# Progress in the study of the association between craniomaxillofacial deformities and the occurrence of cervical vertebra anomalies

# Pahedeng, Xiaerguli · Muhetaer, Yan Li, Subinuer · Yilihamu, Gulibaha · Maimaitili<sup>\*</sup>

Department of Stomatology, The 2nd Affiliated Hospital of Xinjiang Medical University, Urumqi, Xinjiang, 830028, China \*Corresponding author: gulibaha 1972@163.com

Abstract: Progress in the study of the association between craniomaxillofacial deformities and the occurrence of cervical vertebra anomalies the assessment of cervical vertebra structural features is closely related to orthodontic clinical diagnosis and treatment planning. Lateral cephalometric radiographs are commonly used for orthodontic diagnosis, treatment planning and skeletal maturation assessment, and can also provide additional diagnostic data related to the cranial, facial, and upper cervical spine. In recent years many scholars have studied the prevalence of abnormalities in cervical vertebra structures and different malocclusions and other diseases and found a strong correlation between different skeletal malocclusions and abnormalities in cervical vertebra structures. Also the prevalence of cervical spine abnormalities was statistically higher in patients with skeletal malocclusion type. This article reviews the association between various craniomaxillofacial structural abnormalities and different cervical spine structural features and discusses the importance of cervical vertebrae in craniofacial analysis.

**Keywords:** cephalometric radiographs; malocclusion; cervical vertebra abnormalities; skeletal malocclusion

# 1. Introduction

Several congenital and acquired disorders can affect the skull, face and jaws, resulting in a variety of craniofacial deformities, which usually have a higher probability of occurring at birth or in early infancy [1]. Among them, malocclusion is more common deformities in the overall craniomaxillofacial morphological and structural anomalies. Malocclusion encompasses abnormal maxillary and mandibular relationships, steep mandibular angle (SMA), anterior open bite (AOB), high and narrow palate (HNP), posterior cross bite. In 1899, Mr. Angle, the father of orthodontics, proposed his famous classification of malocclusion, and now the World Health Organization estimates that malocclusion is the third most prevalent oral health problem after dental caries and periodontal disease.

#### 2. Characteristics of malocclusion

#### 2.1 Etiology of malocclusion

Many etiologic factors for malocclusion have been proposed. Genetic, environmental and ethnic factors are the main factors in this regard. Certain types of malocclusion, such as Class III malocclusion relationships are familial, which makes a strong relationship between genetics and malocclusion. Likewise, racial factors are also present, for example, bimaxillary protrusion stands at a higher rate in Africans than in other races. Thus, malocclusion can be considered as a multifactorial problem with no specific etiology so far <sup>[3]</sup>.

# 2.2 Prevalence of malocclusion

The prevalence of malocclusion is relatively high in worldwide and occurs early in childhood.

According to recent studies, the worldwide prevalence of malocclusion was found to be 56% (95% CI: 11-99) with no gender differences. The highest prevalence rates were found in Africa (81%) and Europe (72%), followed by America (53%) and Asia (48%). The prevalence score of malocclusion did not vary from the deciduous dentition period to the permanent dentition period, with a common score of 54%. Characteristics of malocclusion, such as Angle classification, deep overjet, deep overbite and asymmetrical midline shift, largely did not change their prevalence across dental stages. Conversely, features such as crossbite and interdental gaps decreased in prevalence during permanent dentition <sup>[4]</sup>.

# 2.3 Classification of malocclusion

#### 2.3.1 Classification according to the molar relationship

Class I: normal position of the maxillary and mandibular dental arches and normal occlusal relationship of the maxillary and mandibular first molars.

Class II: abnormal relationship of the maxillary and mandibular dental arches, with all mandibular teeth in a distal position in the mandible. Angles considers two subclassifications under Class II:

Class II 1 classification - maxillary anterior teeth protrusion;

Class II 2 classification - maxillary anterior teeth inclined towards the lingually;

Class III: abnormal relationship of the maxillary and mandibular dental arches, with all the teeth of the mandible occlude mesially;

#### 2.3.2 Classification by canine relationship

Class I: maxillary permanent canine should occlude directly between the mandibular permanent canine and the first premolar.

