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Original Article

Gross motor dysfunction as a risk factor for aspiration pneumonia in children with cerebral palsy

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Abstract

Background Respiratory problems, such as aspiration pneumonia, are major causes of morbidity and mortality in children with cerebral palsy (CP) and greatly affect the quality of life of these children. Nevertheless, there is limited data on the incidence and risk factors of aspiration pneumonia in children with CP in Indonesia.

Objective To determine the incidence and risk factors of aspiration pneumonia in children with cerebral palsy.

Methods In children with CP aged 1-18 years, incidence of pneumonia was studied prospectively for 6 months and the prevalence of the risk factors was studied cross-sectionally. At baseline, we evaluated subjects' by history-taking, physical examination, risk factors, and chest X-ray to assess the incidence of silent aspiration. Subjects were followed-up for six months to determine the incidence of overt or silent aspiration pneumonia.

Results Eight out of 36 subjects had one or more episodes of aspiration, consisting of silent aspiration (2/36) and clinically diagnosed aspiration pneumonia (7/36). Subjects with more severe gross motor dysfunction experienced more episodes aspiration pneumonia, although it was not statistically significant (P=0.06), while dysphagia (P=0.2) and nutritional status (P=0.11) were not associated with pneumonia or silent aspiration.

Conclusion Twenty-five percent of children with CP experience aspiration pneumonia during the 6-month study period, with gross motor dysfunction as a possible risk factor. **[Paediatr Indones. 2017;57:229-33 ; doi: http://dx.doi.org/10.14238/pi57.5.2017.229-33]**.

Keywords: *aspiration; pneumonia; cerebral palsy*

Cerebral palsy (CP) is the most common disability in children. In the United States, the incidence is 2-2.5 children per 1000 live births. During January-August 2012, 20 new patients were diagnosed with cerebral palsy in Dr. Cipto Mangunkusumo Hospital, Jakarta, Indonesia. Children with CP have many complications related to the disease, such as respiratory problems, oromotor dysfunction, gastrointestinal problems, seizures, and mental disabilities.[1]

Respiratory problems, such as aspiration pneumonia, hypoventilation, sleep apnea, and recurrent respiratory tract infection, are often neglected in children with CP. These problems may affect their quality of life and become major morbidities.[2] Approximately 77% of deaths in children with neurological impairment were caused by pneumonia. Incidence of aspiration pneumonia may be as high as 41.5%.[3] Nevertheless, little is known on the incidence and risk factors for aspiration

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pneumonia in children with CP in Indonesia, where awareness of these respiratory problems is low.

Methods

In this study, incidence of pneumonia was studied prospectively and the prevalence of the risk factors was studied cross-sectionally in children with CP. The subjects ranged from 1 to 18 years of age. Subjects with risk factors for aspiration, such as Down syndrome, facial anomalies, tracheostomy, tracheoesophageal fistules, laryngomalacia, and severe intellectual disabilities, were excluded. This study was approved by the University of Indonesia Ethics Committee.

Forty-four subjects were consecutively recruited at the Pediatric Neurology Clinic and Ward of Dr. Cipto Mangunkusumo Hospital, Jakarta, Indonesia in March 2015. At baseline, we evaluated children by history-taking, physical examination, noting risk factors using a dysphagia questionnaire, and chest X-ray during the first visit to assess the incidence of silent aspiration. Subjects were followed-up for six months from April 1 to September 30, 2015 to determine the incidence of aspiration pneumonia. On each visit, the patient was evaluated for signs and symptoms of aspiration. Diagnosis was established by signs and symptoms of aspiration pneumonia and confirmed by chest X-ray examination. Patients with aspiration pneumonia were treated accordingly.

We used the *Statistical Package for Social Science (SPSS) version 20* in all analyses. Analysis of the risk factors contributing to aspiration pneumonia was done by Fisher's exact and Mann-Whitney tests. Multivariate analysis was done by logistic regression test.

Results

Forty-four patients met the study criteria, but four were excluded. Therefore, a total of 40 subjects were followed-up.

The majority of subjects were male, with a male to female ratio of 1.35:1. The Subjects' median age was 3.5 (range 1.3 to 14.2) years. The most common types of CP were spastic (35/40) and quadriplegic (33/40). Most of the subjects had *Gross Motor Function Classification System* (GMFCS) scale of V (34/40). An almost equal proportion of subjects used a nasogastric tube (NGT) and more than half of them had good nutritional status. The majority of subjects had undergone medical rehabilitation (28/40). Subjects' characteristics are shown on **Table 1**.

