spinal deformity and malocclusion in the orthopedic population and potential effect of orthodontic treatment on the spinal deformity.

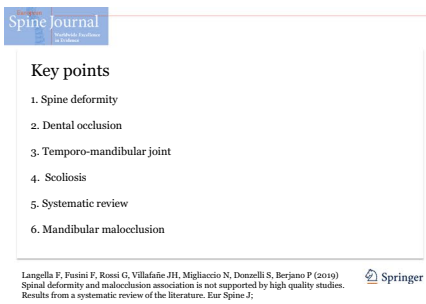
Method The databases: MEDLINE, CINAHL, EMBASE, Cochrane Register, OTseeker and ScienceDirect were searched up to August 2017 for studies reporting on associations between spinal and occlusal conditions. Case series, cohort, case–control studies and randomized clinical trials were considered for analysis. Two reviewers independently selected studies, conducted quality assessment and extracted results. Methodological quality was assessed using MINORS score.

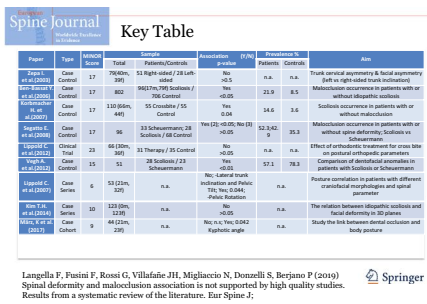
Results Nine publications reporting on 1424 patients were included. Studies were two case series, five case–control studies, one cohort study and one randomized clinical trial. The methodological quality was poor in 8/9 studies.

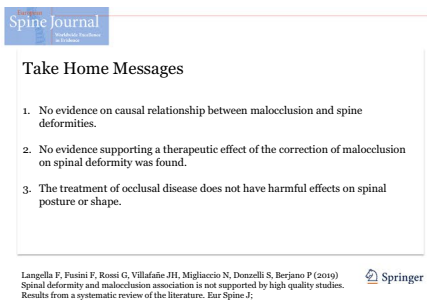
Conclusion Evidence from three low-quality studies suggests an increased prevalence of occlusal dysfunction in patients with known spinal deformity, but the conclusions have a high risk of bias. No evidence of beneficial effects of orthodontic treatment on spinal deformity was found.

Graphical abstract

These slides can be retrieved under Electronic Supplementary Material.







Keywords Spine deformity · Dental occlusion · Temporomandibular joint · Systematic review · Scoliosis · Kyphosis · Scheuermann's disease · Mandibular malocclusion

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Extended author information available on the last page of the article

Introduction

Spine deformity is a common condition in adolescents with two main clinical presentations: scoliosis and Scheuermann's disease. These two spinal deformities have been proposed to be the most influencing facial asymmetry [1, 2]. Abnormal body posture has long been hypothesized to be responsible for various craniofacial orthopedic and orthodontic conditions, but the scientific literature is not able yet to support these assumptions [3]. Pathological curvatures of the spine induce in some cases the development of compensatory curvatures elsewhere along the spine and may result also in compensatory head posture [4]. Therefore, the spine disorders could contribute to the development of different dentofacial anomalies and later to their persistence due to their chronic influence on the head posture. The frequency of different degrees of malocclusion in the orthopedic patients group ranged from 83 to 87% [5]. The two clinical conditions are frequently presented together. Previous studies reported that spine and facial sagittal configurations are related: Subjects with a long face tended to have longer and straighter cervical vertebrae, whereas those with a short face have shorter and more curved cervical vertebra [6].

Correlation between bone deformities of the head and trunk has been advocated by some for years; for example, the position of spine can influence the craniomandibular system and vice versa [6, 7], but never proven [8]. Some studies have been performed with different methods, but usually on small samples due to the invasive methodologies required [9]. Nevertheless, a bone deformity causes external signs that can be checked through tools that were developed for screening purposes [10] and that could be applied in large populations [11].

Though to date the relationship between occlusal dysfunction and spinal deformity has not been widely accepted, increasing pressure is put on adolescents with spinal deformities and their families to seek orthodontic treatment and vice versa for the young with occlusal problems, causing increase in perceived abnormality and potentially unnecessary diagnostic and therapeutic procedures.

The aim of this study is to systematically review the medical literature for associations between spine deformity and occlusal dysfunction in both directions and for effectiveness of the treatment of occlusal dysfunction on the associated spine deformity.

Method

Study design

This is a systematic literature review of studies investigating or reporting an association between malocclusion and spinal deformity in the literature. PRISMA [4] guidelines were followed during the design, search and reporting stages of this systematic review.

Literature search

Our literature search aimed to identify all available studies that evaluated the association between dental malocclusion and spinal deformity. We searched in MEDLINE, CINAHL, EMBASE, Cochrane Register, OTseeker and ScienceDirect database in every language between 1970 and August 2017. The search string was: (((((((((scoliosis) OR kyphosis) OR spine deformity) OR spine posture) OR spinal alignment) OR spinal imbalance) OR defect of posture) OR hyperkyphosis)) AND ((((((malocclusion) OR mandibular joint) OR crossbite) OR temporomandibular joint) OR occlusal dysfunction) OR dental occlusion). Similar search strategies were used in MEDLINE, CINAHL, EMBASE, Cochrane Register, OTseeker and ScienceDirect database.

Two authors (G.R. and F.F.) performed the search and evaluated the abstracts independently for potential eligibility and subsequently full-text publications for eligibility. The search strategies are shown as PRISMA Flow-Chart in Fig. 1. A third author (F.L.) resolved discrepancies. Each researcher reviewed the title and abstract of all the articles and selected the relevant ones according to inclusion and exclusion criteria. The list of references of each article was screened in order to find any additional original articles. In addition to the computer search, an independent hand search including scanning of reference lists from other systematic reviews was performed.