Class II: Maxillary permanent canine occlude mesially to the groove between the mandibular permanent canine and the first premolar.

Class III: The maxillary permanent canine occlude distally to the groove between the mandibular permanent canine and the first premolar <sup>[5]</sup>.

#### 3. Characteristics of the cervical spine

#### 3.1 Structure of the cervical spine

The cervical spine consists of seven cervical vertebrae, from C1 (Cervical Vertebrae 1) to C7 (Cervical Vertebrae 7), which are divided into two main parts: the craniocervical joint (CCJ) and the subcentral vertebrae. CCJ includes vertebrae-Atlas (C1) and the second cervical vertebrae-Axis (C2). The first cervical vertebra (C1) is circumferential in shape, with no vertebral bodies, spinous processes, or articular processes, but consists of an anterior arch, a posterior arch, and two lateral blocks. The second cervical vertebra (C2) is characterized by a cone with an upwardly directed dentition. The third through sixth cervical vertebrae (C3-C4-C5-C6) have a similar morphology, while the seventh cervical vertebra (C7) has a longer posterior arch than the other cervical vertebrae. As a whole, the cervical spine is responsible for supporting the weight of the skull and allowing head and neck movements <sup>[6-7]</sup>.

#### 3.2 Pathologic and physiologic abnormalities of the cervical spine

Abnormal cervical spine morphology occurs in patients with normal occlusal and normal craniofacial morphology, as well as in patients with craniofacial syndromes and severe malocclusion. A recent study found a 14.3% incidence of fusion between the second and third cervical vertebrae (C2-C3) in healthy subjects [8]. Therefore, fusion between cervical vertebrae within this range is considered normal. According to Sandham et al, the cervical spine anomalies (CVA) can be divided into two categories: 1. Fusion anomalies: they can be subdivided into Fusion, Block fusion and Occipitalization. Fusion refers to the fusion of one cervical unit with another vertebral body. The fusion can occur in different parts of the vertebral unit, such as the vertebral body, articular facets, neural arches or transverse processes. Atlanto-occipital fusion refers to the partial or complete union of the first cervical vertebra with the occipital bone. 2. Posterior arch deficiency (PAD), which is divided into vertebral arch cleft and posterior arch hypoplasia. Posterior arch deficiency (PAD) is defined as a fusion of the posterior arch of the cervical bridge (Ponticulus Posticus, PP) is another cervical spine variant,

and Chitroda PK et al. showed that the posterior cervical bridge is an anatomical alteration of the atlantoaxial spine (C1), located in the posterior atlantoaxial arch and associated with the vertebral artery. This is an incidental finding, as seen in the lateral cephalogram of conventional orthodontic treatment. Posterior bridge means "small posterior bridge" in Latin. Other synonyms for posterior bridge are the arcuate foramen, the posterior foramen of the joint and the posterior foramen of the condyle [10]. In addition, there are two types of posterior bridges: partial and complete [11]. Wenger TL et al. found craniosynostosis, midfacial recession, and double handedness with fusion of the second to fourth fingernails in a population with Apert syndrome. A subset of patients had cleft palate and progressive protrusion of multiple bones (skull, hands, feet, wrist bones, tarsus and cervical vertebrae) was also common [12]. Klippel-Feil syndrome (KFS), or congenital cervical fusion, has been considered to be an extremely rare diagnosis. However, recent studies have found an increased prevalence of this condition and a high percentage of asymptomatic individuals. The pathogenesis involves failure of segmentation of the cervical vertebral body segments during embryonic development. Most commonly, the C2-C3 and C5-C6 levels are involved <sup>[13]</sup>.

