

Open Transthoracic Plication of the Diaphragm for Unilateral Diaphragmatic Eventration in Infants and Children

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Background: To evaluate our experience of early surgical plication for diaphragmatic eventration (DE) in infancy and childhood. **Methods:** This study evaluated infants and children with symptomatic DE who underwent plication through an open transthoracic approach in our childhood development department between January 2005 and December 2012. Surgical plication was performed in several rows using polypropylene U-stitches with Teflon pledgets. **Results:** The study included 12 infants and children (7 boys and 5 girls) with symptomatic DE (9 congenital and 3 acquired). Reported symptoms included respiratory distress (91.7%), wheezing (75%), cough (66.7%), and recurrent pneumonia (50%). Preoperative mechanical ventilatory support was required in 41.7% of the patients. The mean length of hospital stay was 6.3 ± 2.5 days. The mean follow-up period was 24.3 ± 14.5 months. Preoperative symptoms were immediately relieved after surgery in 83.3% of patients and persisted in 16.7% of patients one year after surgery. All patients survived to the end of the two-year follow-up and none had recurrence of DE. **Conclusion:** Early diagnosis and surgical plication of the diaphragm for symptomatic congenital or acquired diaphragmatic eventration offers a good clinical outcome with no recurrence.

Key words: 1. Diaphragm
2. Eventration
3. Plication
4. Thoracotomy

INTRODUCTION

Diaphragmatic eventration (DE) is a rare entity characterized by the relaxation and elevation of an aplastic, atrophic, inactive, and paralyzed diaphragm following phrenic nerve injury during or after birth [1]. It may originate from a congenital defect or may be acquired. Congenital eventration is a developmental abnormality characterized by muscular aplasia of the diaphragm [2].

DE usually remains asymptomatic in early life and presents later with respiratory and occasionally gastrointestinal complications [3]. Symptomatic DE is an uncommon condition and

is sometimes impossible to distinguish clinically from paralysis [4].

Early diagnosis is necessary and plication of the diaphragm (PD) is the treatment of choice in cases of progressive dyspnea on exertion and recurrent respiratory tract infection [5]. PD is intended to decrease lung compression, stabilize the thoracic base and mediastinum, and to strengthen the respiratory action of intercostal, perithoracic, and abdominal muscles [6]. The aim of this study was to evaluate our experience and short-term outcomes of plication for diaphragmatic eventration (acquired or congenital) in infants and children.

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Table 1. Preoperative characteristics of infants and children who underwent plication of the diaphragm for diaphragmatic eventration

Characteristic	Patients (n=12)
Gender (male/female)	7/5
Age (yr)	2.3±1.3
Etiology	
Congenital	9 (75.0)
Acquired	3 (25.0)
Idiopathic	0
Symptoms	
Respiratory distress	11 (91.7)
Wheezing	9 (75.0)
Cough	8 (66.7)
Recurrent pneumonia	6 (50.0)
Chest pain	0
Dyspepsia	0
Ventilatory support	5 (41.7)

Values are presented as mean±standard deviation or number (%).

METHODS

1) Patients

The study included infants and children with symptomatic DE who underwent diaphragmatic plication at El-Minia University Hospital from January 2005 to December 2012. The diagnosis of DE was established radiologically, specifically by plain chest radiography followed by ultrasonography and chest computed tomography (CT).

2) Operative technique

All patients were operated upon under general anaesthesia, with single-lumen intubation by the same surgeon. Open transthoracic plication was performed via posterolateral thoracotomy through the 7th or 8th intercostal space. The diaphragmatic plication was performed in several rows using polypropylene U-stitches with Teflon pledgets, starting at the posterior-most portion of the phrenic center and running in a radial fashion until the diaphragm became taught and firm. A single intercostal drain was left in place and the thoracotomy was closed in layers.

After transfer to the intensive care unit, patients were kept warm and given intravenous fluids. Vital signs were monitored closely with regular analyses of blood gases. Ventilatory support was continued with the aim of maintaining the

Table 2. Operative and postoperative outcomes of infants and children who underwent plication of the diaphragm for diaphragmatic eventration

Outcome	Patients (n=12)
Side of plication	
Left	9 (75.0)
Right	3 (25.0)
Preoperative symptoms	
Relieved	10 (83.3)
Persisted	2 (16.7)
Duration of chest tube drainage (day)	3.0±2.1
Hospital stay (day)	6.3±2.5
Follow-up duration (mo)	24.3±14.5
Position of the diaphragm	
Normal	12 (100.0)
Elevated	0

Values are presented as number (%).

partial pressure of oxygen (pO₂) between 80 to 100 mmHg.

3) Data collection

Collected data were retrieved from hospital medical records and follow-up cards including preoperative characteristics, operative notes and postoperative course.

4) Statistical analysis

Statistical analysis was performed using SPSS for Windows ver. 16.0 (SPSS Inc., Chicago, IL, USA). Continuous variables were expressed as mean±standard deviation, and categorical variables were expressed as proportions.

