THE OROFACIAL MUSCULOSKELETAL STRUCTURES AND FUNCTIONS IN CEREBRAL PALSY - A SCOPING REVIEW

By

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Maa, Paa and Tarana
This one’s for you

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ABSTRACT

Background: Bones adapt their shape and structure to best resist the mechanical environment. However, mechanobiological adaptions can backfire in individuals with abnormal biomechanics, such as children with cerebral palsy (CP). Abnormal muscle tone and movement in CP contribute to a progressive musculoskeletal disorder such as lower limb deformities. Many individuals with CP also exhibit orofacial dysfunction, which describes difficulty controlling saliva, swallowing, chewing and speaking. Orofacial literature in CP presents knowledge on the musculoskeletal structures and functions, separately, based on their scientific disciplines. However, there is no report to date relating the deformations in orofacial structures (muscles and bones) to orofacial (dys)functions in CP, that has a multidisciplinary perspective.

Objectives: To aggregate knowledge across all disciples, that quantitatively assessed the orofacial musculoskeletal structures and/or orofacial (dys)functions in Cerebral Palsy, through a scoping review.

Methods: A scoping review was conducted and the PRISMA-ScR checklist was followed. The search was performed in four databases: Medline (Ovid), Embase, CINAHL and Scopus. Titles and abstracts were first screened followed by a full-text review based on the eligibility criteria. From the final list of studies, knowledge was then aggregated and synthesized.

Results: Thirty-nine studies met the inclusion/exclusion criteria. All included studies are cross-sectional in design, with only two including a power analysis to estimate sample size. In terms of discipline, fifteen studies belonged to Dentistry, eleven studies were from Speech & Communication, ten studies involve Dysphagia and three studies focus on Physiology. Of these articles, nine studies quantify skeletal and dental arch characteristics (e.g., cephalometry, model analysis), while no studies investigate muscle morphology. Twelve studies assess orofacial kinematics through motion capture of facial structures. Nineteen studies evaluate muscular activity during functions like speaking or mastication. Six studies used kinetic measures to evaluate bite force and lip pressure. The studies revealed greater path distances, velocity and spatio-temporal indices (STIs) in CP compared to typically developing (TD) individuals for Jaw Kinematics. Muscle Activity in CP was evaluated to be less synchronized, more irregular, and variable but was largely dependent on the type of speech tasks and diets consumed during mastication. Cephalometric characteristics for CP revealed larger SNA (angle describing spatial relationship between the maxilla and cranial base), shorter cranial base length and mandibular ramus height perceived as increased vertical facial dimensions in CP compared to TD. Kinetic analysis revealed lower forces generated across all tasks for CP compared to TD, tracked using various measurement techniques.

Conclusions: From the current scoping review, it is clear that CP is heterogeneous in its oral manifestations. As the head and neck structures are used for a variety of functions, a multidisciplinary approach is required to investigate the implications on health in individuals with CP. The review also identifies the knowledge gaps in the literature regarding the orofacial muscle structure assessment in individuals with CP. This multidisciplinary knowledge will guide future studies in this area.

RÉSUMÉ

Contexte: Les os adaptent leur forme et leur structure pour mieux résister à l'environnement mécanique. Cependant, les adaptations mécanobiologiques peuvent se retourner contre les personnes ayant une biomécanique anormale, comme les enfants atteints de paralysie cérébrale (PC). Un tonus musculaire et des mouvements anormaux dans la PC contribuent à un trouble musculo-squelettique progressif tel que des déformations des membres inférieurs. De nombreuses personnes atteintes de PC présentent également un dysfonctionnement orofacial, qui décrit la difficulté à contrôler la salive, la déglutition, la mastication et la parole. La littérature orofaciale en CP présente les connaissances sur les structures et les fonctions musculo-squelettiques, séparément, en fonction de leurs disciplines scientifiques. Cependant, il n'existe pas à ce jour de rapport de compréhension des déformations des structures orofaciales (muscles et os) aux (dys)fonctions orofaciales dans la PC, qui ait une perspective multidisciplinaire.

Objectifs: Agréger les connaissances de tous les disciples, qui ont évalué quantitativement la structure musculo-squelettique orofaciale et/ou les (dys)fonctions orofaciales dans la plasie cérébrale, par le biais d'un examen de la portée.

Méthodes: Un examen de la portée a été effectué et la liste de contrôle PRISMA-ScR a été suivie. La recherche a été effectuée dans quatre bases de données : Medline (Ovid), Embase, CINAHL et Scopus. Les titres et les résumés ont d'abord été examinés, suivis d'un examen du texte intégral basé sur les critères d'éligibilité. À partir de la liste finale des études, les connaissances ont ensuite été agrégées et synthétisées.

Résultats: Trente-neuf études remplissaient les critères d'inclusion/exclusion. Toutes les études incluses sont de conception transversale, avec seulement deux comprenant une analyse de puissance pour estimer la taille de l'échantillon. En termes de discipline, quinze études appartenaient à la dentisterie, onze études provenaient de la parole et de la communication, dix études concernaient la dysphagie et trois études se concentraient sur la physiologie. Parmi ces articles, neuf études quantifient les caractéristiques des arcades squelettiques et dentaires (par exemple, céphalométrie, analyse de modèles), tandis qu'aucune étude n'étudie la morphologie musculaire. Douze études évaluent la cinématique orofaciale par capture de mouvement des structures faciales. Dix-neuf études évaluent l'activité musculaire lors de fonctions comme la parole ou la mastication. Six études ont utilisé des mesures cinétiques pour évaluer la force de morsure et la pression des lèvres. Les études ont révélé des distances de trajet, une vitesse et des STI plus élevés dans CP par rapport à TD pour Jaw Kinematics. L'activité musculaire dans la CP a été évaluée comme étant moins synchronisée, plus irrégulière et variable, mais dépendait largement du type de tâches de parole et des régimes consommés pendant la mastication. Les caractéristiques céphalométriques pour le CP ont révélé un SNA plus grand, une longueur de base crânienne plus courte et une hauteur de ramus mandibulaire perçues comme des dimensions faciales verticales accrues dans le CP par rapport au TD. L'analyse cinétique a révélé des forces plus faibles générées dans toutes les tâches pour CP par rapport à TD, suivies à l'aide de diverses techniques de mesure.

Conclusions: D'après l'examen de portée actuel, il est clair que la PC est hétérogène dans ses manifestations orales. Comme les structures de la tête et du cou sont utilisées pour une variété de fonctions, une approche multidisciplinaire est nécessaire pour étudier les implications sur la santé des personnes atteintes de PC. La revue identifie également les lacunes dans les connaissances

dans la littérature concernant l'évaluation de la structure musculaire orofaciale chez les personnes atteintes de PC. Ces connaissances multidisciplinaires guideront les futures études dans ce domaine.

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PREFACE TO THESIS FORMAT

This thesis has been written in a traditional format.

CONTRIBUTION OF AUTHORS

Akanksha D Cambala: Master's candidate: Faculty of Dental Medicine and Oral Health Sciences, McGill University, Montreal (QC), Canada: Conceived the study objective, prepared review protocol, performed data collection and interpretation, and wrote the thesis.

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LIST OF ABBREVIATIONS

AC	Akanksha Cambala
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CP	Cerebral Palsy
CONSORT	Consolidated Standards of Reporting Tools
DD	Developmental Delay
EMBASE	Excerpta Medica database
EMG	Electromyography
EZ	Elizabeth Zimmermann
GMFCS.	Gross Motor Function Classification System
MEDLINE	Medical Literature Analysis and Retrieval System Online
MeSH	Medical Subject Headings
MM	Martin Morris
MBF	Maximum Bite Force
MUAP	Motor Unit Action Potential
MVC	Maximal Voluntary Clench
NK	Nguyen Khang
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analysis
PRISMA-ScR	Preferred Reporting Items for Scoping Review Protocol
sEMG	surface Electromyography
TD	Typically Developing
TMJ	Temperomandibular Joint

CHAPTER 1: INTRODUCTION

Cerebral Palsy (CP) is a non-progressive neurological disorder resulting from injury to the immature brain and is considered to be the most common motor disability in childhood [1]. It is characterized by the abnormal muscle tone, posture and movement [2] resulting in secondary musculoskeletal disorders.

CP is heterogeneous in presentation and severity. Children with CP have deficits in the gross motor function [3], which impact their mobility, quality of life and capacity for self-care. Less studied are the effects of CP on the head (orofacial) and neck. These regions are vital for functions like mastication, swallowing, breathing, controlling saliva, facial expression and communication. Orofacial dysfunction describes dysfunction in the tongue, lips, palate, face and throat [4]. Around 85% of children diagnosed with CP have feeding difficulties, 65% have deficient mastication abilities and around 40% have dysfunctions related to the voluntary saliva control [5, 6].

Orofacial dysfunction has a salient impact on the health of individuals with CP [7]. Impaired eating due to a lack of efficient chewing and swallowing in CP leads to nutritional imbalances, impairs growth and may jeopardize respiration [8-11]. The most common causes of death in young individuals with CP are secondary respiratory diseases [10, 11].

Rationale

It is known that the abnormal muscle tone in CP leads to progressive musculoskeletal disorders, including hip and lower limb bone deformities and low bone mass. In CP, due to the abnormal muscle activity and forces, the orofacial (dys)functions could result in *orofacial bone deformities* that could contribute to or worsen comorbidities. To date, the research on CP is conducted in a way that separately investigates the disorder based on different scientific disciplines, for example, speech and communication, medicine- dysphagia or dentistry. The literature includes quantitative measurements of the orofacial bones and quantitative measurements of orofacial (dys)functions (e.g., kinematics, kinetics, muscle activity) in different contexts (e.g., speech, mastication, breathing). Abnormal muscle tone in CP-related dysfunctions may potentially lead to orofacial bone defects.

Objective

This study aims to aggregate knowledge across all disciplines, that quantitatively assessed the orofacial musculoskeletal structures and/or orofacial (dys)functions in Cerebral Palsy, through a scoping review.

CHAPTER 2: LITERATURE REVIEW

According to the literature, the normal development of the human craniofacial complex involves the mutual interaction between the – (i) brain with its associated sensory organs and the basicranium, (ii) the facial and pharyngeal airway systems and the (iii) oral complex. The absence of this interaction would lead to an imbalance in the normal physiological mechanisms and clinically manifest as functional deficits [12-14]. To understand the principles of this equilibrium, the fundamentals of the structure-function relationships are elaborated.

The orofacial region, a constituent of the craniofacial structure, is comprised of the structures in the head and neck, i.e., the muscles, bones, vasculature and neural components.

2.1 Anatomy: Orofacial structures

The facial surface of the skull has 14 bones namely, the zygomatic, lacrimal, nasal, inferior nasal conchae, palatine bone, maxilla, vomer and mandible. All of them occur in pairs except for the vomer and the mandible. The orofacial region is formed by the middle and lower thirds of the face, primarily composed of the maxilla and the mandible.

The orofacial region is comprised of a

(1) Musculoskeletal component and

(2) Dental component.

The musculoskeletal component, as the name suggests, is composed of the muscles and the bones. The dental component includes the structures of the oral cavity like the teeth and its periodontium. All these structures along with the vasculature work as a structural unit to perform various orofacial functions. One can say that the structures of this orofacial complex are highly organized and have distinct shapes based on their location and function [15, 16].

2.1.1 Bones (Musculoskeletal component)

The midface is predominantly formed by the Maxilla. It is located centrally and is known to provide structural support to the viscerocranium (collection of bones that make up the skull). The maxilla not only contributes by providing the fundamental architecture of the face but also provides functional support by separating the oral and nasal cavities from the upper jaw. The maxilla is associated with its surrounding facial structures through four processes namely the *alveolar, frontal, zygomatic and palatine processes*. The maxilla is pyramidal in shape, with its base adjacent to the nasal cavity, the zygomatic process serving as the apex and its body comprising the maxillary sinus. On its superior aspect, the maxilla articulates with the frontal bone, laterally the zygomatic bone, the palatine bone posteriorly and the alveolar process inferiorly which also supports the teeth [17-19].

Forming the lower third of the face, the mandible is the largest bone in the human skull [20]. It comprises of the body, ramus, condylar and coronoid processes. It constitutes the lower dental arch anchoring the teeth and presents a large surface area for the attachment of various muscles, responsible for orofacial functions. The body forms a horizontally curved portion and the external border accounts for the jawline [21]. The rami are two vertical processes that join the body at the angle of the mandible. The ramus contributes to the lateral portion of the mandible on either side. The coronoid process and condyloid process are located at the superior aspect of the ramus. The coronoid process is anterior and the condyloid process is posterior; the two are separated by the mandibular notch. At the superior aspect of each ramus, the coronoid and condylar processes articulate with the temporal bone to create the temporomandibular joint which permits its mobility. The part of the mandible where the body ends and the rami begin, at the angle of the mandible, is known as the gonial angle (angle made between the mandibular ramus and the external mandibular plane) [17-19].

2.1.2 Muscles (Musculoskeletal component)

The muscles of the orofacial region are classified based on their functional demands. However, the muscular system of the orofacial region is complex and performs integral functions, due to the usage of the same muscle groups in multiple functions [15, 16, 19, 21-23].

The masseter, lateral and medial pterygoid and temporalis form the *muscles of mastication*. These muscles bring about the primary functions of mastication, chewing and general movements of the jaws.

The muscles of the tongue include, 1) genioglossus, hyoglossus, styloglossus and palatoglossus (extrinsic muscles) 2) superior longitudinal muscles, inferior longitudinal muscles, transverse muscles and vertical muscle (intrinsic muscles). These muscles are responsible for the movement of the tongue during speech, swallowing and mastication.

These *buccolabial (perioral)* muscles belong to the lips and muscles around the mouth. They bring about functions like facial expressions around the mouth and speech and include the following muscles:

- levator labii superioris, levator labii superioris alaeque nasi, risorius, levator anguli oris, zygomaticus major and zygomaticus minor muscles. – for elevating and everting the upper lip

- depressor labii inferioris, depressor anguli oris and mentalis muscles - for dressing and everting the lower lip
- orbicularis oris muscle - for closing the lips
- buccinator muscle – responsible for compressing the cheek

The *muscles of the pharynx* include, the superior, middle and inferior constrictors, palatopharyngeus, salpingopharyngeus and stylopharyngeus. These muscles line the walls of the oral cavity and the esophagus that form the oropharynx, nasopharynx and laryngopharynx. They support mastication and the subsequent swallowing mechanism as well as the movement of the epiglottis.

The *muscles of the larynx* include extrinsic and intrinsic muscles. The extrinsic muscles (infrahyoid and suprahyoid) are associated with the hyoid bone and bring about its movements. The infrahyoid muscles include the sternohyoid, omohyoid, sternothyroid and thyrohyoid muscle. The suprahyoid muscles include the stylohyoid, digastric, mylohyoid and geniohyoid. The intrinsic muscles are associated with the vocal cords and are responsible for speech functioning. The muscles of the pharynx and larynx, collectively bring about a respiration-swallowing complex through their coordinated movements. This also prevents involuntary aspiration during swallowing [15, 16, 19, 21-23].

2.1.3 Vasculature and Nervous System

The muscles of the face are predominantly perfused by branches of the external carotid artery, particularly branches of the facial artery. The muscles of facial expression are innervated by the facial nerve, whereas the muscles of mastication receive innervation from the mandibular division of the trigeminal nerve [16].

2.1.4 Dental Component

The dental component of the orofacial region is comprised of the teeth and periodontium. The periodontium is composed of four different tissue types that vary in composition, the gingiva, alveolar bone, cementum, and periodontal ligament. The Alveolar bone is that part of the maxilla and mandible which supports the teeth by anchoring the fibres of the periodontal ligament [24].

At the microarchitectural level, the alveolar bone consists of two plates of cortical bone separated by a spongy bone. In some areas, the alveolar bone is thin with no spongy bone. The alveolar bone and the cortical plates are the thickest in the mandible [25]. The spaces between the trabeculae of the spongy bone are filled with marrow, which consists of hematopoietic tissue in early life and fatty tissue later. The shape and structure of the trabeculae may reflect the stress-bearing requirements of the particular site. The surfaces of the bone are lined by *osteoblasts,* which are responsible for bone formation. The cells which become incorporated within the mineral tissue are called osteocytes and maintain contact with each other via canaliculi. The osteoclasts are responsible for bone resorption [24].

2.2 Bone Growth Concepts

2.2.1 Craniofacial Bone Growth

The facial skeleton is generally believed to expand continuously throughout life [26, 27]. Here, we focus on post-natal growth. Facial growth begins with the distinction between the basic kinds of growth movement at a cellular level.

Bone remodeling is the process of spatially coordinated bone resorption followed by the sequential deposition of bone, usually observed for skeletal renewal [28]. The process by which a bone grows is referred to as bone modeling, where the bone tends to change its shapes based on the surface resorption or formation [28]. *Bone modelling* contributes to the growth observed in the craniofacial skeleton [29]. *Displacement* is the movement of the bone as a whole unit. In the process of displacement, as a bone enlarges, it is simultaneously carried away from other bones by creating a "space" within which the bony enlargement takes place. When the bone gets displaced as a result of its growth, it is called *Primary displacement*. If the bone gets displaced as a result of growth and enlargement of the adjacent bone, it is called *Secondary displacement*. Both processes occur simultaneously [29]. In the craniofacial region, displacements are studied in orthodontics and account for the rapid maxillary expansion process [30].

