ORIGINAL ARTICLE

EVENTRATION OF DIAPHRAGM IN ADULTS: ELEVEN YEARS EXPERIENCE

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Background: Eventration of diaphragm is a congenital condition in which there is absence of muscle fibers in the diaphragm while maintaining all the anatomical attachments normally. Surgical treatment is warranted in symptomatic patients so as to reduce the abnormal ascent of diaphragm. The present study was conducted to analyse the perioperative outcome of thoracotomy in adult patients with diaphragmatic eventration. **Methods:** This descriptive case series was carried out in Cardiothoracic Surgery-Unit, Postgraduate Medical Institute, Lady Reading Hospital, Peshawar, Pakistan. Medical records of patients operated upon for eventration from June 2002 to June 2013 were reviewed. Patients of either gender, above 16 years who were operated for symptomatic eventrtaion were included in study. All the demographic data, presenting complaints, baseline and post-postoperative dyspnea grade, forced expiratory volume at 1 second (FEV1) and forced vital capacity (FVC) were recorded on predesigned pro forma and analysed using SPSS-16. **Results:** A total of 38 adult patients underwent surgery for diaphragmatic eventration over the past 11 years in our unit out of which 29 (76%) were males and rest of 9 (24%) were females. Mean age of patients was 41.6±13.84 years. In 31 (81.5%) patients left side was involved. Majority of patients had a dyspnea grade-3 on presentation. Preoperative dyspnoea score (MRC), FEV1 and FVC values were 2.6±0.73, 63.5±13.3 and 67.2±14.6 respectively. Pre-operative and 6-months follow-up values of dyspnoea grade, FEV1 and FVC values showed statistically significant improvement. **Conclusion:** Our study showed that adult patients with symptomatic unilateral eventration of diaphragm significantly benefit from diaphragmmatic plication.

Keywords: Eventration, diaphragm, thoracotomy, Dyspnea score, FEV1, FVC

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INTRODUCTION

Elevation of diaphragm per se is usually by chance finding during chest x-ray (CXR) done for other purposes and patients are usually asymptomatic.[1–3] The cause of this elevation may be diaphragmatic paralysis or diaphragmatic eventration.[4] Eventration of diaphragm is a congenital condition with paucity or absence of muscle fibres in the diaphragm while maintaining all the anatomical attachments normally.[2,5]

Histologically the specimen of diaphragm shows fibro-elastic changes replacing the muscle tissue layer, sandwiched between pleura and peritoneum while still maintaining the 3-layered structure of diaphragm[6] which differentiates it from congenital diaphragmatic hernia and diaphragmatic paralysis.

Eventration was first identified by Jean Louis Petit in 1774 during autopsy studies.[7] Word eventration was first used by Beclard in 1829.[8] Morrison reported the first successful repair of eventration in adults in 1923.[9] Eventration is a rare condition with an estimated incidence of <0.05%[2,10], symptoms estimated to occur only in 25% patients.[11] Most common symptoms are dyspnoea and orthopnoea[1,2,4,12] objectively proved by restricted spirometry values.[1,2,12,13] Orthopnoea is often related with dyspnoea. It is sudden, occurring soon after lying supine secondary to ascent of abdominal viscera.[1,2,4]

Some patients' exhibit nonspecific gastrointestinal symptoms including epigastric discomfort, belching etc.[14] Rarely life-threatening complications are also reported like stomach volvulus[15], rupture of eventrated diaphragm[16] with trivial trauma and acute progressive respiratory distress.[17]

Asymptomatic patients do not need treatment.[2] Treatment in symptomatic patients is surgery, the rationale being to reduce the abnormal ascent of diaphragm during respiration and lying supine.[1,14] Different techniques include hand-sewn plication[18–21], double breasting[1,14], with or without mesh application[1,22] using through thoracotomy[18-21], thoracoscopy[2,22–26], laprotomy[27] or laproscopy.[13,28,29]

The aim of the present study was to analyse the clinical presentation of patients with symptomatic eventration, its effect on ventilation and lifestyle, and

peri-operative outcome of surgery in adult patients with symptomatic diaphragmatic eventration.

