Journal of the Medical Sciences
(Berkala Ilmu Kedokteran)

Volume 54, Number 4, 2022; 384-391
https://doi.org/10.19106/JMedSci005404202209

Family-centered rehabilitation in a high-risk infant: a case report

Lulus Hardiyanti[1]*, Rizky Kusuma Wardhani[2], Aditya Rifqi Fauzi[3], Gunadi[3]

[1] Physical Medicine and Rehabilitation Division, Department of Surgery, Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada/Dr. Sardjito General Hospital, Yogyakarta, [2]Department of Physical Medicine and Rehabilitation, Faculty of Medicine Universitas Indonesia/ Dr. Cipto Mangunkusumo General Hospital, Jakarta, [3]Pediatric Surgery Division, Department of Surgery, Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada/Dr. Sardjito General, Hospital, Yogyakarta, Indonesia

Submitted: 2022-05-23
Accepted : 2022-10-03

ABSTRACT

Preterm infants are categorized as a high-risk group in neonatology. They are physiologically less mature and have limited compensatory responses to the extrauterine environment compared with term infants. Preterm infants need long-term evaluation, monitoring, and follow-up to optimize neonatal care and development through an extensive rehabilitation period. However, the COVID-19 pandemic restricted patient care and follow-up in the outpatient hospital setting. This case report discusses a high-risk infant treated with family-centered rehabilitation (FCR). The patient's rehabilitation issues included delays in gross motor, fine motor, and language development and preventing complications that may arise in a high-risk premature infant. Considering recent occurrences, our approach to rehabilitation programs for high-risk infants needs to be reevaluated and revised, focusing on home programs through family-centered treatment. These techniques may aid in delivering rehabilitation treatments to children with developmental delays during COVID-19.

ABSTRAK

Bayi prematur merupakan kelompok risiko tinggi di bidang neonatologi, karena secara fisiologis belum matur dan kemampuan kompensasi terhadap lingkungan ekstrauterin yang terbatas dibandingkan bayi cukup bulan. Bayi prematur biasanya memerlukan evaluasi, monitoring, dan ditindak lanjuti terus menerus untuk mengoptimalkan perawatan dan perkembangan neonatal melalui periode rehabilitasi jangka panjang. Namun, terjadinya pandemi COVID-19 menyebabkan terbatasnya pelayanan dan tindak lanjut pasien di poli rawat jalan. Di sini, kami melaporkan kasus bayi risiko tinggi dengan tatalaksana melalui *family-centered rehabilitation* (FCR). Masalah rehabilitasi yang didapatkan pada pasien meliputi keterlambatan perkembangan motorik kasar, motorik halus, bicara bahasa, dan mencegah komplikasi yang mungkin timbul pada bayi prematur risiko tinggi. Mengingat kondisi pandemi yang baru terjadi, pendekatan program rehabilitasi pada bayi risiko tinggi perlu dievaluasi dan direvisi kembali, dengan menitikberatkan pada tatalaksana di rumah yang berbasis pada keluarga. Teknik-teknik ini dapat membantu dalam memberikan layanan rehabilitasi medik kepada anak dengan keterlambatan perkembangan selama pandemi COVID-19.

Keywords:
high-risk infant;
family-centered rehabilitation;
COVID-19;
pandemic

*corresponding author: lulus.hardianti@ugm.ac.id

INTRODUCTION

Late preterm infants (LPIs) are born between 34 0/7 and 36 6/7 weeks gestational age. From 2014 to 2016, the LPI birth rate rose from 6.82% to 7.09%, accounting for approximately 72% of all preterm births in the United States. In Indonesia, more than 550,000 late preterm infant babies are born yearly.[1-3] Compared with term infants, preterm infants have an immature central nervous system. Between 34 and 40 weeks of gestational age, cortical volume increases by 50%, and 25% of cerebellar development occurs. These factors place preterm infants at an increased risk for altered brain development, which may influence long-term neurodevelopmental outcomes. Late preterm infants account for 72% of all preterm births, and even the smallest increases in adverse outcomes could causing a large public health burden.[4-6]

It has been suggested that congenital cytomegalovirus (CMV) infection is a risk factor for preterm delivery.[7] Infection by CMV is the most frequent cause of congenital infection globally, affecting 0.7-6.1% of newborns. Among symptomatic patients, 50% presented with sequelae. The most serious sequelae are neurocognitive impairment and sensorineural hearing loss. Symptoms at birth include low birth weight and microcephaly. Microcephaly at birth was the most specific predictor of poor cognitive outcome and major motor disability in children with symptomatic congenital CMV infection. A highly significant positive correlation was found between head size at birth and the intelligence/developmental quotient (IQ/DQ).[8-10]

A long-term evaluation, monitoring, and follow-up of high-risk infants are needed to optimize neonatal care, promote healthy growth and development, and improve human health status. However, the COVID-19 pandemic restricted the patient care and follow-up. Family-centered rehabilitation that provides home-based programs is considered useful in addition to or even replacing center-based therapy in this situation.

