

Impact of congenital heart disease on motor development in children with down syndrome – systematic review

Abstract

Motor development shows us the evolution of children and the integrity of their motor, reflex and voluntary activity. Down syndrome is a genetic alteration where trisomy 21 occurs, with an incidence of 1 in every 700 live births. It is worth noting that 40% to 50% of children with Down syndrome have congenital heart disease, which affects their motor development mainly due to frequent hospitalizations and early surgeries. Objective: To describe the impact on neuropsychomotor development of children with Down syndrome who have congenital heart disease. Methodology: This is a literature review through books and scientific articles using the

Keywords: down syndrome, congenital heart disease, motor development, physical therapy

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Priscila Lanzillotta, Vitoria De Lima Balula

Physiotherapy student at Centro Universitário Lusiada, Brazil

Correspondence: Priscila Lanzillotta, Physiotherapy student at Centro Universitário Lusiada (UNILUS), Praça palmares, n 8, apt 72 - Bairro Macuco, CEP 11015-330, Santos-SP, Brazil, Email prilanzi@hotmail.com

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Introduction

Motor development is due to characteristics that mark the evolution process of babies, showing us the integrity of their motor, reflex and involuntary activity, evolving more and more towards more coordinated and fine movements. Through motor development and its milestones, it is possible to analyze the integration and maturation of the child's nervous system through activities such as playing, rolling, hand and foot activity, walking, among others.¹ At birth, the central nervous system of a child, without alterations, can perceive the world through the senses and in this way, external stimuli can alter the CNS, causing the child to evolve and learn in the environment in which he lives. In a child with Down Syndrome, this evolution and learning take place through biology, behavior, and the environment in which they live, and not exclusively through the maturation of the central nervous system.² Down Syndrome has as its main characteristics muscle hypotonia, hypermobility, more fragile joints, motor alterations, first cervical vertebrae with great instability and mobility, tendency to obesity due to hyperthyroidism, tendency to heart disease due to physiological and structural alterations, among others. Therefore, these specificities end up negatively affecting and hindering the locomotion and stabilization activities of these children.³ Skills such as manipulating objects, reaching and learning are essential for development, and they are started at 4 months of age. By age 2 the typical child demonstrates forward planning, coordination of both hands, and apprehension patterns in fine motor activities. The intrinsic alterations of Down syndrome, such as muscle hypotonia and joint hypermobility, mainly affect these factors and contribute to the child making slower movements and having changes in postural control. Soon there is a decrease in the exploration of the environment and motor experiences, mainly affecting the abilities of the fine motor system.⁴

We call congenital heart disease the condition of children who have abnormalities in the cardiocirculatory structure and function since birth, there are about 10 out of every 1000 live births who manifest congenital heart disease (CHD) and of these 1/3 need surgical intervention.⁵ Its classification is given by the bluish or non-

bluish color of the skin caused by poor oxygenation or by blood flow or decreased, that is, if it is cyanogenic or acyanogenic, it is also classified by obstruction outside the heart or mixed flow. The most common symptoms are cyanosis, tiredness, dyspnea, low weight, tachycardia and repeated respiratory infections. These symptoms end up leading the child to be hospitalized frequently, to use medication, to have various physical and general restrictions, to retake exams, to be absent from school, developmental delay and decreased quality of life.⁵ It is worth noting that through the experiences lived by the child and the maturation of the nervous system, neuropsychomotor development develops through motor, cognitive and language learning. Therefore, children under the conditions of congenital heart disease and its factors, end up having delayed motor development.⁵ It should be noted that 40% to 50% of children with Down syndrome have congenital heart disease, so these children have been undergoing hospitalizations and surgeries earlier and earlier, which ends up affecting their motor development.⁶

Goal

To describe the impact on the neuropsychomotor development of children with Down syndrome who have congenital heart diseases, as well as to highlight the importance of physical therapy acting on the motor development of children with Down syndrome and congenital heart disease, showing the benefits of physical therapy associated with treatment.

Methodology

A systematic review was produced, with a survey of bibliographic data on the following platforms: SciELO, PubMed and Virtual Health Library (VHL) using the following descriptors in Portuguese/English: *Motor development, Down's syndrome, Congenital heart disease*. The inclusion criteria established were: scientific articles published between January 2011 and January 2023, in Portuguese and English (Figure 1). The following exclusion criteria were established: titles that do not match the theme, repeated titles, incomplete texts, data that do not fit the objective of the work, paid works (Table 1).

