

## Bochdalek's Hernia in the Newborn

—6 years' experience—\*)

Takashi YOKOYAMA, Toru ICHIKAWA, Eiso HIYAMA,  
Mitsuaki OKITA and Nobukazu MIYOSHI

*The 1st Department of Surgery, Hiroshima University School of Medicine, 1-2-3  
Kasumi, Minami-ku, Hiroshima 734, Japan*

(Received September 25, 1984)

*Key words: Bochdalek's hernia, Persistent fetal circulation*

### ABSTRACT

We treated 13 cases of neonatal Bochdalek's hernia at the 1st Department of Surgery, Hiroshima University Hospital in the 6 years from 1978 to 1983 and have studied on the factors responsible for mortality of neonatal Bochdalek's hernia.

The following conclusions were obtained in this study.

1. Of the 13 patients, 7 survived and 6 died. The most frequent cause of death was persistent fetal circulation, the cause of 5 of the deaths.
2. All 3 of the premature newborns died.
3. The earlier the time of onset of symptoms, the higher was the mortality.
4. Five of the 7 patients who required respiratory control by intubation before hospitalization died.
5. The mortality rate for patients with pH less than 7.00, base excess less than  $-15$  mEq/liter, and M index higher than 6 on admission was extremely high.
6. Three of the 7 patients persisting a distinct pressure difference on simultaneous measurement of preductal PaO<sub>2</sub> and postductal PaO<sub>2</sub> died. In the surviving patients, it was possible to maintain the postductal PaO<sub>2</sub> at above 60 mmHg.
7. Trazoline was used on 2 patients, and both died. A thorough study of the time and mode of administration is considered to be necessary.

### INTRODUCTION

Poor therapeutic results have been reported for Bochdalek's hernia in the newborn, especially in patients operated on within 24 hr after birth<sup>1,3,9,11</sup>. With progress in the medical care system for newborns in recent years, cases of Bochdalek's hernia in the early postnatal period can be detected and the results of surgery have become increasingly poor owing to the increase in these cases detected in the early postnatal period. Previously, the reason for the bad prognosis in early neonatal Bochdalek's hernia was considered to be hypoplasia of the lung<sup>1,9</sup>. But with the progress in examination techniques in recent years, studies have been made from the viewpoint of hemodynamics and

the poor therapeutic results are now attributed to hypoxemia due to persistent fetal circulation resulting from contraction of the pulmonary artery<sup>2,4,8</sup>. We had 13 patients with neonatal Bochdalek's hernia from 1978 to 1983, of whom 6 died. We wish to report on the studies we have made on these cases and on the factors responsible for the mortality of neonatal Bochdalek's hernia.

### MATERIALS

The study was made on 13 patients with neonatal Bochdalek's hernia admitted to and then treated at the 1st Department of Surgery, Hiroshima University Hospital, in the 6 years from 1978 to 1983. Respiratory care by mechanical ventilation was given to all of the

\*) 横山 隆, 市川 徹, 檜山英三, 沖田光昭, 三好信和: 最近6年間に経験したボーダレック孔ヘルニア症例の検討

patients. Patients No. 1-7 survived and patients No. 8-13 died, giving a mortality rate of 46%. The following abbreviations are used: PFC, persistent fetal circulation;  $\text{FiO}_2$ , fractional inspired oxygen concentration;  $\text{PaO}_2$ , arterial oxygen tension (mmHg);  $\text{PaCO}_2$ , arterial carbon dioxide tension (mmHg); and M index, modified index.

### RESULTS

The risk factors prior to hospitalization to our clinic are shown in Table 1. The mortality rate was higher to low-body weight newborns. There were no survivors among the newborns

with birth weight less than 2500 g.

