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Diagnostic algorithm of cranial deformities in children with severe neurological disorders

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Abstract

Background: Cranial deformities (plagiocephaly) generate various health problems in children, fact that may influence neurophysiological development both craniometrically and functionally. The aim of this study was to develop an algorithm for diagnosis of cranial deformities and dental alveolar appearance in children with delayed neurologic sequelae.

Material and methods: 370 children with neurological disorders were examined by a team (neurologist, orthodontist, oral and maxillofacial surgeon, plastic surgeon). It was recorded in the individual questionnaire: a) the degree of cranial deformity; b) the form of dentoalveolar anomaly c) type of brain dysfunction, which allowed the elucidation of correlations, previously unknown, depending on the nature and intensity of specific clinical and evolutionary manifestation.

Results: Cranial deformities in children were highlighted in 50% of cases. Relation boys:girls was 1:1. Positional cranial deformities in 25% of cases are associated with a different degree (the first degree – 1%, the second degree – 7.3%, the third degree – 50%, the fourth degree – 4.1%, the fifth degree – 1%), and 1.96% of craniostenosis and 21% of other deformities in children with severe cognitive and motor disorders. Dental and maxillar deformities in the sagittal plane were found in 28% of children with and in 25% without cranial deformities. Dental crowdings were found in 54% of children with and in 30% without cranial deformities. While the protrusion of the mandible and dental spaces were found in 8% and 21% in children with and 10% and 18% in children without cranial deformities.

Conclusions: In conclusion we found that cranial deformities in children with neurological disorders are present in 50% of cases. In the diagnosis of dental and maxillar deformities, it is necessary to determine when cranial deformities can lead to installation of malocclusions in children. In order to confirm the results of this study, it is necessary to make studies on anatomic disorders of cranial basis, cranial vault.

Key words: cranial deformities, children, neurological disorders.

Introduction

The United States Public Health Service has launched the campaign "Back to Sleep" to support the supine position for newborns during sleep. As a result of this campaign, the United States Craniofacial Anomalies Center has marked an increase in incidents of craniofacial skeletal deformities in infants [5, 8]. If in 1974 the presence of cranial and facial deformities was reported as 1newborn of 300 newborn children, so in 1996 the incidence increased to 1of 60 living newborn children [2, 5, 10]. Increased incidence of craniofacial deformities generated many aesthetic and functional problems in these children. Collett B. R. (2013), Hutchison B. L. (2012), Miller R. I. (2000) [3, 4, 5] found cognitive disorders, that were more highlighted in subsequent periods of neuropsychological development. 39.7% of children with plagiocephalies, examined during the preschool period, required special care, additional exercises, and individual curriculum. Collet B. R. (2013), Miller R. I. (2000) [3, 6] studying the scores of cognitive development of children with cranial and facial deformities, found a high risk of reduction of IQ coefficient during the school period, compared with healthy children.

The facts that the maxillar is directly joined to 11 and indirectly to the rest of cranial bones, and that skull anatomical changes could influence the development of the dentoalveolar system, are less reflected in the specialty literature [11, 13].

In literature we found no data available on the evolution of cranial deformities, forms of cranial deformities, about frequency of cranial deformities and their correlation with the presence of malocclusions in children of school age and children from various health groups. Thus, several authors found that orthodontic treatment is needed in children of school age from the group of healthy children [2, 14]. In this context it is known that the frequency of malocclusions simultaneously increases [1, 7, 9] in children with neurological disorders. Thus, in studies made on 124 and 381 children with neurological disorders [7] in South Africa, 58% to 74% of cases needed orthodontic treatment. On the other hand, scientific studies reflect only the correlation of behaviour disorders that could influence changes of dentoalveolar system. In this study we mapped out to examine the incidence of cranial deformities and their correlation with the presence of dentomaxillar deformities in school-age children with neurological disorders.

Material and methods

370 children with neurological disorders were examined in 3 residential institutions with special education. The children in the study had various forms of neurological diseases: mental retardation [F70 - F79] – 43 [11.6%] children; cerebral palsy and other paralytic syndromes [G80 - G83] – 45 [12.1%] children; episodic paroxysmal disorders [G40 - G47] – 52 [14.0%] children; sequelae of inflammatory diseases of the central nervous system [G09] – 65 [17.5%] patients; neurotic, stress and somatoform disorders [F40 - F48] – 69 [18.6%] children; congenital malformations, chromosomal deformities and anomalies [Q00 - Q99] – 36 [9.7%] children; other disorders of the nervous system [G90 - G99] – 60 [16.2%] children.

