Successful laparoscopic management of congenital diaphragmatic relaxation: A case report

Giuseppe Di Buono*, Giulia Bonventre, Giuseppe Amato, Federica Ricupati, Giorgio Romano, Antonino Agrusa

Department of Surgical, Oncological and Oral Sciences, Section of General and Urgent Surgery, University of Palermo, Italy

ABSTRACT

INTRODUCTION: Diaphragmatic relaxation is an infrequent condition characterized by a permanent elevation of all or part of an hemidiaphragm which maintains its insertions on the ribs and which does not have continuous solutions but a reduced thickness.

PRESENTATION OF CASE: We studied a 65 years old male patient with mild dyspnea and chest pain occurred during the last 12 months after moderate efforts. He underwent a contrast enhanced CT thorax scan that showed a left diaphragmatic relaxation with the transposition in the cranial sense of the hypochondric abdominal organs associate with an ipsilateral subtotal atelectasia. A laparoscopic plication of the diaphragm was performed to repair the congenital defect.

DISCUSSION: The relaxatio diaphragmatica is probably caused by a congenital defect, but there are also idiopathic causes or cases of acquired relaxation due to phrenic nerve damage because of neof ormations, traumas, thoracic and cardiac surgery. In cases of asymptomatic relaxatio nothing is necessary, but in symptomatic cases it is possible the plication of the diaphragm with a remission of symptoms.

CONCLUSION: The plication can be performed through thoracotomy or laparotomy and recently also in thoracoscopy or laparoscopy. In our experience the laparoscopic repair of the relaxatio was accomplished successfully with a left pneumothorax compatible with the intervention, but the operative strategy should be always individualized with attention on diagnosis, patient characteristics, availability of resources and experience of surgical team.

© 2020 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Diaphragmatic relaxation, also called diaphragmatic elevation, is an anomalous position of the diaphragmatic dome in the chest and represents a rare case of exertional dyspnea. The diaphragm is the most important inspiratory muscle and the other main muscles of inspiration are the scalenes, external intercostals and sternocleidomastoid. The muscles of expiration are the internal intercostals and the muscles of the abdominal wall including the rectus abdominus, internal and external obliques, and transversus abdominus. In addition to its role in the respiratory pump, the diaphragm also serves as a mechanical barrier between the abdominal and thoracic cavities and maintains the gradient pressure between them. When a diaphragmatic relaxation occurs because of congenital, acquired or idiopathic causes, the elevated hemidiaphragm does not participate in respiration resulting in paradoxal movement of the injured hemidiaphragm during inspiration and expiration. In this specific symptomatic cases the surgical treatment needs [1–6]. On the basis of tenacious adhesions found during surgery and the absence of previous trauma or surgery the diaphragmatic relaxation was probably due to a congenital defect. In this case report we evaluated the feasibility and safety of laparoscopic approach in line with the SCARE criteria [7].

2. Presentation of the case

We studied a 65 years old male patient affected by hypertension and abdominal aortic aneurysm with mild dyspnea and chest pain occurred during the last 12 months. He underwent a complete preoperative study [8] with a spirometry that demonstrated reduction in forced vital capacity (FVC) and a contrast enhanced CT thorax scan that showed marked left diaphragmatic relaxation with the transposition in the cranial sense of the hypochondric abdominal organs, in particular of the splenic colic flexure and part of the transverse colon, stomach, spleen and pancreatic tail [9]. As result we found a volumetric reduction of the left pulmonary parenchyma...
with an ipsilateral subtotal atelectasia; the right lung was regular (Fig. 1). In relation to the age of the patient, to his good clinical condition and to the extend of the hypochondriac abdominal organs transposition in the cranial sense because of the anomalous congenital position of the diaphragmatic dome, we decided for a laparoscopic approach. Such as in upper GI laparoscopic surgery the patient was placed in supine position with legs apart (classic French position). We performed pneumoperitoneum by Veress needle with the optical trocar in the supra-umbilical region. Other 4 trocars were positioned respectively in the right and left hypochondrium, in subxiphoid region and in the left flank [10]. The exploration of the peritoneal cavity revealed a wide cranialization of the left hemidiaphragm with thoracic trasposition of the stomach, great omentum, descending and transverse colon, spleen and pancreatic tail. The esophageal jato instead appeared continent with normal endo-abdominal positioning of the esophagus-gastric joint. These intraoperative findings confirmed the absence of a significant jatal hernia and the presence of a left diaphragmatic relaxation (Fig. 2a). With careful maneuvers we proceeded to section the tenacious congenital adhesions between the visceral organs (stomach, colon, spleen, pancreatic tail) and the diaphragmatic dome in order to obtain their repositioning in the normal anatomical place (Fig. 2b). Then we continued with an adequate laparoscopic plication of the left hemidiaphragm with nonresorbable sutures (Fig. 2c). In post-operative course we registered a left basal pneumothorax with maximum size of 5 cm medially, with no clinical signs and compat-
ble with the surgical procedure (Fig. 3a). We chose a conservative management and on POD 7 the patient was discharged after completed resolution of the pneumothorax (Fig. 3b).