Study selection

Eligibility criteria

Types of studies included observational studies (cohort studies, case–control studies and case series), randomized or quasi-randomized clinical with human subjects and without restrictions regarding date of publication.

We excluded from analysis all repeated articles, case reports, editorials, technical notes and review articles. Also, articles not written in English and in vitro, preclinical and animal studies were excluded.

Study assessment

Reviewers used the methodological index for non-randomized studies (MINOR) scores to assess the

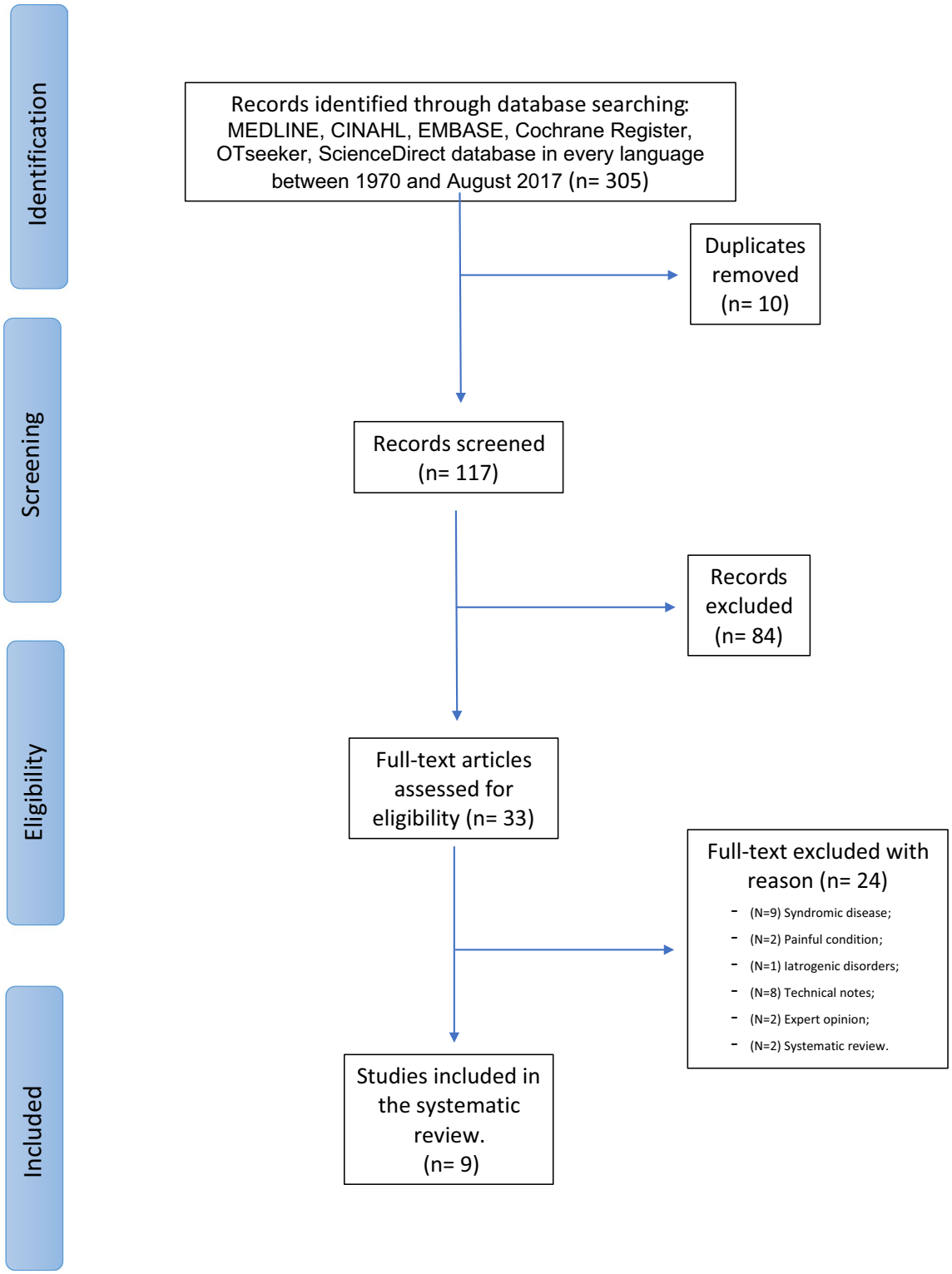

Fig. 1 Flow diagram of studies through the different phases of the systematic review

methodological quality of each clinical article. The MINOR score is a simple and effective tool to rank the strength of results measured in research studies [5].

We extracted a number of variables on study design and methodological characteristics, patient and intervention characteristics and outcomes from all eligible studies.

The quality of studies conducted by the MINOR score ranged between 6 out of 24 [6] and 23 out of 24 [7] with an average score of 14.55 ± 5.25 and median score of 17. Study design included randomized clinical trial (1), case–control studies (5), cohort study (1) and case series (2).

Data synthesis and analysis

Data were analyzed using SPSS version 22.0 (SPSS Inc., Chicago, IL, USA). The level of evidence (LoE) of a given study was assigned based on the 2000 scoring system adopted by the North American Spine Society. The categorization of the studies according to LoE was based on the combined evaluation of two reviewers (F.F. and G.R.) with disagreements resolved by a third author (F.L.).

Results

Study selection

We identified 305 studies through database search. After removing duplicates and screening titles and abstracts of all remaining unique articles, 33 full-text articles needed to be assessed to verify their eligibility for the inclusion in the present study. Ultimately, 24 of these manuscripts were excluded for various reasons ($N=9$ syndromic disease; $N=2$ painful condition; $N=1$ iatrogenic disorders; $N=8$ technical notes; $N=2$ expert opinion; $N=2$ systematic review). Thus, nine studies were finally selected for this review as shown in Fig. 1.