RESULTS

During the study period, 12 infants and children were operated on by the same surgeon through an open trans-thoracic approach. Regarding the preoperative characteristics (Table 1), there were 7 boys and 5 girls, the mean age was 2.3±1.3 years (range, 7 to 9 years). The etiology of the DE was congenital in 9 patients (75%) and acquired in 3 patients (25%). All patients had symptoms related to the disease: respiratory distress in 11 (91.7%), wheezing in 9 (75%), cough in 8 (66.7%), and recurrent pneumonia in 6 (50%). Five patients (41.7%) required preoperative mechanical ventilatory support.

Regarding operative and postoperative outcomes after plica-

tion of the diaphragm (Table 2), the plication was done for left-sided eventration in 9 patients (75%) and right-sided in 3 patients (25%). There were no operative or postoperative deaths related to surgery. Preoperative symptoms were immediately relieved in 10 patients (83.3%) following surgery, while preoperative symptoms persisted in 2 patients (16.7%) one year after surgery. The mean duration of chest tube drainage was 3.0 ± 2.1 days. The mean length of hospital stay was 6.3 ± 2.5 days. The mean follow-up period was 24.3 ± 14.5 months. Postoperative and follow-up chest X-ray showed flattening of the diaphragm in all patients. None of the patients had recurrence of eventration during the follow-up period.

DISCUSSION

Eventration of the diaphragm is defined as an abnormal and permanent elevation of the hemidiaphragm without defects. It may be congenital (muscular hypoplasia) or acquired (injury of the phrenic nerve) [7]. The etiologic investigations of DE in the present study were exhaustive including clinical history, plain chest radiography, ultrasonography and chest CT. The sensitivity of thoracic and abdominal CT in the diagnosis of DE has been highlighted in the literature, as CT scans should be performed in every patient to exclude both neoplastic disease (at the cervical, mediastinal, or pulmonary level) involving the phrenic nerve and subdiaphragmatic abnormalities (abscess, Chilaiditi syndrome) [8].

In the present study, the most common etiology of DE was acquired in adults and congenital in paediatric patients. Congenital eventration is the result of incomplete development of the muscular portion of the diaphragm. The cause of this failure is not known, but cytomegalovirus infection [9], fetal rubella [10], and heterozygous genetic mutation with Marfan syndrome [11] have been reported to be associated with congenital DE. In adults with the acquired form, phrenic nerve palsy from compression or iatrogenesis is the most common cause of DE [1].

Multiple surgical techniques have been proposed to treat diaphragmatic eventration, including excision and suture, diaphragmatic plication, and prosthesis [12,13]. Diaphragmatic plication through standard thoracotomy is the most frequently employed technique. Diaphragmatic plication can be done in

a straightforward manner with a very low complication rate when carefully performed [14]. The aim of plication is to reduce dysfunctional caudal excursion of the diaphragm during inspiration by plication [15].

In the present study, plication of the diaphragm early in infant and childhood was a safe, simple, and suitable treatment for eventration with good results through a mean follow-up of 24.3 ± 14.5 months. It reduced most preoperative symptoms immediately after surgery and improved deteriorated preoperative pulmonary function. Similarly to our findings, other studies have demonstrated significant improvement in symptoms of diaphragmatic eventration or paralysis after plication [5,16-19].

A study by Yazici et al. [5] on 33 infants and children concluded that DE is an important condition associated with significant mortality, thus early diagnosis is necessary and plication is the treatment of choice. The results of the clinical study by Obara et al. [16] on 18 infants and children, ranging in age from 10 days to 6 years, suggest that in order to reduce pathological changes in the lung, early surgical plication should be performed even in patients with acquired DE if respiratory and digestive symptoms are noted. Furthermore, the study by Tsugawa et al. [18] suggests that symptomatic patients who have diaphragmatic eventration should be operated on immediately with an expected dramatic resolution of their respiratory problems.

The report of Jawad et al. [17] with 8 pediatric patients treated for DE included 5 with congenital and 3 with acquired eventration. All patients except one required surgical plication of the diaphragm. Three patients developed minor postoperative complications and all patients were alive and well on follow-up. In a recent study by Wu et al. [19], diaphragm plication surgery led to good results among 177 children (boys, 128; girls, 49; mean age, 10.28 ± 2.35 months) with congenital DE with no recurrence, and those authors concluded that timely accurate diagnosis and treatment of symptomatic congenital DE could effectively resolve respiratory morbidity and reduce complications. Therefore, it appears that diaphragmatic plication is a safe and well-tolerated procedure for symptomatic diaphragmatic eventration.

The transthoracic approach used in the present study was mandatory for optimal access. We aim to avoid injury of the

intra-abdominal organs by using a shallow scoop of the diaphragm during placement of plication sutures. Our findings agree with those of other investigators and suggest that plication of the diaphragm can eliminate the serious complications that result from both prolonged ventilatory support and malnutrition [18].

In conclusion, diaphragmatic plication in infants and children was performed safely and simply in our clinical setting to restore normal pulmonary parenchymal volume. There was immediate relief from the symptoms of DE. During the average follow-up period of more than two years, the location of the diaphragm was normal in most patients, with no paradoxical movement.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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