Examination of malocclusions in children was carried out after the simplest method, which determines the presence or absence of malocclusion. Also, certain information was specified, considering the difficulty of examinations of these children [12]. The simplest were examinations in the anterior segment, determining malocclusion in the sagittal plane, the presence of dental crowding and dental spaces. Data were recorded in questionnaires reflecting: 1 – occlusion (Angle classification); 2 – overjet; 3 – overbite; 4 – correlation dental alveolar in the anterior segment. Angle classification was used to determine interdental relations in the anterio-posterior position. This classification divides occlusions into first class, second class (two subdivisions) and third class, using first molars as a reference point. In the absence of molars canines were used. Examination standards were used for diagnosis of malocclusions. Normal occlusion: the term includes minimal deviations from ideal parts that do not generate aesthetic and functional changes.

Spaces. Segment considered „spaces" is determined when there is no proximal contact between the teeth.

Crowding – the segment that contains overlapping teeth, or lack of spaces in the dental arch for tooth eruption.

Skull shape of all children was examined by manual palpation methods and, in case of detection of cranial deformities, a bandage on deformed skull was applied to confirm the presence of deformities and to determine their form. Children with cranial deformities were divided into three categories: 1 – craniostenosis; 2 – plagiocephalies; 3 – other cranial deformities (microcephalies, hydrocephalies etc.).

The results were analyzed using "Epi-info-2002" and „Excel" from „Microsoft Office" package. The data were interpreted as M ± m (average error) by means of the t-Student test. All statistical methods were obtained from the "Statistics for Windows" version 6. The difference was regarded as conclusive when $p < 0.05$.

Thus, according to the criteria reflected in tabular list of diseases ICD-10-AM (10th Revision of the International Statistical Classification of Diseases and Related Health Problems) 7 groups of children with neurodental health problems were obtained.

Results

In this study the distribution of children by gender was equal [b:g = 1:1]. Table 1 elucidates the frequency of cranial deformities in relation to clinical manifestations of neurological pathology. Thus, in 1.9% of cases (7 children) were found deformities of the skull, specific to craniostenosis with an increase (0.8% of cases) in group of children with cerebral palsy [CP] and other paralytic syndromes. In other 44 children (11.9%) cranial deformities, identical to positional plagiocephalies, were found, especially in children with predominantly neurological disorders and in 10 children [2.7%], mainly with motor disorders, cranial deformities were not included in classifications of plagiocephalies and craniostenosis.

Thus, the distribution of children by cranial deformities, shape and degree of clinical manifestation of neurological pathology at first sight is not statistically authentic (P>0,05). Also, depending on the structure of neurological pathology it is found impressive that cranial deformities, on the one hand, in 48.9% of cases were associated in the late period more frequently with: a) cerebral palsy (CP) and other paralytic syndromes (15 4% of cases); b) congenital malformations (9.5% of cases); c) episodic and paroxysmal disorders (8.4% of cases); d) mental retardation (7.3%) and less frequently with: a) other diseases of the central nervous system (CNS) [4.3% cases]; b) neurotic, stress and somatoform disorders (2.1%) and c) the consequences of neuroinfections (1.9% cases).

On the other hand, cranial deformities in the late period, in children examined more frequently, prevailed in: a) episodic and paroxysmal disorders [68.9%]; b) CP and other paralytic syndromes (65.5%); c) other CNS disorders (48.5%); d) the consequences of neuroinfections (43.6%); e) congenital malformations (41.7%); f) mental retardation (35.1%) and g) neurotic, stress and somatoform disorders (28.6%) (table 1).

Table 1

Number of children with positional cranial deformities and their distribution by neurological disorders

Pathology	Group I without cranial deformities		Group II with cranial deformities		Forms of cranial deformity						Total	
					Craniostenosis		Plagiocephaly		Other cranial deformities			
	n	%	n	%	n	%	n	%	n	%	n	%
G80 - G83	30	8,1	57	15,4	3	0,8	44	11,9	10	2,7	87	23,5
Q00 - Q99	49	13,3	35	9,5	1	0,27	24	6,4	10	2,7	84	22,7
G40 - G47	14	3,8	31	8,4	2	0,6	19	5,1	10	2,7	45	12,2
F70 - F79	50	13,5	27	7,3	0	0.00	7	1,9	20	5,4	77	20,8
F40 - F48	20	5,4	8	2,1	1	0,27	6	1,6	1	0,27	28	7,6
G90 - G99	17	4,6	16	4,3	0	0.00	9	2,5	7	1,9	33	8,9
G09	9	2,4	7	1,9	0	0.00	4	1,1	3	0,8	16	4,3
Total	189	51,1	181	48,9	7	1,9	113	30,4	61	16,5	370	100