Types of study

The type of studies included two case series [9, 12], five case–control studies [13–17], one cohort study [18] and one randomized clinical trial [19], as given in Table 1.

Levels of evidence

The systematic research of the literature provides one high LoE work: a clinical randomized control trial. In most cases, articles reached LoE III as the best result. The review included 55.55% (5/9) LoE III and 33.33% (3/9) LoE IV studies. LoE V papers were excluded from the research as for exclusion criteria. A summary of articles' major findings and LoE can be found in Table 2.

Data from studies

Association between spine deformity and malocclusion

Spine deformity prevalence in population with and without malocclusion:

Korbmacher et al. [14] investigated the prevalence of orthopedic coronal asymmetries in patients with or without unilateral crossbite. Fifty-five children with unilateral crossbites and 55 gender and age-matched children without crossbite, as the control group, were evaluated. Body asymmetry was assessed by examination of the differences in shoulder elevation (threshold for asymmetry set at 5 mm of height difference) and by the Adam's forward bending test (positive when the hump at one side was 8 mm higher than the other or more). Intra-rater agreement was good for both clinical tests. Children with a unilateral crossbite showed more frequently an elevated shoulder (24 vs 10%; $p=0.004$) and scoliosis (8 vs 2%, $p=0.04$). The MINORS quality score of the study was 17/24.

Malocclusion prevalence in patients with or without spine deformity

Segatto et al. [15] studied 33 children with Scheuermann's disease, 28 with scoliosis and a control group of 68 orthopedically healthy ones to map the facial anomalies that might be correlated with the two most frequent spinal deformity entities. The authors compared the sagittal occlusal anomalies (normal, distal molar occlusion, mesial molar occlusion, all of which could be unilateral or bilateral) and found that subjects with normal spinal alignment had more frequently bilateral normal occlusion (64.68%) than those with scoliosis (57.12%) and those with Scheuermann's disease (47.74%), but they did not provide statistic tests for the significance of differences, nor the data were available for analysis. They found statistically significant differences for the mean overjet distance (measured in mm) between patients with Scheuermann's disease (with or without brace treatment) and the control group, for the mean overbite distance between the subgroup of patients with Scheuermann's disease who were treated with brace and the control group, and for the midline deviation mean only between the scoliosis group in brace treatment and the control group. It has to be said that the authors found statistically significant associations in four comparisons out of a total of 12 comparisons made, and that the a priori definition of what was the main outcome parameter was missing. Authors did not report the type of brace that the patients were wearing for treatment of the spinal deformity (i.e., Milwaukee brace). The quality score of the study was 17/24.

Table 1 Description of the studies

Participants	Intervention	Outcome measures and follow-up	Reported results
Zepa et al. [17] 79 subjects Exp 51 Ctrl 28 Mean age 11 Inclusion criteria Trunk asymmetry at Adam's test > = 10 mm Thoracic kyphosis > = 50°	*Exp* 51 patients with right-sided thoracic and/or cervical scoliosis *Ctrl* 28 patients with left-sided thoracic and/or cervical scoliosis	Outcome measures Clinical trunk asymmetry parameters Rib humps Lumbar prominence Cervical inclination Cephalometric parameters Horizontal line Atlas line C4-line True vertical plumb line Amigd line Lower face line Follow-up n/a	Results All postural and morphologic variables were almost similar in the right-sided and left-sided thoracic hump and lumbar prominence Authors' conclusion Trunk asymmetry in young adults clinically expressed as thoracic humps or lumbar prominence may not affect the facial symmetry
Ben-Bassat et al. [13] 802 subjects Exp 96 Ctrl 705 Mean age 13.9 [3.5] Inclusion criteria Idiopathic scoliosis	*Exp* 96 consecutive patients with idiopathic scoliosis *Ctrl* 705 Ashkenazi children	Outcome measures Cobb angle Site of scoliosis Side of scoliosis Stage of dental development Molar relationship Canine relationship Upper and lower midline deviation Anterior crossbite Posterior crossbite Occlusal AP category Follow-up n/a	Results The distribution of the angle classes of malocclusion was significantly different in the two groups ($p = .0001$) because of many class II subdivision patients in the scoliotic group No correlation between the side of scoliosis and the facial asymmetry was found statistically significant Authors' conclusion Patients with idiopathic scoliosis have more asymmetric features of malocclusion than a random group. No significant correlation between the side of scoliosis and the side crossbite.
Korbmacher et al. [14] 110 subjects Exp 55 Ctrl 55 Mean age 7 [2.08] Inclusion criteria Subjects with unilateral crossbite and age-matched children with a symmetric occlusion and no crossbite.	*Exp* 55 patients with unilateral crossbite *Ctrl* 55 age-matched children with a symmetric occlusion	Outcome measures Clinical orthopedic examination: shoulder asymmetry, Adam's test, functional leg length difference and laxity of ligaments Orthodontic examination Occlusal relations in the sagittal, vertical and transversal dimensions, space relations of the maxillary and mandibular frontal segment and the TMJ status and function Follow-up n/a	Results Children with a unilateral crossbite had, statistically more often, an oblique shoulder ($p = 0.004$), an oblique pelvis ($p = 0.007$), functional leg length differences ($p = 0.002$) and a scoliosis ($p = 0.04$) than children with dental symmetry Authors' conclusion Children with an asymmetric upper cervical spine and unilateral crossbite had, significantly more often, an oblique shoulder functional leg length differences and scoliosis when compared with children with a symmetric occlusion.

