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Original Research

Influence of route of delivery on perinatal outcomes in fetuses with myelomeningocele

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Summary

The optimal route of delivery of fetuses with myelomeningocele is controversial. The aim of this study is to determine whether route of delivery predisposes to perinatal complications and influences short-term outcomes in patients with myelomeningocele. The authors performed a retrospective review of the medical records of 26 patients with myelomeningocele admitted to the Neonatal Intensive Care Unit in this hospital from 2001 to 2015. They compared perinatal complications and short-term outcomes of elective cesarean section (n = 21) and vaginal delivery (n = 5) groups. There were no ruptured nor infectious myelomeningoceles cases in either group. No statistically significant difference in ambulation status at two years of age was observed between the two groups. The present data suggest that perinatal complications and short-term outcomes were not associated with route of delivery. Vaginal delivery might be the optimal route of delivery for fetal myelomeningocele if there is no obstetric contraindication.

Key words: Cesarean section; Infection; Myelomeningocele; Rupture; Vaginal delivery; Ambulation.

Introduction

The worldwide incidence of neural tube defects (NTDs) ranges from 1.0 to 10.0 per 1,000 births, with myelomeningocele being the commonest NTD [1]. With the widespread use of maternal serum α-fetoprotein screening, ultrasonography, and magnetic resonance imaging, antenatal diagnosis of myelomeningocele has increased [2]. A review of second-trimester ultrasound examination in a high-risk population reported a detection rate for myelomeningocele of about 95 percent [3]. Fetuses with an antenatally diagnosed myelomeningocele should be delivered at a comprehensive perinatal medical center with neonatal intensive care unit and pediatric neurosurgery services. However, the optimal route of delivery of fetuses with antenatally diagnosed meningomyelocele remains controversial [4]. Although patients with lipomeningomyelocele (in which neural tissue is covered and protected by skin) often have almost normal lower leg function, most newborns with open NTDs exhibit severe neurologic impairment of the lower extremities at birth. These findings suggest that neurologic injury may occur antenatally or at the time of delivery, and that direct injury to the spinal cord may cause damage to and loss of function of the spinal cord [5, 6]. In addition, a previous study suggests that the presence of central nervous system infection is linked to increased mortality and decreased intellectual potential [7]. It is the present authors policy to deliver fetuses with antenatally diagnosed myelomeningocele by cesarean section in order to prevent infection and damage to the myelomeningocele at delivery. This also ensures a smooth transition of care from the obstetrician to the neonatologist and pediatric neurosurgeon after delivery. On the other hand, recent studies propose that vaginal delivery can be considered if there are no obstetrical indications, e.g. breech presentation or cephalopelvic disproportion, for cesarean section. These studies demonstrated no benefit in terms of motor function from delivery by cesarean section, or avoidance of labor in these fetuses [8, 9].

The aim of this study is to determine whether the route of delivery affects perinatal complications and short-term outcomes in patients with myelomeningocele. The authors compared fetuses born by cesarean section to those born by vaginal delivery, including those who were delivered vaginally in other hospitals and transported to the neonatal intensive care Unit (NICU) at this hospital.

Materials and Methods

Forty-two neonates with spina bifida (32 spina bifida aperta and 10 spina bifida occulta) were admitted to the NICU at this hospital between 2001 and 2015. Of the 32 cases of spina bifida aperta, the authors excluded two cases with additional congenital anomalies. They also excluded four cases where myelomeningocele was diagnosed postnatally in other hospitals after cesarean delivery due to obstetric indications. They divided the remaining 26 cases with isolated myelomeningocele into two groups based on

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Table 1. — *Clinical characteristics of patients with myelomeningocele (mean ± SD).*

	Cesarean delivery	Vaginal delivery	*p* value
Number	21	5	-
Gestational age at delivery (week)	37.3±1.1	40.0±0.86	<0.001
Birth weight (Z score)	0±1.12	-0.47±0.99	0.95
Apgar score 1 minute	8±2.27	8±0.55	0.035
Apgar score 5 minutes	9±1.10	9±0.71	0.191
Maternal age	31±6.7	34±6.8	0.396
Head circumference (Z score)	1.53±0.09	0.07±0.59	0.051
Hydrocephalus	20 (95.2%)	1 (20%)	0.0016
Sac diameter (cm)	3.5±1.87	3.0±1.79	0.51
Anatomical level of lesion (index)	21.0±2.97	23.0±2.0	0.08

SD = standard deviation.

route of delivery (cesarean delivery in this hospital, n=21; vaginal delivery in other hospitals, n=5), and assessed differences in the frequency of perinatal complications and short-term outcomes between these two groups. This was done by retrospectively reviewing the medical records. The following clinical information was recorded: gestational age at delivery, birth weight, maternal age, presence of hydrocephalus, sac diameter, anatomical level of lesion, complications at delivery. The gestational ages were determined by the last menstrual period, as well as a crown–rump length measured by ultrasound in the first trimester. To compare body weight and head circumference between the cesarean and vaginal delivery groups, Z-score transformations were used. For Z transformation of body weight and head circumference, the publicly available data (mean and standard deviation per gestational week for each parameter) of Japanese newborns documented by the Ministry of Health, Labor, and Welfare was used (http://www.mhlw.go.jp/houdou/0110/h1024–4c.html#hyo14) [10]. The size of the myelomeningocele defect was defined as the largest diameter of the placode in centimeters, measured at the time of closure. The anatomical level of the vertebral defect was defined as the most caudal intact lamina on computed tomography or magnetic resonance imaging after birth. This vertebral level was converted to a numeric index as described in a previous report (cervical 1 to 8, thoracic 9 to 20, lumbar 21 to 25, sacral 26 to 29, and intact motor function 30) [11]. Perinatal complications of delivery were rupture of the sac, elevated CRP levels at birth, and infection in the first week after birth. Ambulatory status was determined at two years of age. The Hoffer Functional Ambulation scale was used to classify patients into community ambulators (capable of walking indoors and outside), household ambulators (capable of walking indoors only, and requiring equipment and/or a wheelchair for outside mobility), and no ambulation (wheelchair dependent) [12]. Statistical differences were determined by probability values $p < 0.05$ (two-tailed). All statistical analyses were performed by using R properly.