To understand bone growth at a structural level, it is also necessary to understand: (1) the location or growth sites, (2) the type of growth that occurs in the location, and (3) the factors that determine or control growth [29].

Growth sites: The craniofacial complex can be separated [29] into four areas that grow rather differently: (1) the cranial vault, the bones that cover the upper and outer surface of the brain; (2) the cranial base, the bony floor under the brain, which also is the dividing line between the cranium and the face; (3) the naso-maxillary complex, made up of the nose, maxilla, and associated small bones; (4) the mandible. In the cranial vault, the *sutures* are the most important structures where growth occurs. Sutures are the periosteum-lined contact areas between adjacent skull bones [31]. At the Cranial base, the sphenooccipital synchondrosis, the nasal septal cartilage in the nasomaxillary complex, and the condylar cartilage in the mandible play important roles as major *growth sites* for the respective anatomical components [32, 33].

Among these, the condylar cartilage acts as the center of greatest growth in the craniofacial complex and is associated with morphogenesis of the craniofacial skeleton and temporomandibular joint function [34]. Condylar cartilage, which is designated as secondary cartilage, differs from other primary cartilages in the histological organization; modes of proliferation, differentiation and calcification; and response to environmental factors (e.g., biomechanical stress, hormones and growth factors) [35].

Types of bone growth: (1**)** Endochondral ossification- Endochondral ossification is a type of ossification that proceeds through the formation of intermediate cartilage. Generally, this intermediate cartilage is hyaline cartilage. Here, the cartilage only serves as a template. Endochondral ossification is involved in the formation of long bones as well as the bones at the base of the skull [36]; (2) Intramembranous ossification- Intramembranous ossification is the type of ossification in which the compact and spongy bones directly develop on a sheet of mesenchyme. The formation of flat bones in the face, skull and clavicle occurs through intramembranous ossification [31].

2.2.2 Orofacial Bone Growth

(1) Maxilla (Nasomaxillary complex)

The maxilla develops postnatally entirely by intramembranous ossification [31]. Since there is no cartilage involvement and replacement, the growth occurs in two ways: (1) apposition of bone at the sutures that connect the maxilla to the cranium and cranial base, and (2) by surface modelling.

The sutures remain the same width, and the various processes of the maxilla become longer. Bone apposition occurs on both sides of a suture, so the bones to which the maxilla is attached also become larger. Part of the posterior border of the maxilla is a free surface in the tuberosity region. Bone is added at this surface, creating additional space into which the primary and then the permanent molar teeth successively erupt.

Interestingly, as the maxilla grows downward and forward, its anterior surfaces are remodelled where the bone is removed from most of the anterior surface. As displacement proceeds downward and forward, an equivalent amount of maxillary remodeling simultaneously takes place in an opposite upward and backward direction (i.e., toward its contact with the cranial floor)[31].

(2) Mandible

In contrast to the maxilla, both endochondral and modeling activity are important in the growth of the mandible. The displacement created by the movement of the temporomandibular joint (TMJ) plays a negligible role. The cartilage that covers the surface of the mandibular condyle at the TMJ is not like the cartilage at an epiphyseal plate or a synchondrosis that undergoes hyperplasia, hypertrophy, and endochondral replacement. All other areas of the mandible are formed and grow by direct surface apposition and remodeling backward [28, 37]. As this occurs, the condyle and ramus grow upward and backward thereby, “relocating into the space" created by the displacement process [38]. Note that the ramus also remodels in shape and size as it relocates postero-superiorly, i.e., it becomes longer and wider to accommodate (1) the increasing mass of masticatory muscles inserted onto it, (2) the enlarged breadth of the pharyngeal space, and (3) the vertical lengthening of the nasomaxillary part of the growing face [29, 35, 36, 39].

2.2.3 How Bones Respond to Mechanical Forces- Mechano-Adaptation

In healthy bones, there is a regulatory rate of absorption and deposition of bone tissue which plays an important role in maintaining the total bone mass and thereby the strength of the bone.

When strain levels within the bones increase or decrease past a certain level, bones must undergo adaption in their shape and architecture in order to perform their primary function [40], (ie.), act by efficiently providing structural support, conduct as rigid levers for the muscles to pull against, and remain as light as possible to allow efficient movements and locomotion. This is referred to as *Mechano adaptation* of bone [40, 41].

During functional movements and exercise, bones are exposed to mechanical loading, one of the crucial triggering factors for bone remodeling that adapts the bones to the loading condition [37]. According to the amount of strain (i.e., tissue level deformation), bones tend to adjust their size and thus their strength in order to carry the workload without breaking suddenly or in fatigue [42]. Exercise is known to increase bone mass and density; on the other hand, unloading causes a decrease in bone mass [43]. An example of a change in mechanical loading and its impact on the development of the skeleton are reflected in skeletal modifications in athletes as a result of increased loading. Therefore, the fundamental process of bone deposition and absorption begins to align with the mechanical loading/stress patterns [44, 45].

At a cellular level, it is the activity of the osteoblasts and osteoclasts that help in the maintenance of this process through bone formation and bone resorption. However, osteocytes are thought to sense such mechanical changes and coordinate bone formation and resorption. [46, 47]. Thus, bone tissue is remodelled in response to mechanical loading, which is largely dependent on osteocytes [48].

2.2.4 Skeletal and Dental Characteristics of the Craniofacial Bone

Cranial base morphology affects the skeletal form (profile) and occlusal characteristics [49, 50]. To understand the arrangement of the skeletal structures in the craniofacial region, it is fundamental to describe the cephalometric dental and skeletal classifications.

(1) Skeletal Classification: describes the relation of the maxilla and mandible with respect to the cranial base, in the anteroposterior direction

Class I: Maxilla and mandible are in harmony with each other

Class II: Maxilla is ahead of the mandible with reference to the anterior cranial base, or the maxilla is prognathic with a retruded mandible

Class III: Mandible is ahead of the maxilla with reference to the anterior cranial base, or the mandible is prognathic with a retruded maxilla

(2) Angle’s Classification of malocclusion [51]: describes the positioning of the mesiobuccal cusp of the upper first molar occludes with the buccal groove of the lower first molar

Class I: A normal molar relationship is present

Class II: The mesiobuccal cusp of the maxillary first molar occludes anterior to the buccal groove of the mandibular first molar. Class II is categorized into two further parts:

- Class II, Division 1: The anterior maxillary teeth are tilted forward or proclined, presenting a large overjet.
- Class II, Division 2: The anterior maxillary teeth are retroclined, creating a deep overbite.

Class III.: The mesiobuccal cusp of the upper first molar falls posterior to the buccal groove of the lower first molar

2.2.5 Quantitative Assessment Techniques

Cephalometry:

In orthodontics, cephalometry or cephalometric analysis, is used to evaluate the spatial relationships of the dental and skeletal components of the craniofacial region. In this technique, lateral cephalograms are used to evaluate growth or for orthodontic diagnosis and treatment planning. Cephalometric landmarks represent the skeletal tissue landmarks and serve as reference points to analyze and measure the linear and angular dimensions of the skull. There are several types of analytical approaches by various authors that serve as tools for cephalometry, (Steiner's Analysis [52], McNamara's analysis [53] etc) that use different cephalometric landmarks as reference points. Fig. 1a shows the basic cephalometric landmarks. Using these landmarks, linear measurements and angles (Fig. 1B,C, Fig. 2) are used to describe the relative positions and size of the cranial base, maxilla and mandible.

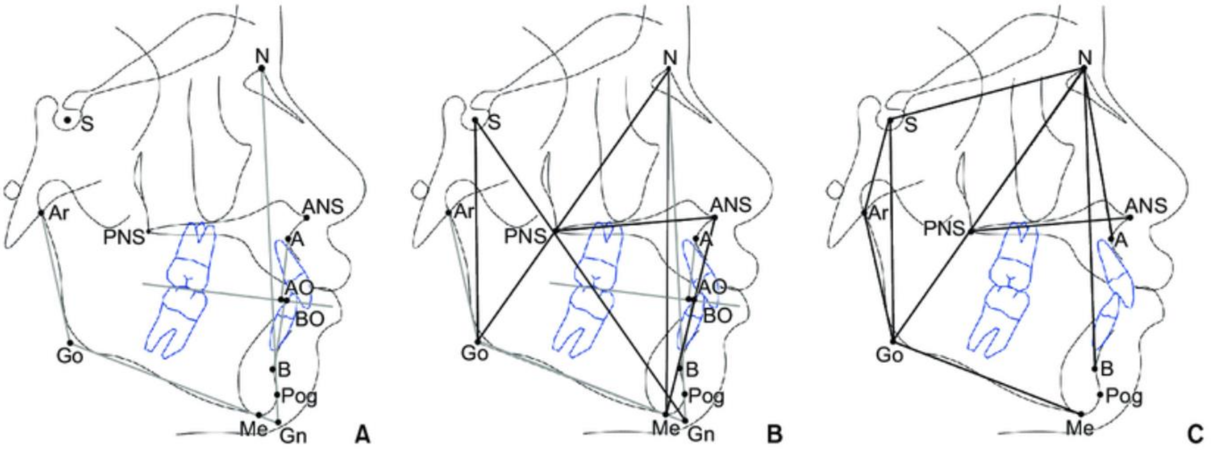


Figure 1: *Cephalometric landmarks and measurements.* **A**. Cephalometric landmarks: S- Sella turcica; N- nasion; Ar- articulare; ANS- anterior nasal spine; PNS- posterior nasal spine; A point- A; B point- B; AO- the point perpendicular to the occlusal plane at point A; BO- the point perpendicular to the occlusal plane at point B; Go- constructed gonion; Me- menton; Pog- pogonion; Gn- mechanical gnathion. **B**. Linear measurements: AO-BO, (Wit's); N-Me - anterior facial height; ANS-Me - lower anterior facial height; S-Go - posterior facial height; N-Go- facial depth; S-Gn- facial length. **C**. Angular measurements: SNA - The relationship of the maxilla to the cranial base; SNB - the relationship of the mandible to the cranial base; ANB - the relationship between the maxilla and the mandible; SN-GoMe, - mandibular plane angle; Ar-Go-Me, -gonial angle; Ar-Go-N,- upper half of the gonial angle; N-Go-Me – the lower half of the gonial angle. Illustration adapted from [54].

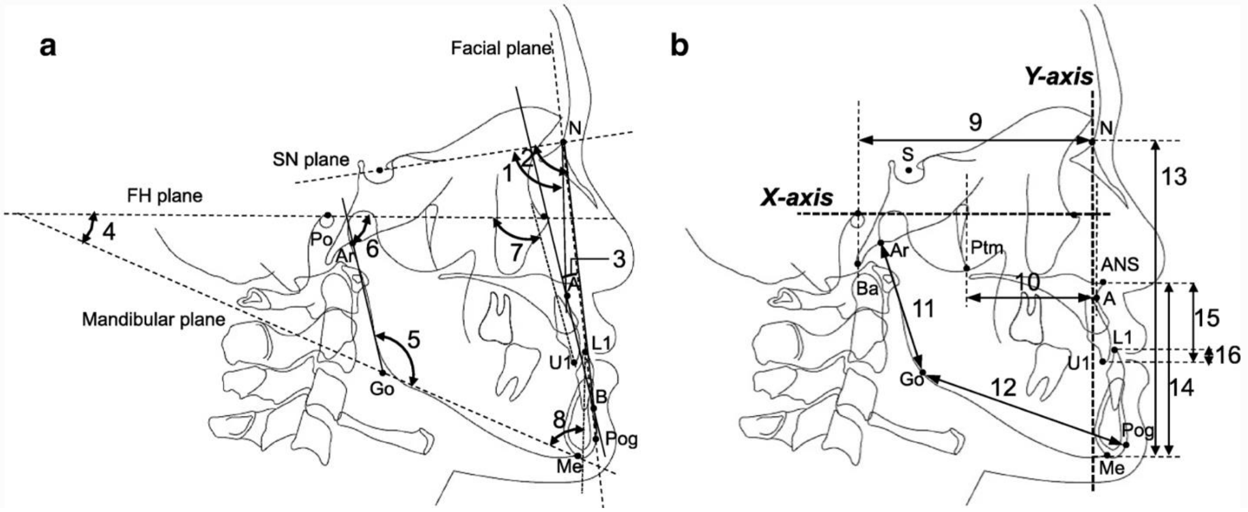


Figure 2 *Cephalometric reference points particular to angular and linear measurements.* **A** 1. ∠ SNA, 2. ∠ SNB, 3. ∠ A-B plane angle, 4. ∠ Mandibular plane angle, 5. ∠ Gonial angle, 6. ∠ Ramus inclination, 7. ∠ U1 to FH plane, 8. ∠ L1 to mandibular plane. **B** *X*-axis: parallel to the FH plane, *Y*-axis: perpendicular to the *X*-axis, 9. Ba-N (perpendicular distance to the *Y*-axis),

10. Ptm-A (perpendicular distance to the Y-axis), 11. Ar-Go, 12. Go-Pog, 13. N-Me (perpendicular distance to the X-axis), 14. ANS-Me (perpendicular distance to the X-axis), 15. ANS-U1 (perpendicular distance to the X-axis), 16. U1-L1 (perpendicular distance to the X-axis). Illustration adapted from [55].

2.3 Structure-Function Relationship

2.3.1 Functional Matrix Theory

In the 1960s, Moss, introduced the “Functional Matrix Theory" of growth [56]. In this theory, neither the cartilage of the mandibular condyle nor the nasal septum cartilage is a determinant of jaw growth. Moss explained the functional matrix theory by stating that “form follows function" [56]. Instead, he theorizes that the growth of the face occurs as a result of functional needs and neurotrophic influences and is mediated by the muscles in which the jaws are embedded. This conceptually means that the bone and cartilage are influenced by the growth of the muscles.

Moss also theorizes that the major determinant of growth of the maxilla and mandible is the enlargement of the nasal and oral cavities, which grow in response to functional needs [56].

According to this theory, the growth of the orofacial bones would be highly influenced by their muscular counterparts and their associated functions.

2.3.2 Structure-function Relationship in Orofacial Region

Muscle-bone interactions in the body follow the concept of Wolff’s law [57] and the “Mechanostat” theory proposed by Frost [58]. Both these theories suggest that bone development is highly influenced by its environmental mechanical loading. Since muscles attach directly to their skeletal counterparts, any changes in the muscle forces or activity would directly translate to the bone.

An example of how mechanical forces in the mouth result in slow and progressive structural changes in orthodontic tooth movement. Orthodontic tooth movement differs significantly from the abnormal or physiological tooth movement, as it entails a biological response of the surrounding tissues of the teeth due to an external force. It usually results in the remodelling of the periodontal ligament and the alveolar bone. Thereby leading to the reorganization of the intracellular and the extracellular matrix. This is due to the form of the osteogenic cells reorganizing themselves and then followed by the local vascularization.

The growth of the craniofacial skeleton, particularly the orofacial region, is influenced by the occlusal relationships, jaw relationships, and orofacial functions [59].

The articulators of the orofacial region - the tongue, jaws, lips and their associated muscles work in coordination to bring about orofacial functions like speech and mastication -Interarticular coordination. The spatial arrangement of the articulators is highly responsible to demonstrate Spatio-temporal coupling-coordinated movements of the jaws, which is a fundamental underlying mechanism for an efficient orofacial functioning [60].

Speech-related mechanisms require timely movements of the orofacial articulators. Here, perioral muscular forces are adjusted to provide adequate forces that bring about the coordination necessary. For the production of speech sounds, labial approximation and the position of the tongue against the dentoalveolar arches. Mechanical properties of the speech articulators influence movement coordination and can be associated with movement efficiency and speech motor control [61].

During the growth period, the jawbones are subjected to certain forces imposed by the surrounding tissues, including the teeth and orofacial muscles (masticatory, lingual and mimetic muscles).

The process of mastication, involves the activation and coordination of the masticatory muscles, the direction of jaw movements and the occlusal forces. With the masseter being the strongest oral muscle, it is also known to cause robust effects on the craniofacial musculoskeletal structures [62, 63] and has been well reported. The resulting facial profile of an adult is believed to be closely associated with these forces, generated during mastication processes [22]. A brachyofacial pattern (short face) was observed in individuals with strong masticatory muscles and a dolicofacial pattern (long face) in individuals with weak masticatory muscles [62, 64]. Apart from determining the vertical facial parameters, the masticatory muscles also influence the dentoalveolar structures, for example, the development of broader dental arches with thicker masseter muscles [65]. Diet and food consistency also determine the efficiency of the mastication process and on a long-term basis affect the development of the orofacial skeletal structures [66, 67]. Oro-muscular forces from the tongue, lips and occlusal/bite forces/pressures [68] and the spatial arrangement of the jaws [69] also account for mastication efficiency. From an orthodontic point of view on craniofacial growth, this constitutes an etiologic basis for dentoalveolar or skeletal adaptations [63, 70].

The role of the perioral forces/lip competency, movements and forces by the tongue and the oropharyngeal coordinated movements are fundamental to swallowing mechanisms. The forces and motility of the tongue and buccal tension are responsible for bolus formation and are pushed into the oropharyngeal space with the tongue.