Table 1 (continued)

Participants	Intervention	Outcome measures and follow-up	Reported results
Segatto et al. [15] 96 subjects Exp 1 33 Exp 2 28 Ctrl 68 Mean age 14.7 [2.3] Inclusion criteria Patients with spine deformity: scoliosis or Scheuermann's disease	*Exp 1* 33 patients with Scheuermann's disease *Exp 2* 28 patients with idiopathic scoliosis *Ctrl* 68 controls	Outcome measures Orthodontic examination Occlusal relations in the sagittal, vertical and transversal dimensions and space relations of the maxillary Examination of the jaw space relations Examination of temporomandibular joint Orthodontic X-ray Clinical orthopedic examination: shoulder asymmetry, Adam's test, functional leg length difference and laxity of ligaments Follow-up n/a	Results More and greater abnormalities among patients in the Scheuermann's disease group than in the scoliosis group. Comparing the values of the two spinal disorder groups and the control group, ($p < .05$) occurred in: frequency of unilateral Cl.II. molar occlusion, overjet and extreme overjet mean value. The bilateral normocclusions occurred in 64.68% of the control group, 57.12% of scoliosis group and 47.74% Scheuermann's group Authors' conclusion The frequency of the asymmetric deviations of posterior region (molar relations) occurring in the sagittal dimension was similar in the groups with spinal deformities, and this value was double than the one registered in the control group
Lippold et al. [19] 66 subjects Exp 31 Ctrl 35 Mean age 7.3 [2.1]; fu 8.3 [2.1] Inclusion criteria Patients to be included were required to have late deciduous and early mixed dentition, unilateral posterior crossbite and functional mandibular asymmetry.	*Exp* (Münster treatment concept) Therapy group for slow expansion of the maxillary bone formation, a bonded palatal expansion appliance (Fig. 2 a) was used. After correction of the maxillary discrepancy, an orthodontic activator treatment was applied to achieve midline coordination and to retain the amount of palatal expansion *Ctrl* Clinical observation	Outcome measures Rasterstereography Kyphotic angle Lordotic angle Lateral deviation Vertebral rotation Pelvic tilt Pelvic torsion Follow-up 1.1 year	Results No clinically relevant differences between the control and the therapy groups at T1 and T2 were found for the parameters of kyphotic and lordotic angle, the surface rotation, lateral deviation, pelvic tilt and pelvic torsion Authors' conclusion This randomized clinical trial demonstrates that in a juvenile population with unilateral posterior crossbite the selected early orthodontic treatment protocol does not affect negatively the postural parameters
Vegh et al. [16] 51 subjects Exp 28 Ctrl 23 Mean age 14.7 and 14.8 Inclusion criteria Patients with recent diagnosis of spine deformity: scoliosis or Scheuermann's disease.	*Exp* 28 consecutive patients with idiopathic scoliosis *Ctrl* 23 consecutive patients with Scheuermann's disease	Outcome measures Orthodontic examination Sagittal incisal relation Vertical incisal relation Transversal incisal relation Examination of the jaw space relations Examination of temporomandibular joint: clicking of the joints, pain, limited mouth opening Follow-up n/a	Results Facial asymmetries significantly higher in Scheuermann's group (78.3%) than SC group (57.1%) Authors' conclusion The majority of dentofacial anomalies are present in patients with Scheuermann's disease. Its necessity of closely monitoring for orthodontic problems the patients with spine deformity

Table 1 (continued)

Participants	Intervention	Outcome measures and follow-up	Reported results
Lippold et al. [12] 53 subjects Mean age 24.6 [9.0] Inclusion criteria Healthy adults (32 women and 21 men; mean age 24.6 years, SD 9.0 years) with skeletal malformations: classes II and III malocclusion.	Aim To relate the differences in the posture of patients with different craniofacial morphologies	Outcome measures Cephalometric analysis Facial axis Mandibular plane Inner gonial angle Lower facial height Facial depth Maxilla position Rasterstereography Lateral trunk inclination Pelvic tilt Pelvic rotation Follow-up n/a	Results Statistically significant differences ($p < .05$) in pelvic torsion were documented with respect to the facial axis and facial depth. Moreover, the differences ($p < .05$) between patients with a skeletal horizontal–vertical facial axis and patients with a basal distal–mesial position for the the facial depth could be determined for the pelvic torsion Authors' conclusion Correlations between the pelvic torsion and the facial axis, between the pelvic torsion and the facial depth. Possible relationships between the vertical and the sagittal position of the lower jaw and the body posture
Kim et al. [9] 123 subjects Mean age 15.9 range 14–28 Inclusion criteria Patients visited for treatment of idiopathic scoliosis and had no previous orthodontic treatment were enrolled in the study	Aim To analyze the relationship between idiopathic scoliosis and facial deformity in horizontal, vertical and anteroposterior planes. To study the correlation between facial deformities and the severity of scoliosis.	Outcome measures Cephalometric analysis Facial axis Mandibular plane Inner gonial angle Lower facial height Facial depth Maxilla position Full standing X-ray Cobb angle Lenke type of scoliotic curve Follow-up n/a	Results The measurements did not reveal any significant association between the Cobb angle and cephalometric measurements and between the curve type based on the Lenke classification and cephalometric measurements Authors' conclusion No apparent relation was observed between the severity of scoliosis and facial form variations in idiopathic scoliosis patients
März et al. [18] 44 subjects Mean age 24.7 range 22–33 Inclusion criteria Good general health, no tattoos or large scars on the back and a natural dentition with bilateral molar support	Aim To investigate the short-term impact of different occlusal positions on spine and body posture parameters as mapped using rasterstereography. Rasterstereography measurement is repeated in seven different mandible positions: habitual intercuspation, habitual intercuspation with maximal force, physiological rest position, cotton rolls on both sides, cotton rolls only on right side, bite elevation at 1 mm and right eccentrically positions.	Outcome measures Cephalometric analysis Facial axis Mandibular plane Inner gonial angle Lower facial height Facial depth Maxilla position Rasterstereography Lateral trunk inclination Pelvic tilt Pelvic rotation Follow-up n/a	Results Statistically significant differences were found for the fleche cervicale and kyphotic angle, comparing data collected for the habitual intercuspation and right eccentrically positions Authors' conclusion Not conclusive association between dental occlusion to an instantaneous impact on the tested rasterstereographic parameters. The posture changes showed may be arisen from individual neuromuscular compensation.