X^2=27,829; P>0,05

Table 2

Maxilla protrusion in relation to cranial deformities

Maxilla protrusion	Forms of cranial deformity										
	Without cranial deformity		With cranial deformity							TOTAL	
			Craniostenosis		Plagiocephaly		Other cranial deformity		Total		
	n	%	n	%	n	%	n	%	n	n	%
Present	46	25	3	37,5	30	31	18	23	51	97	26
Absent	139	75	5	62,5	68	69	61	77	134	273	73
Total	185	100	8	100	98	100	79	100	185	370	100

X^2=2,161; P>0,05

Table 3

Protrusion of the mandible in relation to the cranial deformities

Mandible protrusion	Forms of cranial deformity										
	Without cranial deformity		With other cranial deformity							TOTAL	
			Craniostenosis		Plagiocephaly		Other cranial deformity		Total		
	n	%	n	%	n	%	n	%	n	n	%
Absent	166	90	6	75	92	94	72	91	170	366	91
Present	19	10	2	25	6	6	7	9	15	34	9
Total	185	100	8	100	98	100	79	100	185	370	100

X^2=3,770; P>0,05

In order to emphasize clinical neurodental features, the results of examinations in the oral cavity in relation with or without cranial deformities and delayed neurologic sequelae are revealed. In tables two and three the presence of dental and maxilla deformities in the sagittal plane was identified. Thus, in 370 children with neurological disorders, 185 [50%] children did not have cranial deformities. So, in this group of children cranial deformities were found with a high frequency – 50%.

In this study, together with the detection of frequency of cranial deformities in children with severe neurological disorders, we have analyzed the clinical dentomaxillary condition of the anterior segment of the maxilla and mandible and their correlation with cranial deformities. Table 2 shows reflected data on the anterior displacement in the sagittal plane of the maxilla (maxilla protrusion). From 370 children maxilla protrusion was detected in 97 (26%). In the group of children without cranial deformities from 185 children in 46 [25%] cases maxilla protrusion was found and in 139 (75%) children was not found. At the same time, from 185 children with cranial deformities in 51 (28%) children the anterior displacement of the maxilla in the sagittal plane was found. We want to mention, that in the group of children with cranial deformities of craniostenosis and plagiocephaly type the anterior maxillar displacement in the sagittal plane considerably surpasses (31% to 38%). In the group of children with other cranial deformities, this pathology was found on a smaller percentage of children (23%) (table 2).

The relations of the maxilla to the mandible and cranial deformities were presented in table 3. From 370 children the anterior displacement of the mandible in the sagittal plane (mandible protrusion) was observed in a small number of children in comparison with the maxilla - 34 children (9%). At the same time, from 185 examined children with disorders the mandible protrusion was found in 10 (10%). In those with cranial deformities, from 185 children 15 (8.1%) were children with anterior displacement of the mandible in the sagittal plane, especially 6 children (3.2% cases) presented plagiocephalies, 2 (1.0%) craniostenosis and 7 (3.7%) cases of other deformities with anterior displacement of the mandible were found (table 3).

159 children (43% cases) in the study group were found with Angle class I, 60 children (16.2%) – Angle class II and 23 (9.2%) – Angle class III. We have to mention that in group with Angle class II 20 children (5.4%) with cranial deformities of plagiocephaly type, 3 cases (0.8%) of craniostenosis and 7 cases (1.9%) with other cranial deformities were found. A smaller number of children with Angle class III were found: 23 cases (6.2%), of which 5 children (1.4%) with plagiocephaly, 2 (0.5%) with craniostenosis and 2 (0.5%) with other deformities.

Outcomes of study, aimed to determine the dental bone relation in the anterior segment of maxilla and mandible of the examined children, are shown in Tables 4 and 5. The tables show that dental crowdings were observed in 123 cases (33%) of all examined children: 66 (54%) children from that group

Table 4

Distribution of children with cranial deformities and dental-bone relation (anterior segment)

Dental crowding	Forms of cranial deformity										
	Without cranial deformity		With other cranial deformity							TOTAL	
			Craniostenosis		Plagiocephaly		Other cranial deformity		Total		
	n	%	n	%	n	%	n	%	n	n	%
Absent	128	69	7	87,5	62	63	50	63	119	247	67
Present in maxilla	15	8	0	0	9	9	9	11	18	33	9
Present in mandible	20	11	1	12,5	14	14	11	14	25	46	12
Present in both	22	12	0	0	13	13	9	11	23	44	12
Total	185	100	8	100	98	100	79	100	185	370	100

X^2=4,327; P>0,05

were with cranial deformities and 57 children (30%) without cranial deformities. We noticed that dental crowdings are present more frequently in children with plagiocephaly than with other cranial deformities 36 (55%).

