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Diaphragmatic eventration: long-term follow-up and results of open-chest plication[☆]

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Abstract

Objective: Diaphragmatic eventration is a relatively uncommon entity with a simple surgical correction technique – plication of the diaphragm. This study aims to assess the clinical and ventilatory impact of this technique. **Materials:** From April 1988 to February 2007, we operated on 20 patients (12 men) with diaphragmatic eventration using the postero-lateral approach and correction by radial plication. The mean age of the patients studied was 56.3 ± 15.6 years (range: 13–74 years). A traumatic cause was identified in 13 patients; one patient had a congenital cause and the remainder were of idiopathic origin. Chronic obstructive pulmonary disease and arterial hypertension were present in one-half of the study group, while diabetes mellitus was present in three patients. Dyspnoea was the most common complaint in 75% of the patients, and thoracic pain was present in 25%. The mean forced expiratory volume in 1 s (FEV₁) and vital capacity (VC) were $66.2 \pm 15.3\%$ and $70.4 \pm 16\%$ of the predicted values, respectively. **Results:** There was no operative mortality. Apart from a patient with moderate/severe pain and another who had pneumonia, there were no other important perioperative complications. Average drainage time was 3.3 ± 1.6 days (range: 2–7 days). Hospitalisation time was 6.2 ± 1.6 days (5–10 days). Follow-up was complete, for a mean of 59.6 ± 55.1 months (4–206 months). There were three late deaths (one sudden, one stroke and one trauma). Eight of the 17 survivors (47%) are asymptomatic. According to the MRC/ATS grading system, the dyspnoea score was 2.06 ± 0.97 preoperatively and 1.06 ± 1.14 postoperatively ($p = 0.007$). At follow-up, the FEV₁ was $76.1 \pm 20.1\%$ and the VC was $78.4 \pm 17.3\%$ ($p > 0.1$). Two patients had chronic pain. **Conclusion:** Plication of the diaphragm is a safe and efficient procedure. Most patients experienced significant clinical improvement with enhancement of the FEV₁ and VC. Chronic surgical pain still remains a potential problem with the classical approach.

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1. Introduction

Diaphragmatic eventration is defined as a permanent elevation of a hemidiaphragm without defects of continuity. The muscular insertions are normal, the normal orifices are sealed and there is no interruption of the pleural or peritoneal layers [1]. The eventration may be of congenital origin, with a large unilateral elevation of the diaphragm because of secondary hypoplasia of the homolateral lung, usually presenting with severe cardiorespiratory symptoms in the newborn.

Eventration that occurs in older children and in adults is thought to be caused by paralysis of a diaphragmatic leaf, normally accompanied by dyspnoea. Gastrointestinal symptoms may be present due to dislocation of abdominal viscera [2]. Although it is conceivable that long-lasting diaphragm paralysis leads to cor pulmonale, there is no evidence

substantiating this assumption [3]. Malignancy, trauma, infection, cervical spondylosis and iatrogeny (surgery on the mediastinum/cardiac) and neuromuscular disorders are the most common causes of this condition. There are a number of patients in whom a cause cannot be explained (idiopathic eventration). The exact incidence of this pathology is not known, and can most probably be attributed to under-recognition.

The first report of a surgical repair was published by Morrison in 1923 [4]. Since then, several series were published describing different techniques and results of diaphragmatic plication. With this technique, the muscle is fixed in the position of maximum inspiration, decreasing the ventilatory work. This is particularly important in patients with chronic obstructive lung disease, for it creates a larger space to these hyper-expanded lungs [2].

However, surgical treatment is infrequently performed for unilateral diaphragmatic paralysis in adults. Barriers to wider use of plication of the diaphragm include lack of attribution of symptoms to unilateral diaphragmatic paralysis, uncertainty about the potential benefits of plication and the perceived need for thoracotomy [5]. In

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this work, we describe our experience with 20 patients who had unilateral non-malignant diaphragmatic eventration, focussing the analysis on the symptomatic status, especially with regards to the degree of dyspnoea, and on the pulmonary function during a long-term follow-up.

2. Patients and methods

2.1. Population

Between April 1988 and February 2007, 20 consecutive patients, 12 (60%) males, with a mean age of 56.3 ± 15.6 years (range: 74–13 years), underwent surgical treatment for diaphragmatic eventration. Since most patients had the eventration for at least 1 year, the chance of spontaneous recovery was small, which, in combination with the symptoms, was the indication for surgical treatment.

For inclusion in this study, the patients were identified retrospectively by consulting the surgical reports. The following data had been registered prospectively in the clinical records: age, sex, clinical history, aetiology, symptoms, side (location) of disease, values of spirometry (vital capacity and 1-s forced expiratory volume) in the sitting position, operative findings and procedures, duration of chest drainage, peri- and postoperative complications and duration of hospital stay. Previous roentgenograms were reviewed, if available. Thoracic and upper abdominal computed tomographic (CT) scans were always done. Several patients also had magnetic resonance imaging (MRI) ordered by the referring physician prior to hospitalisation.

