Investigating the Motor Function of School-Age Children Born with Congenital Diaphragmatic Hernia: The Need for Long-Term Efforts to Improve Motor Function.

#### **Key Points**

•This study, investigating the motor function of children aged 6 to 10 years born with congenital diaphragmatic hernia (CDH), revealed a decline in limb muscle strength, balance, and endurance.

•An examination of physical activity duration showed that, despite no specific restrictions from physicians, these children engaged in less physical activity.

•The study also explored the relationship between the severity of CDH and motor function, finding that a lower value of the prenatal severity indicator "o/e LHR" (observed-to-expected lung area-to-head circumference ratio) was associated with lower grip strength in school-age children.

•Ensuring adequate physical activity and providing follow-up and exercise programs focused on improving limb muscle strength, balance, and endurance are expected to enhance motor function.

#### Summary

The research group, including Takamasa Mitsumatsu (first author), Dr. Yuji Ito (corresponding author), Dr. Hiroyuki Kidokoro, and Prof. Yoshiyuki Takahashi of the Department of Pediatrics, Nagoya University Graduate School of Medicine (Dean: Hiroshi Kimura, MD, PhD), and Dr. Yoshiaki Sato and Dr. Tomomi Kotani of the Center for Maternal-Neonatal Care, Nagoya University Hospital, and Dr Tadashi Ito and Dr. Sho Narahara of Aichi Prefectural Mikawa Aoitori Medical and Rehabilitation Center for Developmental Disabilities, investigated the motor function of school-age children born with congenital diaphragmatic hernia (CDH). The study revealed a decline in limb muscle strength, balance, and endurance. Additionally, a survey on physical activity time using a questionnaire showed that, despite no specific restrictions from physicians, the children had lower physical activity time. The group also examined the correlation between the severity of CDH and motor function, finding that a low o/e LHR (observed-to-expected lung area-to-head circumference ratio), an indicator of prenatal severity, was associated with reduced grip strength in childhood.

CDH is a rare congenital anomaly characterized by a defect in the diaphragm,

resulting in herniation of abdominal organs into the thoracic cavity. Recent advances in perinatal care have improved survival rates, but the incidence of long-term complications has increased, and the decline in quality of life related to exercise and walking has become a concern.

Ensuring adequate physical activity and providing follow-up and exercise programs focused on improving limb muscle strength, balance, and endurance are expected to enhance motor function in children born with CDH.

#### Research Background

Congenital diaphragmatic hernia (CDH) is a rare congenital anomaly characterized by a defect in the diaphragm, resulting in herniation of abdominal organs into the thoracic cavity. Recent advances in perinatal care have improved survival rates; however, the incidence of long-term complications has increased. Children born with CDH are at higher risk for neurodevelopmental delays, and the decline in quality of life related to exercise and walking has become a concern. However, few studies have conducted comprehensive assessments of limb muscle strength, balance, endurance, and gait ability, or examined the association between these motor functions and perinatal factors in school-age children born with CDH.

Therefore, we performed motor and functional tests and gait analyses to elucidate comprehensive motor and functional characteristics in school-age children born with CDH to identify motor functions requiring special attention during follow-up and to develop appropriate intervention programs. Additionally, we sought to identify perinatal factors associated with motor function.

### **Research Results**

We assessed motor function in 24 children aged 6 to 10 years who were born with CDH and compared them with 72 age- and sex-matched healthy children who participated in the Okazaki Child Medical Checkup for the Physical Function of Elementary School Students. The assessments included motor and functional tests (grip strength test, five times sit-to-stand test, one-leg standing time, and six-minute walking distance test) and three-dimensional gait analysis (Figure 1). (Figure 1) Motor and functional tests and three-dimensional gait analysis

## Motor and functional tests



### Three-dimensional gait analysis

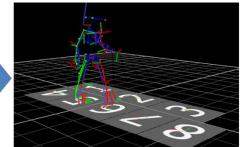


①Markers are attached to lower limbs and pelvis.



28 optical cameras capture motion during walking.





3 Data are analyzed using computers.

The results showed that children born with CDH had significantly worse grip strength, longer five times sit-to-stand test time, shorter one-leg standing time, and shorter 6 min walking distance compared to healthy children. These findings indicate impairments in limb muscle strength, balance, and endurance. The three-dimensional gait analysis revealed no significant differences in gait parameters, including walking speed, step length, and the Gait Deviation Index. Additionally, we used a questionnaire to investigate physical activity time (moderate-to-vigorous physical activity time per week). The results showed that, despite no specific exercise restrictions from physicians, children born with CDH had less physical activity time than healthy children (Figure 2).

	Children born with CDH (n = 24)	Healthy children (n = 72)	p value
Grip strength (kg)	8.8 ± 2.7	11.0 ± 3.3	0.004
Five times-sit-to-stand test (s)	7.4 (3.6-11.1)	6.0 (3.8-9.9)	0.014
One-leg standing time (s)	21.0 (2.7-120.0)	109.8 (9.3-120.0)	<0.001
6 min walking distance (m)	444.1 (317.1-595.4)	510.0 (400.0-750.0)	<0.001
Walking speed (m/s)	1.21 (0.96-1.58)	1.16 (0.80-1.65)	0.379
Step length/length of lower extremities	$0.89 \pm 0.13$	$0.87 \pm 0.10$	0.295
Gait Deviation Index (points)	$91.4 \pm 5.7$	$93.1 \pm 6.0$	0.244
Physical activity time (hours/week)	1.3 (0.0-6.0)	4.9 (0.0—24.0)	<0.001

(Figure 2) Motor function and physical activity time

Results are presented as means  $\pm$  SD or median (range).

The significance level is set at p < 0.05.

Furthermore, we examined the relationship between disease severity (o/e LHR, birth weight, 5 min Apgar score, and duration of oxygen requirement) and motor function in children born with CDH. The analysis revealed that a lower o/e LHR, an indicator of prenatal disease severity, was associated with reduced grip strength in school-age children (Figure 3).

(Figure 3) Correlations between motor and functional tests and perinatal factors

### Children born with CDH (n=20)\*

Children born with CDH (n=20) <sup>*</sup> * Perinatal data were available.					
	o/e LHR (%)	Birth weight (kg)	5 min Apgar score (points)	oxygen requirement (days)	
Grip strength	0.47 (p=0.038)	0.38 (p=0.098)	0.39 (p=0.093)	-0.41 (p=0.071)	
Five times-sit-to-stand test	-0.33 (p=0.154)	0.29 (p=0.209)	-0.26 (p=0.276)	0.16 (p=0.495)	
One-leg standing time	-0.16 (p=0.498)	0.12 (p=0.612)	0.09 (p=0.716)	0.26 (p=0.269)	
6 min walking distance	0.02 (p=0.950)	-0.23 (p=0.324)	0.24 (p=0.305)	-0.32 (p=0.166)	

Data are presented as correlation coefficient (p value).

In conclusion, school-age children born with CDH are at risk of decreased motor function in terms of limb muscle strength, balance, and endurance. Assessing grip strength in children born with more severe CDH is particularly important in clinical settings. Although this study did not investigate the reasons for reduced physical activity time, providing objective data on motor function may help encourage children to participate in physical activity.

# Research Summary and Future Perspective

Follow-up and exercise programs focusing on limb muscle strength, balance, and endurance, as well as ensuring adequate physical activity time, may improve motor function in children born with CDH. In the future, we plan to conduct long-term assessments of motor function and evaluate the effectiveness of targeted follow-up and exercise programs.

# Publication

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