

REVIEW

by assoc. prof. dr. Dobrinka Damyanova, DMD, PhD

on dissertation work for the award of Educational and Scientific Degree „Doctor”

in the field of higher education 7. Health care and sports, Professional direction 7.2.

Dentistry

Dissertation topic:

„ ORAL MANIFESTATIONS IN CHILDREN WITH AUTISM AND SOME SYNDROMES ”

Author of the dissertation: dr. Siyana Atanasova

Academic advisor: prof. dr. Radosveta Stoyanova Andreeva-Borisova, DMD, PhD, DSc

Biographical data for the candidate:

In 2018, she graduated from the MU "Prof. Dr. P. Stoyanov" in the city of Varna as a Master of Dental Medicine. She also has a specialty from 2022 in Pediatric dentistry. Since 2018, she has been an assistant at the MU "Prof. Dr. P. Stoyanov" in the city of Varna, at the Department of "Pediatric Dentistry". Since 2020, Dr. Siyana Atanasova is a PhD student in the Department of "Pediatric Dentistry" at the Faculty of Dental Medicine of the Medical University "Prof. Dr. P. Stoyanov" in the city of Varna.

General characteristics of the dissertation work. Relevance of the scientific problem

A new report, dated 18/05/2022, published by WHO and UNICEF, reveals that more than 2.5 billion people need one or more aids. Nearly one billion people with specific needs and disabilities do not have access to them, especially in low- and middle-income countries, where only 3% of those in need have access to these life-changing products. Nearly 240 million are the number of children with disabilities in the world. The number of people who need one or more means is likely to rise to 3.5 billion by 2050, due to an aging population and the prevalence of some non-communicable diseases increasing worldwide. The estimated number of children with disabilities and developmental difficulties is about 32,000, but there are no complete data on their exact number in the country. Their dental treatment and prevention are very difficult due to their

physical, mental or social disability and inequality. Children with special needs are at high risk of developing oral diseases as a result of: longer feeding, special diets, medication intake, use of carbohydrate food as a reward, oral-motor or general-motor dysfunctions leading to poor oral hygiene, inadequate intake of systemic fluorides, excessive parental or guardian involvement with the child's disability, parental or institutional neglect, neglect of oral health. The study of children with specific health needs and syndromes, their timely and early prevention and treatment algorithms are important and valuable for our clinical practice and the daily activities of pediatric dentistry specialists.

Structure of the dissertation

The dissertation has a volume of 178 pages. Contains an introduction; literature review, purpose, tasks and hypotheses; materials and methods; results and discussion; contributions; clinical perspectives; conclusions; bibliography, 6 detailed appendices. Includes 26 color graphs, 15 figures and 37 tables. The bibliography covers 399 literary sources, of which 366 are in Latin, 33 in Cyrillic.

Content of the dissertation

In the introduction the emphasis is placed on the topicality of the problem and the reasons for creating this dissertation work.

The Literary Review provides a historical and theoretical analysis of children with specific health needs, definitions and the epidemiology of the problem are given.

The first paragraph clarifies the essence of the concepts and diseases: Autism and Asperger's syndrome, Down's syndrome, Silver-Russell syndrome and emphasizes modern concepts, definitions and scientific analyses. In the second paragraph, clinical characteristics of children with disorders of the autism spectrum (autism) are introduced with a description of 3 areas: Area I – Impaired communicative abilities (verbal and non-verbal); Area II – Impaired social skills, limited or absent initiative and interaction; Area III – Limited range of interests, single type games. Autistic disorders are described - Social interactions; Communications; Behavioral relations.

An in-depth review of the pathology of dental caries and periodontal diseases in children with autism spectrum disorders was made. Most of them have a high risk of developing dental caries due to the diagnosis of more than one active caries lesions. 97% of the examined children with autism had gingivitis. According to the review analysis, children with autism reveal a higher

proportion of oral soft tissue lesions, and according to the conducted scientific studies, most of the children with autism have malocclusion, with the most frequently observed class I - being 65%. The author also analyzes research related to *Oral hygiene and dental treatment in children with autism*.

The third paragraph focuses on the description of **Children with Down syndrome and Silver-Russell syndrome**, the clinical characteristics of children with a genetic abnormality (Down syndrome and Silver-Russell syndrome). A literary description of the general medical symptoms and indicators with the general physical and psychological development of these patients was made, as well as the oral and dental and maxillofacial pathology with: Dental, hygienic, periodontal and orthodontic status of children with autism, Down syndrome and Silver-Russell syndrome and dental abnormalities in individuals with Down syndrome are quite high with mandibular retrognathia and relative macrocephaly for children with Silver-Russell syndrome. The review also mentions the modern treatment of children with Silver-Russell syndrome with growth hormone (GH). Increasing the duration of GH therapy by 10 months in the data resulted in a mean increase in DMFT index of 0.70.