The swallowing mechanism shares anatomy with the upper airway; thus, in addition to directing food into the digestive tract, this mechanism also serves to restrict the entry of food into the airway. This mechanism is achieved by nervous tissue reflexes and the coordinated movement. The upper respiratory tract muscles from the pharynx and larynx contribute to speech production. The epiglottis, the oropharyngeal muscles and the swallowing mechanisms contribute to the swallow-respiration coordination.

The physiologic and functional mechanisms of the orofacial tissues (swallowing, speech, mastication, respiration, chewing etc.) are brought about by the coordinated interaction of the orofacial musculoskeletal system, a lack of which results in the development of parafunctions in the orofacial region (mouth breathing, tongue thrusting, clenching/ bruxism etc) and are susceptible to craniofacial bone deformities. These craniofacial deformities manifest as skeletal changes (changing the structure of the skull) or malocclusions (changing the occlusal characteristics).

A study by Lesse et al. [71] stated that children with mouth breathing habits tend to have a higher mandibular inclination and more vertical growth (posterior facial height smaller than the anterior height) compared to nasal breathers. Bruxism, which is a condition of persistent jaw clenching and grinding of teeth during sleep or while awake [72], is related to cortical thickening in the gonial region of the mandible [73]. Moore et al. [74] found significant differences in relation to the maxillary length and prognathism of individuals with thumb-sucking habits [74]

Grippaudo et al. [75] showed the association of the occurrence of increased overjet and overbite with mouth-breathing [75].

2.3.3 Techniques to Quantify Orofacial Musculature Morphology and (Dys)functional Activity

Structure-function relationships in the orofacial region can be evaluated using quantitative techniques that measure the musculoskeletal forces, activity and growth-related patterns of the craniofacial skeleton.

Analysis of the dentoalveolar structures:

1) Cephalometry is performed to determine and analyze the skeleto-dental relationships. It is used in diagnosis and planning in orthodontics as well as orthognathic surgeries (as described above)
2) Model analysis is the formation of dental casts to study the 3D occlusal and dentition characteristics. It is used to analyze the degree and severity of malocclusions. Analytics of the Permanent and Mixed Dentition is performed using several Model analysis tools (eg. Moyer's mixed dentition analysis [76], Pont's Index [77], Bolton's analysis [78] etc.)

Techniques: Dental casts are measured using callipers to perform accurate measurements.

Parameters assessed: Different model analysis techniques use several parameters to measure the dental characteristics. However, the most commonly measured dimensions are: Dental Arch Dimensions- length, circumference, shape; Palatal Dimensions- Height, Length, Breadth; Occlusal characteristics- Class I, Class II, Class III; Malocclusions: overjet, overbite, crossbites, crowding

Analyses that quantify orofacial movements- Kinematics:

Kinematic studies with regard to the orofacial region are performed to assess the spatial characteristic of the articular structures – tongue, jaws, and lips. Most commonly used to study the biomechanics of the jaw, this technique measures the degrees of motion of the rigid bones of the jaw relative to the skull. The main aim of kinematic studies is to create realistic movements corresponding to the anatomical range of motions by varying parameters. In the case of jaw movement analysis, some of the movements include opening, lateral excursions, protrusion or retrusion. The anatomical locations are tracked digitally by placing reflective markers on the face/body which are captured by cameras during different tasks [79, 80]. Theoretically, the movements tracked, are computed using linear mathematical equations to determine the parameters that are used to assess the ranges of motion.

In addition to creating computed movements, the software used for the technique, also helps to validate if the motion ranges are anatomically correct or not by normalizing the linear mathematical equations to determine the parameters. The range of motions are recorded into the software as a 3D model of the corresponding anatomical location over which the parametric measurements are recorded. Kinematic recordings can especially be in the case of complex orofacial functions like chewing, speech or even post-construction surgeries [80].

Instruments: Multiple cameras that are calibrated (intrinsically, extrinsically and synchronized with time), reflective markers, and Motion capture software.

Parameters assessed: some of the parameters analyzed for jaw kinematics are- path distance/displacement, time-domain measures like the duration with the range of movements, velocity/maximum speed of the tongue tip/jaws, and Spatio-temporal indices (STI-used to measure coordination patterns).

Analyses that quantify orofacial Muscular activity:

Electromyography (EMG) is a technique used to record and study the electrical properties of the skeletal muscles’ response to nervous stimulation which determines the contractile properties of the muscle. In the orofacial region, the EMG is used for temporomandibular joint (TMJ) disorders, TMJ dysfunctions, muscles of the head and neck and disorders with oromotor impairments [81]. Generally, there are two methods used to perform EMG. 1) Intramuscular EMG, which employs a needle and fine-wire electrodes that are inserted into muscle tissue through the skin; 2) surface EMG (sEMG), is a non-invasive technique that uses electrodes placed on the skin surface to record the muscle activity. Another difference between the two methods is that the intramuscular EMG detects and reports the single motor unit potential (motor unit action potential (MUAP) and is specific to the muscle morphology, whereas the sEMG identifies superimposed MUAPs recorded from the fibres of many muscles which may contribute to discrepancies and low reproducibility. However, due to the non-invasiveness of sEMG, it can be safely used for children and other individuals who are otherwise uncooperative.

Instruments: the EMG electrodes, signal processing and modelling software.

Parameters: The EMG signals that are recorded are recognized by their waveform patterns. These signals are a summation of the spatial and temporal activities of the motor units and the recordings on an EMG waveform include- the general muscle activity value (amplitude); time-domain measures like duration of active periods/rest periods, period of onset/offset, variability in their activity over time; synchronization or symmetry of EMG waveforms.

EMG techniques in the orofacial region are used to assess muscle activity during functions like chewing, swallowing, the posture of the head etc using muscles like the masseter, temporalis, sternocleidomastoid etc [82-85]

Analyses that quantify orofacial muscle physiology – Kinetics:

Another way to identify structural motor performance is by recording their kinetic characteristics like forces/pressures. Kinetic measurements are often recorded when the structures analyzed are in a state of motion during their physiological functions. This helps to record the magnitude of forces (measured as Newtons) or pressures (in kPa-kilo pascals). The values recorded usually depict the strength of the muscles associated with the structures that are assessed.

With regard to orofacial structures, there are multiple instruments used to measure pressure/forces.

Takahashi et al. [86] used a balloon-based measurement device to record tongue and cheek pressures. This “lip piece” was used to measure the pressures of the perioral muscles of the cheek, upper lip, lower lip and tongue and was tested for its validity and reliability. Shang-Jung.Wu et al. [87] used the Iowa Oral Performance Instrument (IOPI) to measure the labial and lingual strength during swallowing tasks. Palinkas et al. [88] recorded the Maximal Bite Force (MBF) and masticatory muscle development using a 1000 N dynamometer. Different studies performed diverse tests for measuring orofacial kinetics, using different instruments and thereby presenting different outcomes. However, all the tests intend to measure the oromotor performance, but target different orofacial structures.

The presence of normal physiological functions equates to normal equilibrium in the underlying structural mechanism. Any abnormality in the morphological structure would translate to an impairment in the motor function.

2.4 Cerebral Palsy

“Cerebral palsy (CP) describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that is attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, and behaviour, epilepsy, and by secondary musculoskeletal problems” [89]. The definition by itself demonstrates the heterogeneous nature of this disorder and validates the range of impairments that would be exhibited. The definition also corroborates the need for a structured classification and ways to assess the degree of functional and structural (musculoskeletal) impairment.

2.4.1 Prevalence

According to population-based studies from around the world reported by the Centers for Disease Control and Prevention (CDC) [90], CP has a prevalence ranging from 1 to nearly 4 per 1,000 live births or per 1,000 children [91-99]. There is a variation in the prevalence rates across studies and in different parts of the world mainly because population studies are expected to record the "Lifestyle Assessment Score" that measures the severity of disability in a way that only significant developmental impairment is included. In the case of CP, data with regard to children with lower levels of motor impairment might go unreported, which explains the variation in prevalence rates. CP is reported to be more prevalent among children born preterm with or at low birth weights but has decreased due to better healthcare for low birth infants in some parts of the world [91, 92, 97-99].

2.4.2 Etiology

The etiology of CP is best understood based on the timing of the occurrence of disturbances causing the disorder. In this regard, it can be divided into the prenatal, perinatal and postnatal periods.

Prenatal period: referred to as Congenital Cerebral Palsy, these risk factors occur before or during pregnancy. The risk factors present as, Hypoxia, contagious diseases, trauma or bleeding during the third trimester, genetic factors, infections, Maternal thyroid dysfunctions, usage of drugs/alcohol and episodes of maternal epilepsy [100, 101].

Perinatal and period: Perinatal refers to the period during pregnancy up until a year after giving birth.

The most common risk factors during this period are low birth weight, prematurity and Intraventricular hemorrhage. [100]. However, recent studies have shown a decrease in low birth weight and intraventricular hemorrhage related hemorrhage-related CP (advances in mechanical ventilation and improvement) [102].

In the recent times, the most common brain abnormality is the development of Periventricular White Matter Injury in preterm children (PWMI)[103]. PWMI is caused due to many reasons, however, Periventricular Leukomalacia (PVL) is the most prevalent [103, 104]. Other etiological risk factors include jaundice, infections, and asphyxia [101].

Postnatal period: risk factors like head trauma, meningitis, encephalitis, hypoxic Ischemic Encephalopathy, toxicities, shaken baby syndrome and brain infarcts, contribute as risk factors [100, 105, 106].

Overall, prenatal risk factors account for 70-80% of the infants who develop CP [100], 21-40% contribute to the perinatal period and 12-21% contribute as postnatal period risk factors [100, 106]. However, to date, 30% of the estimated CP cases still remain idiopathic [89, 100].

2.4.3 Classification of Cerebral Palsy

The classifications of CP are based on its various sensorimotor impairments manifested by the nature of its heterogeneity [89].

1) Based on the anatomical distribution of the impairments;

Individuals can be classified as- diplegic (impairment of both limbs on either the lower or the upper body), hemiplegia (impairment of one side of the body), quadriplegia (impairments to all four limbs), and the less prevalent forms of monoplegia (impairment in one limb) and triplegia (impairment in three limbs) [89, 100, 105-107].

2) Based on the part of the brain affected (neuroimaging findings);

Individuals can be classified as-

1) Spastic: accounts for 70-80% of the CP cases. It is also referred to as hypertonic cerebral palsy due to the high muscle tone and movements (spasticity). Spastic CP is caused due to the damage in the cerebral cortex of the brain.
2) Athetoid: accounts for about 2.6% of the CP cases. It is also referred as the non-spastic or dyskinetic CP. It is caused due to damage in the cerebellum/basal ganglia of the brain. Athetoid individuals demonstrate hypotonia and hypertonia.
3) Ataxic: accounts for about 2.4% of the CP cases. It is caused due to the damage to the cerebellum of the brain.
4) Hypertonic: accounts for about 2.6% of all CP cases. It is also called atonic CP and is also caused due to the damage of the cerebellum
5) Mixed: accounts to 15.4% of the CP cases. Here, the brain damage is not localized to one area but, the damage is found in several areas of the brain. A child diagnosed with the mixed type usually presents with more than two types of CP, usually spastic and athetoid [107].

The most common clinical phenotype of CP is spastic diplegia which accounts to 35% of the CP cases [89, 90]

2.4.4 Signs and Symptoms

As a consequence of its heterogeneous etiology, the primary feature of CP is the abnormal motor function, posture and balance associated with impaired cognitive development. This is inherently due to the damage to the developing brain. CP has various clinical signs. The primary signs/symptoms are presented as- (in infants) abnormal muscle tone and stiffness of joints and/or muscles is characteristics of CP. In infants, other signs like delays in sitting, walking grasping objects; (in toddlers) involuntary/uncontrolled movements, difficulty/impairments with fine motor skills like eating, standing, speech (dysarthria), swallowing (dysphagia), lip competence (drooling), control of facial expressions, hearing and abnormal posture/gait.

The common neurological symptoms are- autism, epilepsy, and impairments with regard to sensorimotor skills like learning, hearing, visual, speech leading to intellectual disabilities, and behavioural problems. As a consequence of these symptoms, CP individuals may also develop secondary symptoms. Although CP was defined as non-progressive [89] disorder, there is a high incidence of the development of secondary conditions or comorbidities that can potentially occur due to delayed neurological reflexes that occur as a consequence of the immature nervous system [108]. Secondary symptoms are usually airway obstruction and sleep disorders (sleep apnea), dental manifestations, increased frequency of bone impairments and even stunted growth.

2.4.5 Severity of CP

To assess severity of the disorder based on the level of support CP individuals required, there were four classification systems introduced.

The Gross Motor Function Classification System (GMFCS) was introduced in 1997 by Palisano et al. [109] to describe gross motor functions, specifically the ability to walk, for children with CP from 2-18years of age [110]. This system was used to describe the presence of performing self-initiated movements as well as movement with the help of hand-held crutches, canes or wheelchairs [110]. The levels of the GMFCS ranged from level I to level V, ascending in sequence of the severity of impairment.

The Manual Ability Classification System (MACS) was used to describe the use of both hands and upper extremities for children with CP from 4-18 years of age [111]. The Communication Function Classification System (CFCS) is used to describe the ability of persons with CP for daily routine communication (sending or receiving a message) [112]. The CFCS also incorporates methods of communication, manual signs, eye gazes, pictures, vocalizations and speech-generating devices [112]. The Eating and Drinking Ability Classification System (EDACS) is used to assess eating and drinking safety and efficiency [113] i.e., it reports the risks of aspiration or choking and the time taken to eat the defined amount of food respectively [113].

In CP, *motor impairments* are exhibited by the underlying spasticity, incoordination, rigidity and spasms which are characteristics of this disorder. Apart from the general functional deficits displayed by the disorder, sensation, cognition, communication and comprehension, perception, and behaviour are usually altered. This could be attributed to the development of secondary musculoskeletal deficits [89].

Spasticity [114] is the increase in muscle tone along with hyperreflexia (increased reflexes) resulting from the hyperexcitability of stretch reflexes in the muscles. It is velocity-dependent. Spasticity is responsible for the functional impairments, stiffness and atrophy reported in CP and if not treated in a timely manner can lead to complications related to muscular fibrosis, contractures and musculoskeletal deformities.

2.4.6 Structural Adaptations in CP

The systematic review by Barrett and Lichtwark [115] on muscle morphology and structure in children with CP, reported changes in the muscle length, fibre size and type of muscle fibre. The most evident change was the reduced size of muscles in children with CP. Children with CP have reduced muscle strength due to the impairments in the underlying activation and co-contractions of antagonist muscles [116], and are also reported to have prolonged periods of activation and relaxation on stimulation [117]. There is limited evidence on the concepts behind skeletal muscle deformity in children with CP, however, a combination of muscle growth impairments and associated muscle adaptations are suggested [118]. The narrative review by Handsfield et al. [119] suggests altered muscle morphology and growth in individuals with CP to be associated with biomechanical consequences and thereby decreasing opportunities for mechanical loading. He also suggested that the "muscle morphology is a downstream effect of altered neural activation and biomechanics, rather than a direct consequence of the neural lesion" [119].

According to the theories proposed by Wolff [57] and Frost [58], any deficits in the muscle and bone would cause limited mechanical muscle stimulation onto the bone surfaces, which occurs in the case of CP. Hence the magnitudes of strain on the bones would be relatively low in CP due to the deficits in the amplitude of muscle activity, power or slow rates of forces by their muscles. [120, 121]. Henderson et al. [122] reported that in children with CP, the ostopenia observed does not demonstrate a loss of bone rather, it may suggest a failure of normal bone growth patterns.

Most of the CP-related literature on the structural morphology influencing functions in CP, focus on the limbs and posture. However, there is little evidence with respect to the quantitative assessments of the structures and functions in head and neck region.

Quantitative assessments of the orofacial structures, have only reported the oral health-related manifestations of CP. In 2002, Caterina Bensi et al. conducted a systematic review and meta-analysis and reported the incidences of Angle's Class II and anterior open bite in CP [123]. Similarly, Raducanu et al. [124] reported the general and oral features in the the spastic tetraparesis type of CP.

Hence, it is essential to recognize the orofacial musculoskeletal morphology and its adaptations during orofacial function in CP. Considering the heterogeneous clinical manifestation of CP, an integrated multidisciplinary approach is required to understand its effects on the orofacial musculoskeletal structures.

Hence, we developed a broad research question for the current scoping review.

CHAPTER 3: RESEARCH QUESTION AND SCOPING REVIEW

Research Question

"What is known about the orofacial musculoskeletal structures and (dys)functions in Cerebral Palsy?"

The knowledge synthesis for this topic of research is feasible by collaborating the heterogeneous parameters. Given the broad range of scientific disciplines involved in CP-related studies, it is an elaborate process. Hence, following a scoping review methodology will be beneficial in accommodating the breadth of the available knowledge.

Scoping Review

Scoping reviews are conducted to map the key concepts underlying a particular research area especially when the research area is complex and requires comprehensive assessments. The main sources of evidences available can be taken up as separate standalone projects for further knowledge synthesis [125]. As described, although scoping reviews are recognised as methods to "identify and map the literature"[125, 126], they can also be used to clarify multiple concepts/ definitions in the literature, analyse how the research is conducted in specific research areas and also potentially serve as a pre-requite for the development of systematic reviews and the need to develop more literature necessary to fulfil the identified knowledge gaps [127].

