

High recurrence rate in thoracoscopic repair of Bochdalek type congenital diaphragmatic hernia

Torakoskopik onarımın Bochdalek tipi doğuştan diyafragma hernisinde yüksek nüks oranı

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Background: This study aims to determine the possible causes of high recurrence in thoracoscopic repair of congenital diaphragmatic hernia.

Methods: Medical records of 14 children (8 boys, 6 girls; mean estimated gestational age of 38.2 weeks; range 36 to 40 weeks) who underwent thoracoscopic repair of Bochdalek type congenital diaphragmatic hernia in our clinic between August 2006 and August 2010 were retrospectively analyzed. Data including demographic characteristics, intraoperative details and respiratory status of the patients were reviewed in terms of recurrence.

Results: Only one patient died due to pulmonary hypertension. The mean follow-up was four years (range, 3 to 7 years). Recurrence was seen in five patients. No significant difference in the parameters evaluated was found between the groups.

Conclusion: Because of the high recurrence rate after thoracoscopic repair of the congenital diaphragmatic hernia, care must be taken to select the low risk patient for recurrence.

Keywords: Congenital diaphragmatic hernia; recurrence; thoracoscopic repair.

Amaç: Bu çalışmada doğuştan diyafragma hernisinde torakoskopik onarım ardından gözlenen yüksek nüksün olası nedenleri araştırıldı.

Çalışma planı: Bu çalışmada kliniğimizde Ağustos 2006 - Ağustos 2010 tarihleri arasında torakoskopik diafragma hernisi onarımı uygulanmış olan 14 çocuğun (8 erkek, 6 kız; ortalamaları tahmini gebelik yaşı 38.2 hafta; dağılım 36-40 hafta) tıbbi kayıtları retrospektif olarak incelendi. Hastaların demografik özellikleri, ameliyat sırası ayrıntıları ve hastaların solunum parametreleri nüks açısından değerlendirildi.

Bulgular: Yalnızca bir hasta ağır pulmoner hipertansiyon nedeni ile kaybedildi. Ortalama izlem süresi dört yıl (dağılım 3-7 yıl) idi. Beş hastada nüks görüldü. Değerlendirilen parametreler açısından gruplar arasında anlamlı bir fark saptanmadı.

Sonuç: Doğuştan diyafragma hernisi, torakoskopik olarak onarıldığında yüksek nüks oranına sahiptir ve uygun hasta seçimi riskin düşürülmesinde önemlidir.

Anahtar sözcükler: Doğuştan diyafragma hernisi; nüks; torakoskopik onarım.

Minimally invasive surgery for a congenital diaphragmatic hernia (CDH) was first reported in infants with delayed hernias,^[1] and after 2000, this type of surgery began to be the treatment of choice, however, the results are still inconsistent.^[2-5] In a recent report by Tsao et al.^[5] that included the

prospective data from the congenital diaphragmatic hernia (CDH) study group, they found a higher recurrence rate with thoracoscopic repair. Our experience also reveals a good early postoperative outcome with a subsequent high recurrence rate when performing this procedure (a recurrence rate of five



Available online at
www.tgkdc.dergisi.org
doi: 10.5606/tgkdc.dergisi.2014.9271
QR (Quick Response) Code

Received: September 08, 2013 Accepted: November 04, 2013

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out of 14 for the thoracoscopic group and one out of 12 for the laparotomy group; $p=0.04$). The aim of this study was to evaluate the high rate of recurrence after thoracoscopic repair of the Bochdalek-type of CDH to determine the reason for this phenomenon.

PATIENTS AND METHODS

The hospital records of 14 children (8 boys and 6 girls; mean estimated gestational age of 38.2 weeks; range 36-40 weeks) who had undergone thoracoscopic repair of the Bochdalek-type CDH between August 2006 and August 2010 at the Ege University Department of Pediatric Surgery were reviewed retrospectively. All of the patients were treated by a single surgeon for a CDH and were operated on using the thoracoscopic technique. The following data was reviewed and evaluated in terms of recurrence: estimated gestational age, birth weight, gender, side of defect, comorbidities, age at repair, intraoperative details (the presence of a hernia sac and its excision along with the need for rib-anchoring sutures), operation time, pre- and postoperative status of the patient (vital signs, arterial blood gases, and ventilatory parameters), duration of mechanical ventilation, length of hospital stay, and survival. Afterwards, the patients were divided into recurrence and no-recurrence groups.