The fourth paragraph presents a detailed analysis of corrective preventive approaches and teaching strategies, examining the **Oral Health Prevention database in children with autism and rare syndromes**. The PhD student describes successful behavioral techniques of the dental team to build trust in children with autism: **Encouraging communication, Applying the "Say-Show-Do" method, Limitations in touch due to high sensitivity in autistic children, Distraction, Sensory techniques, Using behavioral theory. Mutual consultation** with occupational therapists provides guidance to dental professionals on necessary changes in the dental environment in terms of limiting the harmful influence of certain visual stimuli for children with autism, Down syndrome and other rare syndromes.

In the third chapter, the model of scientific research is presented. The purpose, the tasks (the subject, the object, of the research), the hypotheses of the research are derived. They are presented in a logical sequence, clearly and specifically.

The 4 tasks set are formulated in a causal relationship with the goal, the subject and the object, as well as with the theoretical analysis of the topic in the previous two chapters. Three

hypotheses were put forward that are related to **DMFT index, periodontal status, orthodontic status of children with autism and some syndromes compared to healthy controls.**

The participants in the study are 240 children aged 3-18 years from the Varna region, who were admitted to the University Medical and Dental Center of FDM-Varna for primary examination, control examination, prophylactic examination or treatment for a period of 2 years. The patients were divided into four groups as follows: 60 children with autism spectrum disorders (19 girls and 41 boys), 60 children with Down syndrome (33 girls and 27 boys), 60 children with Silver-Russell syndrome (31 girls and 29 boys), 60 healthy children (control) (36 girls and 24 boys).

The methods and methodologies used are presented strictly and in accordance with scientific and research principles.

Statistical processing - The data were subjected to statistical processing with statistical program SPSS v.20.0. for Windows, including various statistical methods: Analysis of Variance (ANOVA) - for comparison of quantifiable normally distributed indicators in more than two groups; Variation analysis - when describing quantitative indicators - arithmetic mean \pm standard deviation (mean \pm SD); Correlation analysis – to study the relationship between observed phenomena. Pearson's coefficient and Spearman's coefficient were used; Comparative analysis (hypothesis evaluation) - χ^2 , F and t-test. An acceptable level of significance of the null hypothesis $p < 0.05$ was accepted. There is a graphical and tabular method of displaying the results.

Ethical considerations: The research protocols of the dissertation work have been approved by the commission for scientific research - KENI, at the Varna MU.

The fourth chapter demonstrates the analysis of the results and their discussion of the conducted scientific research in the dissertation work. According to the first task: according to the indicator of frequency of tooth brushing, no significant difference was found between the individual studied groups, children from all groups in the largest percentage brush their teeth every day, with a slight advantage in the healthy controls ($\chi^2 = 7.853$, $p = 0.249$). According to the indicator frequency of tooth brushing, there is statistical significance, a moderate correlation has been proven between the groups. No significant difference was observed between the four studied groups for the indicator of duration of tooth brushing. The basis for this is given by the characteristic ($\chi^2 = 1.244$, $p = 0.742$). Regarding the indicator of duration of tooth brushing, it was found that a total

of 180 children (75.0%) brush their teeth for 1-2 minutes, while the remaining 60 (25.0%) - more than 2 minutes, and only 8 children - 2 of each group uses complex prophylaxis. The facts are alarming that out of a total of 240 children, 116 with a relative share of 48.3% have the parent brush the child's teeth and children with autism and Down syndrome have the most carbohydrate meals during the day, with more than one intermediate breakfast per day between the main meals. Healthy children visit a dentist twice a year in a higher percentage compared to the other patient groups studied by the doctoral student. An increase in the plaque index indicates a deterioration of oral hygiene with age in children with autism, of the children with Down syndrome studied, there is the highest prevalence of lesions d3/D3 and d4/D4 in the children of the group aged less than 6 years.

According to task 2, the ANOVA test shows that for the GI Loe-Silness indicator, the differences between the mean values for the three age groups are statistically significant. The PSR index for the four groups of patients in the study/children with autism, Silver-Russell syndrome and healthy children/, obtained values mostly in code 0,1 and 2, which is characterized by the absence or presence of gingival bleeding and dental plaque. For children with Down syndrome, the presence of plaque, bleeding and loss of attachment, with a probing depth of more than 4 mm, or the clinic of periodontal disease is established. For the groups of children with autism, the doctoral student measured a value of 3.47 ± 0.80 , for children with Silver-Russell syndrome – 2.89 ± 0.63 and for healthy children – 2.96 ± 0.59 .