Results

Clinical characteristics of patients with myelomeningocele are shown in Table 1. There was a significant difference in gestational age at delivery (37.3 ± 1.2 weeks *vs.* 40.2 ± 1.0 weeks, $p < 0.001$) between the two groups, because elective cesarean section was performed before the onset of labor. Although two cases in the cesarean section group were delivered at 34 and 36 weeks of gestation due to preterm rupture of membranes in the absence of labor, the indication for cesarean section in all other cases in this group was fetal myelomeningocele.

Table 2. — *Evaluation of ambulatory status in 19 patients with myelomeningocele at 2 years of age.*

	Cesarean delivery	Vaginal delivery	*p*-value
Number	16	3	-
Ambulatory status			
Ambulate independently	3 (18.8%)	2 (66.7%)	0.31
Ambulate with assisting device (orthoses, crutches, or walker)	5 (31.3%)	0 (0.0%)	
Not ambulatory	8 (50.0%)	1 (33.3%)	

There were no statistically significant differences in maternal age, gestational age-adjusted birth weight, and Apgar scores between the cesarean section and vaginal delivery groups (Table 1). On the other hand, the frequency of hydrocephalus was significantly higher in the cesarean section group compared to the vaginal delivery group. Corresponding to the frequency of hydrocephalus, gestational age-adjusted head circumference was marginally larger in the cesarean section group compared to the vaginal delivery group. The size of the myelomeningocele sac was not significantly different between the two groups.

The authors further assessed the frequency of rupture of the myelomeningocele sac, elevation of CRP level on the day of the birth, and incidence of infection in the first week after delivery. There were no patients with ruptured myelomeningocele sacs in either group. The authors found neither significant elevation of CRP level on the day of birth, nor significant incidence of infection in the first week after delivery in the two groups. All patients underwent standard neurosurgical closure within 48 hours of birth in this hospital. There were no signs of infection during admission in the NICU in either of the two groups.

Finally, the authors evaluated the ambulatory status of 19 patients with myelomeningocele at two years of age (16 cesarean sections and three vaginal deliveries). Eight patients who had delivered by cesarean section and one delivered vaginally were not ambulatory at the age of two years (Table 2).

Discussion

The optimal mode of delivery for fetuses diagnosed antenatally with myelomeningocele remains controversial. There is concern that vaginal delivery may damage the spinal cord, leading to a decline in neurologic function in these patients [13]. Therefore, cesarean section tends to be selected as the delivery mode for fetuses with this condition. The rationale is that it will prevent infection and dam-

age to the myelomeningocele at delivery, as well as ensuring a smooth transition from obstetrician to neonatologist and pediatric neurosurgeon after delivery. However cesarean section itself has some risks to the woman. These include surgical complications such as hemorrhage, venous thromboembolism, infection, and injury to other organs. There are also potential complications to subsequent pregnancies due to scarring of the uterus. These include placenta previa, placenta accreta, and uterine rupture. There are also neonatal risks, such as transient tachypnea, respiratory distress syndrome, and persistent pulmonary hypertension [14, 15].

Recently, Greece *et al*. summarized the current literature on the effect of mode of delivery on motor function. Although previous large studies [11, 16, 17] recommended cesarean section for fetal myelomeningocele, they indicated problems with these studies, in that they were more than a decade old, and many of the infants in these studies were diagnosed at the time of birth [9]. Therefore, they suggested that a registry of myelomeningocele patients be kept to compile their data in a standardized fashion with regards to mode of delivery and motor function as specified by Luthy *et al.* [11]. Maternal data points would also be included.

There is no study discussing the optimal route of delivery of fetuses with myelomeningocele in Japan. In this study, the authors assessed rupture of the myelomeningocele sac leading to infection, as this is an important prognostic factor in the management of infants with myelomeningocele in NICUs. In keeping with recent studies on the optimal route of delivery of fetuses with myelomeningocele [8, 9, 11], the present findings showed no apparent association between the route of delivery and perinatal complications and short-term outcomes. However, this study had some limitations, such as case selection and outcome measures. The patients in the vaginal delivery group were not diagnosed antenatally. To avoid sample selection bias, the authors needed to enrol vaginally delivered patients who were antenatally diagnosed with myelomeningocele, as a control group. In addition, they had a limitation in assessment of neurologic function in patients with myelomeningocele. Although they defined ambulatory status at two years of age as representative of neurologic function in patients with myelomeningocele, and showed no statistical difference in ambulatory status between cesarean section and vaginal delivery groups, they could not measure the level of paralysis (motor and sensory level), and could not determine the difference between the anatomical and the motor levels as described by Luthy *et al.* [11]. Further studies are needed to evaluate neurological function of patients with myelomeningocele.

In conclusion, vaginal delivery may be a delivery option for fetuses with myelomeningocele. In order to obtain a better understanding, and to delineate the optimal route of delivery for fetal myelomeningocele, a multicentric retrospective observation study should be performed as a next step.

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