As regards the time of symptoms, all of the patients who died developed cyanosis or apnea immediately after birth. Onset of symptoms was delayed in 3 of the surviving patients and prognosis was better than in those with early onset of symptoms. These seriously ill patients who developed apnea and cardiac arrest and received cardiopulmonary resuscitation all died, and their poor prognosis was considered to be related to contraction of the pulmonary artery due to hypoxia. The time from birth to hospitalization in our clinic was within 6 hr for all patients who died, and all except 1 of the

Table 1. Risk Factors among neonates with Bochdalek's hernia (Before arrival)

No.	Sex	Gestational Weeks	Birth weight (g)	affected side (sac)	Onset of symp. after birth (hr)	symptom	Cardiopulm. resuscitation	Intubation before arrival	Arrival time at our hospital after birth (hr)
1	♀	40	3200	left (+)	Immediately	Cyanosis	(-)	(+)	4
2	♂	40	3850	left	1/4 hr	Cyanosis	(-)	(-)	8
3	♂	40	2821	left	Immediately	Cyanosis	(-)	(+)	13
4	♀	40	3040	right (+)	Immediately	Cyanosis	(-)	(-)	17
5	♂	40	3300	left	12 hr	Cyanosis	(-)	(-)	22
6	♀	38	2870	left	Immediately	Cyanosis	(-)	(-)	23
7	♂	36	2755	left	60 hr	Cyanosis	(-)	(-)	63
8	♀	36	2260	right (+)	Immediately	Cyanosis	(+)	(+)	1½
9	♂	40	2970	left	Immediately	Apnea	(-)	(+)	2½
10	♂	36	2300	left	Immediately	Apnea	(-)	(+)	3½
11	♂	41	3300	left	Immediately	Cyanosis	(+)	(+)	4
12	♀	41	2970	left	Immediately	Cyanosis	(-)	(-)	5
13	♂	37	2300	left	Immediately	Apnea	(+)	(+)	6½

Table 2. Blood gas analysis on admission

Patient	$\text{FiO}_2$	pH	$\text{PO}_2$ (mmHg)	$\text{PCO}_2$ (mmHg)	BE (mEq/liter)	M index
1	1.0	7.034	36.9	91.1	-10.3	16.85
2	1.0	7.170	90.0	61.2	-8.5	7.24
3	0.4	7.317	134.1	31.4	-8.9	1.83
4	0.5	7.113	51.2	60.5	-11.5	5.49
5	0.4	7.182	36.8	56.1	-8.3	5.84
6	0.21	7.382	44.1	36.6	-2.8	2.36
7	0.4	7.360	92.9	38.3	-3.5	2.55
8	1.0	6.929	91.5	76.5	-18.4	6.97
9	1.0	7.028	26.2	66.3	-15.9	24.68
10	1.0	7.154	86.1	59.2	-9.9	7.59
11	1.0	6.775	57.9	106.7	-23.9	10.47
12	1.0	7.112	41.3	74.8	-8.9	15.45
13	1.0	6.780	60.1	103.0	-23.9	10.15

Table 3. Blood gas analysis after operation

Patient	FiO <sub>2</sub>	pH	PO <sub>2</sub> (mmHg)	PCO <sub>2</sub> (mmHg)	BE (mEq/liter)	M index
1	1.0	7.031	59.4	97.1	-9.3	10.37
2	1.0	7.336	499	28.8	-9.1	1.37
3	0.5	7.260	92.5	45.7	-7.1	3.24
4	1.0	7.496	392.9	22.8	-3.0	1.76
5	0.6	7.225	82.5	64.2	-3.0	4.20
6	1.0	7.272	265.5	42.3	-7.9	2.53
7	0.3	7.242	103.3	31.1	-2.8	1.69
8	1.0	6.906	382.7	56.3	-21.0	1.72
9	1.0	7.201	47.2	54.1	-7.9	13.96
10	1.0	7.462	343.3	28.8	-1.6	1.99
11	1.0	6.806	31.8	138.5	-14.8	18.07
12	1.0	7.080	20.1	90.1	-5.0	30.99
13	1.0	7.233	415.2	52.8	-6.8	1.59

surviving patients came to our clinic after a lapse of over 6 hr. This was considered to show that prognosis was poor in patients who presented with serious symptoms and were transferred to our clinic soon after birth.

The results of blood gas analysis made at the time of admission to our clinic are shown in Table 2. The pH was 7.0 or less in 3 patients, all of whom died. These 3 patients had developed cardiac arrest before or during hospitalization in our clinic. Base excess was over -18 mEq/liter, and marked metabolic acidosis was present.