The presence of dental spaces in the anterior segment in children with cranial deformities has been detected less frequently in children with neurological disabilities compared with the presence of dental spaces – 74 (20%) cases. In the group of children with cranial deformities dental spaces were found in 18% of cases and in 21% of cases in children without cranial deformities. In Tables 4 and 5 we find that dental crowdings in children with cranial deformities are encountered more frequently in relation to dental spaces.

Discussion

The presence of cranial deformities has been addressed in literature on the specialty for more than 20 years, with the launch of the campaign "back to sleep", after a considerable increase in the number of children with cranial deformities was noticed [2, 8, 5]. In countries from the former Soviet Union they are less elucidated [15]. Together with this campaign many publications on the side effect of these pathologies in the development of preschool children have appeared.

Considering the high risk of harm to these children during their neuropsychiatric development, a great attention from parents, teachers, physicians [3, 4, 6, 10] is payed to this problem. In this study we tried to make a correlation between children with neurological disorders, cranial deformities and dentomaxillary status. We found the presence of cranial deformities in 50 % of children with neurological disorders. 30.4% of cases were positional plagiocephalies, 2% were craniostenosis and 16.5% were other cranial deformities, which more or less may affect neuropsychological development of a child.

Thus, every second child with severe neurological disorders has cranial deformities in a ratio of 1:2. In specialized literature data on the role of cranial deformities in the structure of neurological entities were not ilucidated. In this study it was observed that some neurological pathologies – CP and other paralytic syndromes (15.4% of cases); b) birth defects [9.5% of cases]; c) and episodic and paroxysmal disorders (8.4% of cases); d) mental retardation (7.3%) – present a higher incidence of cranial deformities.

The frequency of occlusal pathologies of neurological disorders among children has been studied by many authors [9, 1, 7]. Some authors have concluded that dentomaxillar deformities are more common in children with neurological pathologies compared to healthy children [1, 9]. They reasoned

Table 5

Distribution of children with cranial deformities and dental-bone relation (anterior segment)

Dental spaces	Forms of cranial deformity										
	Without cranial deformity		With cranial deformity							TOTAL	
			Craniostenosis		Plagiocephaly		Other cranial deformity		Total		
	n	%	n	%	n	%	n	%	n	n	%
Absent	145	78	8	100	83	85	60	76	151	296	80
Present in maxilla	16	9	0	0	6	6	13	16	19	35	10
Present in mandible	11	6	0	0	2	2	3	4	5	16	4
Prezent in both	13	7	0	0	7	7	3	4	10	23	6
Total	185	100	8	100	98	100	79	100	185	370	100

X^2=11,295; P>0,05

the high frequency of malocclusions with a type of disability, with the frequency of adjacent pathologies and behavioral vices. Onyeaso C.O., (2002) [7] comparing the frequency of malocclusions in children with neurological disorders and healthy children found no statistically authentic difference of occlusion disorders. At the same time, Birgit Theander and coauthors (2001) [12], Fabio Ciuffoli and coauthors in 2005 [14] doing a literature review, found that the incidence of abnormalities of occlusion in healthy children of school age ranges from 39% to 93%. Also these authors in their studies have found that only 11.9% of children examined had ideal occlusion criteria. The results of different authors vary, probably, because of the geographical location of the country where the study was made, its socio-economic condition, and examination methods used in each case [12,14]. In this study we found that in children with neurological disabilities Angle Class I was present in 21.4% and Angle Class II in 9.5% of cases. This study results showed a frequency of Angle index similar to Onyeaso C.O. and coauthors and lower than at other authors – Utomi I. L. 2009 [9]. It is to note in this context that data can be unclear because of examination difficulties of children with various forms of behavior. Also, in this study we found that the high number of children with deformities of jaws of Angle type I and II is more common in children with cranial deformities (21% of children diagnosed with plagiocephalies and 35% of children with craniostenosis). Changes in sagittal occlusion (protrusion of the maxilla and mandible) were made by several authors trying to make a priority in all dentoalveolar anomalies [7, 9, 12, 13, 14]. Onyeaso C.O. and coauthors [7] in their conclusions found a similar relation in the group of children with disorders and also found no differences in percentage in the group of healthy children and those with disorders. In this study we found out that the protrusion of the maxilla prevails (29%) compared with the mandible (9%). Also, protrusion of the maxilla was higher among children with cranial deformities (28%) compared to children without cranial deformities (25%). They were found most frequent in children with craniostenosis.

