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Osteopathic Evaluation and Positional Plagiocephaly: A Descriptive Study on a Population of Children with ASD

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Abstract: The discipline of osteopathy is a whole person approach that incorporates medical and scientific knowledge using an array of manipulative techniques for diagnosis and treatment of several types of diseases (WHO). The ostheopathic examination allows to locate somatic dysfunctions which are the hallmarks of health imbalance caused by stressful events, external or internal to the body, such as trauma and / or other pathologies. The objective of osteopathic treatment is to improve posture and motor skills, which are the prerequisites for a balanced and harmonious development of the body. The osteopathic evaluation of children with autism spectrum disorder allows the identification of dysfunctional aspects at a somatic level that can enrich the understanding of the child's health and behavior, starting from the structure / function relationship, including craniofacial dimorphisms and plagiocephaly. The present exploratory research has made it possible to detect the presence of signs of plagiocephaly in about half of a sample of 250 preschool and school age children with autism spectrum disorder; the 44% of these, shows signs of craniofacial dysmorphism, which indicates a continuous morphostructural adaptation not yet sufficiently considered as an interferent element in the overall development of the child. The observed incidence is consistent with the incidence of plagiocephaly in the pediatric population and supports the hypothesis that in children with autism spectrum disorder it may be useful to integrate the osteopathic expertise with the other health professionals involved in the diagnostic and therapeutic process. The authors conclude that osteopathic observation can contribute to the definition of the functioning profile of the children with autism spectrum disorder and their needs, in a global perspective of taking charge and individualization of care.

Keywords: Osteopathy, Plagiocephaly, Autism Spectrum Disorders, Evaluation, Treatment

1. Introduction

Autism spectrum disorders (ASD) are included in the DSM-5 [1] (APA, 2013) among neurodevelopmental disorders; the two domains involved by the disorder are divided into social (socio-communicative deficit) and non-social component (restricted and repetitive behaviors and interests, altered sensory processing and perception); due to its pervasiveness, the set of symptoms compromises daily functioning and the onset usually occurs in early childhood.

In recent decades, in literature, have increased those studies investigating the coexistence of alteration of sensory and motor features [2-4] and their involvement in the pathogenetic process of some forms of autism. In consideration of this, if an osteopath was present within the multidisciplinary teams, a careful diagnostic palpation could also be carried out, to better understand the perceptual and postural-motor structure of the subject and promote health through the use of specific manual techniques aimed at normalizing somatic dysfunctions.

In fact, the contribution of osteopathy integrates the specialist medical clinic from a preventive perspective, especially when somatic dysfunction contributes to generate a fertile ground for a motor, sensory, somatovisceral disorder.

For example, over the years, the osteopathic manipulative treatment (OMT) has been shown to produce an improvement in gastroesophageal reflux symptoms,

including cervical spine mobility and pain [5]. In children with ASD, gastrointestinal symptoms are obviously not included among the diagnostic criteria, but they represent an area of growing interest and could be studied as they often coexist with stereotypies or core behaviors of autism. There are also some clinical conditions (peculiar abdominal pain relief positions, atypical evacuative position with increased abdominal press) or specific eating habits (food selectivity for consistency, high intake of liquids, self-reduction of dairy products and / or carbohydrates) for which the gastroenterological genesis should be carefully evaluated with respect to the neuropsychiatric one and, subsequently, an adequate therapy should be indicated. From this differential perspective, osteopathic palpatory diagnosis plays a supporting role for the clinician and the patient himself.

Furthermore, children with ASD often also present sensory-perceptual and neuropsychological alterations (including dyspraxia) which, although are not predictive or pathognomonic signs of the autism spectrum disorder, are frequently part of the clinical picture and are accompanied by somatic dysfunctions [6]. Some of these are compensatory forms of the organic or neuropsychological disturbance, others are instead determined by structural disharmonies or by alterations in the kinetics such as the reduction of cranial-cervical mobility, the minor extension of the vertebral column, the asymmetry of the shoulder blades or pelvic girdle [7]. Orienting among several types of motor asymmetries and atypia will be useful for the purposes of the differential diagnosis and the therapy to be adopted.

The observation of the general dynamics of the child always highlights crucial elements of evaluation to be integrated with the palpatory diagnosis, such as the general posture and that of the head, walking, motor behavior through the motor patterns implemented in relation to the surrounding environment, or to the people and the stimuli received.