MATERIAL AND METHODS

This descriptive case series was carried out in Cardiothoracic Surgery Unit, Postgraduate Medical Institute, Lady Reading Hospital, Peshawar, Pakistan. Computerized medical records of patients operated upon for eventration from June 2002 to June 2013, were reviewed. All patients of age 16 years and above, of either gender, with the diagnosis of unilateral eventration of diaphragm causing symptoms for at least one year, were included in the study. Patients with history of trauma, tumour, parenchymal lung disease, heart failure, myopathy and neuropathy were excluded from the study. Obese (BMI >30) and symptomatic patients with symptoms of less than one year duration were also excluded from the study.

Demographic data, clinical presentation, dyspnoea grading, laterality (left or right), CXR findings, cervicothoracic and upper abdominal CT-scan with intravenous contrast, spirometry values (FVC, forced expiratory volume at first second (FEV1)), surgical procedure and outcome were noted on a predesigned *pro forma*. Supine spirometry values, ultrasound and fluoroscopy reports were also noted where available. Follow-up data at 6-months was noted including dyspnoea grade, spirometry values and complications; and compared with preoperative values.

Detailed history and thorough clinical examination were performed in all the subjects. Surgery was performed after taking informed consent from the patients. Pre-operatively Nasogastric tube was put in all patients to empty stomach. All the patients were operated upon by standard posterolateral thoracotomy through 7^{th} or 8^{th} intercostal space under general anaesthesia (GA) with single lumen endotracheal tube. Lung, mediastinum and phrenic nerve were examined for any pathology. Plication was done in continuous running sutures using polypropylene until diaphragm became flat and taut in full inspiratory position. Diaphragm was opened by a small incision in some patients where chance of abdominal viscera injury could not be excluded safely.

Dyspnoea score was recorded according to medical research council (MRC) dyspnoea score[30] while FEV1 and FVC values were recorded as percent of predicted values.

Mortality was defined as any death within first 30 post-operative days. Morbidity was defined as wound problems, pleural space problems, failure or recurrence of procedure, pulmonary, cardiac and abdominal complications.

All the data was analysed using SPSS-16. Continuous data were presented as mean±standard deviation (SD) while categorical data was presented as frequencies and percentages. Follow-up data were compared with preoperative values. Study data was summarized using descriptive statistics (number, mean, range, and standard deviation). All tests were two-sided and statistical significance was set at $p<0.05$.

RESULTS

A total of 38 adult patients underwent surgery for diaphragmatic eventration over the past 11 years in our unit out of which 29 (76%) were males and rest of 9 (24%) were females. Mean age of patients was 41.6±13.84 years. In 31 (81.5%) patients left side was involved. Majority of patients had a dyspnea grade-3 on presentation. Patients' demographics and baseline clinical characteristics are given in table-1.

The most common clinical presentation was dyspnoea with mean duration of dyspnoea being 37.3 months (range 21–57 months). Preoperative dyspnoea score (MRC), Forced expiratory volume at one second (FEV1) and Forced vital capacity (FVC) values were 2.6±0.73, 63.5±13.3 and 67.2±14.6 respectively. In 31 (81.6%) patients the symptoms interfered with the routine daily activities while 26 (68.4%) patients had compromised work capacity and had compromised their jobs.

In all patients standard continuous layer plication was performed with usually requiring at least 6 rows (range 4–8 rows). Diaphragm was opened by a 5cm incision in 5 patients to ensure safe plication and prevent abdominal viscera injury.

Mean operative time was 72±13.6 minutes. There was 5.2% morbidity including one superficial surgical site infection (SSI) and surgical emphysema in one patient which settled with conservative treatment. One patient died in our study on second postoperative day (mortality 2.6%) secondary to fatal arrhythmia.

Pre-operative and 6-months follow-up values of dyspnoea grade, FEV1 and FVC values and their comparison are given in Table-2. The difference between the preoperative and follow-up values was statistically significant.