Alveolar artery oxygen pressure difference as an indicator of efficiency of oxygen transport in the lung is not always a reliable indicator, because it is likely to change with FiO<sub>2</sub> where as the modified index is less affected by changes in FiO<sub>2</sub> and is therefore more reliable. For that reason we used the modified index (M index)<sup>12)</sup>. The normal range of the M index is 1.36-1.45 in newborns<sup>16)</sup>. The M index was higher than 6 in all patients who died but in only 2 of the patients who survived. In view of this, a preoperative disturbance in the efficiency of oxygen transport was believed to have the principal effect on the prognosis.

The results of blood gas analysis after operation were as shown in Table 3, with many showing improvement compared with the results at the time of hospitalization. Two patients with base excess of less than -10 mEq/liter and 3 out of 4 patients with an M index of higher than 10 died. Decreased efficiency of

oxygen transport and persistent metabolic acidosis due to tissue hypoxia were found to affect the prognosis. Change in FiO<sub>2</sub>, preductal PaO<sub>2</sub>, postductal PaO<sub>2</sub> and PaCO<sub>2</sub> within 60 hr in patients under postoperative mechanical ventilation for respiratory care together and the special drugs used are shown in Figs. 1 and 2.

The postoperative blood flow ratio between the left and the right lungs as determined by pulmonary blood flow scintigrams is shown in Fig. 1. The weights of the left and right lungs, presence or absence of cardiac deformity and causes of death in the fatal cases as determined at autopsy are shown in Fig. 2.

In patients who survived, postductal PaO<sub>2</sub> did not decrease to less than 60 mmHg and in all of the 5 patients who showed a difference of more than 20 mmHg between preductal PaO<sub>2</sub> and postductal PaO<sub>2</sub>, the difference disappeared within 42 hr. By studying the surviving patients, it was considered necessary to maintain the postductal PaO<sub>2</sub> over 60 mmHg. Furthermore, not a single case of retrorenal fibrosis was found among the surviving patients in postoperative examinations.

Respiratory care was given to patients 11, 12 and 13 while only the preductal PaO<sub>2</sub> was measured, and patients 11 and 12 died with no elevation in PaO<sub>2</sub> levels, which made us consider the involvement of a ventilation disturbance but the cause of death was believed to be PFC.

In patients 8, 9 and 13, in whom PaCO<sub>2</sub> levels were considered to be normal, postductal PaO<sub>2</sub>