The observation must obviously consider the age of the child to better understand the level of development achieved [8]. Motility, sensory and perception have a circular relationship; the sensory development proceeds with the motor one (sensory-motor development), where the motor component conditions and is conditioned by the sensory [6, 9]. Sensory, which is dysfunctional in most people with ASD, can also be altered by a structural difficulty as often occurs in children with plagiocephaly [10]. In these cases, the reduced mobility of the head with respect to the neck, can contribute to the establishment of a motor asymmetry of the upper limbs, but also of the lower limbs, which in the acquisition of motor stages conditions some developmental steps by adapting them through compensatory motor schemes.

The perception in a state of well-being usually generates an adequate motor behavior, so it is likely that if the child moves in a disorganized way he expresses an internal condition of discomfort and otherwise the perception deriving from an altered motor organization will itself be altered and this may cause further discomfort. In the case of children with kinetic anomalies, it is always useful to investigate whether these constitute a structural constraint or a response attributable to clinical conditions. We believe that sensoriality finds expression in morphology and body dynamics, which are highlighted through posture; while diaphragmatic dynamics is an adaptive and homeostatic form of the deepest sensoriality, morphology can be present from birth and can interfere with sensoriality. This perspective helps to interpret the presence of plagiocephaly.

1.1. The Plagiocephaly

The term Plagiocephaly derives from the Greek *plagios* (oblique) and *kephalè* (head) which means head distortion and clinically refers to asymmetries of the head [11]. Plagiocephaly is a quantitative descriptive morphological adaptation of cranial asymmetries involving the splanchnocranium in particular, the viscerocranium and the functionality of the organs there included [12].

To date, cranial macro deformities are diagnosed and defined as "plagiocephaly", leaving out all those asymmetries which are less evident in morphological terms, but which always give rise to dynamic compensation both of the bones themselves and of the body in general. Plagiocephalies can be divided into synostotic, nonsynostotic and syndromic: the synostotic plagiocephaly is due to premature closure of the sutures of the cranium; the nonsynostotic plagiocephaly is caused by extrinsic forces acting on the growing skull in the pre or postnatal period (deformational or positional plagiocephaly) [13].

In this study we will deal with the positional plagiocephaly, which do not involve the premature closure of the sutures of the cranium, and which are not associated with other organic / genetic alterations of known origin. According to a 2013 study, the incidence of plagiocephaly in the general population is now estimated at around 46% [14].

In 1992 the American Academy of Pediatrics began the “Back to Sleep” campaign for the prevention of the “sudden infant death syndrome” (SIDS), which indicates, as a fundamental element, the maintenance of the supine position of children during sleep, with the slogan "to sleep tummy up, to play tummy down". Despite of this slogan, the use of the prone position in the first months of life has not been sufficiently considered and supported. This actually led to a reduction in lethal events but caused a marked increase in plagiocephaly. This occurs when the infant is in the same supine position all day and night [15-17].

In addition to determining asymmetries and dysfunctions in the craniofacial district, plagiocephaly also conditions the movements of the head with respect to the neck and subsequently of the neck with respect to the spine, being able to determine, over time, if not adequately managed, scoliotic attitudes (descending scoliosis). Through the atlanto-occipital joint, it gives rise to postural compensations that are highlighted in the vertebral column. Distinguishing the local effects of plagiocephaly from the more general and topographically more distant ones is purely descriptive in consideration of the functional integration of the being but is useful for a better understanding.

As is well known, the development of the skeletal structure and motor control follow a top-down pattern: the articular

limitations of the upper districts can compromise the development of later more evolved functions [18-21]. During the first months of life, the effects of plagiocephaly are mainly highlighted at the craniofacial level, involving many important functions of neurodevelopment, such as vision, balance, sensoriality, orientation of the head in space, sucking, swallowing, breathing, mucus drainage in the middle ear [10, 11]. If there is no alternation of postures and the newborns are always positioned in the same way, the cranial bones, both those of the vault and the cranial base, malleable in the newborn and infant, are deformed.

1.2. The Osteopathic Observation

Osteopathy developed in the middle of the 19th century by Andrew Taylor Still, physician and surgeon of the USA, founder of the first independent osteopathic medical school at the end of the 19th century [22]. Osteopathy is based on physical manipulation of the body's muscle tissue and bones, both in the diagnostic and treatment phase. It respects the relationship between body, mind and spirit in conditions of health and disease; the emphasis is placed on the structural and functional integrity of the organism and on the intrinsic tendency of the latter to self-healing. Osteopaths use a wide range of manual therapeutic techniques, aimed at improving physiological function and / or supporting homeostasis that has been altered by a somatic dysfunction (body structure), for example impairment or alteration of component functions related to the somatic system; skeletal, arthrodial and myofascial structures; and the relative vascular, lymphatic and neural elements. Osteopaths, then, use their knowledge on the relationship between structure and function to optimize the body's self-regulation and self-healing abilities.