2.1.1. Aetiology and associated pathology

All patients with malignant diaphragmatic paralyses/eventrations were excluded from the study. After a careful analysis of the anamnesis and physical examination of all patients, trauma was identified as the only objective cause of diaphragmatic eventration in 13 patients (65%). A 13-year-old patient had an eventration that was clearly of congenital origin. The remaining six were classified as idiopathic (Table 1). The aetiological assessment was performed by the assistant clinician and presented for decision by the pneumologist and the thoracic surgeon.

Obstructive chronic pulmonary disease and arterial hypertension were identified in half of the study group (10 patients). Diabetes mellitus was diagnosed in three patients and one patient had coronary disease. Five patients (25%) had no associated pathology.

2.1.2. Pulmonary function tests

Vital capacity (VC) and 1-s forced expiratory volume (FEV_1) were measured in all patients in the sitting position, preoperatively and at follow-up, according to the guidelines of the European Respiratory Society (ERS), and are expressed as a percentage of the predicted values.

2.1.3. Dyspnoea index

Assessment of dyspnoea was done using the Medical Research Council/American Thoracic Society (MRC/ATS) grading system [6], which correlates the patient's functional

Table 1
Patient data.

Patient	Sex	Age	Aetiology	Side of eventration	Year of operation
1	M	40	Traumatic	Left	1997
2	M	74	Unknown	Left	2001
3	M	52	Traumatic	Left	2002
4	M	68	Unknown	Left	2003
5	F	65	Traumatic	Right	2004
6	F	67	Traumatic	Right	2004
7	M	60	Unknown	Right	2004
8	F	62	Unknown	Left	2005
9	M	50	Traumatic	Right	2005
10	M	51	Traumatic	Right	2006
11	F	28	Traumatic	Right	2006
12	F	56	Traumatic	Left	2006
13	F	73	Traumatic	Left	2006
14	F	68	Unknown	Left	1988
15	M	66	Traumatic	Right	1988
16	F	65	Unknown	Right	1996
17	M	56	Traumatic	Right	1990
18	M	43	Traumatic	Left	2007
19	M	69	Traumatic	Right	2003
20	M	13	Congenital	Right	1999

Table 2
MRC/ATS system of grading dyspnoea^a.

Grade	Label	Description
Grade 0	NONE	No trouble with breathing except with strenuous exercise
Grade 1	MILD	Shortness of breath when hurrying on level or walking up a slight hill
Grade 2	MODERATE	Walk slower than people of same age on the level or has to stop for breath walking at own pace on the level
Grade 3	SEVERE	Stop for breath when walking up to 100 m or after a few minutes on the level
Grade 4	VERY SEVERE	Too breathless to leave the house or breathless when dressing or undressing

^a Adapted from Mahler et al. [6].

impairment with the magnitude of task needed to evoke dyspnoea (Table 2).

2.2. Surgical procedure

Surgery was performed under general anaesthesia, with single-lumen intubation and limited postero-lateral thoracotomy through the seventh or eighth intercostal space (Fig. 1). The thoracic cavity and its structures (lung, mediastinum and phrenic nerve) were carefully examined to rule out any unsuspected pathology. The diaphragm was then plicated in several rows (normally four to five), until it became taught and firm, using polypropylene U-stitches buttressed with Teflon pledgets to prevent cutting through the tissue. Usually, the first row starts at the posterior-most portion of the phrenic centre and runs in a radial fashion. Then, other suture layers are used to complete the shortening as required (Fig. 2).

Redundant tissue was flattened both anterior and posterior to the plicature by using running sutures. Extreme care was exerted to avoid injuring abdominal viscera with the sutures. A single intercostal drain was left in place and the

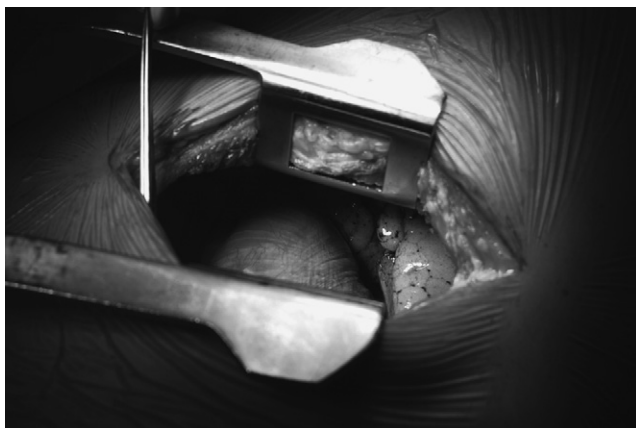


Fig. 1. The 6–7 cm muscle-sparing thoracotomy.