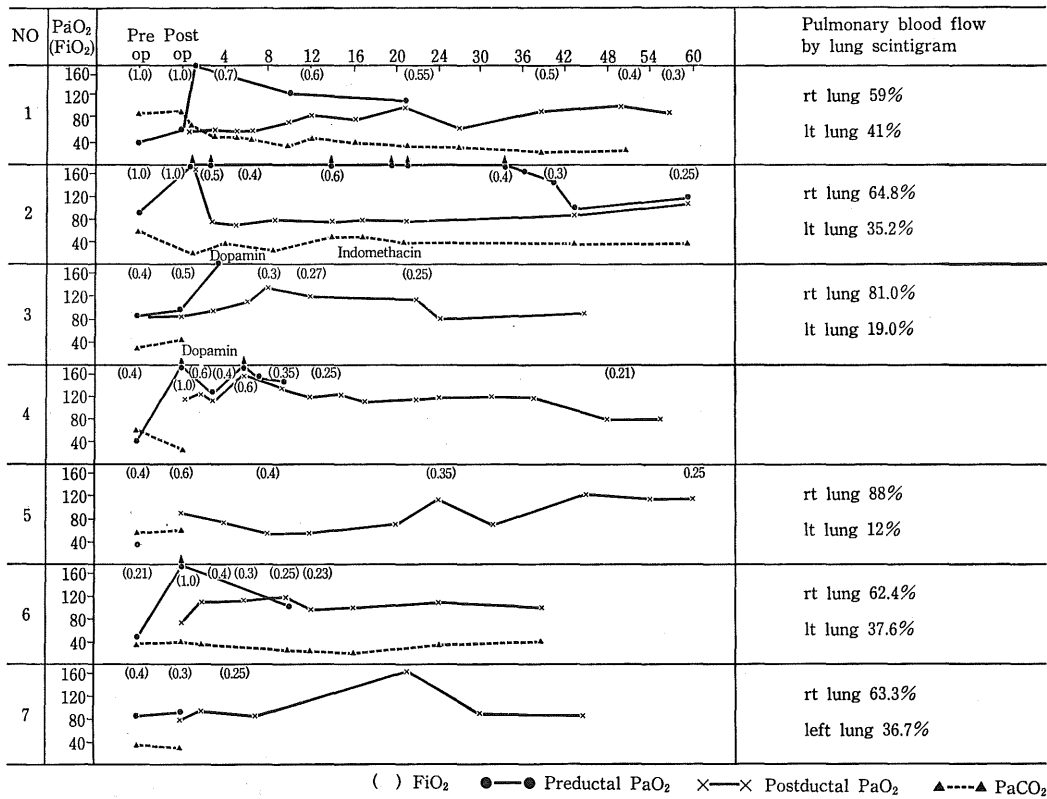


Fig. 1. Clinical course of Bochdalek's hernia (No. 1)

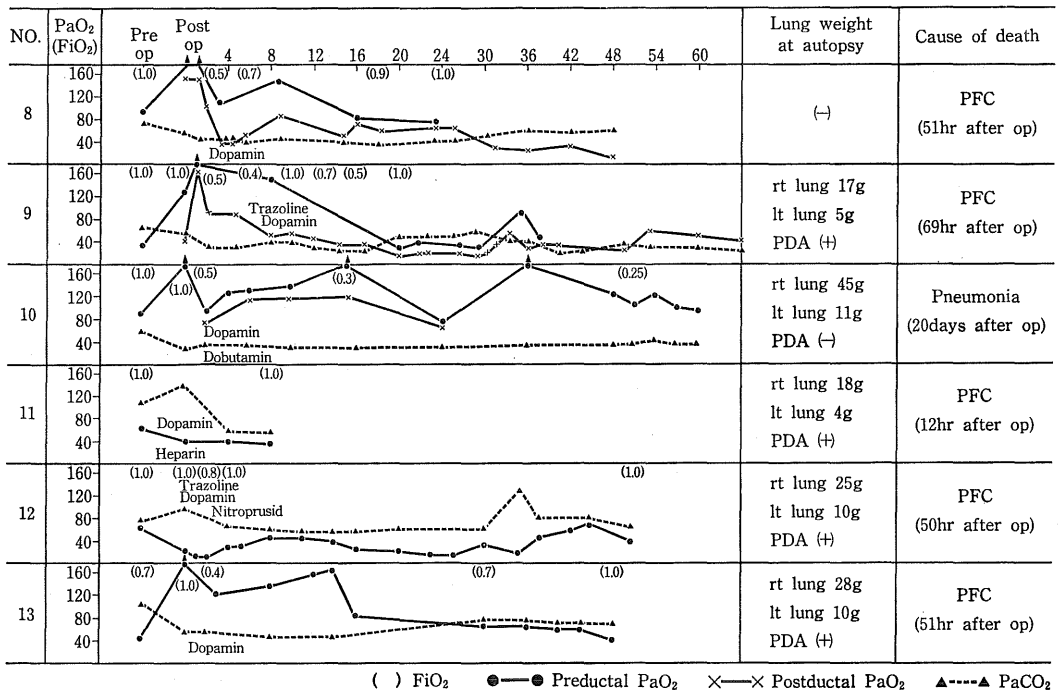


Fig. 2. Clinical course of Bochdalek's hernia (No. 2)

was over 100 mmHg in the early postoperative period, within the so-called honeymoon period. As the  $\text{FiO}_2$  decreased, a precipitous decrease in  $\text{PaO}_2$  occurred. This is presumably due to respiratory care in which preductal  $\text{PaO}_2$  was used as the index for respiratory care. Respiratory care with postductal  $\text{PaO}_2$  as the index should perhaps be given in such cases. Trazoline was administered into the right arm in cases 9 and 12, but there was no pronounced improvement.