Table 2 Results

Paper	Type	MINOR score	Sample		Association (Y/N) *p* value	Prevalence%		Quest	Aim	Main result
			Total	Patients/controls		Patients	Controls			
Zepa et al. [17]	Case control	17	79 (40m, 39f)	51 right-sided/28 left-sided	No > 0.5	n.a.	n.a.	3	Trunk cervical asymmetry and facial asymmetry (left vs right-sided trunk inclination)	"When compared inclination of cervical spine and cranial morphology, all postural morphologic variables were almost similar in the left-sided and right-sided thoracic hump and lumbar prominence groups… trunk asymmetry in young adults clinically expressed as thoracic humps or lumbar prominence may not affect the facial symmetry"
Ben-Bassat et al. [13]	Case control	17	802	96 (17m, 79f) Scoliosis/706 control	Yes < 0.05	21.9	8.5	2	Malocclusion occurrence in patients with or without idiopathic scoliosis	"Patients with idiopathic scoliosis have more asymmetric features of malocclusion than a random group… No correlation between the side of scoliosis and the side of posterior crossbite or lower midline deviation was found statistically significant"
Korbmacher et al. [14]	Case control	17	110 (66m, 44f)	55 crossbite/55 control	Yes 0.04	14.6	3.6	1	Scoliosis occurrence in patients with or without malocclusion	"The dental asymmetries correlate with orthopedic asymmetries in the frontal plane. Children with an asymmetric upper cervical spine and unilateral crossbite had, significantly more often, an oblique shoulder functional leg length differences, and scoliosis when compared with children with a symmetric occlusion"

Table 2 (continued)

Paper	Type	MINOR score	Sample		Association (Y/N) *p* value	Prevalence%		Quest	Aim	Main result
			Total	Patients/controls		Patients	Controls			
Segatto et al. [15]	Case control	17	96	33 Scheuermann; 28 scoliosis/68 control	Yes (2) <0.05 No (3) >0.05	52.3;42.9	35.3	2 and 3	Malocclusion occurrence in patients with or without spine deformity and scoliosis versus Scheuermann	"The frequency of the asymmetric deviations of posterior region (molar relations) occurring in the sagittal dimension was similar in the groups with spinal deformities and this value was double than the one registered in the control group…. The mean asset of the midline deviation was significantly higher in the scoliotic group". The bilateral normocclusions: the control group (64.68%), Scoliosis group (57.12%) and Scheuermann (47.74%)"
Lippold et al. [19]	Clinical trial	23	66 (30m, 36f)	31 therapy/35 control	No >0.05	n.a.	n.a.	4	Effect of orthodontic treatment for crossbite on postural orthopedic parameters	"This randomized clinical trial demonstrates that in a juvenile population with unilateral posterior crossbite the selected early orthodontic treatment protocol does not affect negatively the postural parameters"
Vegh et al. [16]	Case control	15	51	28 Scoliosis/23 Scheuermann	Yes <0.01	57.1	78.3	3	Comparison of dentofacial anomalies in patients with scoliosis or Scheuermann	"The visual record of facial asymmetries showed asymmetry in 78.3% of the Scheuermann disease group, which is significantly higher than that recorded in the Scoliosis group (57.1%)….Comparing the parameters of the subgroups on the basis of the severity of spinal deformities, significant differences were seen only in the midline shifts and in facial asymmetries"

Table 2 (continued)

Paper	Type	MINOR score	Sample		Association (Y/N) *p* value	Prevalence%		Quest	Aim	Main result
			Total	Patients/controls		Patients	Controls			
Lippold et al. [12]	Case series	6	53 (21m, 32f)	n.a.	No Lateral trunk inclination and Pelvic tilt Yes 0.044 Pelvic rotation	n.a.		1	Posture correlation in patients with different craniofacial morphologies and spinal parameter	"Correlations between the pelvic torsion and the facial axis as well as the pelvic torsion and the facial depth indicate possible relationships between the vertical and the sagittal position of the lower jaw and the body posture"
Kim et al. [9]	Case series	10	123 (0m, 123f)	n.a.	No > 0.05	n.a.		3	The relation between idiopathic scoliosis and facial deformity in 3-D planes	"Based on the results of this study, no apparent relation was observed between the severity of scoliosis and facial form variations in idiopathic scoliosis patients… The null hypothesis was rejected because occurrence of facial form deviations are not increased in proportion to the severity of scoliosis"
März et al. [18]	Case Cohort	9	44 (21m, 23f)	n.a.	No n.s. Yes 0.042 Kyphotic angle	n.a.		1	Study the link between dental occlusion and body posture	"…we could not conclusively associate dental occlusion to an instantaneous impact on the tested parameters (Rasterstereography). The posture changes that we detected could also have arisen from individual neuromuscular compensation"

1: Spine deformity occurrence in population with and without malocclusion

2: Malocclusion occurrence in patients with or without spine deformity

3: Relationship between a specific spinal deformity and malocclusion

4: Treatment of malocclusion in patients with spinal deformity

*Not significant: n.s

***Not available: n.a

Vegh et al. [16] studied the prevalence of occlusal alterations in 23 children with Scheuermann's disease and 28 with scoliosis, all of them referred by the orthopedic department after diagnosis. They found in the Scheuermann's group that a greater prevalence than in the scoliosis group for the following abnormal occlusal patterns: abnormal overjet (21.7 vs 3.6%) upper midline deviation from the facial midline (60.9 vs 42.8%) and lower midline deviation from the facial midline (60.9 vs 32.1%). The study did not have a control group of patients with no spinal deformity. Authors did not report how many of the patients were under treatment for scoliosis with brace or the type of brace (i.e., Milwaukee brace). The quality score of the study was 15/24.