Plagiocephaly and dysmorphism in neurodevelopmental disorders.

There are several studies in the literature that have investigated the incidence of dysmorphic features in children with neurodevelopmental disorders [23-26], but these usually refer to minor physical anomalies (MPAs), defined as subtle, abnormal morphological features, such as deviations in morphology of the head, eyes, ears, mouth, hands, and feet, while the presence of plagiocephaly is still little investigated. According to Shapira and colleagues, the presence of multiple dysmorphic features in some children with ASD might identify distinct ASD phenotypes and serve as potential markers for understanding causes and prognoses. Their study shows that about 17% of children with ASD had dysmorphism, and these data are consistent with what has been reported by some studies (25.5% in [24]; 15.8% in [27]), but slightly higher than reported by others (10.8% in [25]; 5.6% in [28]).

In a recent study, Tian and colleagues [29] report that dysmorphisms are more common among individuals with ASD, disability, schizophrenia, hyperactivity than in the typically developing population [23, 30, 31]; for which they studied the relationships between dysmorphic characteristics in children with ASD and their cognitive and behavioral development. Their data suggest that the presence of dysmorphism is associated with decreased language production and comprehension skills in children with ASD, an association not observed in the control population. On the other hand, no associations were found between the presence of dysmorphism and the level of severity of ASD symptoms, as already described by Flor and colleagues [28]. Taken together, these findings suggest that dysmorphisms are only related to a global cognitive functioning of neurodevelopment for children with ID, regardless of ASD status.

Objective of the present research is:

Verify the incidence of positional plagiocephaly in ASD children, the level of severity of the plagiocephaly and the presence of differences due to the age of the children.

2. Methods

2.1. Participants

250 children with ASD, aged between 1.6 and 13.7 years, were recruited. Of these, 120 children (48%) showed signs of plagiocephaly. Children with plagiocephaly were aged from 1.6 to 13.6 years (mean 4.04 years; sd = 2.19 years). At the time of the research, 97% of children were under the age of 7. 90% of the children were male (N = 108). At the time of data collection for the research, the average score at the ADOS-2 was 17.40 (sd = 6.94).

2.2. Procedures

The children in the sample were recruited between 2016 and 2020. All children had received a diagnosis of autism spectrum disorder from public and private territorial services affiliated with the National Health System. The diagnosis was confirmed at the Institute of Ortofonologia (IdO) in Rome, where the research was conducted by a multidisciplinary team, with decades of experience, which includes various professionals including as well as osteopaths. All children who showed evident signs of neurological damage or sensory deficit, children who presented craniostenosis and myogenic torticollis were excluded from the sample. The osteopath has been present at the IdO for several years and participates in the global and multidisciplinary assessment of children with ASD.

2.3. Measures

Argenta Classification of Positional Plagiocephaly

Although various methods have been used to quantify and classify positional plagiocephaly, such as computed tomography (CT) or anthropometric measurements, the clinical observation is the simplest and most reliable method. As part of the integrated clinical evaluation shared with other professionals at the IdO, it was decided to use this observational protocol as it was non-invasive and easily usable with children, moreover corroborated by the palpatory examination, and therefore it proved to be a suitable tool for the research.

The Argenta is a clinical scale based on the morphological classification of positional plagiocephaly into 5 types,

depending on the severity of the asymmetry of the skull, the position of the ear and the appearance of the face, which is asymmetrical in the forms of greater severity, without taking into account the pathogenetic mechanism. of the shape [32] (Figure 1):

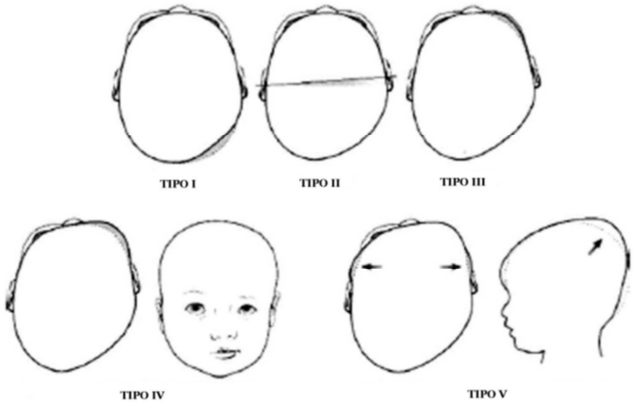

Figure 1. *The Argenta classification, a clinical 5-point scale of DP.*

Type 1: The cranial asymmetry is limited to the back of the skull. The degree of posterior flattening may vary, but the deforming action is limited to this anatomical region. There is no ear asymmetry assessed by measuring the nose-to-ear distance. The frontal squama is symmetrical, there are no anomalous temporal protrusions or vertical elongation of the skull. This represents the mildest form of positional plagiocephaly.