#### 4. Craniomaxillofacial deformities and the occurrence of cervical vertebra anomalies

#### 4.1 Relationship between malocclusion and structural abnormalities of the cervical spine

Sonnesen et al. recently studied that cervical spine skeletal anomalies can occur in healthy individuals with neutral occlusion and normal craniofacial morphology <sup>[14,15]</sup>, followed by Kim et al. who studied cervical spine morphological deviations in patients with severe skeletal misalignment, bony opening, bony anterior maxillary protrusion, and bony mandibular hypoplasia [16-18]. Anusuya V et al. found that partial cervical spine cleft (PC), fusion and complete cleft were the most frequently found cervical spine anomalies (CVA), while spina bifida (SB) was only found in individuals with a hypodifferentiated growth pattern. Compared to the vertical growth pattern (CVA) tends to be found more often in patients with sagittal growth malocclusion<sup>[19]</sup>.Bedoya A found a relationship between the cranial base structures that determine the sagittal position of the maxilla, mandible and jaw and the cervical vertebral complex and hyoid bone after evaluating 24 images of young patients with Class II, Division 1 malocclusion using spiral CBCT <sup>[20]</sup>.Di Venere D et al. studied the prevalence of PP (posterior cervical bridge) in patients with misalignment and found an 8% prevalence of PP (12 out of 150 cases), with 4.7% and 3.3% of complete and partial forms, respectively. Although females were more affected (9.6%), no significant gender-related differences were found. Analysis of lateral cephalometric films showed that half of the patients with PP were sagittally growing bony class I patients <sup>[21]</sup>. Adisen MZ et al. in the same study found PP to be a relatively common anomaly in patients with dental malocclusion. Although patients with An's class III showed a higher incidence of PP, no statistically significant differences were found between the various malocclusion groups <sup>[22]</sup>. Aranitasi L et al. investigated the relationship between different bony malocclusions and cervical spine anomalies and found that cervical fusion, cleft and partial cleft were the most common cervical spine anomalies. Cleft and partial cleft were found to be statistically significantly different between Class I, II and III. Cervical spine abnormalities and head posture are associated with fusion<sup>[23]</sup>. Bayrakdar IŞ et al. investigated the relationship between posterior atlantoaxial bridge and different bony malunion deformities and found that PP is a common abnormality in Turkish population, associated with different sagittal facet types, and the highest frequency of PP was found in patients with An's class III<sup>[24]</sup>.

#### 4.2 Relationship between cleft lip and palate and cervical vertebra anomalies

Patients with cleft lip and palate are at a higher risk of developing cervical vertebra anomalies than the general population. Rate Recent studies on the possible development of cervical spine abnormalities (CVA) in patients with cleft lip and palate found a higher overall incidence of CVA in patients with cleft lip and palate found that the incidence of cervical spine abnormalities was 20. 3% in the cleft lip and palate group compared to 6.4% in the control group. In addition, the prevalence of cervical spine abnormalities was 16.6% in the CPO group, 19.1% in the BCLP group, and 22.2% in the unilateral cleft lip and palate (UCLP) group. It was concluded that the prevalence of cervical spine abnormalities is higher in patients with cleft lip and palate <sup>[28]</sup>.Bayrakdar IŞ studied the occurrence of ponticulus posticus (pp) in patients with cleft lip and palate and found that although 12 of 54 (22.3%) patients with surgically repaired cleft lip and/or palate in the study group were identified as having PP, 108 (9.2%) of the control group only 10 of the patients had PP. the distribution of the presence of PP between the two groups was statistically significant <sup>[29]</sup>.

# 4.3 Relationship between obstructive sleep apnea, upper airway stenosis and cervical vertebra anomalies

Adisen SR DDS et al. examined the prevalence of cervical spine morphological abnormalities in patients with different bony misalignments and evaluated the relationship between upper airway dimensions and cervical vertebra anomalies. The results of the study showed that the prevalence of CVA was 45.7%. The prevalence was not significantly different from that of skeletal malocclusion (P = 0.89) [<sup>30-32]</sup>. Zheng ZH et al. studied the structural characteristics of the upper airway in patients with different sagittal malocclusion and found a significant relationship between pharyngeal airway (PA) volume and Min-CSA and sagittal malocclusion (P < 0.05). Class I and III subjects had Nasopharyngeal airway (NA) volume was significantly greater than that of Class II subjects (P < 0.05) [<sup>33]</sup>. Claudino LV et al. found that the minimum and mean areas (hypopharynx, velopharynx, and oropharynx) were smaller in Class II subjects than in Class III, and the velopharynx was significantly less homogeneous in morphology than in Class I and Class III groups. A negative correlation was observed between ANB values and airway volumes in the hypopharynx and velopharynx (both males and females) and the oropharynx (male subjects only) [<sup>34</sup>].