Ben-Bassat et al. [13] evaluated the prevalence of occlusal alterations in 96 children with idiopathic scoliosis and compared it with a control group of 706 children without spinal deformity from a previously published study. The examiners were aware of the spinal deformity status of the children. The differences in prevalence of asymmetrical molar relationship between scoliotic and non-scoliotic children (26.6 vs 28.9%) were not statistically significant. The prevalences of asymmetrical canine relationship (31.5 vs 22.1%), upper midline deviation (21.1 vs 9.5%), lower midline deviation (53.7 vs 32.9%), anterior crossbite (16.6 vs 9.3% and posterior crossbite (8.1 vs 18.2%) were significantly higher in the scoliosis group. The authors did not compute the number of patients under treatment with brace or their types, but provide as an example one figure showing a participant in the study who is wearing the Milwaukee brace. The quality score of the study was 17/24.

Relationship between a specific spinal deformity and malocclusion

Zepa et al. [17] evaluated 79 adults who had trunk asymmetry on clinical evaluation from a cohort of 430 young Finnish adults. Though they did not specifically evaluate occlusal alterations in these subjects, their cranial radiographical study showed no differences in the sidedness of craniofacial asymmetries depending on the sidedness of the trunk asymmetry. A similar study by Kim et al. [9] evaluated 129 young females referred for treatment of idiopathic scoliosis classifying the severity of the spinal deformity by the magnitude of the Cobb angle and the type of deformity by the main categories of the classification by Lenke. The craniofacial morphology was evaluated by front and lateral radiographs (cephalograms). The authors could not demonstrate any association between severity or type of scoliosis and craniofacial dysmorphism. The quality score of the study was 17/24.

Lippold et al. [12] evaluated 53 healthy adults with class II and class III malocclusion using cephalometric radiographical analysis and rasterstereography (a photometric 3D reconstruction of the shape of the trunk). The authors could not demonstrate associations between facial radiographical patterns and lateral trunk deviation from the midline or coronal inclination of the pelvis. The quality score of the study was 6/16.

Effect of the treatment of malocclusion on spinal alignment

A randomized clinical trial by Lippold et al. [19] provided data on the effect of orthodontic treatment for crossbite on postural orthopedic parameters. Sixty-six children with lateral crossbite and no spinal deformity were randomized to receive or not orthognathic treatment of the occlusal condition. At the beginning of treatment and after on average 1 year, spinal surface was scanned by rasterstereography. No differences of spinal postural and shape parameters (lateral deviation, kyphotic angle, coronal tilt of the pelvis and trunk torsion) were found between groups at the beginning and at the end of treatment. The authors concluded that no harmful effect on spinal alignment could be demonstrated as a result of orthognathic treatment. The quality score of the study was 23/24.

Effect of the modality of occlusion on postural parameters of the spine

März et al. [18] evaluated 44 young adults with rasterstereography of the trunk surface and repeated for each subject in seven different mandible positions. Statistical analysis was performed for 72 combinations of parameters. Significant ($p < 0.05$) variations were found for six combinations of dental mandibular position and trunk position parameters. The authors considered the results inconclusive assuming that the posture changes detected could also have arisen from individual neuromuscular compensation. No subject in the study had spinal deformity. The quality score of the study was 9/16.

Discussion

The purpose of this systematic review was to evaluate the evidence on relationship between occlusal alterations and spinal deformity and its quality. The vast majority of the published studies refer to patients with idiopathic spinal disorders and are constructed on clinical case reports and expert opinions that we excluded from the review due to their low level of evidence.

Regarding the first research question, i.e., whether a higher prevalence of spinal deformity could be found in subjects with malocclusion, one low-quality study suggests a higher prevalence of shoulder asymmetry and scoliosis in the crossbite group [14]. The assessment of spinal

deformity was based only on clinical evaluation without radiographical confirmation. The radiographical examination is the standard examination for confirming diagnosis of spine deformities, and therefore, what was found was a higher prevalence of postural alterations and not diagnosed spinal deformities. This may lead to misinterpretation of the results and incorrect generalization to the population of adolescents affected by spine deformities.

The quality score of the study is not high. Thus, low-quality evidence from only one study suggests an increased incidence of spinal deformity in subjects with occlusal alterations, but this conclusion is exposed to substantial risk of bias.

Our second research question was whether in patients with spinal deformity a different prevalence of occlusal alterations can be found. One low-quality study [15] reported a higher prevalence of sagittal occlusal alterations in patients with known Scheuermann's disease, especially in the subgroup on brace treatment and a higher prevalence of occlusal asymmetry in patients with brace treatment for scoliosis compared to controls. The numbers in this study were modest.

A second low-quality study [16] compared only Scheuermann's disease (SD) patients with scoliosis patients and found that SD patients had greater incidence of both sagittal and frontal plane occlusal alterations than scoliotic patients. The study did not have a control group. No information was provided on the type and prevalence of brace treatment in these patients.

A third low-quality study with a greater number of subjects in the control group [13] found a greater prevalence of occlusal alterations in patients with scoliosis compared to controls.

In all of these articles, the potential bias induced by a direct effect of brace treatment on altered occlusion was not discussed or analyzed. In fact, one of the possible side effects of some scoliosis and kyphosis braces (i.e., the Milwaukee brace or its derivations) is alteration of the jaw function and morphology, due to direct contact to the anatomical area. The Milwaukee brace and its derivations have been for years the brace of choice for kyphotic deformities (Scheuermann's disease or scoliosis combined with kyphosis) and for major curves with apex at T7–T8 or above [20, 21]. Thus, moderate evidence from low-quality studies supports an increased incidence of occlusal alterations in patients with spinal deformity, but the risk of confounding and bias of these studies is substantial.