We also found big differences in results of authors on the presence of dental crowding and spaces in children. Some authors found the prevalence of dental crowding [14,12] while others [9,7] of dental spaces. Onyeaso C. O. (2002) [7] is the author who considers there is no percentage differences of these abnormalities in the group of healthy children and those with neurological disorders. We found that of 370 children examined, 33% had dental crowdings and 20% - dental spaces. In 185 children with deformities 36% were found with dental crowdings and 18% with dental spaces, compared with those who did not have cranial deformities 31% and 22%.

Conclusions

In conclusion we found that cranial deformities in children with neurological disorders are present in 50% of cases. A prevalence of abnormalities of occlusion was found in children with cranial deformities. When determining malocclusions, their prophylaxis and treatment, it is necessary to take into account the changes that occur in the cranial vault and the base. In order to confirm these findings, it is necessary to clarify when cranial deformities may influence the development of the dentomaxillary system.

References

1. Oliveira Ana Cristina, Paiva Saul Martins, Torres Martins Milene, et al. Prevalence and determinant factors of malocclusion in children with special needs. Eur J Orthod. 2011;33(4):413-418.
2. Argenda LC, David LR, Wilson JA, Bel WO. An increase in infant cranial deformity with supine sleeping position. J. Craniofacial Surg. 1996;7:5-11.
3. Collett BR, Gray KE, Starr JR, et al. Development at age 36 months in children with deformational plagiocephaly. Pediatrics. 2013;131(1):e109-15. doi: 10.1542/peds.2012-1779. Epub 2012 Dec 24.
4. Hutchison BL, Stewart AW, de Chalain T, Mitchell EA. Serial developmental assessments in infants with deformational plagiocephaly. J Paediatr Child Health. 2012;48(3):274-8. doi: 10.1111/j.1440-1754.2011.02234.x. Epub 2011 Nov 14.
5. Littlefield TR, Saba NM, Kelly KM. On the current incidence of deformational plagiocephaly: an estimation based on prospective registration at a single center. Semin Pediatr Neurol. 2004;11(4):301.
6. Miller RI, Clarren SK. Long-term developmental outcomes in patients with deformational plagiocephaly. Pediatrics. 2000;105(2):E26.
7. Onyeaso CO. Malocclusion pattern among handicapped children in Ibadan, Nigeria. Nigeria J. of Clincal practice. 2002;5(1):57-60.
8. Turk AE1, McCarthy JG, Thorne CH, Wisoff JH. The „back to sleep campaign" and deformational plagiocephaly: is there cause for concern? J Craniofac Surg. 1996;7(1):12-8.
9. Utomi IL, Onyeaso CO, Chukwudi Ochi. Malocclusion and Orthodontic Treatment Need of Mentally Handicapped Children in Lagos, Nigeria Pesquisa Brasileira em. Odontopediatria e Clinica Integrada. 2009;9(1):7.
10. Van Vlimmeren, van der Graaf Y, Boere–Boonekamp MM, et al. Risk factors for deformational plagiocephaly at birth and at 7 weeks of age. A prospective cohort study. Pediatrics. 2008;119:e408-e418.
11. Captier G, Leboucq ĆN, Bigorre ĆM, et al. Plagiocephaly: morphometry of skull base asymmetry. Surg Radiol Anat. 2003;25:226-233. DOI:10.1007/s00276-003-0118-x.
12. Thilander Brigit, Pena Lucia, Infante Clementina, et al. Prevalence of malocclusion and orthodontic treatment need in children and adolescents in Bogota, Colombia. An epidemiological study related to different stages of dental development. Europ J. of Orthodontocs. 2001;23(2):153-167.
13. Silkman Raymond DDS. Is it mental or is it dental? http.//www.westonaprice.org/holistic-healthcare. March 30 20061/13-13/13.
14. Ciuffolo Fabio, Manzoli Lamberto, D"Attilio Michele, et al. Prevalence and distribution by gender of occlusal characteristics in a sample of Italian secondary school students: a cross-sectional study. European J. of Orthodontics. 2001;23(2):153-167.
15. Belichenko VA. Cherepno-litsevaya khirurgia [Cranio-facial surgery]. 2006;338.