Fig. 2. First rows of sutures.

thoracotomy was closed in layers. Pain control was achieved with dorsal/lumbar epidural analgesia for 24–48 h or by intercostal blockade using 7.5% ropivacaine. All patients were extubated in the operating theatre immediately following completion of the procedure.

2.3. Follow-up

Patients were contacted telephonically and invited to come for a consultation at our institution where a careful evaluation of the dyspnoea score was done, chronic pain or its equivalent assessed and a spirometry was performed. As some patients were unable to attend this consultation, the interview was conducted telephonically and the spirometry solicited from the patient's assistant physician. All patients were accounted for.

2.4. Statistical analysis

Continuous data are expressed as mean \pm standard deviation. Values of VC and FEV₁ are expressed as a percentage predicted for age and gender and body surface. Follow-up data were compared with preoperative values using the Wilcoxon signed-rank test.

3. Results

3.1. In-hospital mortality and morbidity

There were no deaths in-hospital or within 30 days following the procedure. The mean hospital stay was of 6.5 ± 1.3 days (range: 5–10 days), with a median of 6 days, and drainage time was 3.1 ± 1.4 days (range: 2–7 days). The patient with a longer drainage time had an emphysematous lung and a prolonged air-leak. Most of the patients had the drain for 2 or 3 days.

One patient had a nosocomial lobar pneumonia requiring prolonged oxygen therapy. He was successfully treated with antibiotics and was free from respiratory symptoms at the time of discharge. With the exception of some nausea related to medication, there were no gastrointestinal complications. Almost all patients experienced a feeling of tightness/slight pain in the lower chest/upper abdominal area following the procedure, which subsided with analgesics.

3.2. Follow-up

The follow-up was complete for an average length of 59.6 ± 55.1 months (range: 4–206 months). Nineteen patients had a follow-up period greater than 12 months. There were three late deaths (one sudden, one due to stroke and one traumatic); eight of the 17 survivors (47%) are asymptomatic. There are two patients with chronic pain and another one with paraesthesia in the scar area. The remainder have different degrees of dyspnoea, as discussed below.

Two patients presented, 75 and 117 months after surgery, with recurrence of the eventration on the follow-up chest X-ray. The first patient requires nightly oxygen therapy, is on treatment for chronic pain and has dyspnoea grade 3. He refused to undergo surgery again. The second patient had the same dyspnoea score as before surgery plus the feeling of fullness after meals. He was identified at follow-up and has since been re-operated, and since then he is asymptomatic.

3.3. Dyspnoea score

According to the MRC/ATS grading system (Table 2), one patient had no dyspnoea before surgery. Seven patients had dyspnoea grade 3, and seven grade 2. The remaining five patients had shortness of breath when hurrying on level or walking up a slight hill. At long-term follow-up, grade of dyspnoea improved 1 point in 10 patients, 2 points in two, 3 points in two and remained unchanged in another two. It worsened 3 points in one patient. The mean dyspnoea score was 2.06 ± 0.97 preoperatively and 1.06 ± 1.14 postoperatively ($p = 0.007$; Table 3).

3.4. Spirometry

The results of spirometry are summarised in Tables 3 and 4. Although the VC and FEV₁ mean values were higher after surgery, the non-parametric Wilcoxon signed-rank test showed a non-significant result ($p > 0.05$), probably because of the small size of the sample.

Table 3
Measured respiratory values and grade of dyspnoea.

Patient	VC pre	VC post	FEV ₁ pre	FEV ₁ post	Dyspnoea pre	Dyspnoea post
1	90	89	87	80	0	3
2	53	72	57	41	2	1
3	94	100	92	111	2	2
4	71	64	73	65	3	0
5	93	73	80	78	3	2
6	64	95	67	88	1	0
7	42	71	71	68	1	0
8	64	62	57	55	3	2
9	77	64	73	69	3	2
10	54	56	62	59	2	0
11	62	60	53	55	1	0
12	57	84	53	80	2	1
13	83	102	100	108	3	2
14	55	^a	49	^a	1	^a
15	65	^a	56	^a	2	^a
16	71	^a	57	^a	2	^a
17	52	82	45	68	1	0
18	88	65	50	89	2	0
19	80	77	78	70	3	3
20	93	117	63	110	3	0

VC: vital capacity; FEV₁: forced expiratory volume in 1 s; pre: preoperative; and post: postoperative.

^a Late death.

Table 4
VC and FEV₁ values, expressed as mean ± standard deviation of predicted.

	VC	p value	FEV ₁	p value
Preoperative	70.4 ± 16.0		66.2 ± 15.3	
Postoperative	78.4 ± 17.3	0.21	76.1 ± 20.1	0.31

VC: vital capacity; and FEV₁: forced expiratory volume in 1 s.