According to task 3, orthodontic analyzes of the occlusal relationships observed, normal occlusion or Angle Class I were observed in 110 of the examined children - 32 (29.1%) of them were children with autism, 31 (28.2%) were with Silver-Russell syndrome, 11 (10%) with Down syndrome and 36 (32.7%) of the healthy controls. Angle Class II (distal occlusion) was obtained in the analysis of 103 studied patients. The largest share of them were children with Silver-Russell syndrome - 29 (28.2), in second place were children with Down syndrome - 26 (25.2), children with autism and healthy controls - 24 (23.3%). Of the studied groups of children on the subject, 70 children had hypodontia, 45 of them had Down's syndrome (64.3%), 11 had the diagnosis of Silver-Russell syndrome (15.7%), 6 had autism (8.6%) and 8 were from healthy controls (11.4%). 20 (44.44%) boys and 25 (55.56%) girls were diagnosed with hypodontia with Down's syndrome. According to Petrunov's classification, the calculations show that for children with autism 28 children (46.7%) had a mild orthodontic deformity, moderately severe orthodontic deformity was

observed in 15 of the patients (25%), and severe deformity with hypodontia of single teeth are observed in 6 of the examined (10%).

32 children with Silver-Russell syndrome (53.3%) have a slight deformity: a deviation in the position of the teeth is present in 23 children (38.3%); a distal bite of $\frac{1}{3}$ to $\frac{1}{2}$ was observed in 13 children (21.67%); 18 children (30%) have a crossbite in the lateral area. Moderately severe deformity was present in 10 children (16.67%): a distal bite of $\frac{1}{2}$ to 1 measurement unit was observed in 8 children (13.3%); 6 children (10%) had a unilateral crossbite in the lateral region of more than 2 pairs of antagonists. Severe deformity was demonstrated in 11 children (18.3%) with hypodontia of single teeth, 3 children (5%) had distal occlusion above 1 unit of measurement in a mixed dentition, and 7 children had deep occlusion with crown coverage of the lower incisor (11.67%).

In the fourth task, the results of the conducted survey are presented, as the doctoral student examines the awareness of dentists and parents of children with autism and some syndromes. Updated protocols and motivational materials for the prevention and treatment of children's oral diseases are being prepared. The following conclusions were drawn: Dentists do not feel confident in treating these patients; A large percentage of physicians desire additional information related to the treatment of these patients. Collaboration between treating doctors and dentists is needed in order to better conduct effective oral prophylaxis and treatment for children with autism and some syndromes.

The dissertation presents **general conclusions and valuable recommendations for work in our clinical practice.**

The abstract accurately and correctly reflects the content of the dissertation work.

The author presents 4 publications on the scientific topic of the dissertation.

The following **contributions** are made to the theory and practice of research and work with representatives of childhood patients with autism, Down syndrome, Silver-Russell syndrome and some other syndromes:

Original contributions:

1. For the first time in our country, orthodontic status and dental anomalies in children with autism and some syndromes are examined.

2. For the first time in our country, the oral health (intensity of dental caries, the level of oral hygiene, gingival inflammation and periodontal status) was examined in children with Silver-Russell syndrome.

The dissertation also contains Confirmatory contributions of a practical-applied nature and confirmatory contributions.

I accept the contributions and believe that they are reasonably refined based on the dissertation development.

Dr. Siyana Atanasova has presented 4 publications on the topic of the dissertation, 2 of which in specialized periodicals and 2 in representative collections.


The proposed dissertation is the first of its kind in the field of pediatric dentistry, and is a valuable clinical resource for future research, for the work of professionals in social care homes, and for parents of children with syndromes and specific health needs.

The dissertation development is distinguished by a precise interpretation of complex dental problems, with an easy-to-read style, with comprehensively presented evidence from the development and conclusions of 4 tasks for the author's thesis. The results of the dissertation can be used both in future research and in the application of work methods in dental clinics with children with Down syndrome.

In conclusion, I give my positive assessment regarding the dissertation work, the overall research and the clinical teaching activities of the doctoral student. The dissertation meets all the minimum scientometric requirements of Varna University of Medical Sciences according to Varna Medical University's Regulations for the development of the academic staff, meets Law for the Development of the Academic Staff and Regulations for the implementation of the Law on the development of the academic staff in the Republic of Bulgaria. I propose to the Honorable Scientific Jury to vote positively for the educational and scientific degree of Educational and Scientific Degree "PhD" of Dr. Siyana Atanasova in the field of higher education 7. Health care and sports, Professional direction 7.2. Pediatric Dentistry.

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Varna

Author of the review: 

/Assoc. prof. dr. Dobrinka Damyanova, DMD, PhD/