#### 4.4 Relationship between abnormal condylar morphology and cervical vertebra anomalies

Kim JR et al. investigated the association between temporomandibular disorders (TMD) and cervical vertebra anomalies and found that subjects with condylar hypoplasia had significantly more cervical spine morphological deviations and deviations from the pattern compared to normal craniofacial morphology, as evidenced by the positive correlation between increased cranial base angle, cervical lordosis, upper cervical tilt, and cranial base angle and cervical column fusion. [35]. The results of the study by Hong SW et al. suggest that degenerative cervical changes were associated with changes in head posture and the development of active TrP in craniocervical musculature in older adults with myofascial TMD. In addition to this Kang JH et al. found an association between the duration of orofacial pain, the number of active TrPs in the trapezius muscle, the intensity of neck pain, the distance between the occipital bone and the atlas and migraine in adolescent patients with TMD. [36].

In summary, orthodontists often take cephalometric X-rays to diagnose cranial and maxillofacial abnormalities in order to determine the specific maxillofacial deformity of a patient, and cervical spine growth and development is often observed from cephalometric X-rays to estimate whether the patient has growth potential, and based on these aspects orthodontists often find cervical spine deformities first. The relationship between cervical spine deformities and craniomaxillofacial deformities is gradually being elucidated. Exploring the relationship between craniomaxillofacial deformities, and secondly, it can help to diagnose cervical spine deformities and thus prevent and avoid complications caused by cervical spine deformities through early treatment.

#### References

[1] Woolley EJ, Richardson D, May P. Management of craniofacial abnormalities. Hosp Med. 2005 Jul; 66(7): 405-10.

[2] D'Onofrio L. Oral dysfunction as a cause of malocclusion. Orthod Craniofac Res. 2019 May; 22 Suppl 1(Suppl 1): 43-48.

[3] Alhammadi MS, Halboub E, Fayed MS, Labib A, El-Saaidi C. Global distribution of malocclusion traits: A systematic review. Dental Press J Orthod. 2018 Nov-Dec; 23(6): 40.e1-40.e10.

[4] Lombardo G, Vena F, Negri P, Pagano S, Barilotti C, Paglia L, Colombo S, Orso M, Cianetti S. Worldwide prevalence of malocclusion in the different stages of dentition: A systematic review and metaanalysis. Eur J Paediatr Dent. 2020 Jun; 21(2): 115-122.

[5] Ramakrishnan, KarthikScott, CynthiaSarda, Pavithra K.Vivek, NarayananSaravanan, Chandran. Association between shape of the mandibular condylar head and the occurrence of unilateral condylar fracture - A retrospective computed tomographic study[J]. Journal of Cranio-Maxillofacial Surgery, 2021, 49(6).

[6] Fei O, Shi-Jun D, Jin-Ge Z, et al. Clinical application of individual digital reconstruction technology in cranio-maxillofacial deformities[J]. Journal of Clinical Rehabilitative Tissue Engineering Research, 2010, 14(22):4032-4035.DOI:10.3969/j.issn.1673-8225.2010.22.010.

[7] Pastor-Pons I, Hidalgo-García C, Lucha-López MO, Barrau-Lalmolda M, Rodes-Pastor I, Rodríguez-Fernández ÁL, Tricás-Moreno JM. Effectiveness of pediatric integrative manual therapy in

cervical movement limitation in infants with positional plagiocephaly: a randomized controlled trial. Ital J Pediatr. 2021 Feb 25; 47(1): 41.