4. Discussion

Atrophy, thinned wall and progressive distension of the diaphragm represent the main characteristics of diaphragmatic eventration. Newborns usually present with acute respiratory failure, but in adulthood the most common symptoms are dyspnoea and thoracic pain. Simansky et al., in 2002, reported that the indication for surgery for all children was failure to wean from ventilatory support, while the indications for surgery in the adult group were ventilator dependency ($n = 4$) and symptomatic dyspnoea ($n = 7$) [7]. Earlier, Graham et al. also indicated shortness of breath as the main symptomatic feature of diaphragmatic paralysis [8].

Although diaphragmatic eventration and paralysis are two different entities, the symptomatic consequences and the therapeutic procedures are very similar, and the two groups of patients are quite analogous. DeVries et al. clearly state that phrenic nerve injury – inducing chronic diaphragmatic paralysis – leads to eventration [9]. A paralysis of the hemidiaphragm can produce significant alterations in the respiratory physiology. These include atelectasis and ventilation–perfusion mismatch, worsening of pulmonary spirometry, preferential redistribution of pulmonary blood flow to the contralateral lung, shift of the mobile mediastinum to the contralateral side and paradoxical

movement of the affected hemidiaphragm [5]. For practical purposes, in most situations, the differentiation of these entities is of limited value. The pathophysiology of these entities is equivalent, hence comparison between both groups of patients with regards to symptomatology, therapy and outcomes is possible.

Diaphragmatic plication is a standard, well-described technique to treat diaphragmatic eventration [2] but there have been only a limited number of reports published, most with small series and short follow-ups. Higgs et al. described a group of 19 patients with long-term follow-up in 15 [10] and Versteegh et al. reported on 22 patients with a follow-up in 17 [3]. Both clearly proved that the main and probably most important result is the significant and long-lasting improvement in the subjective symptomatology. To our knowledge, these are the largest series published with long follow-ups. In this work, we describe the long-term follow-up of 20 patients, with a maximum of up to 17 years. Most of our patients had an important impairment of their daily activities preoperatively, and all except three improved their physical status. One patient referred worsening of the COPD, one had recurrence of the eventration (probable technical failure) and the third complained of persistent disabling pain.

The reports by Higgs et al. [10] and Versteegh et al. [3] demonstrated statistically significant differences between preoperative and postoperative values of VC and FEV₁. Our patients achieved similar improvements, although we could not demonstrate a statistical significance, probably attributable to the small number of patients analysed. Also, with such a severe symptomatic disease, one should expect lower values than predicted, preoperatively, hence with a larger difference to postoperative values. In any case, our values are within the range reported by most other surgical groups (65–70% of predicted, preoperatively, and 70–79% of predicted, postoperatively).

Recently, diaphragmatic plication has also been performed through minimally invasive techniques, either laparoscopic [11] or thoracoscopic [5,12–14]. Most of the published papers on the use of this technique concern case reports or small series, except one that was published by Freeman et al. [5] which reports on 22 patients. Our series dates from 20 years ago when thoracoscopic techniques were not yet widely used, hence we describe the classical approach in this article. We strongly believe that the diaphragm should be rendered as taut as possible, which may not be possible with a video-assisted technique because of the lack of full tactile feedback. The risk of damage to the abdominal viscera through such a thin diaphragm is present and is probably greater with a less invasive approach. To inspect the entire raised diaphragmatic cupula may be quite difficult. In any case, the incidence and intensity of post-thoracotomy pain does not seem to be very different [15]. Obviously, there are no head-to-head comparisons to date [3], and the answer to this dilemma will only come with further study.

In conclusion, this work demonstrates that adult patients with chronic dyspnoea due to unilateral, non-malignant eventration significantly benefit from diaphragmatic plication. We showed that there is a significant and long-lasting clinical improvement in this group of patients, with return to normal life after the surgical correction. Subjective assess-

ment in order to establish the surgical indication and to ascertain the clinical results of the procedure has been of much more value than the objective data from the spirometric studies.

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Appendix A. Conference discussion

Dr S. Elia (Rome, Italy): You said you had one sudden death. What was that due to?

Dr Calvino: No, I can't say it was sudden death. It was during the follow-up, almost 6 years after the surgery. He was at home and he just didn't wake up.

Dr Elia: So it was long after the procedure.

Dr Calvino: Yes, 6 years after the procedure.

Dr Elia: How did you treat the three patients who didn't do well? Did you reoperate on them?

Dr Calvino: No. One had worsening COPD, so he was not a surgical patient. In the other one, we had to replace the diaphragm with a Marlex net because the diaphragm just tore off by the knots. It was probably a technical failure. The last patient is being followed in pain clinic at our institution and is doing better.