### DISCUSSION

Results of therapy for Bochdalek's hernia are poor in patients operated on within 24 hr of birth<sup>1,3,9,11</sup>. Involvement of PFC has been reported to be able to cause of the bad prognosis. PFC in Bochdalek's hernia is caused by delayed contraction of the pulmonary blood vessels due to marked hypoxia resulting from respiratory disturbance<sup>2,4,8</sup>. As diagnostic criteria for PFC in Bochdalek's hernia, Nagaya et al.<sup>10</sup> gave 1) shunt ratio of more than 20% as determined by preductal  $\text{PaO}_2$ , 2) a difference of more than 20 mmHg between preductal  $\text{PaO}_2$  and postductal  $\text{PaO}_2$ , 3) pulmonary arterial pressure and systemic arterial pressure either almost equal or with the former higher and 4)  $\text{PaCO}_2$  less than 60 mmHg.

On the other hand, Ogawa et al.<sup>16</sup> reported that the practical method of diagnosis is based only on the pressure difference between preductal  $\text{PaO}_2$  and postductal  $\text{PaO}_2$ . Although pulmonary arterial pressure was not measured in our cases, as viewed from the shunt ratio determined by preductal  $\text{PaO}_2$  and the pressure difference between preductal  $\text{PaO}_2$  and postductal  $\text{PaO}_2$ , PFC was the cause of death in 3 of the 6 patients who died. In 2 other cases, although only preductal  $\text{PaO}_2$  was measured, PFC was considered to be the cause of death in view of the clinical course in these patients. In cases of Bochdalek's hernia in the postnatal period, aggravation of PFC can be said to be the greater cause of death. The mortality rate was high in our patients weighing less than 2500 g at birth. Gastzman<sup>6</sup> reported that PFC is more frequent in mature newborns when the cases are not complicated by respiratory diseases detected roentgenologically, but in patients with respiratory diseases it was more frequent in premature newborns. In Bochdalek's hernia,

which is a respiratory disease, PFC was considered to be more aggravated in premature newborns. The earlier the time of onset of symptoms the poorer was the prognosis but PFC is an adaptation disorder occurring within 24 hr of birth and it is considered that the earlier the onset of symptoms the more it is aggravated. Patients in whom intubation was performed for apnea of serious respiratory disturbances prior to their hospitalization presented with marked metabolic acidosis at the time of hospitalization. PFC was found to have been caused by tissue hypoxia due to marked respiratory disturbance attributable to Bochdalek's hernia.

It has been stated that the reason why PFC develops so easily in patients with Bochdalek's hernia are not only tissue hypoxia but also an increase in pulmonary arterial smooth muscle<sup>10</sup> and a decrease in the number of pulmonary vessels<sup>10,14</sup>.

From the therapeutic viewpoint, which is the best index to use during respiratory care? On the basis of our studies, it is believed to be important to monitor both the preductal  $\text{PaO}_2$  and the postductal  $\text{PaO}_2$ . It is practical to base the diagnosis of PFC on the pressure difference between preductal and postductal  $\text{PaO}_2$  and the shunt ratio determined by preductal  $\text{PaO}_2$ . It is considered to be important for respiratory care to maintain the postductal  $\text{PaO}_2$  at higher than 60 mmHg.

As treatment for PFC, mechanical ventilation especially hyperventilation, drug treatment<sup>6,13</sup>, PDA ligation<sup>8</sup> and extracorporeal circulation<sup>5</sup>, are used at present. Mechanical ventilation was used for all of our patients, but Peckham<sup>17</sup> was stated that hyperventilation is more effective. As drug treatment, Trazoline has been used as the drug of first choice and cases in which it was effective have been reported. We also used it in 2 cases, but its effectiveness perhaps was not fully demonstrated because it was administered into the right arm and used after the  $\text{PaO}_2$  had drastically decreased. We believe it should be administered directly into the pulmonary artery by inserting a catheter, while monitoring the pulmonary artery pressure.

Goto et al.<sup>7</sup> have reported on PDA ligation, but there are arguments for and against it, and it should be studied further. Since our cases

are still few and the various factors cannot be said to have been sufficiently monitored, we wish to continue our study.