The operative technique included three trocars and diaphragmatic repair using interrupted 2/0 silk sutures. No staples or non-resorbable mesh were used. Furthermore, an extra small incision (1 cm) was made so that additional posterolateral diaphragmatic stitches could be passed around the ribs and tied extracorporeally when there was no identifiable rim.¹⁶ All of the operations were performed by the same surgeon, and statistical analyses were performed using either Fisher's exact test and the Mann-Whitney U test.

RESULTS

The mean follow-up period was four years (3-7 years), and there were five recurrences. The mean birth weight was 3570 (2490-4000) grams. No differences were found when the demographics were examined in the recurrence and no-recurrence groups, so we also reviewed the operative database and medical records of the patients to determine the possible reasons for the recurrences.

All five patients with a recurrence had normal chest X-rays just before they were discharged, and all of the recurrences happened after the initial hospitalization. In four of the patients, they occurred

at between one and four months postoperatively (mean 1.5 months) while it was discovered after 51 months in the other patient. Four of these patients were symptomatic and presented with discomfort and dyspnea or nausea and vomiting. The fifth was asymptomatic, and the recurrence was detected in the follow-up assessment the first month after surgery.

Nine patients were diagnosed antenatally while three had no antenatal diagnosis but presented with respiratory symptoms in the newborn period. Furthermore, two of the patients were diagnosed late (at 5 and 10 months old) with respiratory symptoms (repeated attacks of pneumonia). Two of the patients in the recurrence group were antenatally diagnosed, two were diagnosed in the newborn period, and one at the postoperative fifth month. However, we detected no statistically significant relationship between early or late presentation and recurrence ($p=0.176$).

There were only two patients with comorbidities (symptomatic malrotation in one patient and an inguinal hernia in another), and the patient with malrotation underwent a laparotomy for obstruction of the ileus on postoperative day 12. Interestingly, this patient had experienced no recurrence during the three-year follow-up period. There were no other early postoperative complications, and we found no statistically significant association with respect to comorbidities ($p=0.712$) or the presence of complications ($p=0.539$) and recurrence.

Eight patients required postoperative mechanical ventilation (only one was not on mechanical ventilation before the operation). Our clinic's approach is to keep patients with a markedly increased tension of the diaphragm under neuromuscular blockage and controlled mandatory ventilation for at least three days after surgery, regardless of their respiratory status, and we noted that this had been done for five of the patients in this study. The mean duration of mechanical ventilation (excluding the patient who died) was 11.7 days (range 2-38). Three of the children had severe pulmonary hypertension. Two of these responded to inhaled nitric oxide (NO) and high frequency oscillation ventilation, but the other patient died. In addition, one of these patients with severe hypertension was in the recurrence group. The preoperative (after stabilization) mean oxygenation index was 9.1 for the recurrence group and 10.4 for the non-recurrence group. To determine whether there was a relationship with respiratory status, parameters such as the need for mechanical ventilation, preoperative

oxygenation index of the patients, total duration of mechanical ventilation, and need for inhaled NO application were considered, but no statistically significant relationship between the respiratory status of the patients and recurrence was noted (Table 1).

To evaluate the effect of anatomical differences, intraoperative details were examined by reevaluating the operation notes and operative videos. We also looked at the patient data, including the side of the defect, presence and excision of the sac, and need for rib-anchoring sutures. Unfortunately, no data regarding the size of the defect or tension of the repaired diaphragm could be evaluated since no objective comparable data was available. However, the use of rib-anchoring sutures with no identifiable rim was taken into consideration as an indirect sign of a large defect. There were three cases of right-sided and 11 cases of left-sided hernias. In the recurrence group, one patient had a right-sided hernia and four had left-sided hernias. After our evaluation, we determined that the side of the defect was not related to recurrence ($p=0.670$). In addition, there was a hernia sac in nine patients, and four were excised. In the recurrence group, three of the patients had a sac, and two were excised. However, once again no statistically significant differences could be found regarding the presence of sac ($p=0.545$) or its excision ($p=0.405$) and recurrence. Rib-anchoring sutures were needed in one patient in the recurrence group and three in the non-recurrence group, but there was also no significant correlation between the requirement for rib-anchoring sutures and recurrence ($p=0.689$).