Table 1 A total of 27 articles were found and after filtering by the exclusion criteria, 5 studies were selected in the search.

Author/year	Title	Goals	Methodology	Result	Conclusion
Visoosak, Mahle, Kirshbom, Huddleston, Caron-Besch, Ransom, and Sherma ⁷	Neurodevelopmental outcome in children with Down syndrome and congenital heart defects	The objective of this study was to compare the developmental domains (cognitive, language, and motor) of children with DS and congenital heart disease with children with DS without heart defects.	A total of 29 children were separated into groups, 12 with CHD and 17 without heart disease. Evaluation and comparison between the groups: - Bayley Tests of Child and Child Development III (Bayley III).	The only domain that showed a statistically significant difference between the groups was the motor score ($p < 0.05$), but in all scores, such as cognitive pattern and language scores, they were lower in the cases of children with CHD compared to controls without WC.	Children with CHD had greater developmental deficits in the motor domain compared to children with DS without heart disease.
Hoepers and Schivinsk ⁸	Heart disease and motor development in Down syndrome: case series	To show the assessment of motor development of children with DS, with and without CHD, and to relate the results to motor development milestones.	Participants were 8 children, with a mean age of 13.72 months, 5 without WC and 3 with heart disease. Evaluation: - Brunet and Lézine scale. - The parents answered a questionnaire regarding socioeconomic data and pre-, peri- and post-natal conditions.	Children without WC were rated much lower in almost all areas of the scale, with the exception of 2 cases, who performed slightly better in the postural, social, oculomotor and global areas. In the group of heart patients, one of the cases did not present any result below normal and one child demonstrated a level above very low only in the social item.	The study showed that children with DS and associated WC had motor development very similar to that of children without associated heart disease, but the parents' education as well as regular financial condition and housing conditions may have influenced the result.
Alsaied, Marino, Esbensen, Anixt, Epstein and Cnota ³	Does congenital heart disease affect neurodevelopmental outcomes in children with Down syndrome?	The objective was to evaluate the relationship between CHD that required surgery in the first year of life and the results of neurodevelopment, behavior, and emotional issues in children with DS.	A review of 1,902 medical records of children (0-18 years) with DS who visited a single institution from 08/2008 to 08/2013 was conducted. Children who underwent at least one of the nine neurodevelopmental tests were divided and evaluated by: Infants/Toddlers - Children's and children's Bayley scale III. Preschoolers - Peabody Developmental Motor Scales (PDMS-2) -adolescents The test scores of children with DS who underwent cardiac surgery in the first year of life were compared with children with DS without CHD.	Infants/toddlers with cardiac surgery had lower scores on language only compared to those without CC, the gross and fine motor development scores were not statistically significant. Preschool children with cardiac surgery had lower language scores and motor scores, but not statistically significant.	Children with Down syndrome undergoing cardiac surgery during the first year did demonstrate worse neurodevelopmental outcomes as infants/children, in contrast they had no differences in development at school age compared to children with Down syndrome without CHD.

Table 1 Continued...

Author/year	Title	Goals	Methodology	Result	Conclusion
Gonzaga, André, Oliveira, Damasceno, Guerrero, Carletti, Alves, Carvalho and Bofi ⁶	The influence of congenital heart malformations in children with chromosome 21 syndrome	The objective was to compare the motor development of infants with DS from 0 to 4 months of age, who had or did not have WC.	Participants were 26 children with DS, and 15 had WC. Evaluation: - Operationalized Portage Inventory (IPO) – Infant stimulation. - Anamnesis with parents	All children had delayed neuropsychomotor development corresponding to their chronological age, of those with heart disease, 53.33% had significant delays (below what was expected for their age) and of those who did not have heart disease, 75% had significant delays.	Congenital heart disease was not a determinant for the delay in neuropsychomotor development. However, negative developmental outcomes depend not only on one, but on a combination of several risk factors. Therefore, it is of paramount importance to ascertain the nature of these risk factors since they are directly affected by the development.
Amaral, Corrêa and Aita. ⁹	Profile of independence in self-care of children with Down Syndrome and congenital heart disease	To outline the profile of independence in self-care activities of SD children with WC in the age group of 3 to 7 years who receive outpatient follow-up at a cardiology referral unit in the city of Belém, Pará State, Brazil.	A study was conducted with 18 children being followed up at the cardiology reference outpatient clinic in the state of Pará. For data collection, an interview with the parents was carried out with personal information and part I of the Pediatric Disability Assessment Inventory (PEDI) regarding the child's abilities.	The study showed that children with DS and WC had significant impairment in the area of fine motor skills, which interfered with the performance of activities such as handling objects, opening and closing faucets, and using utensils, among others.	Both DS and WC are conditions that interfere with the development of skills necessary for independence in activities. Thus, the study demonstrated that the functional performance in the activities of the children studied is inferior to that of children with typical development.