Home-based programs provide a unique opportunity to train continuously. These programs enable parents to incorporate training into their daily routine with their childs. Separated training is not needed. In addition, increased training may facilitate the retention of established intervention effects.[11,12]

When parents become therapy providers, the relationship between parents and the health professional changes: the health professional becomes the parents' coach. Depending on the role of parents and their specific needs, the way and amount of coaching can vary from limited instruction only at the beginning of the program to extensive demonstration, feedback, and coaching throughout the entire program. The coaching mode can vary from home visits by the therapist to remote coaching by e-mail or telephone consultation.[11]

Family-centeredness has been the cornerstone of service delivery for children with developmental difficulties in pediatric health care for decades. Services that provided family-centered care were associated with improved developmental outcomes and adjustment for the children, better family functioning, parental wellbeing, parental perceptions of competency and satisfaction, and more efficient use of services.[13] Here, we report a case of a high-risk premature infant treated with FCR.

Case

In October 2020, a male infant patient was referred from the Pediatric Outpatient Clinic, Dr. Sardjito General Hospital with a perinatal history of

congenital infection of cytomegalovirus (CMV), herpes simplex virus (HSV), respiratory distress, hypoglycemia after delivery, left parieto-temporal epidural hemorrhage, left temporal intracerebral hemorrhage, bilateral intraventricular hemorrhage, umbilical cord hemorrhage due to suspected vitamin K deficiency, neonatal anemia, and extrahepatic cholestasis due to sepsis.

Considering fetal distress, intrauterine growth restriction, and oligohydramnios, the patient was delivered late preterm (35 weeks of gestational age) via cesarean section at Dr. Sardjito General Hospital. The body weight at birth was 1270 g (extreme low), body length was 39 cm, and head circumference was 28 cm. The patient was admitted to the Neonatal Intensive Care Unit (NICU) for 35 days. Before being discharged home, the patient could breastfeed orally, and his body weight reached 1810 g.

The patient is the first child of his parents, live together daily. His mother is his primary caregiver, and his father earned 1.8 million rupiahs monthly as a private employee. The medical treatment for this patient was covered by national healthcare insurance. There was no history of developmental problems or serious sickness in the family. The patient's mother was pregnant at the age of 33 y.o. During her pregnancy course, she received regular antenatal care from a midwife. She consumed vitamins and iron supplements daily during her pregnancy.

When the patient arrived at the Medical Rehabilitation Outpatient Clinic, he was 6th months 16th d.o. (correction age 5th months 9th d.o.) and had a complete medical examination. His body weight was 6.4 kg, his height was 63.5 cm, his body mass index (BMI) was 15.87 kg/m^2, and his head circumference was 38.5 cm. Temperature, pulse, and blood pressure were all within normal limits. The respiratory system and other internal organs were all functioning normally. During the examination of the extremities, movements in both hands and feet were equally active, there was hypertonus on the trunk extensor and extremity, physiological reflexes (biceps, triceps, and patella) were increased +3/+3, and pathological reflexes (Hoffman-Trommer and Babinski) were positive. The patient could roll to prone, but it was still on rare occurrence, and he could not return. The patient could elevate his head in a prone position despite having poor neck control, turning his head left and right, and holding a toy that is touched to his hand (movement/physical development match to 3 mo.o. baby). Primitive reflex showed central nervous system maturation at the midbrain level (asymmetrical tonic neck reflex (ATNR) positive, neck righting reflex positive, foot placement reflex negative, and parachute reflex negative).

The patient made a sound (mumble) in a low voice, reacted to the sound of the bell, and did spontaneous cooing (language/communication match to a 2-3 mo.o. baby). He also could recognize his mother and smile spontaneously (a social/emotional match to 4 mo.o. baby). The patient received enough breast milk, could suck adequately, and had no previous choking or shortness of breath episodes during breastfeeding. The patient has also begun receiving supplementary food, organic baby porridge, given twice a day in amounts of up to 2 tablespoons; there have been no complaints of coughing or choking while swallowing. Oromotor function examination showed inadequate lip seal with drooling, good oral hygiene, and baby teeth have not grown yet.

