EDITORIAL

Reshaping the Evidence for Surgical Correction of Pectus Excavatum Using Cardiopulmonary Exercise Testing

Ashok Kar ២, MB BChir; Max Baghai, MBBS, PhD; Ian Hunt ២, MBBS

Pectus excavatum is the most encountered chest wall deformity¹ yet there is ongoing debate as to the physiological and cardiopulmonary impact of this disease and significantly if surgical repair may offer benefit to patients.² In the United Kingdom, the controversy around the physiological and psychological impact of pectus deformities led the National Health Service to decommission (not for routine) pectus surgery treatment.³ This judgment has had far-reaching and significant effects and led to questions being asked in the Houses of Parliament as well as formal review of the policy (decision pending).

See Article by Jaroszewski et al.

There have been 275 papers published in the past 10 years on "functional" changes before and after corrective pectus surgery. Many of those papers are small cohort and retrospective case series^{4–6} and are, as reflected by National Health Service's decision in the United Kingdom, heterogenous in nature and quality of evidence presented.

In this issue of the *Journal of the American Heart Association (JAHA)*, new work by Jaroszewski et al.⁷ investigates using cardiopulmonary exercise testing (CPET), the cardio-respiratory effects of minimally invasive "Nuss" surgical repair (MIRPE).

A prior study by the same group using transesophageal echocardiography following pectus repair demonstrated immediate and significant improvements in anatomic and functional cardiac parameters (including right and left ventricular dimensions, stroke volume, and speckle tracking strain).⁸ Indeed, the authors previously published a detailed review paper summarizing the available literature on cardiopulmonary outcomes along with quality of life and patient satisfaction after pectus excavatum repair.⁹

Among surgeons it is clearly recognized that although subjective improvement in exercise tolerance is often reported by patients following MIRPE,¹⁰ few studies have robustly investigated this potentially functional benefit, especially in such a large cohort of patients, taking into consideration physiological parameters as opposed to anatomic assessment.¹¹

The authors in the current issue share the findings performed from a retrospective study among patients operated between 2011 and 2020 and identified 392 undergoing CPET before MIRPE of whom 68% had abnormal peak oxygen consumption. Of these, 130 patients also underwent CPET immediately before bar removal at a mean time interval of 3.4 years post repair with a significant demonstrable improvement in outcomes including peak VO₂, oxygen consumption at anaerobic threshold, and maximal ventilation. In a small subgroup of 39 patients with available pre- and

Key Words: Editorials = cardiopulmonary outcomes = CPET = MIRPE = pectus excavatum = pectus surgery

The opinions expressed in this article are not necessarily those of the editors or of the American Heart Association.

Correspondence to: Ian Hunt, MBBS, Department of Thoracic Surgery, St Georges' Hospital NHS Foundation Trust, Blackshaw Road, London SW17 0QT, United Kingdom. Email: ian.hunt@stgeorges.nhs.uk

© 2022 The Authors. Published on behalf of the American Heart Association, Inc., by Wiley. This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

JAHA is available at: www.ahajournals.org/journal/jaha

For Disclosures, see page 2.

postrepair transesophageal echocardiography studies of the right heart, improvement in stroke volume was also demonstrated. Notably, there was also an improvement in patients with normal baseline cardiopulmonary function and without severe anatomical defects. No significant association was found in a univariate analysis between the improvement in VO₂ max and anatomical variables such as Haller Index, sternal tilt, or cardiac compression index.

When trying to generalize the findings, it is important to note that all operations were performed by a single, high-volume surgeon experienced in repair of adult pectus excavatum. A 2-bar technique was used in 69% of patients and 3-bar in 31% with an average age of 30 years old. The average age is interesting in that it certainty reflects the "adult" nature of the practice presented as most published studies^{12,13} on MIRPE are in a younger age group. The retrospective nature of this study also meant that 78 patients (78/262, 29.78%) during the study period declined postoperative CPET evaluation (before bar removal) for personal reasons, particularly during the COVID-19 surge, when they did not want to prolong hospital admission. Moreover, at the end of the current study period, of the 262 patients without a postoperative CPET, the bar removal procedure had not yet been performed for 184 cases (184/262, 70.22%).