3. Discussion

In diaphragmatic relaxation all or part of the muscle fibers are replaced by fibroelastic tissue, leading to a thinned and pliable central portion of the diaphragm. It may be congenital or acquired. The congenital form reflects a failure of the fetal diaphragm to muscularize and the incidence is two- to threefold higher in males than in females. The acquired form is associated with phrenic nerve injuries (prevalence 0.3%–0.5%) or is observed after surgery, especially open heart surgery (prevalence 0.5% and 2.2% in adults). In some cases it is idiopathic [11].

In this disease the continuity of the diaphragm and normal attachments to the costal margin are maintained. In adults, diaphragmatic eventration is rarely symptomatic. Principal symptoms can include dyspnea, palpitations, chest pain, dyspepsia, and recurrent pneumonia. The diagnosis of diaphragmatic relaxation is usually made on the presence of an elevated diaphragmatic dome at a chest X-ray. CT scan of thorax and abdomen and MRI can be helpful for differential diagnosis with other diseases [12]. The area of elevation coincides to the weakened or fibrotic area of the hemidiaphragm which is displaced into the thorax. The differential diagnosis in these patients includes unilateral diaphragmatic paralysis, diaphragmatic anterior hernias (Morgagni hernia) or posterior one (Bochdalek hernia) which also present a radiologic finding of an elevated hemidiaphragm, subdiaphragmatic or diaphragmatic masses and pleural effusion [13]. Most patients with diaphragmatic relaxation are asymptomatic and require no treatment, but sometimes they complain dyspnea or other symptoms, already described. In these cases we can take into consideration the surgical treatment, especially when we note a great decreased in physical daily activity or a severe lung disease [14].

According to the literature data the thoracotomy was the traditional approach for surgical diaphragmatic plication. Today the video-assisted thoracic surgery (VATS) offers a less invasive treatment because it avoids the incision of the lower intercostal muscles. Even if there is no consensus regarding which minimally invasive approach (thoracoscopic versus laparoscopic) is preferred for this repair, the choice is dependent on the experience of the surgical team [15–17] and it is certainly related to the extent of the transposition of the abdominal organs in the thorax and to the presence of tenacious adhesions. In our case report the diaphragmatic relaxation seemed to be congenital and the laparoscopic approach resulted the most appropriate technique to obtain the repositioning of abdominal organs and diaphragmatic plication. Nowadays robotic surgery has features which can overcome the difficulties of conventional laparoscopy and can also introduce new surgical options but the cost is higher and operative time is typically longer [18–20].