Table 1. Subjects' characteristics

Characteristics	N=40
Gender, n (%)	
Male	23 (58)
Female	17 (42)
CP type, n (%)	
Diplegia	6 (15)
Quadriplegia	33 (82)
Monoplegia	1 (3)
Spastic	35 (60)
Hypotonic	5 (40)
Dysphagia, n (%)	
Yes	39 (98)
No	1 (2)
GMFCS level, n (%)	
I	0 (0)
II	1 (2)
III	0 (0)
IV	5 (13)
V	34 (85)
Nutritional status, n (%)	
Severe malnutrition	4 (10)
Mild to moderate malnutrition	13 (33)
Good	21 (53)
Overweight	1 (2)
Obese	1 (2)
Using NGT, n (%)	
Yes	19 (48)
No	21 (52)
Had undergone medical rehabilitation, n (%)	
Yes	28 (70)
No	12 (30)

One subject died in the third month of follow-up (probably due to meningitis) and another subject died in the fifth month of follow-up (probably due to sepsis). Thus, they were considered to have dropped out from this study. Two other subjects were lost to follow-up because the family moved to another city. Hence, 36 subjects completed the follow-up for six months. Two subjects had silent aspiration. Aspiration pneumonia was clinically diagnosed in 7 episodes of 36 subjects. These episodes were not evident in the radiological examination, but 6 out of 7 chest X-rays were done within less than 6 hours of the predicted aspiration event.

Risk factors for aspiration analyzed in this study were GMFCS scale, dysphagia, and nutritional status. None of the risk factors was found to be significantly associated with aspiration (**Table 2**).

Table 2. Risk factors of aspiration in children with CP

Risk factors	Aspiration (n=8)	No aspiration (n=28)	P value
GMFCS level			
II	1	0	0.06
IV	2	3	
V	5	25	
Oromotor dysfunction			
Yes	7	28	0.22
No	1	0	
Nutritional statu			
Wasting	1	13	0.12
Good	7	15	
Using NGT			
No	3	13	0.70
Yes	5	15	
Physiotherapy			
No	4	5	0.09
Yes	4	23	

Discussion

Almost all subjects completed the follow-up and the drop out or loss to follow-up rate was minimal. Subjects were followed up for six months to detect at least one episode of pneumonia in children with CP. This study is the first prospective cohort study to evaluate the incidence and risk factors of aspiration pneumonia in children with CP in Indonesia. A previous study had shown that a retrospective study was not a reliable design for detecting pneumonia, thus, we chose a prospective cohort as the best design.[4]

A limitation of this study was our small sample size and that dysphagia was evaluated using a modified questionnaire, which only gave the general picture of dysphagia. There are various validated surveys for patients with developmental delay and intellectual disability, such as the *Dysphagia Disorder Survey*.[5] Only two patients underwent flexible endoscopic evaluation of swallowing to evaluate for dysphagia. A second limitation of this study was that chest X-ray performed only once, during the first visit. As such, we could not evaluate for silent aspiration occurring during the second to sixth months. Furthermore, gastroesophageal reflux as another risk factor for aspiration was not evaluated in this study.

Most subjects in our study were male (23/40). Male biological sex is a risk factor for CP, as explained by a previous study.[6] The median age of subjects was 3.5 years. Another study in Indonesia found that the mean age at CP diagnosis was 28.8 months.[7] The majority of subjects in this study had GMFCS level V (30/36) and spastic quadriplegic type of CP. This study was conducted in the top tertiary hospital which explained why most study subjects had the severe form of CP and gross motor function level. Almost all subjects had dysphagia (35/36). Silent aspiration was found in 2 out of 36 subjects. Subjects did not undergo a videofluoroscopic swallowing study to detect aspiration; therefore, it is possible that more subjects experienced silent aspiration. There were 7 episodes of clinically diagnosed aspiration pneumonia in 36 subjects. Other studies reported that the incidence of aspiration pneumonia in children with CP was estimated to be 27-41.5%, and could even be as high as 90%.[3,8-9] One subject experienced two episodes of aspiration pneumonia in the six months of follow-up.

Aspiration is often undetected in children with CP. Three out of six subjects had witnessed episodes of aspiration, but almost all subjects had no pathognomonic chest X-ray to confirm aspiration pneumonia, probably due to the chest X-rays being performed within less than six hours after the aspiration episodes. Thus, the diagnosis of aspiration pneumonia was made based on clinical findings.

Risk of aspiration may decrease after nutritional, medical, and behaviorial modification. Patients with dysphagia were given liquid or modified solid food. A nasogastric tube is needed in patients with known risk factors of aspiration, after nutritional and behaviorial modification.[10] Almost half of the subjects in this study had apparently undergone nutritional modification, which could decrease the incidence of aspiration in this study. The majority of subjects had also undergone physical and oromotor rehabilition. These steps could help airway clearance and, as a result, prevent aspiration.[2]

The GMFCS shows the level of motor developmental delay. We found that subjects with more severe gross motor dysfunction experienced more episodes of aspiration pneumonia, although it

was not statistically significant (P=0.06). Gross motor function may worsen or improve over time. Children with CP who underwent physical rehabilitation for 6, 12, and 18 months showed improved gross motor function measures.[11,12] The follow-up of risk factors for aspiration pneumonia should be done continuously, as the level of gross motor function is dynamic, especially after physical rehabilitation. Unfortunately, we did not monitor whether the risk factors evolved during the six months of study.