Table-1: Baseline clinical characteristics of patients with eventration (n=38)

Variable	Frequency
Mean Age	41.6±13.84
Male **Female**	29 (76%) 9 (24%)
Laterality (Left sided)	31 (81.5%)
Clinical presentation	
Dyspnoea	
Grade 1	2
Grade 2	12
Grade 3	21
Grade 4	1
Orthopnoea	23
GI symptoms	9
Palpitations	4

Table-2: Comparison of preoperative and follow-up dyspnoea grade and spirometry values (n=38)

Variable	Preoperative value	Postoperative value	*p*-value
Dyspnoea grade[a]	2.6±0.73	0.56±0.47	<0.05
FEV1[b]	63.5±13.3%	75.2±18.1%	<0.05
FVC[c]	67.2±14.6%	78.7±12.8%	<0.05

[a] graded according to MRC scale, [b] Forced expiratory volume at first second, [c] Forced vital capacity

DISCUSSION

Diaphragmatic eventration is an uncommon condition[1,10] which is often found on CXR of asymptomatic patients as raised hemidiaphragm.[1–3,10] Careful evaluation to exclude other causes of raised diaphragm is necessary as eventration of diaphragm is diagnosis of exclusion.[1,4,12] Symptoms, when present, are attributed to eventration only after other possible aetiologies are excluded.[4] Asymptomatic patients with mere elevated diaphragm and no other pathology do not require any treatment apart from follow-up[2] and plication is only performed in symptomatic patients.[1,14] In one study symptoms were present in 25% patients with diaphragmatic eventration.[11]

In present study age distribution (41.6±13.84), male predominance (76%) and laterality (left side 81.5%) are in accord with most previous reports by other authors[1,4,10,12,22] regarding eventration of diaphragm in adult patients. Though some reports show contradictory observations.[2,3]

The main symptom was dyspnoea and orthopnoea in our study, as previously reported.[1,2,12,14] It ranged from uphill task to disabling dyspnoea in one patient who could not lie supine at all. Dyspnoea interfered with routine daily active life of 34 patients. Twenty six patients had compromised work capacity and had compromised their jobs. This fact is also noted by some other authors.[3,12,19,25] Collectively MRC dyspnoea score was 2.6±0.73.

At 6-months follow-up, dyspnoea was improve in most patients (37 patients, 97%) while one patient having grade-1 dyspnoea had the grade unchanged while none of the patient worsened. The dyspnoea score, mean FEV1 and FVC values preoperatively were 2.6±0.73, 63.5±13.3% and 67.2±14.6% and at follow-up were 0.56±0.47, 75.2±18.1% and 78.7±12.8% respectively. The difference was statistically significant. This subjective and objective evidence of preoperative respiratory compromise and postoperative improvement is also noted by many other authors.[2,4,12,13,19–22,25,27] Calvinho *et al* didn't get statistically significant improvement in spirometry values and suggested the reason to be the rarity of disease and small sample size.[3] Most of the patients in our study, who had compromised daily activities, improved and majority returned to their work as well, also observed in some previous reports.[12,19]

Plication was performed by standard posterolateral thoracotomy through 7th or 8th intercostal space in our study using polypropylene continuous running sutures. This approach is used by many authors.[2,18,21] While others prefer abdominal route[27] and some have reported using minimally invasive approaches as well with comparable results.[2,13,25-28] Whatever the approach and technique is used, the basic principle is to make the diaphragm flat and taut in position of full inspiration avoiding abdominal visceral injury.[3,12] In present study diaphragm was opened by a 5cm incision in 5 patients in order to avoid injury to abdominal organs. This technique is routine for some authors[12] while others do not adovocate it.[4] In our view, the diaphragm can usually be picked up easily because it is thin and pliable so opening it is not necessary. But sometimes there is high pressure on diaphragm from abdominal viscera or there are adhesions of viscera to diaphragm. In these situations opening the diaphragm is safe and should be done.

No operative mortality is reported regarding the procedure in adults. In our study one patient (40 years female) died on second post-operative day secondary to fatal arrhythmia resistant to treatment, the cause of which could not be determined. Arrhythmia was also reported by Groth *et al*[13] in one patient postoperatively in his series of 25 patients.

Morbidity in our study was 5.2% (2 patients). There are variable reports of morbidity in literature ranging from 5% to 32% in different series[12,13,22,25] but most authors have reported low morbidity. Some authors have reported the infrequent yet possible complications related to the procedure including splenic injury[31], abdominal compartment syndrome[32] etc.

CONCLUSION

Our study showed that adult patients with symptomatic unilateral eventration of diaphragm significantly benefit from diaphragmatic plication. The procedure carries low morbidity and mortality in the adult population.

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