#### REFERENCES

1. **Adelman, S. and Beusa, C. D.** 1976. Bochdalek hernia in Infants: Factors determining mortality. *J. Ped. Surg.* 11 : 569-573.
2. **Bloss, R. S., Arande, J. V. and Beaudmore, H. E.** 1981. Vasodilator response and prediction or survival in congenital diaphragmatic hernia. *J. Ped. Surg.* 16 : 118-121.
3. **Collins, D. L., Pomerance, J. J., Travis, K. W., Turner, S. W. and Pappelbaum, S. J.** 1977. A new approach to congenital posterolateral diaphragmatic hernia. *J. Ped. Surg.* 12 : 149-156.
4. **Ein, S. H., Barker, G., Olley, P., Shandling, B., Simpson, J. S., Stephens, C. A. and Filler, R. M.** 1980. The pharmacologic treatment of newborn diaphragmatic hernia. A 2-year evaluation. *J. Ped. Surg.* 15 : 384-394.
5. **German, J. C., Gazzaniga, A. B., Amlie, R. and Huxtable, R. F.** 1977. Management of pulmonary insufficiency in diaphragmatic hernia using ECMO. *J. Ped. Surg.* 12 : 905-912.
6. **Goezman, B. V., Sumshine, P., Johnson, J. D., Wennberg, R. P., Hackel, A., Merten, D. F., Bartoletti, A. L. and Sliverman, N. H.** 1976. Neonatal hypoxia and pulmonary vasospasm. Response to trazoline. *J. Pediat.* 89 : 617-621.
7. **Goto, T., Hatta, M., Horiba, K., Hashimoto, S. and Miyano, H.** 1978. A case of congenital diaphragmatic hernia with extreme hypoxia. *J. Intensive Care Medicine ICU & CCU* 2 : 671-678.
8. **Haller, J. A., Singer, R. D., Golladay, E. S., Inon, A. E., Harrington, D. P. and Shermeta, D. W.** 1976. Pulmonary and ductal hemodynamics in studies of simulated diaphragmatic hernia of fetal and newborn lambs. *J. Ped. Surg.* 11 : 675-680.
9. **Johnson, D. C., Dearen, R. M. and Koop, C. E.** 1967. Diaphragmatic hernia in infancy: Factors affecting the mortality rate. *Surgery* 62 : 1082-1091.
10. **Levin, D. L.** 1978. Morphologic analysis of the pulmonary vascular bed in congenital left-sided diaphragmatic hernia. *J. Pediat.* 92 : 805-809.
11. **Livaditis, A. and Nordstrand, A.** 1971. Congenital posterolateral diaphragmatic hernia in infants. *Scand. J. Thoracic. Cardiovasc. Surg.* 5 : 67-73.
12. **Marukawa, S.** 1977. An appraisal method of the efficiency of oxygen transport in the lung-Respiratory M index and its concepts. *Jap. J. Anesthesia* 26 : 1501-1510.
13. **Moodie, D. S., Talander, R. L., Kleinberg, F. and Feldt, R. H.** 1978. Use of trazoline in newborn infants with diaphragmatic hernia and severe cardiopulmonary disease. *J. Thorac. Cardiovasc. Surg.* 75 : 725-729.
14. **Naeye, R. L., Shochat, S. J., Whitman, V. and Maisels, M. J.** 1976. Unsuspected pulmonary vascular abnormalities associated with diaphragmatic hernia. *Pediatrics* 58 : 902-906.
15. **Nagaya, M., Ito, T., Sugita, T., Niinomi, N., Yamada, N., Ishiguro Y., Tsuda, N., Ando, H., Watanabe, Y. and Iyomasa, Y.** 1981. Persistent fetal circulation associated with congenital diaphragmatic hernia. *Jap. J. Ped. Surg. Associat.* 17 : 851-859.
16. **Ogawa, T., Takahashi, S., Kimura, K. and Kohno, S.** 1982. Evaluation of Trazoline treatment on PFC of congenital diaphragmatic hernia. *Jap. J. Neonat.* 18 : 758-763.
17. **Peckham, G. J. and Fox, W. W.** 1978. Physiologic factors affecting pulmonary artery pressure in infants with persistent pulmonary hypertension. *J. Pediat.* 93 : 1005-1010.
18. **Tunell, R.** 1975. The influence of different environmental temperatures on pulmonary gas exchange and blood gas change after birth. *Acta Pediat. Scand.* 64 : 57-68.