Scoping reviews can be conducted by the five-stage framework that was outlined by Arksey & O'Malley, 2005 [125]. The stages include (1) Identifying the research question; (2) identifying relevant studies; (3) study selection; (4) charting the data; and (5) collating, summarizing and reporting the results. The methodological integrity of a scoping review can be guided by the PRISMA-ScR checklist illustrated by Tricco et al., 2018 [128] It is also important to note that this review methodology seeks to evaluate the quality of evidence of the included studies

In this scoping review, the focus was to

a) identify the knowledge gaps present in the literature and highlight the potential effects of the abnormal muscle tones expressed in CP during orofacial functions on the orofacial musculoskeletal structures
b) aggregate the available knowledge of CP-related studies spanning all scientific disciplines
c) serve as a pre-requite for future studies to be developed on the orofacial region in CP individuals

CHAPTER 4: METHODOLOGY

The PRISMA-ScR checklist illustrated, guided this scoping review. (see Appendix Table 1).[128]

4.1 Identifying the Research question

The first step was to identify the research question. [125] The following research question was used to guide this review: "What is known about the orofacial musculoskeletal structures and (dys)functions in Cerebral Palsy?"

4.2 Identifying the Relevant Studies

A health librarian (MM), trained in searching for knowledge syntheses developed an initial comprehensive scoping review strategy for Medline (Ovid), which was then translated for Embase (Ovid), CINAHL and Scopus. The initial searches were run from inception to January 2021. Additional articles were identified by tracking references. The resulting strategies comprised of a combination of Medical Subject Headings (MeSH) or their equivalent, title/abstract keywords, truncations and Boolean operators, and included the concepts of cerebral palsy, orofacial anatomy, (i.e., muscles and bones of the face), and various terms linked to related activities (such as eating, chewing, speech, expressions) and orofacial morbidities associated with cerebral palsy.

As searching in scoping reviews is iterative, [129] a second, updated search for Medline (Ovid) was conducted in March 2021 following screening and identification of additional relevant subject headings and vocabulary. Final updates to the searches were performed in January 2022. The final search was performed on consult with the Thesis Advisory Committee to also include 'sleep apnea' as an associated orofacial comorbidity. This search was again translated to the same databases as before, the final search strategy is given in Appendix 1. Given the search yield, there were no filters and limits relating to language. All the studies were imported to a reference management software, Endnote. The duplicates were identified electronically and were excluded.

4.3 Study Selection

The selection of the studies addressing the research questions was performed with the help of the inclusion and exclusion (*Eligibility*) criteria, which are as follows:

4.3.1 Inclusion Criteria

1. Studies involving a population of individuals with a medical diagnosis of Cerebral Palsy.

2. Studies assessing the quantitative (objective, physical, and instrumental) measurements of orofacial musculoskeletal structures (i.e., morphology, spatial arrangement) or orofacial function.

4.3.2 Exclusion criteria

1. Studies assessing the prevalence rates of the various CP-related health manifestations

2. Studies on treatment, intervention, or training program for patients.

3. Studies evaluating the reliability of tools or instruments

4. Methodological studies

5. Animal studies.

For this scoping review, the study selection stage was iterative and the eligibility criteria were revised as the reviewers became more familiar with the literature.

To ensure a blinded screening, all the studies were imported to Rayyan to help with the study selection process. Rayyan is a web-based software that serves as a valuable tool for researchers in the process of screening and selecting articles for knowledge synthesis. This helped attain a non-biased decision on study selection. Three blinded review authors (AC, NK and EZ) scanned the titles and abstracts of the studies. After the initial screening and exclusion of studies based on the titles and abstracts, two of the authors (AC and EZ) performed the full text screening to ensure a match to the eligibility criteria.

4.3.3 Limits

As mentioned, there were no limits set to foreign languages or publication dates in the search and screening criteria. Studies in a foreign language were translated using the software- *Google Translate* and were chosen based on whether they could be easily translated using the translation software.

4.4 Charting the Data

The data extraction step was done using the database programme *Microsoft Excel*. The data from each of the included studies was corroborated by the review question (stated in section 4.1).

Firstly, the studies were categorised based on the type of quantitative measurement of the orofacial structure and/or function described. This led to the formation of 6 categories by which helped to simplify reporting the results. Secondly, the variables to be extracted from the included studies were determined to extract, synthesize and chart the data. This iterative process led to derive specific data pertaining to the following headings: (1) Author(s) and Year of Online Publication; (2) Study Population Characteristics like; (a) Study Design; (b) Sample Size; (c) Age (mean or range); (d) Sex; (e) CP Type (f) CP Severity; (3) Outcome Measures like; (a) Methodology/Technique; (b) Target Tissue(s); (c) Target Function(s); (4) Discipline of Science.

After the completion of charting, both reviewers (AC and EZ) confirmed if the data was adequately attained and ensured that the language and abbreviations were uniform.

4.5 Collating, Summarizing and Reporting the Results

The results of this review were reported in different sections. Firstly, the search results were described along with the exact numbers of the studies included and excluded at every step of screening. This is supported with the help of the Prisma diagram. (Figure 1)

Following this, the data extracted and analysed from the tabulation (Table 1) were discussed in multiple sections. The first section described the overall characteristics of all the studies were discussed based on the specific variables pertaining to the general demographics, participant characteristics as well as the scientific discipline. Then the results were reported based on the 6 categories used to identify the type of qualitative measurement of the orofacial structure and/or function for the studies. This helped in providing comprehensive results to answer the research question and most importantly understand the persistent knowledge gaps in the literature.

4.6 Quality Assessment

The quality assessment of each of the included studies was performed using an integration of the critical appraisal tools: 1) AXIS (the Appraisal tool for Cross- Sectional Studies tool) [130] and the 2) MMAT (Mixed Methods Analysis Tool) [131]. Much like the rating criterion of the AXIS and MMAT tools, the results of this scoping review discuss the rating of each methodologic criterion instead of discussing the quality of the included studies based on their total score. This in turn provides a sensitivity analysis (i.e, the consideration of the quality of the studies by contrasting their results)[131]. Even though most scoping reviews do not perform a quality appraisal, the research team decided for it. The quality appraisal for this review was mainly done to highlight the existent limitations of the available literature for CP related studies and how it can be improved to ensure adequate knowledge synthesis.

CHAPTER 5: RESULTS

5.1 Search Results

The outcomes of the undertaken methodology are reported in the Prisma diagram (**Fig. 3**). The first search was performed in January 2021 and identified 5863 studies. A second search was performed in March 2021 to include more terms (e.g., describing functions, parafunctions, orofacial muscles, etc.), which identified 1920 more studies. A third search was performed in January 2022 to capture any articles published in the preceding year and this search identified 469 studies. Furthermore, reference tracking identified 12 additional studies.

The studies were deduplicated resulting in a total of 5746 original articles that were identified in the four databases (Embase, Ovid, CINAHL, Scopus) and via reference tracking. Next, titles and abstracts were screened by three reviewers (AC, TKN, EZ) on Rayyan, and 5461 studies were excluded based on the eligibility criteria. Then, a full text review was performed by two reviewers (AC, EZ) on 285 studies and 246 were excluded with reasons. Thus, based on the eligibility criteria, 39 studies were identified as the final set of records to be included in the scoping review.

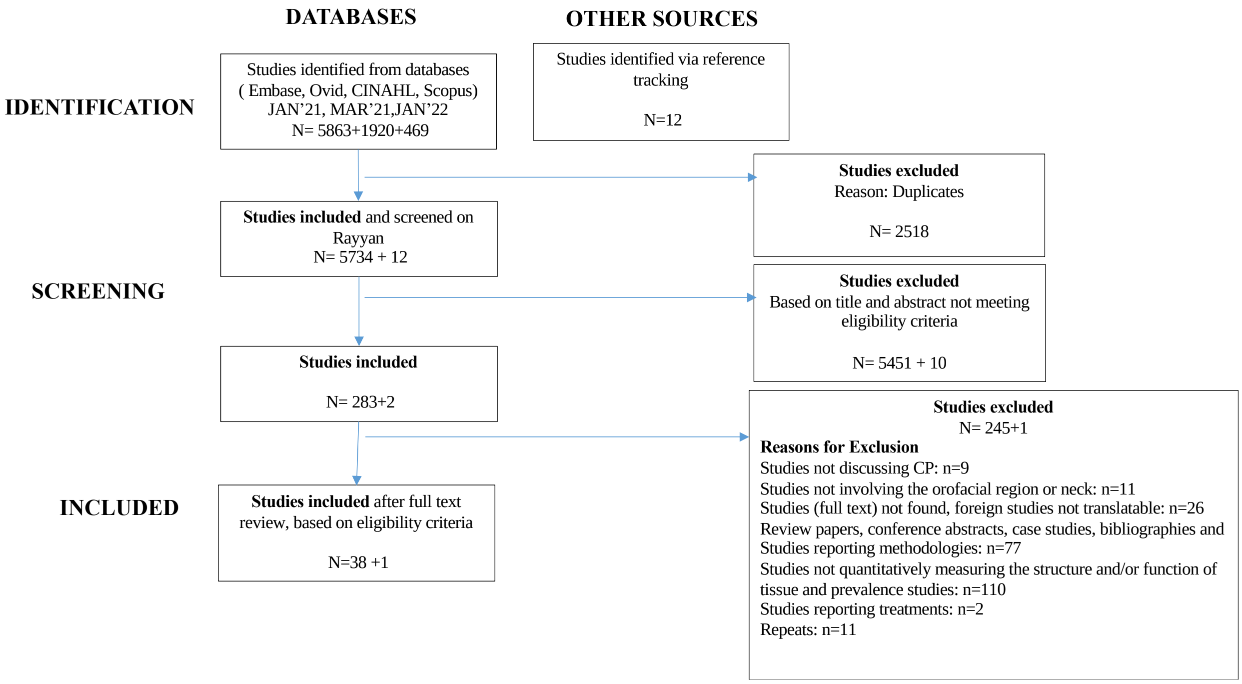
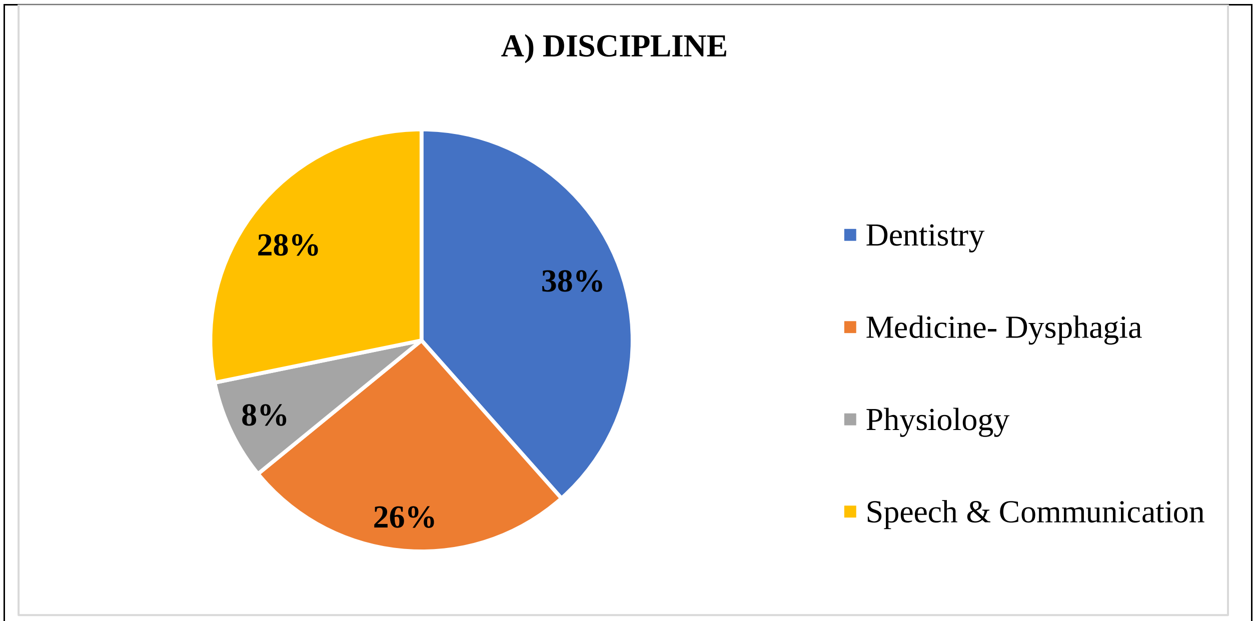


Figure. 3 *PRISMA diagram*

5.2 Characteristics of the studies

The included studies came from the disciplines of Dentistry, Speech & Communication, Physiology and Medicine- Dysphagia (see **Fig. 4A**). This categorization of the disciplines was done based on the function assessed, methodology, journal discipline, author affiliation and/or the study’s stated objective. Methodological techniques to quantitatively assess structure and function (see **Fig. 4B**) included cephalometry (skeletal structures), model analysis (dental structures), motion capture (jaw and tongue kinematics), ultrasound imaging (tongue movement), electromyography (EMG, muscle activity) and pressure-sensitive transducers/sensors (lip or occlusal forces). In addition to these main quantitative measuring techniques, other techniques like electromagnetic articulography (jaw and tongue kinematics), respiratory inductance plethysmography (pulmonary muscle activity/movement), gnathodynamometer (occlusal force-kinetics) and force and movement transducers were also used, but in fewer studies.

The data extraction table is shown in **Figure 5**. The studies are grouped based on methodological techniques used to assess the structures and functions with **Group 1** encompassing analyses on hard tissues like the skeletal and dental structures using cephalometry and/or model analysis techniques, **Group 2** is comprised of studies quantifying jaw or tongue kinematics, **Group 3a** includes muscle activity using electromyography, **Group 3b** are studies reporting muscle activity along with kinematics, **Group 4a** includes studies reporting kinetics using pressure sensitive transducers, **Group 4b** reports studies with muscle activity and kinetics, and **Group 4c** reports studies with kinetics and kinematics techniques.



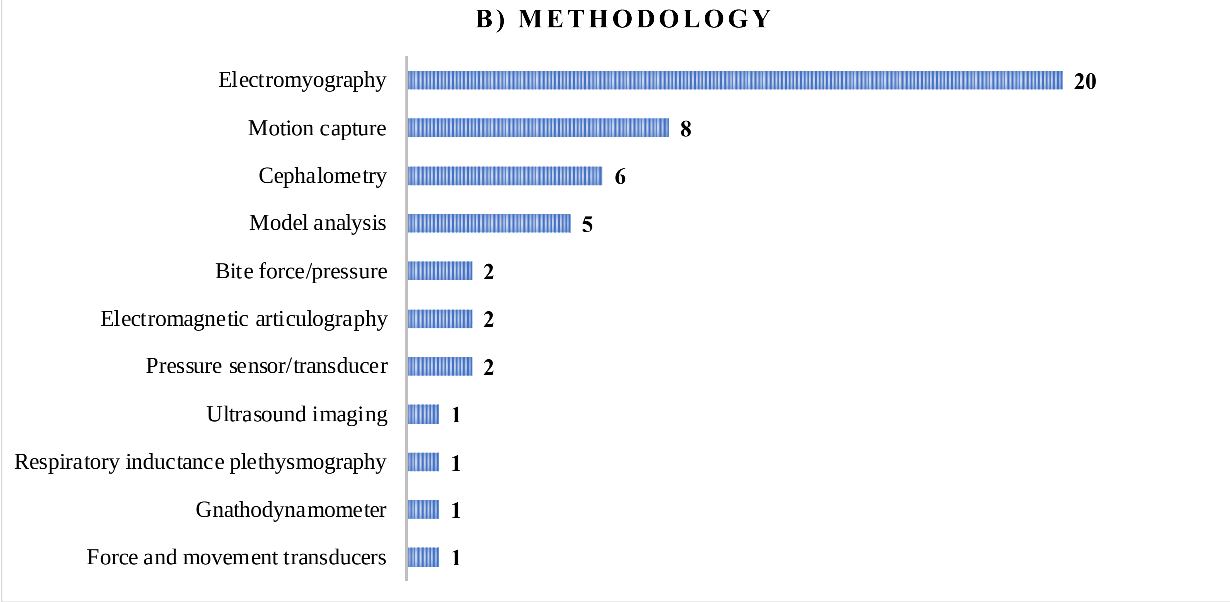


Figure. 4 *Characteristics of the studies*. A) The studies span four disciplines: dentistry, dysphagia, physiology and speech and communication research. B) The studies included a range of methodological techniques to quantitatively assess structure and function.