4. Conclusion

Surgical plication of the affected hemidiaphragm provides excellent results in carefully selected patients. This procedure can be performed using an open, thorascopic or laparoscopic approach and in some centers the robotic technology. The surgical procedure involves creating folds in the diaphragm and suturing them in place to reduce mobility of the hemidiaphragm. Plication usually results in an improvement of lung function, exercise endurance, dyspnea and the other symptoms. Before plication, the healthy hemidiaphragm generates negative intrathoracic pressure that is transmitted to the affected side. This pressure pulls up the affected portion of the diaphragmatic dome and with it parts of the abdominal contents. This ineffective motion does not expand the ipsilateral lung and results in poor gas exchange in that portion of the thorax. After plication, the abdomen is not pulled upwards and the adjacent lung segments expand themselves with better ventilation and improved gas exchange. Furthermore, the unhampered hemidiaphragm now performs less work, rendering it less susceptible to fatigue and decreasing the need to recruit accessory muscles of ventilation [16]. In this case report we show the feasibility and safety of laparoscopic approach for diaphragmatic relaxation with all the advantages that the minimally invasive surgery offers in terms of rapid mobilization and less pain decreasing the incidence of respiratory, circulatory and cardiac postoperative complications.

Declaration of Competing Interest

The authors report no declarations of interest.

Funding

Di Buono Giuseppe and other co-authors have no study sponsor.

Ethical approval

Ethical Approval was not necessary for this study. We obtained written patient consent to publication.

Consent

We obtained written patient consent to publication.

Author contribution

Di Buono Giuseppe: study design, data collections, data analysis and writing.
Bonventre Giulia: study design, data collections, data analysis and writing.
Amato Giuseppe: data collections.
Ricupati Federica: data collections.
Romano Giorgio: study design, data collections, data analysis and writing.
Agrusa Antonino: study design, data collections, data analysis and writing.

Registration of research studies

This article is part of a supplement entitled Case reports from Italian young surgeons, published with support from the Department of Surgical, Oncological and Oral Sciences – University of Palermo.

Guarantor

Di Buono Giuseppe.
Agrusa Antonino.

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.
Acknowledgement

This article is part of a supplement entitled Case reports from Italian young surgeons, published with support from the Department of Surgical, Oncological and Oral Sciences – University of Palermo.

References

[1] S.C. Fell, Surgical anatomy of the diaphragm and the phrenic nerve, Chest Surg. Clin. N. Am. 8 (May (2)) (1998) 281–294.

[2] A. Mirjello, C. D’Angelo, S. De Cosmo, G. Addolorato, R. Landolfi, A rare cause of shortness of breath: relaxatio diaphragmatica, Intern. Emerg. Med. 10 (December (8)) (2015) 1031–1032, http://dx.doi.org/10.1007/s11739-015-1257-x, Epub 2015 May 28.

[3] L. Pellegrini, Partial “relaxatio” of the right hemidiaphragm, Rass. Int. Clin. Ter. 30 (September (39)) (1959) 899–910.

[4] F. Nettesheim, K. Koster, Partial relaxatio diaphragmatic following compression of the phrenic nerve; its differential diagnosis, Tuberkoloseart 6 (October (10)) (1952) 607–609.

[5] A. Giovinetto, P. Rinella, O. Busso, F. Basile, T. Bianca, The diaphragmatic relaxatio, Chir. Ital. 31 (August (4)) (1979) 446–473.

[6] G. Granata, C. Lambertini, Relaxation of the diaphragm, Minerva Pediatr. 9 (October (42)) (1957) 1137–1143.

[7] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshly, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical Case Report (SCARE) guidelines, Int. J. Surg. 60 (2018) 132–136.

[8] G. Novo, E. Corrado, E. Tortorici, A. Novo, A. Agrusa, V. Saladino, I. Marturana, R. Lentini, M. Ferrandes, C. Visconti, F. Massenti, M. D’Arienzo, F. Vitale, G. Gulotta, S. Novo, Cardiac risk stratification in elective non-cardiac surgery: role of NT-proBNP, Int. Angiol. 30 (June (3)) (2011) 242–246.

[9] G.J. Gibson, Diaphragmatic paresis: pathophysiology, clinical features, and investigation, Thorax 44 (November (11)) (1989) 960–970, http://dx.doi.org/10.1136/thx.44.11.960.

[10] A. Agrusa, G. Di Buono, S. Buscemi, G. Cucinella, G. Romano, G. Gulotta, 3D laparoscopic surgery: a prospective clinical trial, Oncotarget 9 (April (25)) (2018) 17325–17333, http://dx.doi.org/10.18632/oncotarget.24669, eCollection 2018 Apr 3.