Type II: In this type of deformity, there are varying degrees of posterior cranial asymmetry. The effect on the medio cranial line and on the skull base, which determines the displacement of the ear on the involved side, forward or downward or in both directions, is quite significant. The asymmetry is usually more evident when examining the child from above. The anterior skull is not involved, and the frontal bone is symmetrical. There is no facial asymmetry. There are no compressive deformities of the skull. This type identifies a more severe form of positional plagiocephaly affecting not only the posterior skull but also the skull base and the central temporal fossa.

Type III: This type of deformity includes posterior flattening, ear shift and forehead asymmetry. This leads to cranial deformity, typically resulting in a parallelogram-shaped skull, characteristic of positional plagiocephaly and more easily found by examining the child directly from above. The face is symmetrical.

Type IV: This deformity, which includes posterior flattening, ear shift, forehead asymmetry and frontal and / or parietal sloping, begins to affect the position of the eyes, cheekbones, jaw, and nasal septum, causing asymmetricity in the baby's face. Facial asymmetry is the result of the displacement of the adipose tissue of the cheek or, less frequently, hyperplasia of the ipsilateral zygomatic area. This deformity reflects the progressive nature of the cranial asymmetry that comes to involve the anterior region causing deformation of the face.

Type V: In these patients the deformity includes posterior flattening, ear shift, forehead asymmetry, orbital, cheek, face or jaw deformity, frontal and / or parietal sloping, temporal bossing and increased cranial vault height.

All patients are clinically examined in four positions.

The first involves the observation of the child while he has a straightforward gaze position. This allows the observer to determine if there are asymmetries of the forehead and face. The second examination position takes place with the child seated, and the head is observed from above, while the child looks straight ahead. This allows for evaluation of forehead asymmetry, posterior cranial asymmetry, and ear malposition. The third position of the clinical examination takes place by looking at the back of the child's skull. This position allows confirmation of ear position and posterior asymmetry. The fourth examination position takes place by observing the child from a lateral position. This allows the observer to determine any degree of abnormal vertical growth of the skull, which can occur in severe plagiocephaly. Abnormalities are clinically visible or classified as present or not. For each of the five observed types of plagiocephaly, the practitioner assigns a score ranging from Level 0 (indicating no clinical sign) to Level 3 (significant presence of plagiocephaly signs).

3. Results

3.1. Descriptive

Of the 250 children with ASD who had the osteopathic examination, 120 children (48%) were found to have at least one sign of plagiocephaly. 90% of children with plagiocephaly are boys (N = 108) and for this reason gender differences have not been analyzed. At the time of assessment, they were 18 months to 164 months (mean 48.4 months; sd = 26.3 months); 97% were under the age of 7. As can be seen in Table 1 (Total column), among the 5 types of plagiocephaly the most detected is Type V.

Table 1. *Frequency (and percentage) of children showing signs of plagiocephaly, in each of the 5 types (N = 120).*

Type	*Level 1*	*Level 2*	*Level 3*	*Total on 250*
III	2 (0.8%)	1 (0.4%)	1 (0.4%)	4 (1.6%)
IV	3 (1.2%)	2 (0.8%)	0 (0%)	5 (2%)
V	59 (23.6%)	47 (18.8%)	5 (2%)	111 (44.4%)

3.2. Gender and Age Differences in the Distribution of Plagiocephaly Typology

No differences emerged in the frequency of the different types of plagiocephaly based on the age of the children (Chi Square = 3.617; P = .46) (see Table 2).

Table 2. *Number of children (divided by age) with different types of plagiocephaly (N = 120).*

Type	*< 3 years old (N = 39)*	*3-4 years old (N = 33)*	*>4 years old (N = 48)*
III	0	1	3
IV	1	1	3
V	39	31	42

4. Discussion

The main objective of this research was to verify the frequency of nonsynostotic plagiocephaly, also called positional plagiocephaly, in children with ASD.

This survey was an integral part of the neuropsychological assessment of the children in the sample examined. The results showed that in the sample of ASD children, 48% had signs of plagiocephaly, in particular the fronto-occipital form (type III, IV and V of the Argenta classification). About 44% had Type V, which is plagiocephaly with posterior cranial asymmetry, ear malposition as well as forehead and face asymmetry (with temporal protrusion and / or occipito-parietal anomaly).