Another determinant cited for the high recurrence rate associated with thorascopies is the technical challenge and steep learning curve of the procedure.

All of the operations in our series were performed by the same surgeon, and no improvement in the recurrence rate was detected during our study period. We also evaluated the length of the procedures between the groups as an indirect sign of the surgeon's experience. The mean length of the procedure (from the moment the patient was taken from the bed to the point of stabilization in the bed after the operation) was 164 minutes (range 100-240) for the recurrence group and 203 minutes (range 120-300) for the non-recurrence group, and this difference was not statistically significant ($p=0.142$).

We determined that there were no identifiable statistically significant differences between the recurrence and non-recurrence groups with regard to any of the parameters (Table 1). At reoperation, the lateral stitches were found to have been pulled through in all cases, and there were no sutures that had come loose, which would have reflected improper flat knot tying. Furthermore, there were no obvious major technical mistakes. Moreover, we performed the thoracotomies and re-repair without using any non-resorbable mesh in the reoperation.

DISCUSSION

Despite being considered as the therapy of choice in most of the centers that are capable of performing thorascopies, controversy still exists with regard to the results of this type of surgery for a CDH.^[2-5] Our clinical results revealed a high recurrence rate; therefore, we stopped performing thorascopies for this fragile patient group and undertook this study to determine the possible cause of the recurrences or at least determine which patients were good candidates for thorascopic repair. Our study mainly focused on the operative findings and respiratory status of the

Table 1. Determinants for the recurrence and non-recurrence groups

	Recurrence group	Non-recurrence group	<i>p</i>
Number of patients	5	9	
Antenatal diagnosis	2	7	0.245
Late diagnosis	1	1	0.176
Comorbidities	1	1	0.712
Need for mechanical ventilation	3	4	0.326
Preoperative oxygenation index	9.1	10.4	0.442
Nitrous oxide requirement	1	2	0.189
Right-sided defect	1	2	0.670
Presence of sac	3	6	0.545
Excision of sac	2	2	0.405
Rib-anchoring sutures	1	3	0.689
Mean length of procedure (minutes)	164	203	0.142

patients because these had been assumed to be the most likely reasons for recurrence in other published reports.^[7,8] However, although every comparable parameter was taken into consideration, we could identify no statistically significant differences between the recurrence and non-recurrence groups.

Some published reports have supported the use of thoracoscopic repair for selected patients, such as those with minimal ventilatory support.^[7] Despite the limited number of patients in our study, our findings contradict this popular opinion. The current common approach at our clinic is to perform a thoracoscopy after a stable respiratory and cardiovascular status has been achieved.^[8] All of our patients must be examined for associated malformations, and appropriate preoperative preparation must be established. We do not consider a patient's respiratory status when deciding whether or not to perform a thoracoscopy because we could not identify any statistically significant difference with respect to respiratory status between the recurrence and non-recurrence groups in our study. In fact, one of the patients with recurrence had an excellent respiratory status and required no mechanical ventilation before or after surgery.

Shah et al.^[3] presented a series of 17 patients with Bochdalek-type diaphragmatic hernias and declared that the initial placement of lateral stitches can minimize the propensity for recurrence by decreasing the tension in this area. However, the importance of lateral stitches was not analyzed in our series.

Keijzer et al.^[9] also evaluated a series of 17 cases and determined that the use of mesh avoids undue tension, thereby preventing recurrence. We could not make such a comparison since none of our patients were operated on using mesh. However, a review of the videos showed that tension of the repaired diaphragm was not a sign of recurrence in any of our study participants. In addition, rib-anchoring sutures, which were also evaluated as a sign of a large defect (and therefore tension), were also not associated with recurrence.

A surgeon's experience could be a determining factor in the surgical outcomes, but after reviewing our results, we found that it was not a viable factor that could be used to explain the high recurrence rate in the thoracoscopic repair of a CDH. A surgeon who is experienced in minimally invasive surgery is capable of performing the procedure by paying strict attention to the selection of the proper port in spite of the limited working space.