The patient's rehabilitation issues included delays in gross motor with postural control abnormality, fine motor, language development, and preventing complications that may arise in a high-risk premature infant (growth failure, neurologic impairments, developmental

delay, and lower cognitive functioning). The rehabilitation management goals were for parents to understand the importance of the rehabilitation program provided for the patient, to be able to stimulate their child according to the child's developmental stages, to optimize the child's growth and development, and to prevent the risk of neurodevelopmental disabilities.

Because of the COVID-19 pandemic that has been impacted since May 2020, most non-emergent practices, including outpatient rehabilitation services, have been restricted by hospital regulations based on government regulations. For this patient, we applied family-centered rehabilitation combined with hospital-based rehabilitation treatment.

The patient visited the outpatient rehabilitation clinic every two weeks for physiotherapy treatments and evaluation by a physiatrist (including the application of the FCR program and achievement of the child's development). For a one-hour therapy session, parents were coached and guided on how to correctly position and stimulate their child at home to improve postural control development and facilitate normal movement patterns. Parents' training includes actions during which health care professionals guide parents and demonstrate how to apply intervention strategies clearly and strictly. During the treatment session, we also ensured whether parents could do the therapeutic technique correctly as a daily routine, i.e., motor stimulation in the prone position with hand support to assist neck and trunk control.

As a daily routine, parents applied a massage method for tactile and proprioceptive stimulation for 10-15 min. Play therapy for 10-15 min at least three times daily to develop reaching and grasping functions by employing colorful toys and appealing sounds. Parents were also advised to maximize engagement and communication with the child (for example: conduct a chit chat for every diaper change, dressing, and bathing, tell a tale while massaging the child, play peek-a-boo, sing, and play with appealing toys according to the child's developmental stage).

At four months follow-up after the rehabilitation program, when the patient was 11 months 3 d.o. (correction age 9 mo 26 d.o.), his body weight was 9.1 kg (weight to age -1<Z<0), body height 74.2 cm (height to age 0<Z<2), BMI 16.5 kg/m^2, and head circumference 41.7 cm (-3<Z<-2, microcephaly). The patient was able to sit independently and crawl. In terms of fine motor, the patient could reach and hold a toy before moving it from one hand to the other, but he couldn't pinch a small object. The patient could smile or laugh when playing peek-a-boo and show several facial expressions (happy, sad, and surprised). During playtime with the mother, the patient could make sounds or babble in a low voice, but it was uncommon. The patient turned toward the bell sound but didn't respond when his mother called his name. Brainstem evoked response audiometry (BERA) examination showed moderate-severe hearing loss in the right ear (60 dB) and moderate hearing loss in the left ear (42.5 dB). Movement/physical development and social-emotional milestones have reached his age's expectations, but we need more attention to language and speech development. The hearing aids and a new strategy would be provided to develop a wide range of listening skills and optimize speech/language development.

DISCUSSION

The patient, in this case, was born at 35 wk of gestation; hence he was categorized as a LPI. Although late preterm infants may be close to term, losing the last 6 weeks of gestation is vital to their physiologic and metabolic maturity. These LPIs have higher morbidity and mortality rates

than term infants. The most common morbidities experienced by LPIs include respiratory complications, feeding difficulty, hypoglycemia, temperature instability, hyperbilirubinemia, and neurodevelopmental delays.[1,3] The perinatal history of congenital infection of CMV and HSV that cause very low birth weight and microcephaly further add to the problems and place the patient at a high risk of developmental delay and cognitive impairment.

During the COVID-19 pandemic, the World Health Organization (WHO) urges individuals of all ages to take precautions against the virus. Current advice emphasizes the necessity of avoiding public areas and remaining at home.[14] The family-centered rehabilitation approach that provides a home-based program might be a good solution to solve this problem. Parents with adequate resources and support and physical and psychological health can provide positive caregiving environments for children with physical disabilities.[15,16]

For this patient, we gave daily programs at home and face-to-face training to parents to take on a therapeutic role as a co-therapist. The patient's mother is a housewife and has enough hours daily to perform the program next to her daily activities. A systematic review reported coaching parents is a key element of home-based programs. When parents are effectively coached and guided throughout the training period, parents become more confident in carrying out the home-based program, find it easier to implement the program in their daily routine, and enjoy seeing their children improve.[11,16]

After four months, the patient has reached the expected motor development of his age. It could be due to no upper motor neuron sign, such as spasticity or congenital deformity that could prevent normal motor development. High compliance of parents in implementing the rehabilitation program plays an important role. A systematic review has indicated that parental involvement in early intervention is associated with a better outcome for infants and families.[11,17]