		Study Population					Outcome Measures			
Study	**Study Design**	**Sample Size**	**Age** (range or average)	**Sex** (Male/Female)	**CP type**	**CP Severity**	**Methodology**	**Target Tissue(s)**	**Target Function(s)**	**Discipline**
Group 1: Orofacial Hard Tissue Structures										
Album, et al. (1964)	Cross sectional	CP: 47 TD: 47	7 - 10 y	CP: 24 / 23 TD: 24 / 23	Spastic, extrapyramidal, mixed, CP & cord injury, motor and mental	n.d	Cephalometry, model analysis	Dental arch, palate, skeletal tissues	N/A	Dentistry
Brown & Sharma (1967)	Cross sectional	CP: 82	4 - 12 y	n.d	Spastic, athetoid, ataxic, unclassified	n.d	Cephalometry	Skeletal tissues	N/A	Dentistry
Karwetzky & Tielsch-Keuthen (1972)	Cross sectional	CP: 150	1 - 17 y	n.d	n.d	n.d	Cephalometry	Dental and skeletal tissues	N/A	Dentistry
Oktay (1972)	Cross sectional	CP: 20	4 - 8 y	n.d	Spastic hemiplegia, diplegia, paraplegia	Very mild to very severe	Model analysis	Dentition, dental arches	N/A	Dentistry
Foster, et al. (1974)	Cross sectional	CP:33 TD: 33	3 - 28 y	n.d	Spastic, athetoid, dystonic, ataxia, myopathy, diplegia, hemiplegia, tetraplegia	Severe	Cephalometry	Skeletal tissues	N/A	Dentistry
Strodel (1987)	Cross sectional	CP: 30 TD: 30	4y5mo - 22y7mo	CP: 16 / 14 TD: 16 / 14	Spastic	n.d	Cephalometry, model analysis	Dentition, dental arches, skeletal tissues	N/A	Dentistry
Poshtaru, et al. (2019)	Cross sectional	CP: 30 TD: 1345	9 - 15 y	n.d.	n.d	n.d	CT-based lateral cephalometry	Dental and skeletal tissues	N/A	Dentistry
Dummett (1975)	Cross sectional	CP: 98 TD: 76	3 - 20 y	CP: 59 / 39 TD: 40 / 36	Spastic, athetoid, ataxic, mixed, undiagnosed, diplegia, hemiplegia, paraplegia, quadriplegia	n.d	Model analysis	Dentition, dental arches	N/A	Dentistry
Mamaghani, et al. (2008)	Cross sectional	CP: 62	18 - 78 y	CP: 36 / 26	Infantile CP	n.d	Model analysis	Palate	N/A	Dentistry
Group 2: Orofacial Kinematics										
Hong, et al. (2007)	Cross sectional	CP:10 TD: 10	CP: 8.2y ± 1.6 TD: 8.1y ± 1.8	n.d	Spastic quadriplegia	n.d	Motion capture	Lip	Speech	Speech and communication
Chen, et al. (2010)	Cross sectional	CP: 10 TD: 10	4.8 - 7.5 y	CP: 7 / 3 TD: 6 / 4	Spastic	GMFCS: I, II	Motion capture	Lip	Speech	Speech and communication
Hong, et al. (2011)	Cross sectional	CP: 12 TD: 12	7 – 11 y	CP: 7 / 5 TD: 7 / 5	Spastic quadriplegia	n.d	Motion capture	Lip, jaw	Speech	Speech and communication
Nip (2013)	Cross sectional	CP: 4 TD 7y: 11 TD 10y: 9 TD 16y: 9 TD adult: 9	CP: 6y11m - 21y10m TD 7y: 7.6y ± 0.29 TD 10y: 10.4y ± 0.27 TD 16y: 16.4y ± 0.32 TD adult: 23.9y ± 3.2	CP: 2 / 2 TD 7y: 5 /6 TD 10y: 4 / 5 TD 16y: 4 / 5 TD adult: 5 / 4	Spastic, hemiplegia, quadriplegia	GMFCS: I, II, IV, V	Motion capture	Lip	Speech	Speech and communication
Nip, et al. (2017)	Cross sectional	CP: 4 TD: 4	9 – 14 y	CP: 4 / 0 TD: 4 / 0	Spastic, diplegia, hemiplegia, quadriplegia	GMFCS: II, III	Electromagnetic articulography, electromyography	Tongue, jaw	Speech	Speech and communication
Nip (2017)	Cross sectional	CP: 12 TD: 12	CP: 4y8m - 15y0m TD: 4y7m - 15y7m	CP: 7 / 5 TD: 7 / 5	Spastic, hemiplegia, diplegia, quadriplegia, Dandy Walker	GMFCS:II-V	Motion capture	Lips and jaw	Speech	Speech and communication

Remijn, et al. (2015)	Cross sectional	CP: 2 TD: 4	CP: 23.1 - 25.8 y TD: 23.6 - 47.2 y	CP: 2 / 0 TD: 1 / 3	Spastic, tetraplegic	GMFCS:III	Ultrasound imaging	Tongue	Mastication	Dysphagia
Nip, et al. (2018)	Cross sectional	CP: 11 TD: 11	3 - 11 y	CP: 7 / 4 TD: 7 / 4	Spastic-hemiplegia, diplegia, quadriplegia, Dandy Walker Var	GMFCS: II – V CFCS: I - IV	Motion capture	Jaw	Mastication	Dysphagia
Group 3a: Orofacial Muscle Activity										
Yoshiyasu (1988)	Cross sectional	CP: 20 TD: 10	CP: 7.5 - 16.3 y TD: 7.9 - 16.4 y	CP: 13 / 7 TD: 5 / 5	Athetoid, spastic, mixed, other	nd	Electromyography	Bilateral masseter and temporal muscles	Occlusion, mastication	Dentistry
Santos, et al. (2010)	Cross sectional	CP: 22 TD: 29	CP: 11.0y ± 2.8 TD: 10.1y ± 0.5	CP: 16 / 6 C: 20 / 9	Spastic, dystonic, athetosis, ataxia, mixed, quadriplegia, diplegia, hemiplegia	n.d	Electromyography	Bilateral anterior temporalis and masseter	Maximum bite force	Dentistry
Serafinowska, et al. (1981)	Cross sectional	CP: 20 TD: 20	5- 15 y	n.d.	spastic paresis	n.d	Electromyography, chewing efficiency	Mandibular, temporal and specific mouth muscles	Mastication, movement	Dysphagia
Shamansurov, et al. (1982)	Cross sectional	CP: 40 TD: 15	CP: 7 - 14 y	n.d.	Spastic diplegia, hyperkinetic	n.d	Electromyography	Anterior temporal, masticatory, facial and chin muscles	Mastication, swallowing	Dysphagia
Sochaniwskyj, et al. (1986)	Cross sectional	TD: 12 CP - non-droolers: 12 CP - droolers: 12	TD: > 6 y CP nondroolers: 6 - 19 y CP droolers: 6 - 19 y	TD: n.d. CP nondroolers: 7 / 5 CP droolers: 10 / 2	Quadriplegia	n.d	Electromyography	Masseter, orbicularis oris, infrahyoid group	Swallowing liquids	Dysphagia
Briesemeister, et al. (2013)	Cross sectional	CP: 16 TD: 16	7 - 13 y	CP: 7 / 9 TD: 11 / 5	Spastic, hemiplegia, diplegia, quadriplegia	GMFCS: I - IV	Electromyography	Bilateral masseter and temporal muscles	Mastication	Dysphagia
Ries & Bérzin (2017)	Cross sectional	CP: 8 TD: 12	CP: 9.5y ± 2.5 TD: 7.91y ± 0.99	n.d	Spastic	n.d	Electromyography	Bilateral masseter and temporalis	Mastication and maximum bite force	Dysphagia
Mishra, et al. (2019)	Cross sectional	CP: 11 TD: 10	CP: 7.7y ± 2.4 TD: 7.6y ± 2.0	CP: 8 / 3 TD: 5 / 5	Spastic hemiplegia, diplegia,quadriplegia	GMFCS: I–V	Electromyography, respiratory inductance plethysmography	Suprahyoid muscle group, airflow	Swallowing, cough	Dysphagia
Neilson & O'Dwyer (1984)	Cross sectional	CP: 5 TD: 5	CP: 19 - 34 y TD: 20 - 30 y	n.d	Athetoid	Severely disabled	Electromyography	Orbicularis oris superioris, depressor labii inferioris, anterior genioglossus, geniohyoideus, medial pterygoideus, anterior belly of digastricus	Speech	Speech and communication
O'Dwyer & Neilson (1988)	Cross sectional	CP:5 TD: 5	CP: 19 - 34 y TD: 20 - 35 y	n.d	Athetoid	Severely disabled	Electromyography	Orbicularis oris superioris, depressor labii inferioris, anterior genioglossus, geniohyoid, medial pterygoid, anterior belly of the digastric	Speech	Speech and communication
Neilson, et al. (1979)	Cross sectional	CP: 7 Stutterer: 4 TD: 6	20 - 30 y	n.d	Spastic, Athetoid	Severly disabled	Electromyography	Orbiculatis oris inferior and superior, levator labii superior, zygomaticus major, risorius, depressor labii inferior, depressor anguli oris, mentalis, buccinator, genioglossus, geniohyoid, mylohyoid, tongue intrinsic, styloglossus, temporalis, masseter, internal pteryygoid, anterior belly of digastric.	Jaw, tongue and lip stretching	Physiology

O'Dwyer, et al. (1983)	Cross sectional	CP: 6 TD: 6	CP: 19 - 34 y TD: 20- 30 y	CP: 5 / 1 TD: 3 / 3	Athetoid, Spastic	Severely disabled	Electromyography	Levator labii superioris, zygomaticus major, buccinator, risorius, orbicularis oris superioris and inferioris, depressor anguli oris, depressor labii inferioris, mentalis, anterior genioglossus, geniohyoideus, anterior belly of the digastricus, medial pterygoideus	Non-speech facial gestures	Physiology
Vaughan, et al. (1988)	Cross sectional	CP: 10 TD: 10	CP: 19 - 32 y TD: 20 - 34 y	n.d.	Spastic, Athetoid	n.d	Electromyography	Masseter, orbicularis oris superioris	Voluntary isometric contraction	Physiology
Group 3b: Orofacial Muscle Activity and Kinematics										
Itakura (1997)	Cross sectional	CP: 4 TD: 6	CP: 29 - 53 y TD: 25 - 34 y	CP: 2 / 2 TD: 4 / 2	n.d	n.d	Electromyography, motion capture	Jaw, bilateral masseter, anterior digastric, sternocleidomastoid	Occlusion	Dentistry
Rong, et al. (2012)	Cross sectional	CP: 3 TD: 3	22 - 43 y	CP: 2 / 1 TD: 3 / 0	Spastic	n.d	Electromagnetic articulography, electromyography	Tongue, jaw, orbicularis oris superior and inferior, anterior belly of digastric, mylohyoid, platysma	Speech	Speech and communication
Connaghan & Moore (2013)	Cross sectional	CP: 3 DS: 7 TD: 11	CP: 4y4m - 6y0m DS: 4y0m - 8y5m TD: 4y3m - 10y0m	CP: 2 / 1 DS: 5 / 2 TD: 8 / 3	Spastic	n.d	Motion capture, electromyography	Jaw, bilateral temporalis, bilateral masseter, suprahyoid	Speech	Speech and communication
Group 4a: Orofacial Kinetics										
Chigira, et al. (1994)	Cross sectional	CP: 10 DD: 11 TD: 104	CP: 3 - 6 y DD: 3 - 6 y TD: 5mo - 5 y	n.d	n.d	n.d	Pressure sensor	Lips	Lip closing pressure during feeding.	Dysphagia
Santos, et al. (2015)	Cross sectional	CP grinding: 42 CP nongrinding: 53	CP grinding: 9.3y ± 2.5 CP nongrinding: 10.5y ± 3.1	CP grinding: 28 / 14 CP nongrinding: 36 / 17	Spastic	GMFCS I - V	Gnathodynamometer	Dentition	Mastication, clenching	Dentistry
Group 4b: Orofacial Kinetics and Muscle Activity										
Lespargot, et al. (1993)	Cross sectional	CP droolers: 10 CP nondroolers: 10 TD: 10	6-14 y	Combined: 16 / 14	Spasticity, feebleness, mixed, hemiplegia, quadriplegia	n.d	Electromyography, pressure transducer at hard palate	Mouth floor muscles and hyoid moving muscles	Swallowing, drooling	Dysphagia
Nakajima, et al. (1988)	Cross sectional	CP group 1: 15 TD group 1: 21 TD group 2: 7 TD group 2: 7	CP group 1: 6 - 15 y Con group 1: 6 - 15 y CP group 2: 13 - 17 y Con group 2: 18 y	n.d	n.d	n.d	Group 1: Bite pressure, masticatory efficiency Group 2: Electromyography, masticatory efficiency	Group 1: Dentition Group 2: Bilateral temporal and masseter muscles	Mastication	Dentistry
Matsui, et al. (2017)	Cross sectional	CP: 30 TD: 30	20 - 35 y	CP: 15 / 15 TD: 18 / 12	Quadriplegia	GMFCS: I - IV	Electromyography, bite force	Bilateral masseter and temporalis	Maximum mouth opening, maximum bite force	Dentistry
Group 4c: Orofacial Kinetics and Kinematics										
Barlow & Abbs (1986)	Cross sectional	CP: 5 TD: 6	21 - 35 y 18-24 y	n.d TD: 6 / 0	Spastic quadriplegia, diplegia, hemiplegia	Mild - severe	Force and movement transducers	Lips, tongue, jaw	Speech	Speech and communication

Figure 5: Synthesized Table of Extracted Data from Included Studies (CP- Cerebral Plasy, TD- Typically Developing, DD- Developmental Delay, nd- not described, mo- months)

5.3 Characteristics of the study design

The data extraction table displays information regarding the study design, study population, methodology, target tissue and target function. All studies are cross-sectional in design. Of the studies, 24 focus on pediatric study populations, 11 on adults, and 4 on populations with adults and children. The distribution of males vs females in the study population is not described in 16 studies, while 20 studies have study populations with a greater proportion of males than females, and three studies had equal numbers of males and females. Epidemiological studies have observed higher rates of CP in males than females [132, 133].

As for the type of CP and limb involvement, 33 studies provide some description. Interestingly, 28 of the studies include individuals with spastic CP, which is the most common type of CP [134, 135]. Only 10 studies describe the severity of CP using the GMFCS scale.

5.4 Jaw Kinematics

The literature search yielded 12 studies on orofacial kinematics in CP. These studies are listed in the data extraction table (**Table 1**) under Groups 2, 3b and 4c. While all of these studies capture movement of facial structures, they are investigated in different contexts: nine studies address speech intelligibility, speech production and/or articulation, two studies quantify jaw or tongue movements during mastication and one study is in the field of dentistry addressing kinematics during occlusal tapping. Of the twelve studies, eight use motion capture [136-143], two use electromagnetic articulography [144, 145], one uses force and movement transducers [146] and one uses ultrasound imaging to measure tongue displacements [147]. With these techniques to measure movement of facial structures, the lips (n = 6 studies), jaw (n = 8) and tongue (n = 4) are investigated to quantify velocity, displacement and spatiotemporal indices (synchrony of movements in time and space of lips, tongue and jaw).

5.4.1 Displacement

Displacement of the lips, tongue and jaws was quantified during speech and mastication tasks. Generally, individuals with CP have greater jaw displacements than TD during speech [137, 144, 145] and mastication [141] than control groups. Rong et al. [145] measured larger jaw amplitude in individuals with CP during speaking of single words, while Hong et al. [137] measured the same during monosyllable and polysyllable tasks. Nip et al. [141, 144] measured kinematics during speech and mastication using the parameters path distance and working space to characterize the kinematics. The path distance describes the distance traveled by the jaws during the complete mastication or speaking process, while the working space describes the volume of space used for mastication produced by vertical and lateral jaw movements. Nip et al. [141, 144] observed greater path distance and working space in CP than the TD in speech tasks (sentence) as well as for soft and solid food consistencies (but not purees).

In contrast, Chen et al. [136] observed no difference in jaw displacement during monosyllable and polysyllable speaking tasks. However, the study by Chen et al. [136] was on a population of children with GMFCS I and II; therefore, the children in this study may have lower severity also in the orofacial region. Itakura [143] measured jaw displacements during repeated tapping of the teeth, which is essentially the participant making repeated occlusal contact. This study found no differences in opening displacement during tapping between the TD and CP groups; however, the CP group did have a greater variability in position of the jaw during occlusal contact.

Conflicting evidence was observed for lip and tongue movements. A greater range of lip movement was observed by Nip et al. [139] on a small number of subjects during monosyllable, polysyllable and sentence speaking tasks; in contrast, Hong et al.[138] observed no difference in lower lip displacement during monosyllable speaking tasks between the TD and quadriplegic children with CP. There may be differences in data analysis, placement of markers and study populations that make a comparison between these two studies difficult. Nip et al. [144] using electromagnetic articulography and found larger tongue tip movements in children with CP during repetition of sentences, while Rong et al. [145] observed lower tongue tip displacement in the adult CP group compared to TD during the formation of alveolar consonants. However, this could be due to the difference in speech tasks. Remijn et al. [147] using ultrasound imaging found larger vertical tongue movements in the coronal plane but not the sagittal plane in adults with CP during mastication. The differences in ages, tasks and low sample sizes (n <5 / group) make conclusions difficult.