There were no conversions from a thoracoscopy to open surgery in our study group. However after being confronted with so many recurrences, we observed a large defect in before performing our last thoracoscopy for a CDH, and the patient then underwent a laparotomy without any attempt for intervention via a thoracoscopy. In this particular patient, we observed a remarkable difference in how the diaphragmatic rim looked from both sides (Figures 1a and b). Furthermore, the lower rate of recurrence in the laparoscopic group compared with the thoracoscopic group in the Tsao et al.^[5] study

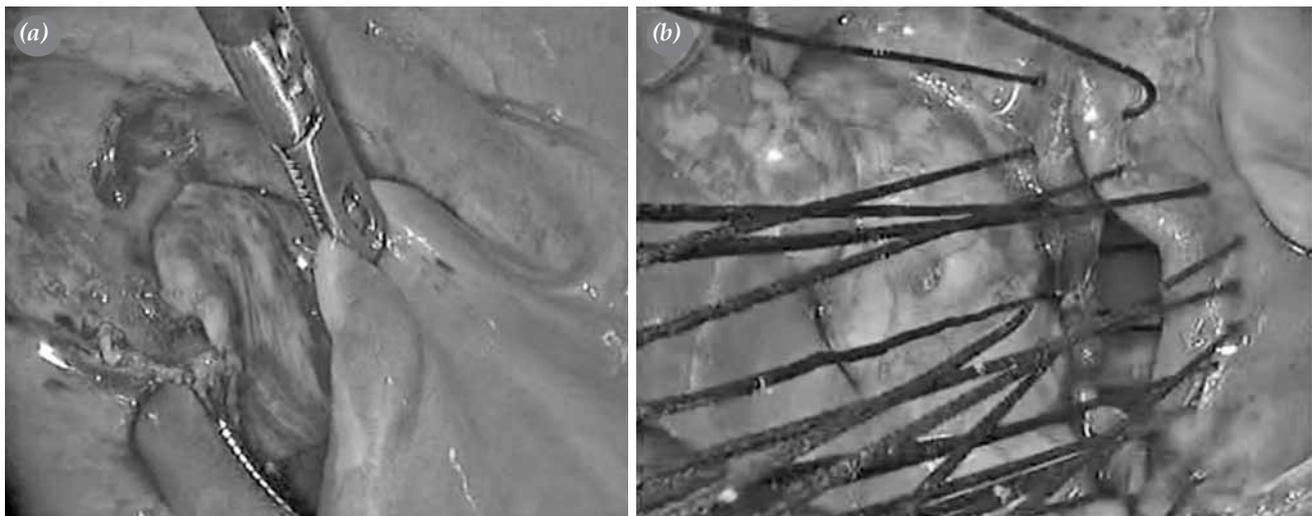


Figure 1. (a) Thoracoscopic view of the diaphragmatic defect. Note the absence of the posterior rim. (b) The same diaphragmatic defect from the abdominal point of view.

also favors this finding. Although not a point of emphasis in our study, there seems to be a smaller difference in the recurrence rate between patients who undergo open surgery and those for whom a laparoscopy is performed. We also admit that there is a technical limitation in performing a thoracoscopy with respect to the mobilization of the rim and we acknowledge that performing the operation from the thorax presents an additional challenge.

Our study has limitations. It is a retrospective study analyzing relatively small series. However, the follow-up time is relatively long. Although we were also not able to provide scientific data, we would also like to draw attention to the remarkable differences in the exposure of the diaphragmatic rims from the thoracic and abdominal points of view with the hope that further studies can clarify this issue.

Conclusion

Thoracoscopic repair offers a remarkable functional and cosmetic outcome; however, current results reveal a higher risk of recurrence. Further studies should be carried out before accepting thoracoscopic repair as the routine approach for Bochdalek-type CDHs.

Acknowledgements

The authors would like to thank Rasheedah Mullings Dağkiran (Sıtkı Koçman Yabancı Diller Yüksek Okulu, Muğla) for improving the literary quality of the paper.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

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