[8] Sonnesen L, Pedersen CE, Kjaer I. Cervical column morphology related to head posture, cranial base angle, and condylar malformation. Eur J Orthod. 2007 Aug; 29(4): 398-403.

[9] Dogan E, Ergican GO, Dogan S. Evaluation of the cervical vertebral anomalies in patients with cleft lip and palate in Aegean region of Turkey. J Pak Med Assoc. 2021 Jan; 71(1(B)): 215-218.

[10] Chitroda PK, Katti G, Baba IA, Najmudin M, Ghali SR, Kalmath B, G V. Ponticulus posticus on the posterior arch of atlas, prevalence analysis in symptomatic and asymptomatic patients of gulbarga population. J Clin Diagn Res. 2013 Dec; 7(12): 3044-7.

[11] Elliott RE, Tanweer O. The prevalence of the ponticulus posticus (arcuate foramen) and its importance in the Goel-Harms procedure: meta-analysis and review of the literature. World Neurosurg. 2014 Jul-Aug; 82(1-2): e335-43.

[12] Wenger TL, Hing AV, Evans KN. Apert Syndrome. 2019 May 30. In: Adam MP, Mirzaa GM, Pagon RA, Wallace SE, Bean LJH, Gripp KW, Amemiya A, editors. GeneReviews®[Internet]. Seattle (WA): University of Washington, Seattle; 1993–2023.

[13] Litrenta J, Bi AS, Dryer JW. Klippel-Feil Syndrome: Pathogenesis, Diagnosis, and Management. J Am Acad Orthop Surg. 2021 Nov 15; 29(22): 951-960.

[14] Sonnesen L, Jasemi A, Gjørup H, Daugaard-Jensen J. Upper cervical spine and craniofacial morphology in hypohidrotic ectodermal dysplasia. Eur Arch Paediatr Dent. 2018 Oct; 19(5): 331-336.

[15] Oh E, Ahn SJ, Sonnesen L. Ethnic differences in craniofacial and upper spine morphology in children with skeletal Class II malocclusion. Angle Orthod. 2018 May; 88(3): 283-291.

[16] Kim P, Sarauw MT, Sonnesen L. Cervical vertebral column morphology and head posture in preorthodontic patients with anterior open bite. Am J Orthod Dentofacial Orthop. 2014 Mar; 145(3): 359-66.

[17] Bedoya A, Landa Nieto Z, Zuluaga LL, Rocabado M. Morphometry of the cranial base and the cranial-cervical-mandibular system in young patients with type II, division 1 malocclusion, using tomographic cone beam. Cranio. 2014 Jul; 32(3): 199-207.

[18] Kamak H, Yildırım E. The distribution of cervical vertebrae anomalies among dental malocclusions. J Craniovertebr Junction Spine. 2015 Oct-Dec; 6(4): 158-61.

[19] Anusuya V, Sharan J, Jena AK. A study of cervical vertebra anomalies among individuals with different sagittal and vertical facial growth patterns. J Craniovertebr Junction Spine. 2020 Apr-Jun; 11(2): 75-80.

[20] Bedoya A, Landa Nieto Z, Zuluaga LL, Rocabado M. Morphometry of the cranial base and the cranial-cervical-mandibular system in young patients with type II, division 1 malocclusion, using tomographic cone beam. Cranio. 2014 Jul; 32(3): 199-207.

[21] Di Venere D, Laforgia A, Azzollini D, Barile G, De Giacomo A, Inchingolo AD, Rapone B, Capodiferro S, Kazakova R, Corsalini M. Calcification of the Atlanto-Occipital Ligament (Ponticulus Posticus) in Orthodontic Patients: A Retrospective Study. Healthcare (Basel). 2022 Jul 2; 10(7): 1234.

[22] Adisen MZ, Misirlioglu M. Prevalence of ponticulus posticus among patients with different dental malocclusions by digital lateral cephalogram: a comparative study. Surg Radiol Anat. 2017 Mar; 39(3): 293-297.