SD, down syndrome; CC, congenital heart disease; CHD, congenital heart disease



Figure 1 Scientific articles published between January 2011 and January 2023.

Discussion

The objective of the present study was to research and report the impacts caused by congenital heart disease on the motor development of children with Down Syndrome and as well as to compare the motor development of children with trisomy 21 without heart disease. We know that cardiac dysfunctions can generate frequent hospitalizations for a prolonged time, dyspnea at the slightest exertion (which can generate hypoactivity), immobility and as a consequence even

limitations in functional motor activities. In this way, it is easier to trigger a significant delay in motor development than in the cases of children with Down Syndrome, which is already more pronounced.

In the study by Hoepers et al.,⁸ it was mentioned that heart disease in children with Down Syndrome causes restrictions in motor activities, immobility and also greater hypoactivity. In this study, all children had delayed motor development, however, it was not more evident in children with congenital heart disease. Similar to this study, Gonzaga et al. compared the motor development of children with DS with heart disease and non-heart disease, finally 60% of children with heart disease had delay and 88% of non-heart disease children had delay, thus showing that children with DS who were not cardiopathic had a greater delay in development when compared to those with heart disease, considering that congenital heart disease was not a determining factor for developmental delay. As Alsaied et al.¹⁰ reported and compared motor development of infants and adolescents with DS and heart disease, it was shown that, although not statistically significant, the infants had delayed gross and fine motor development and statistically significant language scores. On the other hand, surprisingly, adolescents with DS and heart disease did not show differences in comparison with the motor development of children with DS without heart disease in any of the tests that were applied.

Visootsak et al.¹¹ and Amaral et al.¹ also reported changes in gross and fine motor development scores. Visootsak et al.¹¹ conducted a study

with children with DS with and without heart disease and showed that there was a greater delay in the development of gross motor skills in the DS group with CHD when compared to the group without heart disease. Heart diseases can result in greater fatigue and low endurance, so this delay can be explained by several possible factors, both perioperative and postoperative, the main factor being tone asymmetry, resulting in gross motor deficits. With this, the importance of physical therapy after birth is highlighted, with an emphasis on adjusting tone, improving muscle strength and coordination. Similarly, Amaral et al.¹ conducted the same study, but the children in this study had a significant delay in the development of fine motor skills, interfering with skills such as handling objects, opening and closing taps or doors, using eating utensils, among others. The prevalence of congenital heart disease in patients with trisomy 21 is 40% to 50%, but in recent years with early diagnosis and more effective surgical treatments, there have been significant improvements in the life expectancy of these patients. However, we cannot discard the consequences still caused by congenital heart disease or its postoperative period, it is extremely important that there is a multidisciplinary work focused on the motor development of these children. Despite the limitations of the low number of studies published regarding the motor development of children with DS+CHD, this research increases the understanding of the effect of congenital heart disease on children with Down syndrome, also showing that there is still a relativity when comparing the motor development of children with DS without and with CHD.¹² Some children may present alterations mainly in gross and fine motor skills, on the other hand, other children may present the same motor development as a child with DS without WC.

Conclusion

Children with Down syndrome and congenital heart disease may present alterations in motor development, especially in gross and fine motor coordination skills, however, the risk factor of congenital heart disease alone is not a determinant for long-term motor development delay in children with trisomy 21. In short, it is important to highlight that there are still open gaps in the subject under discussion, there is a need for deeper and more comprehensive future research in order to have a more concrete understanding of the topic addressed.

Acknowledgments

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Conflicts of interest

The authors have no conflicts of interest to declare.

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