5.4.2 Velocity

In the studies that focus on speech motor control, the participants performed mono syllable, poly syllable and diadochokinetic tasks. Four studies found greater velocities of the lips in individuals with CP compared to control groups during speaking tasks. Two studies found greater jaw velocity [137, 145] during monosyllable and polysyllable speaking tasks in individuals with CP, while two studies observed no difference in lower jaw velocities between CP and TD groups [136, 142]; however, Chen et al. [136] was on a group of children with mild CP and Connaghan et al. [142]

only contained two participants with CP. Two studies [145, 146] found lower velocity of the tongue in CP vs TD. Barlow et al. [146] observed lower velocities in the lips, jaw and tongue in the context of fine motor control tasks and not function (i.e., speech or mastication)

5.4.3 Spatio-temporal indices

Spatio-temporal indices (STIs) describe the consistency of speech patterns, which require spatial and temporal motor control of the lips, tongue and jaw: greater STI values describe poor consistency in movement patterns. STIs were consistently greater in CP groups compared to TD [136, 138]; however, Chen et al [136] only found greater STIs in polysyllable tasks and not monosyllable. Nip et al. [144] also measured speech movement stability and found the CP group to have reduced movement stability compared to the TD group, which is a similar measure to STI. Two studies differentiate the spatial and temporal components. Interestingly, there was no difference in spatial coupling between the lips and jaw for the CP and TD groups [137, 140]. However, Nip [140] did observe reduced spatial coupling between the upper and lower lips. Furthermore, reduced temporal coupling was observed between the lips as well as between the jaw and lips in CP compared TD groups [137, 140].

5.5 Muscle Activity

In total, nineteen studies measured muscle activity. In the data extraction table, all nineteen studies are described in Group 3a (Orofacial muscle activity), Group 3b (Orofacial muscle activity and kinematics) and Group 4b (Orofacial kinetics and muscle activity). All studies [142, 143, 145, 148-163] measured muscle activity through electromyography (EMG). One of these studies [151] discussed respiration and swallowing characteristics using respiratory inductance plethysmography and EMG. Various orofacial muscles were chosen for the muscle assessments and were linked to the function analyzed. However, the majority (13 out of 19 total studies) assessed the masseter and temporalis muscles (Table 1).

5.5.1 Muscle activation measures

To record the muscle activity at rest, in response to stimulus or during a function, the amplitude was measured. Twelve articles discuss EMG amplitude. Many of the studies observed that individuals with CP have muscle activity when at rest [148-150, 159-161]. Four studies [150, 157, 158, 160] measured muscle activity (mean RMS) during maximal voluntary clench. Three of these studies [150, 158, 160] observed a greater muscle amplitude during maximum voluntary clench in the masticatory muscles (masseter and temporalis) in TD compared to CP participants. However, Ries and Berzin [157] did not find any difference in amplitude of muscle activity during maximum voluntary clench between TD and CP groups.

Three articles assessed muscle activity in the context of speech [145, 154, 155]. In the study by Rong, et al. [145], the amplitude of the EMG signal was normalized to the baseline values and the peak EMG signal (percent maximum amplitude). Spastic CP participants showed a reduced % maximum amplitude in submental muscles during speech compared to their TD peers [145]. Neilson and O'Dwyer [154, 155] measured greater signal power (mean square of EMG waveform) in athetoid CP participants than TD. Furthermore, the energy of the EMG waveform (integral of square waveform combining time and amplitude) was consistently greater across every syllable in the lips (orbicularis oris superioris and depressor labii inferioris), tongue (anterior genioglossus), geniohyoid, and the medial internal pterygoid but not the anterior belly of the digastic [155].

A number of studies also measured muscle activity during swallowing and mastication [148, 151, 152, 157, 159-161, 163]. Shamansurov et al. [160] found that the maximum amplitude of the masseter and temporalis during mastication was lower in CP than in TD; additionally, in contrast to the control group, the facial muscles (lips, chin) in individuals with CP have higher levels of activity during swallowing than the mastication muscles. Other measurements express an amplitude relative to the maximum voluntary clench (which is significantly lower in CP than control). Mishra et al. [151] observed that participants with CP had greater muscle activity as expressed by % maximum amplitude during consumption of puree and chewable solid food, but not liquids. Similarly, Briesemeister et al. [148] found greater normalized RMS values (% of maximum clench) in CP group in right masseter and left temporalis during mastication compared to non-CP. Ries and Berzin [157] found no difference in RMS activity (normalized by mean and maximum muscle activity during clench) during non-habitual chewing cycles. Sochaniwskyj et al. [161] found that muscle amplitudes (integrated EMG waveform) during drinking tasks were similar in control, CP non-droolers and CP droolers but here there was no normalization to maximum clench.

Muscle activity has also been investigated in the context of non-speech tasks. In the study by O'Dwyer, et al. [156] muscle activity was measured during facial gestures. Here, mean amplitude of muscle activity was greater in CP than TD in a majority of the facial gestures. The study by Itakura [143] showed a greater amplitude of the sternocleidomastoid and masseter muscles during tooth tapping movements in the participants with CP compared to TD.

5.5.2 Time domain measures

EMG patterns can also be measured based on time domain components. One of the most commonly recorded time parameters was the mean duration of muscle activity. This can be measured as the difference between the time of onset and offset of the movement. Based on these durations, the variability in the waveform onset-offset timing/fluctuation patterns are also reported. It is interesting to note that, out of nine studies discussing time domain measures, five studies [148, 154-156, 163] reported that individuals with CP had prolonged durations of muscle activity in comparison to the TD group. O'Dwyer et al. [156] reported a prolonged duration of muscle activity during 9 out of 14 facial gestures. Similar results were stated by Neilson and O'Dwyer [154, 155], where the authors report a greater waveform duration, time between syllables and variability in the time between syllables in the CP groups. In the study by Briesemeister et al. [148] the duration of active and inactive mastication was measured in the masseter and temporalis muscles. While there was no differences in the duration of the inactive period, the duration of active muscle period was greater in CP than the TD group. Similar results with respect to duration and interval time of muscle activity during the chewing cycle mastication were presented by Nakajima et al. [152]; the coefficient of variation in the duration and interval times were significantly greater in the CP group, which indicates greater variability in the CP group.

Connaghan and Moore [142] reported the- EMD (electromechanical delay) measurements defined as the interval time between the onset of muscle activation and onset of movement. The EMD measurements showed no differences among the groups. Sochaniwskyj et al. [161] the author reported similar durations of muscle activity for the infrahyoid muscles during swallowing between the groups of droolers with CP, non-droolers with CP and TD children.

5.5.3 Synchronization and symmetry

Sychronization, coordination and symmetry are generally recorded as the timing difference in muscle groups. Patterns of synchronization/coordination are higher among the TD group than CP in six studies [143, 156, 157, 160, 161, 163]. Sochaniwskyj et al. [161] found differences in coordination among droolers with CP, non-droolers with CP and typically developing children. Specifically, the controls activate the masseter and infrahyoid during swallow, whereas the CP non-drooler group activated the orbicularis oris and masseter. Similarly, the CP non-droolers group had difficulty synchronizing the masseter and infrahyoids and lacked synchrony of the orbicularis with either infrahyoids or the masseter. O’Dwyer and Nielson [156] in non-speech facial gestures found that the CP group recruited more muscle groups during gestures but not in consistent patterns with less synchrony than the non-CP group. Symmetry describes the activation of both left and right muscle groups. Ries and Berzin [157] reported more asymmetry in the CP group during isotonic and isometric contractions, while Itakura [143] found less symmetry in the temporalis muscle activity in CP compared to TD. Similarly, Shamansurov et al. [160] found weak correlation coefficients between bilateral muscle activity. Yoshiyasu [163] also reported lesser synchronization in the anterior temporalis and masseter muscles during the chewing cycles among CP compared to TD.

5.5.4 Other Muscle Measures

A series of studies by O’Dwyer and Neilson seek to understand the underlying patterns of dysfunction in the control of muscles. Athetoid CP is characterized by involuntary movements. However, O’Dwyer and Neilson [154] find that this involuntary activity does not affect the formation of syllables but occurs between syllables. In a later study, O'Dwyer & Neilson [155] used spectral density curves to quantify the frequencies present in the EMG waveforms. The athetoid CP group had uniform spectral density curves across all muscles characterized by a peak at low frequency compared to the TD group which showed a higher variation between muscles. In general, the TD group had higher frequencies and broader bandwidth compared to the CP group. Neilson et al. [153] recorded EMG characteristics during measurements of the tonic stretch reflex (TSR) in the lip jaw and tongue muscles. The results of this study showed similar characteristics in both the CP and TD groups, specifically, that tonic stretch reflexes are present in the jaw-closing muscles but not lip and tongue muscles. Vaughan et al. [162] the participants were asked to track the movement of a target marker by activating certain facial muscles (masseter, orbicularis oris superioris) linked to the tracking marker to determine whether muscle spindle reflexes associated with spasticity affect the control of the masseter (contains spindles) and orbicularis oris superioris (no spindles). The CP group had less success tracking the marker than the control group with either muscle group. In the spectral analysis, there was no difference in the frequency range but greater magnitudes of power in the CP group.

5.6 Orofacial Hard Tissue Structures

For the ease of evaluating the literature present on the hard tissue structures of the head/skull, the articles were categorized based on the methodology. There were two main categories, namely, cephalometric evaluation to assess skeletal characteristics, which was performed in six of the nine studies [164-169], and cast model analysis assessing dental characteristics, which was performed in five of the nine studies [164, 169-172]. Unfortunately, the studies do not consistently report the same cephalometric parameters.

5.6.1 Cephalometric Analysis

5.6.1.1 Cranial base

The cranial base was discussed in two studies [166, 168]. Foster et al. [166] measured the length of the anterior (N-S), posterior (S-Bo) and total (N-Bo) cranial base. It is important to note that the cephalometric landmarks of the cranial base in this study, were calculated using Bolton's point. Here, the CP group was stratified according to severity. The lengths of the anterior and total cranial base were significantly higher in the TD group compared to the most severe CP group. Cranial base lengths were not significantly different from controls in the intermediate severity CP group, while total cranial base length was significantly less than controls in the least severely affected CP group [166]. Poshtaru et al. [168] also observed that the length of the anterior cranial base (N-Se) was significantly lower in CP than TD, while there was no difference in the posterior cranial base (Se-Ba). Furthermore, Poshtaru et al.[168] observed that the cranial base angle (NSBa) was significantly lower in CP than TD group; however, Foster et al. [166] did not observe any significant differences in cranial base angle (NSBo).

5.6.1.2 Maxilla

The anatomy of the maxillary bone was studied based on its position/angle with respect to the cranial base and its length. The angles SNA or AMP-S-N describe the maxillary position with respect to the cranial base. From the search, four articles [165-167, 169] discuss the position of the maxilla and all found no significant differences between the CP and TD groups. In the study by Foster et al. [166], the author reported no difference between the control and CP groups in S-N-AMP. Strodel [169] observed a trend towards greater SNA in the CP group, which was not significant. Karwetzky and Tielsch-Keuthen [167] measured the angle of the maxilla with respect to the cranial base (NSe-NA) and did not find a significant difference. Lastly, Brown & Sharma [165] measured SNA in 8-12 year old children and found no significant difference between the CP and non-CP groups. The study populations in three of the articles had a large age range and it is not always clear how age-related changes are taken into account in the analyses [166, 167, 169]. An aggregation of data over this large age range could bias the results because of the growth of the bones and sex-dependent timing of growth. If the individuals with CP do in fact have a shorter the cranial base, then a larger SNA may be expected.

The length of the maxilla was measured in two studies [166, 168]. Foster et al. [166] found a significantly greater mandibular body length (AMP-PMP) in severely affected individuals with CP compared to TD, but no significant difference was observed for the intermediate and least severely affected groups. Karwetzky & Tielsch-Keuthen [167] observed sagittal extension of the maxilla in 57% of the CP cases. Poshtaru et al. [168] found no significant difference between the CP and control groups in A'-Snp and Strodel [169] observed no significant differences in the maxillary bony arch length.

5.6.1.3 Mandible

The mandible bone anatomy can be studied based on its position with respect to the cranial base, length of the mandibular body, ramus height, divergence of the mandibular plane and gonial angle. The angle SNB depicts the mandibular position with respect to the cranial base. Two studies [165, 169] measured SNB and found no difference in the SNB angle in CP compared to TD groups. However, in the study by Brown & Sharma [165] the SN-Pog was found to be reduced in 4, 5, and 6-year-old boys with CP compared to the TD group.

The divergence of the mandible (Yaxis – SN, SN-GOGn) is another important parameter because it measures the changes in the vertical height of the face. These measures describe the forward and downward movement of the mandible in relation to the angle of the cranial base (SN). Two studies [165, 169] measured the divergence of the mandible. Hyperdivergent growth of the mandible results in face appearing longer when compared to the norms. Brown & Sharma [165] found a greater divergence angle among the CP groups compared to the TD group. However, Strodel [169] did not measure a significant difference between the CP and TD groups. These two studies differed in their approach: Strodel takes the mean value from children aged 4-22 years, while Foster stratified the data by age. Another parameter that contributes to an increase in vertical height is the Gonial angle or mandibular plane angle, which was measured in three studies [165-167]. Foster found no difference in the mandibular angle (M-G-Con) between the CP and non-CP groups. Brown and Sharma [165] found a greater mandibular plane angle (MP-SN) in 5 and 6 year-old boys with CP compared to controls. Karwetzky et al. [167] found that all CP cases either matched or exceeded the mean gonial angle for TD.

In addition to this, the length of the mandibular ramus contributes to the vertical facial height. Height of the ramus is discussed in four studies.[164, 166-168] Karwetzky & Tielsch-Keuthen [167] found that the mandibular ramus was shortened among the CP group. Similarly, Foster et al. [166] report a significantly smaller ramus height in the intermediate and severe CP groups. Poshtaru et al. [168] also measured a smaller ramus height (Go-Co) in the CP group.

Foster et al. [166] measured the length of the mandibular body (menton-gonion) and the total mandibular length (menton – condyle) and found it to be significantly smaller length in severely affected CP than non-CP; however, there was no significant difference for the intermediate or least severe CP groups. Karwetzky & Tielsch-Keuthen [167] also found that the mandible length was smaller than average in 82% of the cases with CP. Furthermore, Poshtaru [168] measured the length of the apical base of the lower jaw (B’-mi) and found it to be significantly smaller in the CP group than in TD; however, the length of the mandibular body (Pg-Go) was not significantly different.

5.6.2 Model Analysis

5.6.2.1 Palate

Palatal characteristics were measured on dental casts and discussed in three studies [164, 170, 171]. Mamaghani, et al. [170] measured palatal height index (percentage ratio of palate height / posterior dental arch width) in two age groups (18-36 and >36 years) of adult participants with CP and compared the data to normative values. There were no significant differences between the age groups in the CP population; however, in comparison to normative data, the palatal height index indicated a flatter palate in CP population. Dummett [171] found no significant diffrences in the platal height and palatal vault angle in children with and without CP. However, Dummett [171] reported that the anteroposterior length of the palate was greater in the quadriplegic group and greater in the spastic group during the permanent dentition stage. Album, et al.[164] reported that the palate breadth measured on dental casts was greater in the TD group compared to the CP group.

5.6.2.2 Dental Arch

The dental arch characteristics were reported in two studies [169, 172]. Strodel [169] reported a significantly greater dental arch circumference and dental arch length in the maxilla and mandible in the CP group than compared to the TD group. Oktay [172] reported an increase in the circumferential dental arch lenth from the ages 4 to 8 as a result of the growth of the alveolar arches in both spastic CP and TD groups; however, no comparison was described between the groups.

5.5 Muscle Physiology- Kinetics

Orofacial kinetics were recorded in six studies [146, 149, 150, 152, 173, 174]. Two studies analyzed exclusively the kinetics of orofacial structures [173, 174], whereas three analyzed orofacial kinetics and muscle activity [149, 150, 152] and one analyzed kinetics and kinematics [146]. Three of these studies [150, 152, 173] quantified maximum bite force, one study measured lip pressure [174], another article discussed intraoral pressure/suction during oral stages of swallowing [149], and another study used force transducers on the lips, tongue and jaw [146]. Even though the techniques and measuring parameters were different among the studies, it was observed that the force and/or pressure generated by the CP group is less than the TD group.

Santos et al. [173] used an electro gnathodynamometer device to measure maximum bite force in children with CP who were teeth grinders or non-grinders. Children with CP who were teeth grinders had a lower maximum bite force when compared to the non-grinding CP group. The groups were further stratified by oral motor function assessment scale (functional vs subfunctional) and it was found that the subfunctional groups have a lower maximum bite force than the functional groups in both grinders and non-grinders [173]. Similarly, Nakajima et al. [152] measured masticatory efficiency and biting pressure in children with and without CP. Maximum bite force along with mastication efficiency were both observed to be higher among the non-CP participants compared to CP participants. The study also reported a significant relation between mastication efficiency and biting pressure in the non-CP group; however, no significant relation was found in the CP group. Matsui et al. [150] also report maximum biting force in adults with CP. The maximum bite force was not significantly different between the CP and TD groups; however, oral motor function was measured with the Orofacial Motor Function Assessment Scale and the participants were classified as slightly impaired or moderately impaired. Therefore, in addition to differences in age of the study populations, the impairment may have been less severe in Matsui et al. [150] in comparison to Nakajima et al. [152] and Santos et al. [173].

Chigira et al. [173] measured lip pressure during lip closure around a spoon during eating. Here, the non-CP group had greater peak height of lip pressure compared to the CP participants. The coefficient of variation of the pressure recorded on 10 repeated trails showed that the TD participants had more pressure uniformity than CP. Lespargot et al. [149] measured intra-oral (suction) pressure on the palate during the oral phase of swallowing liquids. There was a lot of scatter in the measurement of intra-oral pressure but the authors observed that droolers had incomplete lip closure, residual liquid in the oral cavity after swallowing and abnormal suction. Lastly, in the study by Barlow and Abbs [146], participants were asked to reach a target force by generating force in their lips, tongue and jaw interfacing with specially designed force transducers. The rate of change in force was lower in the CP than the TD groups, which indicates their reduced capacity to recruit fine force control.