But this patient has not yet reached his expected speech and language development age. Our patient's moderate-severe bilateral hearing loss may restrict access to speech and language input and significantly impact oral communication development. Unlike children with severe and profound loss, our patient still has some access to speech. However, this access will be highly dependent on the amplitude and spectrum of the speech signal and the presence of noise.[18,19]

Home-based programs have become the practice of choice in pediatric rehabilitation and early childhood intervention programs. Our fundamental goal in health service delivery is, and indeed must be, improved quality of life for the affected child and family. Family-centered care provides an evidence-based means to objectively achieve this goal. Evidence of the beneficial effects of this method on outcomes in varying domains exists, including better demonstrable child development, improved child psychological adjustment, enhanced parental psychological wellbeing (reduced stress, anxiety, depression), more robust parental perceptions of competency and control, and higher levels of actual satisfaction with the care provided.[20]

Even though parents are great importance in home-based programs, a survey among parents has shown that they do not have an unfavorable opinion concerning home programs because these programs may induce or enhance stress in parents. Parents may experience pressure to comply, especially when the program is demanding. Furthermore, the altered parent-child interaction during training may cause additional tension. As the role of parents changes to that of a therapy provider, this may

cause a conflict between their parenting style and their approach as a therapy provider. Consequently, the loss of motivation by parents and/or children to complete training activities may affect compliance and probably the effectiveness of the intervention. Because of the factors mentioned above, home-based interventions need to be carefully developed and implemented.[11,21]

Prior study in Turkey pioneered the development of emergency remote training programs for young children with Down syndrome, learning disabilities, and serious health issues.[22] They assessed the effectiveness of the "applied emergency remote training program" designed to meet the requirements of parents of children with Down syndrome and provide them with at-home care. It was an evaluative case study with 11 parents of Down syndrome children aged 11 to 35 mo. The findings showed that the program could be used in the home, enhanced parents' and children's interactional behavior, decreased the number of challenging routines, and could be used as an educational, instructional, and band-aid solution.

During the pandemic, some countries used telemedicine as their major method of providing chronic disease services, while others relied on Virtual Reality and Video Games. Low- and middle-income nations and distant places with limited access to rehabilitation treatments benefit from telemedicine.[23]

Family-centeredness has been the cornerstone of service delivery for children with developmental difficulties in pediatric care.[11,13] However, this approach needs parents and caregivers to be fully involved and adequately educated to attend to the children's needs. Clinicians must ensure the family is ready and can stimulate children according to prescribed treatment. The development of FCR in Indonesia, especially in remote places, may need a long way to go. This program may need further look to be incorporated into national healthcare insurance to include follow-up visits by healthcare providers to ensure optimal children's care at home.

CONCLUSION

Considering recent occurrences, our approach to physical therapy and rehabilitation management of infant with a high risk of developmental delay must be reevaluated and revised, focusing on family-centered programs. These techniques may aid in delivering rehabilitation programs to children with developmental delays during COVID-19.

ACKNOWLEDGMENTS

We would like to thank all staffs and nurses that involved in the patient's care.

REFERENCES

1. Natarajan G, Shankaran S. Short- and long-term outcomes of moderate and late preterm infants. Am J Perinatol 2016; 33(3):305-17. https://doi.org/10.1055/s-0035-1571150
2. Sharma D, Padmavathi IV, Tabatabaii SA, Farahbakhsh N. Late preterm: a new high risk group in neonatology. J Matern Fetal Neonatal Med 2019; 34(16):2717-30. https://doi.org/10.1080/14767058.2019.1670796
3. Handayani KD, Irwanto, Masturina M, Etika R, Harianto A, Sauer PJJ. Duration of breastfeeding in LPIs maternal and infant factors. J Hum Lact 2021; 37(4):795-802. https://doi.org/10.1177/0890334420978380
4. Kinney HC. The Near-term (late preterm) human brain and risk for periventricular leukomalacia: a review. Semin Perinatol 2006; 30(2):81-8. https://doi.org/10.1053/j.semperi.2006.02.006