Oromotor dysfunction is common in children with CP.[13] Recurrent aspiration due to oromotor dysfunction in children with CP may cause chronic cough, breathing problems during sleep, airway colonization by pathogens, and progressive lung parenchymal destruction, which leads to death.[14] Almost all subjects in our study had oromotor dysfunction, which may explain why we found no significant association between oromotor dysfunction and aspiration in our study.

Malnutrition in children with CP is caused by swallowing problems, gastroesophageal reflux, and increased energy expenditure.[15,16] These conditions result in muscle catabolism, including respiratory muscles, which decreases lung function and makes children with CP prone to pneumonia.[2,17] Previous studies in Myanmar and Indonesia found high prevalences of malnutrition in children with CP.[18,19] In contrast, we found that more than half of the subjects had good nutritional status. This finding may be due to many of our subjects having received nutritional modification training. We also did not find a significant association between aspiration pneumonia and nutritional status.

In conclusion, 25% of subjects with CP in our 6-month study have episodes of aspiration, either silent aspiration or aspiration pneumonia. Gross motor dysfunction may be associated with aspiration pneumonia in children with CP.

Conflict of Interest

None declared.

References

1. Krigger KW. Cerebral palsy: an overview. Am Fam Physician. 2006;73:91-100.
2. Seddon PC, Khan Y. Respiratory problems in children with neurological impairment. Arch Dis Child. 2003;88:75–8.
3. Polednak AP. Respiratory disease mortality in an institutionalised mentally retarded population. J Ment Defic Res. 1975;19:165-72.
4. Calis E. Correlates of lower respiratory tract infections and nutritional state in children with severe generalized cerebral palsy and intellectual disability [dissertation]. [Rotterdam]: Erasmus Universiteit; 2011.
5. Sheppard JJ. Dysphagia disorders survey and dysphagia management staging scale, users manual and test forms revised. New Jersey: Nutritional Management Associates; 2002. p.???
6. Chounti A, Hägglund G, Wagner P, Westbom L. Sex differences in cerebral palsy incidence and functional ability: a total population study. Acta Paediatr. 2013;102:712-7.
7. Rahmat D, Mangunatmadja I, Tridjaja B, Tambunan T, Suradi R. Prevalence and risk factors for epilepsy in children with spastic cerebral palsy. Paediatr Indones. 2010;50:11-7.
8. Arvedson J, Rogers B, Buck G, Smart P, Msall M. Silent aspiration prominent in children with dysphagia. Int J Pediatr Otorhinolaryngol. 1994;28:173–81.
9. Rogers B, Arvedson J, Buck G, Smart P, Msall M. Characteristics of dysphagia in children with cerebral palsy. Dysphagia. 1994; 9: 69–73.
10. Marik PE. Aspiration pneumonitis and aspiration pneumonia. N Engl J Med. 2001;344:665-71.
11. Stark C, Nikopoulou-Smyrni P, Stabrey A, Semler O, Schoenau E. Effect of a new physiotherapy concept on bone mineral density, muscle force and gross motor function in children with bilateral cerebral palsy. J Musculoskelet Neuronal Interact. 2010;10:151-8.
12. Ketelaar M, Vermeer A, Hart H, van Petegem-van Beek E, Helders PJ. Effects of a functional therapy program on motor abilities of children with cerebral palsy. Phys Ther. 2001;81:1534-45.
13. Reilly S, Skuse D, Poblete X. Prevalence of feeding problems and oral motor dysfunction in children with cerebral palsy: a community survey. J Pediatr. 1996;129:877-82.
14. Lefton-Greif MA, Carroll JL, Loughlin GM. Long-term follow-up of oropharyngeal dysphagia in children without apparent risk factors. Pediatr Pulmonol. 2006;41:1040–8.
15. Bell KL, Boyd RN, Tweedy SM, Weir KA, Stevenson RD, Davies PS. A prospective, longitudinal study of growth,

nutrition and sedentary behaviour in young children with cerebral palsy. BMC Public Health. 2010;10:179-91.

16. Rogers SL, Coe CL, Karaszewski JW. Immune consequences of stroke and cerebral palsy in adults. J Neuroimmunol. 1998;91:113-20.
17. Kuperminc MN, Stevenson RD. Growth and nutrition disorders in children with cerebral palsy. Dev Disabil Res Rev. 2008;14:137–46.
18. Sjakti HA, Syarif DR, Wahyuni LK, Chair I. Feeding difficulties in children with cerebral palsy. Paediatr Indones. 2008;48:224-9.
19. May WL, Win H, Linn K, Oo K, Kyi S, Phyu AH, Pau DL. Nutritional status of children with cerebral palsy in cerebral palsy clinic, Yangon children's hospital. Myanmar Health Sci Res J. 2014;26:22-7.