[23] Aranitasi L, Tarazona B, Zamora N, Gandía JL, Paredes V. Influence of skeletal class in the morphology of cervical vertebrae: A study using cone beam computed tomography. Angle Orthod. 2017 Jan; 87(1): 131-137.

[24] Bayrakdar IŞ, Miloğlu Ö, Yeşiltepe S, Yılmaz AB. Ponticulus posticus in a cohort of orthodontic children and adolescent patients with different sagittal skeletal anomalies: a comparative cone beam computed tomography investigation. Folia Morphol (Warsz). 2018; 77(1): 65-71.

[25] de Rezende Barbosa GL, Pimenta LA, Tyndall DA, Allareddy TV, Sousa Melo SL. Three-Dimensional Assessment of Cervical Vertebrae Anomalies in Patients with Cleft Lip and Palate. Cleft Palate Craniofac J. 2021 Sep; 58(9): 1102-1109.

[26] Berrocal C, Terrero-Pérez Á, Peralta-Mamani M, Fischer Rubira-Bullen IR, Honório HM, de Carvalho IMM, Alvares Capelozza AL. Cervical vertebrae anomalies and cleft lip and palate: a systematic review and meta-analysis. Dentomaxillofac Radiol. 2019 Dec; 48(8): 20190085.

[27] Uğar DA, Semb G. The prevalence of anomalies of the upper cervical vertebrae in subjects with cleft lip, cleft palate, or both. Cleft Palate Craniofac J. 2001 Sep; 38(5): 498-503.

[28] Datana S, Bhalla A, Kumar P, Kumar Roy S, Londhe S. Comparative evaluation of prevalence of upper cervical vertebrae anomalies in cleft lip/palate patients: a retrospective study. Int J Clin Pediatr Dent. 2014 Sep-Dec; 7(3): 168-71.

[29] Bayrakdar IŞ, Yasa Y, Duman ŞB, Karaturgut UE, Ocak A, Günen Yılmaz S. Cone beam computed tomography evaluation of ponticulus posticus in patients with cleft lip and palate: a retrospective radio-

anatomic study. Folia Morphol (Warsz). 2018; 77(1): 72-78.

[30] Adisen SR DDS, Adisen MZ DDS, PhD, Ozdiler FE DDS, PhD. The evaluation of the relationship between cervical vertebral anomalies with skeletal malocclusion types and upper airway dimensions. Cranio. 2020 May; 38(3): 149-157.

[31] Chokotiya H, Banthia A, K SR, Choudhary K, Sharma P, Awasthi N. A Study on the Evaluation of Pharyngeal Size in Different Skeletal Patterns: A Radiographic Study. J Contemp Dent Pract. 2018 Oct 1; 19(10): 1278-1283. PMID: 30498186.

[32] Sheppard SE, Quintero-Rivera F. Wiedemann-Steiner Syndrome. 2022 May 26. In: Adam MP, Mirzaa GM, Pagon RA, Wallace SE, Bean LJH, Gripp KW, Amemiya A, editors. GeneReviews® [Internet]. Seattle (WA): University of Washington, Seattle; 1993–2023.

[33] Zheng ZH, Yamaguchi T, Kurihara A, Li HF, Maki K. Three-dimensional evaluation of upper airway in patients with different anteroposterior skeletal patterns. Orthod Craniofac Res. 2014 Feb; 17(1): 38-48.

[34] Claudino LV, Mattos CT, Ruellas AC, Sant' Anna EF. Pharyngeal airway characterization in adolescents related to facial skeletal pattern: a preliminary study. Am J Orthod Dentofacial Orthop. 2013 Jun; 143(6): 799-809.

[35] Kim JR, Jo JH, Chung JW, Park JW. Upper cervical spine abnormalities as a radiographic index in the diagnosis and treatment of temporomandibular disorders. Oral Surg Oral Med Oral Pathol Oral Radiol. 2020 May; 129(5): 514-522.

[36] Kang JH. Neck associated factors related to migraine in adolescents with painful temporomandibular disorders. Acta Odontol Scand. 2021 Jan; 79(1): 43-51.