5. Cheong JL, Doyle LW, Burnett AC, Lee KJ, Walsh JM, Potter CR, *et al*. Association between moderate and late preterm birth and neurodevelopment and social-emotional development at age 2 years. JAMA Pediatr 2017; 171(4):e164805. https://doi.org/10.1001/jamapediatrics.2016.4805
6. Tripathi T, Dusing SC. Long-term neurodevelopmental outcomes of infants born late preterm: a systematic review. Res Rep Neonatol 2015; 2015:91-111. https://doi.org/10.2147/RRN.S44062
7. Turner KM, Lee HC, Boppana SB, Carlo WA, Randolph DA. Incidence and impact of CMV infection in very low birth weight infants. Pediatrics 2014; 133(3):e609-15. https://doi.org/10.1542/peds.2013-2217
8. Lanzieri TM, Leung J, Caviness AC, Chung W, Flores M, Blum P, *et al*. Long term outcome of children with congenital CMV infection. J Perinatol 2017; 37(7):875-80. https://doi.org/10.1038/jp.2017.41
9. Lanzieri TM, Dollard SC, Bialek SR, Grosse SD. Systematic review of the birth prevalence of congenital cytomegalovirus infection in developing countries. Int J Infect Dis 2014; 22:44-8. https://doi.org/10.1016/j.ijid.2013.12.010
10. Noyola DE, Demmler GJ, Nelson CT, Griesser C, Williamson WD, Atkins JT, *et al*. Early predictors of neurodevelopmental outcome in symptomatic congenital cytomegalovirus infection. J Pediatr 2001; 138(3):325-31. https://doi.org/10.1067/mpd.2001.112061
11. Beckers LWME, Geijen MME, Kleijnen J, Rameckers EAA, Schnackers MLAP, Smeets RJEM, *et al*. Feasibility and effectiveness of home-based therapy programmes for children with cerebral palsy: A systematic review. BMJ Open 2020; 10(10):e035454. https://doi.org/10.1136/bmjopen-2019-035454
12. An M, Palisano RJ. Family-professional collaboration in pediatric rehabilitation: a practice model. Disabil Rehabil 2014; 36(5):434-40. https://doi.org/10.3109/09638288.2013.797510
13. Pekcici EBB, Akin EO, Akpinar F, Hayran G, Keleş C, Yağbasan B, *et al*. Family-centeredness of services for young children with down syndrome: an observational study from turkey. Turk J Med Sci 2021; 51(1):246-55. https://doi.org/10.3906/sag-2009-76
14. Wu Z, McGoogan JM. Characteristics of and important lessons from the Coronavirus disease 2019 (COVID-19) outbreak in China: summary of a report of 72314 cases from the Chinese Center for Disease Control and Prevention. JAMA 2020; 323(13):1239-42. https://doi.org/10.1001/jama.2020.2648
15. King G, Chiarello L. Family-centered care for children with cerebral palsy: conceptual and practical considerations to advance care and practice. J Child Neurol 2014; 29(8):1046-54. https://doi.org/10.1177/0883073814533009
16. Almasri NA, An M, Palisano RJ. Parents' perception of receiving family-centered care for their children with physical disabilities: a meta-analysis. Phys Occup Ther Pediatr 2018; 38(4):427-43. https://doi.org/10.1080/01942638.2017.1337664
17. Ziegler SA, Hadders-Algra M. Coaching approaches in early intervention and paediatric rehabilitation. Dev Med Child Neurol 2020; 62(5):569-74. https://doi.org/10.1111/dmcn.14493
18. Tomblin JB, Harrison M, Ambrose SE, Walker EA, Oleson JJ, Moeller

MP. Language outcomes in young children with mild to severe hearing loss. Ear Hear 2015; 36(Suppl 1):76S-91S.
https://doi.org/10.1097/AUD.0000000000000219
19. Korver AMH, Smith RJH, van Camp G, Schleiss MR, Bitner-Glindzicz MAK, Lustig LR, *et al*. Congenital hearing loss. Nat Rev Dis Primers 2017; 3:16094.
https://doi.org/10.1038/nrdp.2016.94
20. Shevell M, Oskoui M, Wood E, Kirton A, van Rensburg E, Buckley D, *et al*. Family-centred health care for children with cerebral palsy. Dev Med Child Neurol 2019; 61(1):62-8.
https://doi.org/10.1111/dmcn.14053
21. Kokorelias KM, Gignac MAM, Naglie G, Cameron JI. Towards a universal model of family centered care: A scoping review. BMC Health Serv Res 2019; 19(1):564.
https://doi.org/10.1186/s12913-019-4394-5
22. Çelik S, Tomris G, Tuna DM. The COVID-19 pandemic: The evaluation of the emergency remote parent training program based on at-home support for children with down syndrome. Child Youth Serv Rev 2022; 133:106325.
https://doi.org/10.1016/j.childyouth.2021.106325
23. Chandolias K, Tsigaras G, Besios T, Chalkia A, Trevlaki E. The impact of the COVID-19 pandemic on the rehabilitation of children with chronic diseases and disabilities. J Health Sciences & Research 2021; 11(1):190.