A different low-quality study [18] investigated in subjects without spinal deformity and the effects of different mandibular positions on the trunk alignment, with inconclusive results.

Some hypotheses have been made to explain the relation between dental occlusion and spine deformity. This relationship was already suggested by some authors through observational studies [22], and it is based on the anatomical and functional link between the stomatognathic apparatus and the spinal column. There is an anatomical connection between the jaw and the cervical spine since the cranium and the mandible have muscular and ligament attachments to the cervical area; moreover, cervical spine, head and jaw take part together in a complex functional system [22]. In spite of the degree of elaboration of these hypotheses, there is little experimental support to provide confirmatory evidence on the mechanism that correlates malocclusion and deformity of the trunk.

Festa and D'Attilio [22, 23] found in Caucasian adult females a relationship between jaw length and cervical lordosis angle on lateral X-ray, with a decrease in cervical lordosis along with the increase in jaw body length. Using this association as a demonstration of how variations of the cervical spine may correlate with variations on the mandibular shape or function, they hypothesized that dental malocclusions could be responsible for functional asymmetry of trunk muscles. Saccucci et al. [24] hypothesized a crucial role for the atlas, which is linked to occipital condyles and thus would affect the rest of the spine alignment. According to the author, any problem in masticatory function might lead to activation of compensatory mechanisms in the spine which might give raise to spinal pathology. Despite some authors reported an association between TMD and the imbalance of the whole body [25–28] probably inducing flexion of the first cervical vertebra and an hyperlordosis of the cervical spine [25] and a positive impact of treatment with occlusal splint application on spine mobility and symptoms [28], Rocha et al. [29] concluded that the evidence between TMD and spinal morbidity was still not clear. Those studies did not establish the causal factor between TMD function and spinal shape but just hypothesized their association and the potential benefits that would follow the treatment with splint by means of a supposed the reduction of cervical antalgic contractures, but the conclusions of those reports were speculative.

The only RCT included in our review stated that in a juvenile population with unilateral posterior crossbite, early orthodontic treatment protocol does not affect negatively postural parameters [19]. By design, it was just a pilot study, and to date we were not able to identify a definitive report of the final study. While some studies supported his conclusions and reported an association [13, 14], other reviews did not find any association [9, 17] or at least any strong association [18]. In particular, Zepa et al. [17], comparing the inclination of the cervical spine and cranial morphology, found that all facial morphologic variables were almost similar in the left-sided and right-sided scoliotic patients. According to the author, thoracic humps or lumbar prominence do not affect the facial symmetry. März et al. [18] performed an optical body scans of 44 subjects, asking them to maintain

seven different mandible positions. Though they found variations of the trunk posture induced by different occlusion modalities applied to patients by splints, no substantial conclusions were drawn.

However, a small group of authors reported a higher incidence of facial and bite anomalies in patients affected by scoliosis [13, 15, 16]. These results seem even more evident in patients with Scheuermann's disease [16] (Scheuermann's disease vs idiopathic scoliosis; 78.3 vs 57.1%, $p=0.00068$), but symptoms related to TMD were more frequently associated with scoliosis. In particular, Scheuermann's disease creates a pathological kyphotic curvature, which is responsible for the forward-tilted head posture and a hunchback appearance, primarily pronounced and stabilized in the dorsal spinal region [30]. The role of head posture tilted forward and backward that evolved as a consequence of the pathological curves of the sagittal plane has been mainly examined in the development of the sagittal and vertical jaw anomalies [31].

Two interesting animal studies recently reported in the literature tested the role of an induced malocclusion in rats. After some weeks, the spine of rats demonstrated some scoliotic degeneration, which remained stable overtime after reversal of the induced dental malocclusion. However, no relationship between the side of malocclusion and the side of curvature was found [32, 33]. Though the findings are appealing, generalizability of the conclusions of this study in an animal model so different to humans is not granted.

This systematic review has limitations. The majority of the studies included in the study did not report cohorts including consecutive patients and a priori sample size calculation. Classification bias might be present as the criteria for the diagnosis of scoliosis, kyphosis and Scheuermann's disease were not described in the studies, and the spinal deformity was frequently diagnosed by clinical examination or surface analysis with optical devices. The gold standard of the assessment of malocclusion and spine deformity, that is full-spine standing radiographs in long cassette and cephalogram, was performed in only two studies. The remaining ones performed mainly clinical examination or instrument of indirect detection of spine deformity such as rasterstereography, which is not reliable for diagnosis of spinal deformity and its severity. Most of the included studies have low quality. The only study included with a high-quality score was a pilot study for a randomized controlled trial, but the goal of the study was to investigate whether the treatment of malocclusion might cause spinal deformity. All these biases, along with the retrospective nature of studies, may influence the evaluation of results.

Conclusion

There is low-quality evidence suggesting higher prevalence of occlusal alterations among patients with spinal deformity than in controls. The studies providing this information have substantial risk of bias. No evidence on causal relationship between the two entities (in both directions) was found. No evidence supporting a therapeutic effect of the correction of malocclusion on spinal deformity was found. Evidence from one good-quality study suggests that the treatment of occlusal disease does not have harmful effects on spinal posture or shape.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