[11] K. Irie, T. Okatsu, T. Iijima, M. Fuji, M. Kurihara, Successfully treated case of relaxatio diaphragmatica in an infant, Geka Chiryo 21 (October (4)) (1969) 501–505.

[12] A. Agrusa, G. Romano, D. Chianetta, G. De Vita, G. Frazzetta, G. Di Buono, V. Sorce, G. Gulotta, Right diaphragmatic injury and lacerated liver during a penetrating abdominal trauma: case report and brief literature review, World J. Emerg. Surg. 9 (April) (2014) 33, http://dx.doi.org/10.1186/1749-7292-9-33, eCollection 2014. Review.

[13] C.M. Kneepkens, Abdominal pain and vomiting: a 4-years old boy with relaxation of diaphragm, Ned. Tijdschr. Geneeskd. 145 (November (46)) (2001) 2252–2253.

[14] H. Sandvik, J. Straand, Roboratio or relaxatio? Clinical theory and practice in the 19th century, Tidsskr Nor Laegforen 114 (March (8)) (1994) 918–921.

[15] D.R.I. Graham, D. Kaplan, C.C. Evans, C.R. Hind, R.J. Donnelly, Diaphragmatic plication for unilateral diaphragmatic paralysis: a 10-year experience, Ann. Thorac. Surg. 49 (February (2)) (1990) 248–251, http://dx.doi.org/10.1016/0003-4975(90)90146-w, discussion 252.

[16] A. Agrusa, G. Romano, L.J. Dominguez, G. Amato, R. Citarella, L. Vennuccio, G. Di Buono, V. Sorce, L. Gulotta, M. Galia, P. Mansueto, G. Gulotta, Adrenal cavernous hemorrhage: which correct decision making process? Acta Med. Medit. 32 (2016) 385–389.

[17] G. Di Buono, S. Buscemi, A.L. Lo Monte, G. Geraci, V. Sorce, R. Citarella, E. Gulotta, V.D. Palumbo, S. Fazzotta, L. Gulotta, D. Albano, M. Galia, G. Romano, A. Agrusa, Laparoscopic adrenalectomy: preoperative data, surgical technique and clinical outcomes, BMC Surg. 18 (April (Suppl. 1)) (2019) 128, http://dx.doi.org/10.1186/s12893-018-0456-6.

[18] T.P. Huttel, M.W. Wichmann, B. Reichart, T.K. Geiger, F.W. Schildberg, C. Meyer, Laparoscopic diaphragmatic plication: long-term results of a novel surgical technique for postoperative phrenic nerve palsy, Surg. Endosc. 18 (March (3)) (2004) 547–551, http://dx.doi.org/10.1007/s00464-003-8127-8.

[19] G. Cucinella, G. Calagna, G. Romano, G. Di Buono, G. Gugliotta, S. Saitta, G. Adile, M. Manzone, G. Accardi, A. Perino, A. Agrusa, Robotic versus laparoscopic sacrocolpopexy for apical prolapse: a case-control study, G Chir. 37 (May–June (3)) (2016) 113–117.

[20] A. Agrusa, G. Romano, G. Navarra, G. Conto, G. Pantuso, G.D. Buono, R. Citarella, M. Galia, A.L. Monte, G. Cucinella, G. Gulotta, Innovation in endocrine surgery: robotic versus laparoscopic adrenalectomy. Meta-analysis and systematic literature review, Oncotarget 8 (October (60)) (2017) 102392–102400, http://dx.doi.org/10.18632/oncotarget.22059, eCollection 2017 Nov 24.

Open Access
This article is published Open Access at sciencedirect.com. It is distributed under the IJSCR Supplemental terms and conditions, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.

Please cite this article in press as: G. Di Buono, et al., Successful laparoscopic management of congenital diaphragmatic relaxation: A case report, Int J Surg Case Rep (2020), https://doi.org/10.1016/j.ijscr.2020.09.055