In summary, the studies have a wide variation in objective; however, overall most studies investigating the kinetics of the orofacial region found lower forces produced in individuals with CP than TD. Indeed, the level of force produced may depend on the severity of oral motor function.

CHAPTER 8: METHODOLOGIC QUALITY ASSESSMENT

For the critical appraisal of this scoping review, the AXIS and the MMAT tools were corroborated into making the final list of appraisal questions used to assess the included studies. (see below: Table:) the The scoring was done based on answering the appraisal questions as ‘Yes” or ‘No” and in turn scoring them, with ‘1’ and ‘0’ respectively.

Q No.	**Quality Criteria**	**Yes (1)**	**No (0)**
Q1	Was the research question or objective in this paper stated?		
Q2	Was a sample size justification, power description, or variance and effect estimates provided?		
Q3	Was the study population defined (regarding the Age, Sex, Type and severity of CP)		
Q4	Were the inclusion and exclusion criteria stated in the study?		
Q5	Were the outcome measures (dependent variables) clearly defined, valid and reliable?		
Q6	Were the outcome assessors blinded to the exposure status of participants?		
Q7	Did the authors report bias or discuss potential sources of bias?		
Q8	Did the author(s) report the statistical analysis?		
Q9	Were the results presented as mean and +/- (std) or if applicable in the form of tables/graphs?		
Q10	Were the results described?		
Q11	Were the authors' discussions and conclusions justified by the results?		
Q12	Were the limitations of the study discussed?		

Table 1: Questions for Methodologic Assessment

Studies Reviewed	**Q1**	**Q2**	**Q3**	**Q4**	**Q5**	**Q6**	**Q7**	**Q8**	**Q9**	**Q10**	**Q11**	**Q12**	**Total Score**
Album, et al. (1964)	1	0	0	1	0	0	0	1	1	1	1	0	6
Brown & Sharma (1967)	1	0	0	0	0	0	0	0	0	1	1	0	2
Karwetzky & Tielsch-Keuthen (1972)	0	0	0	0	0	0	0	0	1	1	1	0	3
Oktay (1972)	1	0	0	1	0	0	0	0	0	0	1	0	3
Foster, et al. (1974)	1	0	0	1	0	0	0	1	1	1	1	0	6
Strodel (1987)	1	0	0	1	1	1	0	1	0	1	1	0	7
Poshtaru, et al. (2019)	1	0	0	1	0	0	0	1	1	1	1	0	6
Dummett (1975)	1	0	0	1	1	0	0	1	1	1	1	1	8
Mamaghani, et al. (2008)	1	0	0	1	0	0	0	1	1	1	1	0	6
Hong, et al. (2007)	1	0	0	1	0	0	0	1	1	1	1	0	6
Chen, et al. (2010)	1	0	1	1	0	0	0	1	1	1	1	1	8
Hong, et al. (2011)	1	0	0	1	0	0	0	1	1	1	1	1	7
Nip (2013)	1	0	1	1	0	0	0	0	1	1	1	0	6
Nip, et al. (2017)	1	0	1	1	0	0	0	1	1	1	1	1	8
Nip (2017)	1	0	1	1	0	0	0	1	1	1	1	1	8
Remijn, et al. (2015)	1	0	1	1	1	0	0	1	1	1	1	1	9
Nip, et al. (2018)	1	0	1	1	0	0	1	1	1	1	1	1	9
Yoshiyasu (1988)	1	0	0	1	0	0	0	0	1	1	1	0	5
Santos, et al. (2010)	1	0	0	1	0	0	0	1	1	1	1	1	7
Serafinowska, et al. (1981)	1	0	0	1	0	0	0	0	1	1	1	0	5
Shamansurov, et al. (1982)	1	0	0	1	0	0	0	0	0	1	1	0	4

Sochaniwskyj, et al. (1986)	1	1	0	1	1	0	0	0	1	1	1	0	7
Briesemeister, et al. (2013)	1	0	1	1	0	0	0	1	1	1	1	0	7
Ries & Bérzin (2017)	1	0	0	1	0	0	0	1	1	1	1	0	6
Mishra, et al. (2019)	1	0	1	1	1	1	0	1	0	1	1	1	9
Neilson & O'Dwyer (1984)	1	1	0	1	1	0	0	0	0	1	1	1	7
O'Dwyer & Neilson (1988)	1	0	0	1	1	0	0	1	1	1	1	0	7
Neilson, et al. (1979)	0	0	0	0	0	0	0	0	1	1	1	0	3
O'Dwyer, et al. (1983)	1	0	1	1	0	0	0	1	1	1	1	0	7
Vaughan, et al. (1988)	1	0	0	1	0	0	0	1	0	1	1	0	5
Itakura (1997)	1	0	0	1	0	1	0	1	1	1	1	0	7
Rong, et al. (2012)	1	0	0	1	0	0	0	1	1	1	1	1	7
Connaghan & Moore (2013)	1	0	0	1	0	0	0	1	0	1	1	1	6
Chigira, et al. (1994)	1	0	0	1	1	0	0	1	1	1	1	1	8
Santos, et al. (2015)	1	0	1	1	0	0	0	1	1	1	1	1	8
Lespargot, et al. (1993)	1	0	0	1	0	0	0	0	0	1	1	0	4
Nakajima, et al. (1988)	1	0	0	1	0	0	0	1	1	1	1	0	6
Matsui, et al. (2017)	1	1	1	1	1	0	0	1	1	1	1	1	10
Barlow & Abbs (1986)	1	0	1	1	0	0	0	1	1	1	1	0	7

Table 2: Scoring from Methodologic appraisal.

Table 2 shows the results of the quality appraisal. Only three studies [150, 154, 161] performed a power analysis (based on Q2) to determine or justify the sample size. The inclusion and/or exclusion criteria (based on Q3) were stated in majority of the studies. However, it is relevant to discuss that the three studies [153, 165, 167] that did not discuss the eligibility criteria were published before the 1980s. The validity and reliability of the outcome measures, (based on Q5) was reported only in 9 studies [147, 150, 151, 154, 155, 161, 169, 171, 174]. The internal validity (based on Q6) scores were low in a majority of the studies as the analyses or examiners were not blinded to the condition of the participant. Only three studies [143, 151, 169] reported internal validity, stating that the outcome assessors were blinded to the exposure status. The other sources of potential bias (based on Q7) was only discussed in one study [141]. Twenty-eight of the thirty-eight included studies described or reported a statistical analysis of the results (based on Q8). The results were presented as mean and +/- (std) or in the form of graphs and table in twenty-nine studies (based on Q10). The limitations of were discussed in fifteen studies (based on Q12). Among all the limitations in these studies, the most frequently reported limitations were: 1) small sample size, 2) no validation of outcomes measures, 3) poorly described population characteristics. The study by Matsui, et al. (2017)[150] had the overall highest scoring, which indicates good methodological reporting.

CHAPTER 9: DISCUSSION

Bones can adapt their shape, morphology, and possibly material properties to their mechanical environment. This is thought to occur at the bone cell level, where osteocyte bone cells within the bone matrix sense deformation and coordinate bone formation or resorption. Indeed, bone formation occurs in response to greater levels of strain, while bone resorption occurs in response to lower levels of strain. In individuals with cerebral palsy, deformities of the lower limb and hip occur as bones maladapt to the abnormal movement and muscle tone.

It is unknown in Cerebral Palsy whether bones in the head and neck region maladapt with respect to their adjacent muscles and muscular forces. Abnormal muscle tone and movement also occur in the head and neck in CP. 80% of children with CP have orofacial dysfunction affecting their ability to chew, speak, retain saliva, etc. In 2021, a scoping review assessing the oral health outcomes for people with CP was published by Lansdown K et al. [175] and in 2020, authors Bensi et al. [123] conducted a systematic review and meta-analysis on the oral health status and dental characteristics of children with CP. Lansdown K et al. [175] reported that CP individuals have a higher susceptibility to poor oral health with a risk of dental caries, tooth wear and bruxism. He also reported a lack of research conducted on people with CP over the age of 18 years [175]. Bensi et al. [123] reported the greater risk of dental caries among CP individuals with primary dentition and the occurrence of dental characteristics like Angle's Class II occlusion, anterior open bite and a lower gingival status [123].

However, our scoping review aims to aggregate knowledge across all disciplines that quantitatively assessed the effects of CP on orofacial structures (i.e., hard and soft tissue, airway) in response to orofacial (dys)functions. To enable this, a scoping review protocol was chosen to identify knowledge gaps and synthesize knowledge in the literature spanning disciplines investigating individuals with CP in the head and neck. To the best of our knowledge, this is the first such study. Thirty-nine studies met the inclusion criteria and were reviewed. The included studies, assessed orofacial dysfunctions, most commonly speech motor impairments, drooling and the impaired ability for mastication and swallowing in CP.

9.1 Orofacial Skeletal and Dental Presentation in CP

The following section discusses the skeletal and dental characteristics observed in individuals with CP.

The Cranial Base

The literature in our scoping review reported that the cranial base length was shorter [166] and the cranial base angle was smaller [168] among the individuals with CP compared to controls. The SNA (position of the maxilla with respect to the cranial base) was larger among the CP than in TD with no significant differences observed in the SNB (position of the mandible with respect to the cranial base). However, the SN-Pog was larger among the CP population. In general, a shorter cranial base length is consistent with a greater SNA angle, as observed in CP vs TD. These findings are similar to the results reported by Järvinen [176], stating that the SNA is dependent on the cranial base characteristics. So a decrease in the cranial base angle or length could lead to an increase in the SNA based on the geometrics in the bone structures [176].

Vertical Facial Dimensions

In normal growth patterns, unlike the maxilla which grows in association with the cranial base, the mandible demonstrates independent growth patterns as it is not associated with the cranial base

[177]. The usual downward and forward movement of the mandible during growth is actually associated with posterior and superior growth and remodeling of the ramus and condyle. This results in the ramus growing deeper in the anteroposterior direction with an increase in the vertical facial height [178]. Our scoping review reported that, the vertical facial dimensions evaluated cross-sectionally in the CP group had larger values of mandibular divergence and gonial angle in comparison to the TD group. It follows that the mandibular ramus height was shorter in the CP group. This is seen in other studies suggesting the association of weak masticatory muscles and soft diets affecting the skeletal structure. Kiliaridis et al.[179] that stated that individuals with myotonic dystrophy had weak masticatory muscles and presented with large angles between the mandibular and palatal plane and a steep mandible. This could suggest the presence of abnormal muscular forces and/or activity in CP that are causal and the possible presence of a "long face" (increased anterior vertical facial height in comparison to TD).

The Cranial base associated with Class II

The common occlusal characteristics in CP are open bite, greater overjet and overbite in CP and the classical presentation of Angle's Class II occlusion [123]. A study reporting normative data, demonstrated the presence of short and flat posterior cranial bases with backward and upward mandibular condyles associated with the tendency for occlusal class II [180]. This is suggestive of the reason behind the characteristic class II occlusal characteristic to the presence of the short cranial base length in CP participants. (as mentioned above.)

The palate

In this review, the height of the palate in the CP group was either "flat" or showed no significant differences which are contradictory to the classical "high arched palates" also observed in CP [181]. However, this contradictory finding could be because of the limited number of studies discussing palatal features, in this review.

A hypothesis can be made of an association between the above-mentioned skeletal and dental characteristics, with the abnormal muscle activity and the corresponding abnormal functional patterns of the orofacial(masticatory) muscles, lips and the tongue in CP. This association may imply that that the size, morphology or relative positioning of orofacial structures, like the bones, muscles or airway, adapt to abnormal muscle tone and/or dysfunctions in physiological processes like mastication, swallowing, breathing etc. When these adaptations occur during growth of bone structures, they can be more extreme.

A few descriptions of the mechanisms elementary to bone adaptations in response to forces corresponding to bone development in CP, have to be recognized. The underlying physiological presentations and mechanisms contributing to the disorder are discussed below.

The focus was to relate these skeletal architecture findings to subjacent muscular activity and functional patterns.

9.2 Biomechanical adaptations in CP

Animal models exploring mechanical interactions between muscle and bone highlight adaptations by demonstrating that disruptions in the muscle forces do in fact lead to subsequent bone changes [182]. The mechanical relationship observed between muscles and bones is due to muscular activity and forces generated during function causing active deformation in the bone structure [119]. This muscular change occurs due to various mechanical stimuli. In the case of neurogenic disorders, due to the nerve dysfunctions, the mechanical stimuli bringing about muscular changes may affect the bone tissues adjacent to them pathologically.

“Cerebral palsy is known to phenotypically manifest via muscle dysfunction, reduced skeletal loading and bone degradation” [182-184]. However, this is not adequately measured quantitatively and described for the orofacial region in CP individuals. The following section attempts to aggregate knowledge describing the effects of CP-related muscular activity and forces on bone morphology.

In the case of the orofacial region, there are studies that show that CP-related deficits in recruiting muscle forces at normal rates, lead to impairments in the oromotor performances, especially affecting the fine force control in CP [146]. But our primary interest was to further relate these impairments to bone adaptations.

Presentation of Open Bite: As mentioned above, the increased anterior vertical facial height characteristics in CP compared to TD could be suggestive of the presence of malocclusions like an open bite. From the studies included in this review, forces and pressure-related dysfunctions with regard to the lips in CP participants are manifested as- incomplete lip closure with abnormal suction and scattered measurement of intra-oral pressure in droolers [149], lower rates of forces in the tongue, jaws and lips in CP [146], as well as reduced peak height of lip pressure in CP, compared to the TD participants [174]. Insufficient perioral forces leading to incompetent lips could be a possible explanation for the presence of an open bite in CP. The abnormal forces and movement of the tongue, along with the prevalence of tongue thrusting in CP [185] could also lead to the presentation of an open bite.

Presentation of Small Palatal Arches: From our results, Album et al. [164] discussed the presence of small palatal breadth in the CP group and related it to the characteristics of the tongue. Album suggested that the small palatal arches could be due to habits like mouth-breathing where the tongue would fail to exert its myometric function of linguo-buccal pressures, thereby leading to transverse expansions of the palate. However, it is not clear whether this lack in myometric function occurs as a concomitant of growth or whether it occurs due to the forces and muscle activity patterns observed during habits like mouth breathing.

Presentation of Short Mandibular Ramus Height: The mandibular ramus is the region of insertion for the superficial and deep masseter and the depressor labii inferioris, depressor anguli oris. In addition to this, the buccinator originates from the oblique line present on the ramus of the mandible. The masseter is one of the most important muscles for mastication. Forces and muscular activity that occur due to the movement of the masseter during mastication, clenching and speech could account for the reduced height of the ramus. In our interest, the EMG characteristics of the masseter were reported in various studies. The results from our scoping review revealed that the masseter presented with lesser EMG amplitudes during clench[150, 158, 160] and mastication[160], greater variability in durations of muscle activity and interval time [152] and fewer patterns of synchronization [143, 156, 157, 160, 161, 163] in CP than in TD individuals. Our review also reported contrasting results on the presence of increased masseter muscle amplitude during MVC [148] and tooth-tapping [143] , presence of muscle activity during rest [148-150, 159-161], prolonged duration of muscle activity [148, 154-156, 163], greater active muscle period [148], and greater magnitudes of power associated to masseter muscle spindles [162], in CP individuals compared to TD. Due to the differing results, it is impossible to draw exact conclusions on the reasons for short ramus height in CP. However, it can be assumed that the presence of reduced muscle amplitudes and irregular masseter muscle activity patterns in CP individuals, cause motor impairments during mastication, leading to the short mandibular ramus height. This statement can also be supported by the lower force recruitment in the jaws [146] and the lower MBF [152, 173] causing the motor impairments leading to the effect on the mandible. Hence it is more experiments need to be developed in order to assess the relationship between orofacial muscle activity and bone structures.

9.3 Physiological imbalances in CP

Reiterating the "Functional matrix theory" by Melvin Moss, "form following function", essentially dictates the skeletal growth pattern [56]. Apart from the bone changes that occur due to growth-related physiological mechanisms (like suckling in infants), the prevalence of parafunctional habits, could bring about additional changes to the tissues. With this regard, any alterations in the orofacial functional physiological mechanisms or parafunctions observed in CP are even more likely to translate to deformations in the orofacial skeletal and dental growth [186-188]. The following section discusses the effects of CP on general physiological functions and parafunctions brought about due to muscular activity and forces.

9.3.1 General physiological functional changes

Suckling: Suckling seen in infants and neonatal growth requires the facial musculature to generate negative pressures, and the tongue to generate a tight seal in TD individuals. In CP, abnormal oral sucking habits during infancy could lead to acquired non-nutritive sucking habits (like thumb-sucking and tongue thrusting) and eventually cause malocclusions [189, 190]. The sucking habits observed in CP could be attributed to the decreased perioral muscle tone, masticatory muscle tone and tongue motor control [191]. Even the "tongue-jaw lowering" mechanism [190] that underlies sucking, could be affected due to motor impairments in CP.

Drooling and Swallowing complex: Drooling occurs due to improper voluntary motor activity required during swallowing and also deficits in the oral sphincter. Swallowing dysfunctions occur if there is an impairment in the synchronous movement of the muscles in the lip, tongue, palate, jaws, pharynx, larynx and respiratory muscles. Swallowing and drooling impairments also occur due to the lack of the forces the muscle groups recruit.