References

1. Isgro M, Buraschi R, Barbieri C et al (2014) Conservative management of degenerative disorders of the spine. J Neurosurg Sci 58(2 Suppl 1):73–76
2. Arienti C, Villafane JH, Donzelli S et al (2017) Trunk and craniofacial asymmetry are not associated in the general population: a cross-sectional study of 1029 adolescents. Eur J Med Res 22(1):36
3. Kouwenhoven JW, Bartels LW, Vincken KL et al (2007) The relation between organ anatomy and pre-existent vertebral rotation in the normal spine: magnetic resonance imaging study in humans with situs inversus totalis. Spine 32(10):1123–1128
4. Villafane JH, Bissolotti L, Zaina F et al (2018) Thoracic hyperkyphosis non invasively measured by general practitioners is associated with chronic low back pain: a cross-sectional study of 1364 subjects. J Bodyw Mov Ther 22(3):752–756
5. Korbmacher H, Eggers-Stroeder G, Koch L et al (2004) Correlations between dentition anomalies and diseases of the of the postural and movement apparatus—a literature review. J Orofac Orthop 65(3):190–203
6. Goldberg CJ, Dowling FE (1991) Idiopathic scoliosis and asymmetry of form and function. Spine 16(1):84–87
7. Ohlendorf D, Seebach K, Hoerzer S et al (2014) The effects of a temporarily manipulated dental occlusion on the position of the spine: a comparison during standing and walking. Spine J 14(10):2384–2391
8. Hong JY, Suh SW, Modi HN et al (2011) Correlation between facial asymmetry, shoulder imbalance, and adolescent idiopathic scoliosis. Orthopedics 34(6):187
9. Kim TH, Kim JH, Kim YJ et al (2014) The relation between idiopathic scoliosis and the frontal and lateral facial form. Korean J Orthod 44(5):254–262
10. Grivas TB, Wade MH, Negrini S et al (2007) SOSORT consensus paper: school screening for scoliosis. Where are we today? Scoliosis 2:17
11. Rainoldi L, Zaina F, Villafane JH et al (2015) Quality of life in normal and idiopathic scoliosis adolescents before diagnosis: reference values and discriminative validity of the SRS-22. A cross-sectional study of 1,205 pupils. Spine J 15(4):662–667

12. Lippold C, Danesh G, Hoppe G et al (2007) Trunk inclination, pelvic tilt and pelvic rotation in relation to the craniofacial morphology in adults. Angle Orthod 77(1):29–35
13. Ben-Bassat Y, Yitschaky M, Kaplan L et al (2006) Occlusal patterns in patients with idiopathic scoliosis. Am J Orthod Dentofacial Orthop 130(5):629–633
14. Korbmacher H, Koch L, Eggers-Stroeder G et al (2007) Associations between orthopaedic disturbances and unilateral crossbite in children with asymmetry of the upper cervical spine. Eur J Orthod 29(1):100–104
15. Segatto E, Lippold C, Vegh A (2008) Craniofacial features of children with spinal deformities. BMC Musculoskelet Disord 9:169
16. Vegh A, Fabian G, Jianu R et al (2012) Orofacial characteristics of adolescents with diagnosed spinal disorders. Biomed Tech (Berl) 57(1):65–69
17. Zepa I, Hurmerinta K, Kovero O et al (2003) Trunk asymmetry and facial symmetry in young adults. Acta Odontol Scand 61(3):149–153
18. Marz K, Adler W, Matta RE et al (2017) Can different occlusal positions instantaneously impact spine and body posture?: a pilot study using rasterstereography for a three-dimensional evaluation. J Orofac Orthop 78(3):221–232
19. Lippold C, Moiseenko T, Drerup B (2012) Spine deviations and orthodontic treatment of asymmetric malocclusions in children. BMC Musculoskelet Disord 13:151
20. Luedtke GL (1970) Management of the dentition of patients under treatment for scoliosis using the Milwaukee brace. Am J Orthod 57(6):607–614
21. Olin W, Ponseti I (2011) Orthodontic considerations for the patient wearing a Milwaukee Brace. Iowa Orthop J 31:22–29
22. D'Attilio M, Epifania E, Ciuffolo F et al (2004) Cervical lordosis angle measured on lateral cephalograms; findings in skeletal class II female subjects with and without TMD: a cross sectional study. Cranio 22(1):27–44
23. Festa F, Tecco S, Dolci M et al (2003) Relationship between cervical lordosis and facial morphology in Caucasian women with a skeletal class II malocclusion: a cross-sectional study. Cranio 21(2):121–129
24. Saccucci M, Tettamanti L, Mummolo S et al (2011) Scoliosis and dental occlusion: a review of the literature. Scoliosis 6:15
25. de Farias Neto JP, de Santana JM, de Santana-Filho VJ et al (2010) Radiographic measurement of the cervical spine in patients with temporomandibular dysfunction. Arch Oral Biol 55(9):670–678
26. Flores HF, Ottone NE, Fuentes R (2017) Analysis of the morphometric characteristics of the cervical spine and its association with the development of temporomandibular disorders. Cranio 35(2):79–85
27. Munhoz WC, Hsing WT (2014) Interrelations between orthostatic postural deviations and subjects' age, sex, malocclusion, and specific signs and symptoms of functional pathologies of the temporomandibular system: a preliminary correlation and regression study. Cranio 32(3):175–186
28. Walczynska-Dragon K, Baron S, Nitecka-Buchta A et al (2014) Correlation between TMD and cervical spine pain and mobility: is the whole body balance TMJ related? Biomed Res Int 2014:582414
29. Rocha CP, Croci CS, Caria PH (2013) Is there relationship between temporomandibular disorders and head and cervical posture? A systematic review. J Oral Rehabil 40(11):875–881
30. Palazzo C, Sailhan F, Revel M (2014) Scheuermann's disease: an update. Joint Bone Spine 81(3):209–214
31. Lippold C, Segatto E, Vegh A et al (2010) Sagittal back contour and craniofacial morphology in preadolescents. Eur Spine J 19(3):427–434
32. D'Attilio M, Filippi MR, Femminella B et al (2005) The influence of an experimentally-induced malocclusion on vertebral alignment in rats: a controlled pilot study. Cranio 23(2):119–129
33. Ramirez-Yanez GO, Mehta L, Mehta NR (2015) The effect of dental occlusal disturbances on the curvature of the vertebral spine in rats. Cranio 33(3):217–227

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