The incidence of positional plagiocephaly before the 1992 "Back to Sleep" campaign of the American Academy of Pediatrics (APP) for the prevention of sudden infant death syndrome (SIDS), was about 1/300 live births (0.33%); following the recommendations of the AAP, the incidence has been reached between 1/68 and 1/72 live births [33]. Current prevalence rates are higher, ranging between 8.2% and 48% of newborns [14, 34, 35]. These data indicate that the percentages observed in the sample of children with ASD of the present research (48%) are completely comparable to those found in typically developing children.

In recent years, osteopathic therapy has also assumed a key role in the treatment of cranial malposition, which contributes to the optimization of vertebral alignment and mobility of the head and neck with normalization of the skull base, intraosseous sutures and strains (abnormal ligament tension).

There are no data relating to the use of osteopathic observation and therapy techniques for plagiocephaly in the population with ASD, also because of the age of diagnosis compared to the onset of plagiocephaly. However, in consideration of the number of ASD children with positional plagiocephaly, comparable to that of the general population, it seems important to reflect on the possible implications that this condition could have on the psychophysical well-being of children with ASD. This seems even more valid if we consider two elements: 1. ASD children have greater difficulty, if not impossibility, to verbally express their discomfort and even physical discomfort, therefore it is up to professionals (as well as parents) to find a way to identify states of dysfunction and / or structural conditions that could also negatively interfere with behavioral expressiveness. 2. There are some studies and researches that correlate postural and motor asymmetry with ASD, which for some scientists such as Gallese would explain the dyspractic origin of some forms of ASD [36]. Consequently, we believe that osteopathic observation can give a great contribution within the multidisciplinary team, both in the assessment and in the therapeutic planning. Particularly in the first years of life, when the child learns mainly from sensory-perceptive-motor experiences and in ASD children where atypical sensory and motor asymmetries often contribute to the structuring of dysfunctional behaviors.

Regarding the age of children, there is now a wide international literature that underlines the importance of osteopathic intervention in the neonatal period, which highlights the effectiveness of osteopathic treatment in the presence of symptomatic and idiopathic asymmetries in the first months of life [37, 38]. In this research, children have been involved from the age of 18 months, the age in which the first signs and symptoms of autism are usually observed, so intervening through osteopathic treatment in the first months of life means to be able to prevent the onset of dysfunctions (not related to autism) and their consequences; furthermore, this treatment could favor the development of other areas of functional regulation of the child, for example those related to sleep and feeding, reducing the tension and / or dysfunctional states already present, so continuing the prevention of further dysfunctions..

Specifically, working on the structural blocks and / or perceptual-motor dysfunctions of the oral district means making the child more responsive to therapy and reducing the discomfort he feels, especially in the moments of feeding and sleep, which represent two fundamental areas of physiological regulation. In fact, there are evidences of effective osteopathic interventions in subjects with malocclusion [39], facial [40] and postural asymmetries [41].

Another important correlation is that between auditory attention, typically impaired in children with ASD, and dysfunctions of the auditory system, another area in which there is evidence of the efficacy of osteopathic intervention [12]. Often, as occurs in the first years of life, children produce a lot of mucus and phlegm, which tend to stagnate in the ear canals due to drainage problems related to structural factors. In these cases, osteopathic techniques are highly effective in allowing better drainage, promoting better breathing and auditory processing, as well as greater well-being.

5. Conclusion

Plagiocephaly is a craniofacial dimorphism that does not resolve spontaneously, it evolves with bone growth. The results of this research lead to an important reflection in clinically re-evaluating plagiocephaly within a neonatal assessment of the very first months of life. This observational study highlights not only the high incidence of plagiocephaly even in children with ASD, but above all the degree of severity; the sample under examination does not fall within the age group in which it is possible to model the shape, and none of the children examined received osteopathic or other treatments to resolve plagiocephaly. The fact that the percentage of detected cases is comparable to that of the general population, underlines the need, as happens for children with typical development, to intervene early especially with children with ASD. In typically developing peers, there is numerous evidence of the benefits associated with osteopathic treatment in the presence of somatic dysfunctions; if we consider how much these are related to sensory, perceptual and motor alterations, it is clear that in

children with an already altered profile, these can amplify dysfunctional behaviors and states of discomfort related to autism. It therefore seems important to us to guarantee children with ASD a global care, which does not neglect these aspects within a multidimensional assessment and an integrated therapeutic / care project.

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