In this scoping review, the scatter in intra-oral pressure measurements and the presence of residual liquid among the droolers in CP, reduced insufficient perioral forces leading to incompetent lips [149] and the failed synchronization in the orbicularis oris with either the masseter or the infrahyoid muscles [161], could be the reason for drooling and poor retention of the swallow contents. Authors Sochaniwskyj et al. [161] attributed the lack of synchronization of the muscle activities to the differences in the EMG values of the masseter and the infrahyoids.

Respiration-swallowing coordination:

Swallowing can also be integrated to respiratory impairments in CP to understand the swallowing-respiration coordination due to the same muscle groups. The infrahyoid group of muscles play an active role in the swallowing function through the movement of the larynx required for respiration. From our scoping review, Mishra et al. [151] only reported the prevalence of a greater number of

CP children than TD, with CP children demonstarting post-swallow inhalation and impaired voluntary cough functions during in feeding and swallowing tasks.

Mastication, Chewing and Clenching:

The results of this scoping review show evidence to support the reduced masticatory abilities in CP. The reports on the CP group having greater path distances and jaw displacements [141, 144], lower muscle activity in the masseter and temporalis during mastication [150, 158, 160], increased variation in the duration and interval times of muscle activity during the chewing cycle mastication [152] and the presence of asynchronous muscle activity [143, 156, 157, 160, 161, 163], could be suggestive of motor impairments and lack of muscle coordination in the mastication of CP individuals. Additionally, the lower recruitment of lip, jaw and tongue force rates [146] also suggests the impaired ability of fine forces to perform the functions of mastication, chewing and clenching. Diet and Food consistency also plays a major role in determining the masticatory ability. To support this, there were reports CP individuals having greater maximum amplitudes during consumption of puree and chewable solid food but not for liquids [161] , greater path distances and working space for soft and solid food but not for puree during mastication [141, 144], and lower tongue velocities [146], which suggests the requirement of greater effort required to process food of harder consistencies. Additionally, the presence of a prolonged duration of active muscle period in CP [148] could be attributed to the increased random head movements and involuntary muscle activity in CP during feeding tasks.

Speech

Majority of the results in this scoping review present speech-related changes at a structural level in CP. It is evident that speech, articulation and communication is an intricate process and should involve the integration of multiple kinetic, kinematic and muscle activity-related parametric discussions. Our scoping review reported diverse results on the orofacial structural activity and movements. However, from our results, the effects of abnormal muscle tone in CP affected speech through the presence of greater - jaw displacements [137, 144, 145], jaw amplitudes during single word tasks [145], path distance and working space [141, 144], velocities in lip [137, 140] and jaw [137, 145], STIs [136, 138] in upper and lower lips [140] and reduced temporal coupling (coordination) in jaws and lips [137, 140], in CP individuals compared to TD. Individuals with CP also exhibited greater signal power [154, 155], duration, time and variability in the recorded EMG waveforms [154, 155] across syllables compared to TD and the reduced ability to target forces to their lips and tongue [146] in CP. All these results could be suggestive of the impaired motor functioning of CP individuals during speech and communication. The results are also largely dependent on the type of speech tasks.

9.3.2 Parafunctional changes in CP

Sleep apnea: On the context of respiration, it is important to discuss that although CP has a high prevalence for obstructive sleep apnea, most studies on CP related apnea were not eligible for this scoping review because of their focus on assessing the efficacy of treatments. Sleep apnea in CP-related orofacial effects is a severely under-investigated area in literature.

Bruxism: Sleep bruxism observed in CP is associated with the constant activity of masticatory muscles. The teeth grinding caused due to sleep bruxism often leads to temporomandibular joint complications and could also affect facial muscles and cause deleterious dental effects. However, this scoping review presented CP individuals to have low Maximum Bite Forces during teeth grinding [173] and these low forces recruited by the masticatory unit could be attributed to motor adjustments resulting from the jaw spasticity [173].

Mouth breathing and Tongue thrusting: These parafunctions are assumed to be associated with the lack of perioral forces (in the case of mouth breathing) and irregularities in the voluntary control of the tongue that could potentially lead to the development of open bite and small dental arches. (as discussed in above)

There were no studies relating muscle activity or quantification of function to orofacial structures. What is still unclear, is the extent to which abnormal muscular forces and activity in CP affect and regulate the physiological mechanisms underlying adaptation of orofacial structures, like bone.

9.4 Impaired Motor Control

The following section discuss theories concerning motor dysfunction in CP, that lead to abnormal reflexes on motor control. This section attempts to integrate multiple orofacial functions (multidisciplinary) due to the commonalities in their theories behind the motor control processes in the orofacial region.

9.4.1 Interarticular coordination deficits

Impairment of the speech mechanism (dysarthria) in CP is manifested in several ways. These features could attribute to impaired force control and can be supported by the fact that a reduced force control could lead to the creation of "ballistic movements" during speech, thereby causing slow speaking rates among CP individuals [139]. Among all the characteristics, reduced coordination of movements during speech is a key characteristic of dysarthria in CP. Mature speech production is associated with increased interarticular coordination and speech movement stability [60, 192, 193]. Spatial and temporal coupling in young TD individuals from 1-6 years begins with low levels of speech intelligibility but gets refined into adulthood [60]. In growing children with CP, articular coordination could also impact general motor development [194]. From our review, Nip. [139, 140] also reported the association of interarticular coordination with speech intelligibility and suggested that linguistic task demands often influence the speech motor control for CP children than for TD. This is also supported by the study by Chen et al. [136] stating high STIs and variability on utterance durations, that increased with the speech task in CP participants. Hong et al. [137] reported reduced temporal coordination between the lower lip and jaw among the CP individuals than TD. Rong et al.[145] suggested that the F2 depends on the independent tongue movements. On this context, tongue tip and jaw coordination account for speech movement stability [142, 144] and a "*compensatory strategy*" exists i.e. the jaw movements could be compensated by the reduction in lingual movements. Connaghan et al. [142] also described the presence of characteristic *lingual hypo-articulation* in the speakers with CP due to the reduced lingual movements. In growing children with CP, articular coordination could also impact their overall motor development [194].

9.4.2 Coactivation of agonist-antagonist muscle groups

Individuals with CP usually have abnormal muscle activity, particularly excessive or involuntary activities and in turn, have difficulties in inhibiting the excessive muscle activity due to the spasticity [195]. This would eventually lead to an increase in muscle tension and fatigue. In response to this, jaw muscles undergo an inhibitory mechanism called "*muscle co-activation*". In our scoping review, Connaghan et al. [142] reported muscular coactivation by observing the electromechanical delay (EMD) in EMG patterns during speech. Briesemeister et al. [148] observed muscular coactivation in agonist (jaw opener) and antagonist (jaw closers) muscles during mastication and associated it to the impaired masticatory performance seen in CP individuals. O'Dywer et al. [156] accounted for the co-activation of the facial muscles in CP individuals.

Several motor control deficits in basic physiological mechanisms among the structural complexes have been discussed. These motor control deficits occur in response to the abnormalities in the muscles and could lead to maladaptation of bones. However, it could be interesting to also relate the skeletal deformations caused by the muscular deficits, to the degree of motor impairment clinically observed.

CHAPTER 11: LIMITATIONS

The results interpreted from the included studies are limited mainly by the small sample sizes. Often, the studies reported the results of heterogenous CP populations or only focused on one CP population. Additionally, all the CP participants are exposed to different etiological factors resulting in a heterogeneous presentation of the disease. None of the included studies measured bone in relation to muscle activity, kinematics or kinetics; thus, the association between the changes in bone and musculature due to functions and/or forces is unknown.

CHAPTER 12: KNOWLEDGE GAPS

Our study has identified numerous knowledge gaps in this research area. The most important findings of the general study characteristics of the scoping review are:

There are very few studies describing the orofacial anatomical structure morphology in cerebral palsy. We found a handful of studies using cephalometry to investigate cerebral palsy. However, no studies were uncovered addressing muscle anatomy, including the size and shape of orofacial muscles. Furthermore, we did not uncover any studies addressing the shape or size of the airway and oral cavities, which are so important for respiration. Abnormalities in the airway or oral cavities could relate to obstructive sleep apnea which is prevalent in children with cerebral palsy.

CP is a very heterogeneous disorder in terms of type, presentation, and severity. Future studies should account for the type and severity of CP. The GMFCS scale was introduced in 1997 [196] and is the most widely used scale currently to describe gross motor function; while it is not directly related to the oral function, it is a standard marker of severity in individuals with CP. However, future studies should also incorporate measures of severity of orofacial motor function (such as Nordic Orofacial Test-Screening) [197], which could be used to associate deficits in orofacial function with bone characteristics.

All studies were cross-sectional in nature. Longitudinal studies following the growth of orofacial structures in relation to abnormal muscle tone would clarify the relationship between abnormal muscle tone and bone adaptation, especially during growth. This is important given the heterogeneity of the disorder.

CHAPTER 13: FUTURE OUTLOOK

With regard to CP, it is fundamental to understand the abnormal muscular activity and forces generated in the head and neck during functional movements such as mastication, swallowing, speech etc. as they may be related to comorbidities. However, there is inadequate literature regarding orofacial muscular activities and forces associated with orofacial bone deformities in CP.

Hence, more large-scale studies accommodating more participants and measured longitudinally, are required to further assess the structure-function adaptations in this heterogeneous disorder.

Newer techniques incorporating a multidisciplinary approach to quantitatively measure the orofacial tissue anatomy, also have to be introduced to understand the condition better. This would subsequently help in providing timely treatment options for CP associated with the orofacial region.

CHAPTER 14: CONCLUSION

Musculoskeletal adaptations in the orofacial region are complex interactive processes that can be understood by evaluating the underlying structure-function relationships. Any abnormality in the muscle adaptation can express skeletal abnormalities, thereby affecting the structure-function relation. Clinically, this may lead to deficits in functional abilities that can be assessed by various quantitative techniques.

Cerebral Palsy is a heterogeneous disorder with various clinical manifestations. Due to the abnormal muscle activity, motor function impairments are a characteristic feature of this disorder. The impact of the level of motor impairment is determined by the severity of the disease. In the orofacial region, the various dysfunctions demonstrated by CP can lead to secondary conditions that may even inhibit the efficiency of (?) respiration. Hence, CP needs to be studied at a fundamental level based on their musculoskeletal structure and adaptations that may occur in the orofacial region. There is no sufficient literature on how the orofacial structures respond to orofacial (dys)functions in CP, studied based on their structure-function relationships with a multidisciplinary approach. Our scoping review helps to aggregate relevant quantitative data, having a multidisciplinary approach.

Using the principles on conducting scoping reviews, thirty-nine studies were retrieved from the databases and were found to belong to 4 scientific disciplines namely- dentistry, dysphagia, physiology and speech & communication. Each discipline assessed were further categorized into the type of quantitative assessment technique used to evaluate the orofacial structures/functions studied. Based on the quantitative techniques used, the following results from the studies included are highlighted;

- All the studies were cross-sectional with 38% from dentistry, 28% from speech & communication, 26% from medicine-dysphagia and 8% from physiology, that were assessed using various quantitative techniques.
- Jaw kinematic (tongue, jaw, lips) characteristics revealed greater path distances and working spaces, velocity for speech tasks and STIs in CP than in TD.
- Muscle activity (EMG) characteristics revealed greater activity at rest in CP than in TD and less activity during MVC tasks. However, during speech and mastication tasks, the results were variable. Time domain measures revealed prolonged muscle activity periods and a lot of variability in the onset/offset timings and fluctuation patterns in CP than in TD. CP individuals demonstrated lower synchronization and irregular EMG characteristics and reflexes during other muscle measurements.
- Hard tissue measurements using cephalometry revealed that most studies reported short cranial base, greater SNA and mandibular divergence along with short mandibular ramus height in CP. Model analysis results were diverse in characteristics.
- Kinetic measurements revealed that CP had lower forces generated across various tasks and the measurements were performed using various quantitative assessment tools.

The discussion of the results is done to better understand the changes in the musculoskeletal structures that CP demonstrate with respect to the various (dys)functions. Therefore, this study helps to provide a deeper understanding on the underlying concepts and suggest possible explanations, but mainly to highlight the knowledge gaps present in the literature. The following are the key concepts discussed;

- The short cranial base in CP can be related to the large SNA angle.

- The large vertical dimensions in CP follows a short mandibular ramus height and is suggestive of the presentation due to the effects of the abnormal masticatory muscle forces as well as the role of diet.
- The characteristic open bite in CP is possibly caused by integrated effects of the incomplete lip closure and lower perioral muscular forces.
- The physiological changes in CP with regard to suckling, swallow-respiration complex, drooling & swallowing complex, mastication and speech etc. are suggested to be presented because of the impaired interactions and incoordination of the muscles of the respective systems that are demonstrated as insufficient forces/abnormal muscle activity.
- Mastication, chewing and clenching in CP show differences in their kinetic, muscle activity and kinematic parameters that are influenced by diet and motor reflexes
- Speech in CP shows differences in their kinetic, muscle activity and kinematic parameters that are influenced by nature of speech tasks.
- CP-related orofacial parafunctions are also assumed to be associated with dental deformities like open bite and small dental arch presentation.

In conclusion, our scoping review is the first to explore the effects of CP on the orofacial musculoskeletal structures and functions, studied quantitatively by a multidisciplinary approach. It also identifies the inconsistencies present in CP-related literature and encourages the need for a longitudinal design and more studies related to orofacial individual muscle morphology, to assess the structure -function relationships and relate them to the musculoskeletal deformities caused in the disorder.

Finally, it also proposes the need for multidisciplinary research to be conducted in CP to target the development of early and efficient treatment modalities which also helps to stop the progression of the comorbidities.

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APPENDICES

APPENDIX 1: Search Strategy for this Scoping Review

1. exp Cerebral Palsy/
2. exp Dyskinesias/
3. ("cerebral palsy" or "cerebral palsie?").tw,kf.
4. ("spastic diplegia?" or ataxia? or dyskine* or chorea).tw,kf.
5. 1 or 3
6. exp Skull/
7. (skull? or crani* or stomatognath* or orofacial* or oropharyn* or dental or odontolog* or cephalometr* or mandib* or maxill* or jaw or "temporal process*" or temporomandib* or zygomatic* or ethmoid or palate or palatal or palatine or "nasal bone?" or "dental arch" or vomer or lacrimal or "mental foramen" or oromotor* or "oral motor").tw,kf.
8. (occipitofrontalis or procerus or nasalis or "depressor septi nasi" or orbicularis or ((corrugator or depressor) adj1 supercilii) or ((orbicularis or "depressor anguli" or "levator anguli") adj1 oris) or ((levator or depressor) adj1 labii) or risorius or buccinator or mentalis).tw,kf.
9. exp Eating/ or (masticat* or chew or chewing or bite or biting or eat or eating or drink* or bruxism or occlusion or malocclusion or apn?ea or "mouth breath*" or "tongue thrust*" or (head? adj5 (postur* or position?))).tw,kf.
10. exp Stomatognathic System/
11. exp Stomatognathic Diseases/
12. exp Maxillofacial Abnormalities/
13. or/6-12
14. 5 and 13
15. exp Pterygoid Muscles/
16. pterygoid.tw,kw.
17. (skull? or crani* or stomatognath* or orofacial* or oropharyn* or dental or odontolog* or cephalometr* or mandib* or maxill* or jaw or "temporal process*" or temporomandib* or zygomatic* or ethmoid or palate or palatal or palatine or "nasal bone?" or "dental arch" or vomer or lacrimal or "mental foramen").tw,kf.
18. exp Eating/ or (masticat* or chew or chewing or bite or biting or eat or eating or drink* or bruxism or occlusion or malocclusion or apn?ea or "mouth breath*" or "tongue thrust*" or (head? adj5 (postur* or position? or oromotor* or "oral motor"))).tw,kf.
19. exp Masseter Muscle/
20. exp Temporal Muscle/
21. (masseter or temporalis).tw,kf.
22. exp Eating/ or exp Sialorrhea/ or exp Deglutition/ or exp Speech/ or (clench* or drool* or sialorrhea or hypersalivat* or swallow* or speech or speak* or talk* or facial expression? or masticat* or chew or chewing or bite or biting or eat or eating or drink* or bruxism or occlusion or malocclusion or "mouth breath*" or "tongue thrust*").tw,kf.
23. 6 or 8 or 10 or 11 or 12 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22
24. 5 and 23
25. 24 not 14

APPENDIX Table 1: Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	(i)
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	1-5
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	12-13
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	13
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	No protocol followed
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	44
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	43
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	45
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	43-45
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	45,46
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	45,46

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	46,47
Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	46
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	48,49
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	49-55
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	7-073
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	55-59
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	55-59
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	74-85
Limitations	20	Discuss the limitations of the scoping review process.	87
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	90-93
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	n.a

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.

* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.

† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g., quantitative and/or qualitative research, expert opinion, and policy documents) that may be eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).

‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.

§ The process of systematically examining research evidence to assess its validity, results, and relevance before using it to inform a decision. This term is used for items 12 and 19 instead of "risk of bias" (which is more applicable to systematic reviews of interventions) to include and acknowledge the various sources of evidence that may be used in a scoping review (e.g., quantitative and/or qualitative research, expert opinion, and policy document).

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