Despite most patients being symptomatic before surgery, the authors in the study were unable to perform a formal, objective survey of changes to patient symptoms before bar removal, which could have supported further the reported improvement in CPET outcomes. We therefore look forward to the authors sharing their mid- and long-term outcomes related to symptoms in patients with upcoming CPET testing as well as those who have had bars removed and are willing to have repeat CPET testing at 6 months to 1 year post bar removal. And particularly because following bar removal, 10% to 15% of patients with pectus excavatum can have recurrence, it would be interesting to therefore evaluate the physiological impact based on CPET in this group.

Overall, the authors should be congratulated for conducting this study in such a large number of patients. Following the take-up of MIRPE by the surgical community, early work investigated changes to lung function (spirometry), oxygen pulse, and incremental exercise testing but in small patient cohorts.¹⁴ Further assessment of cardiac function based on echocardiography¹⁵ or imaging¹⁶ (cardiovascular magnetic resonance) have also been reported in patients undergoing corrective pectus surgery, but again the number of patients is small, whereas there are other reported studies that have shown improvement in cardiopulmonary response.¹⁷

Pectus excavatum is too often viewed as a cosmetic disorder with a known associated psychological impact¹⁸⁻²⁰ but no definitive physiological measures that are reproducibly found to show that surgery can be of benefit.

As the authors of the study point out, this can affect the ability of patients to obtain insurance coverage to undergo surgical correction. In publicly funded health care systems such as the UK National Health Service, a cited lack of evidence that MIRPE can improve cardiopulmonary outcomes has resulted in the withdrawal of this treatment for patients. This timely and very interesting study provides support for the surgical correction of pectus excavatum and future studies are sought by clinicians involved in the care of this patient population.

ARTICLE INFORMATION

Affiliations

Department of Thoracic Surgery, St Georges Hospital NHS Foundation Trust, London, United Kingdom (A.K., I.H.); and Department of Cardiothoracic Surgery, King's College Hospital NHS Foundation Trust, London, United Kingdom (M.B.).

Disclosures

None.

REFERENCES

- Biavati M, Kozlitina J, Alder AC, Foglia R, McColl RW, Peshock RM, Kelly RE, Garcia CK. Prevalence of pectus excavatum in an adult populationbased cohort estimated from radiographic indices of chest wall shape. *PLoS One.* 2020;15:e0232575. doi: 10.1371/journal.pone.0232575
- Maagaard M, Heiberg J. Improved cardiac function and exercise capacity following correction of pectus excavatum: a review of current literature. Ann Cardiothorac Surg. 2016;5:485. doi: 10.21037/acs.2016.09.03
- NHS England. Clinical commissioning policy: surgery for pectus deformity. NHS England Specialised Services Clinical Reference Group for Radiotherapy and Specialised Cancer Surgery: NHS England; 2019.
- Pawlak K, Gąsiorowski Ł, Gabryel P, Gałęcki B, Zieliński P, Dyszkiewicz W. Early and late results of the Nuss procedure in surgical treatment of pectus excavatum in different age groups. *Ann Thorac Surg.* 2016;102:1711. doi: 10.1016/j.athoracsur.2016.04.098
- Gurkan U, Aydemir B, Aksoy S, Akgöz H, Tosu AR, Öz D, Güngör B, Yılmaz H, Bolca S. Echocardiographic assessment of right ventricular function before and after surgery in patients with pectus excavatum and right ventricular compression. *Thorac Cardiovasc Surg.* 2014;62:231. doi: 10.1055/s-0033-1342941
- Kelly RE, Mellins RB, Shamberger RC, Mitchell KK, Lawson ML, Oldham KT, Azizkhan RG, Hebra AV, Nuss D, Goretsky MJ, et al. Multicenter study of pectus excavatum, final report: complications, static/exercise pulmonary function and anatomic outcomes. J Am Coll Surg. 2013;217:1080. doi: 10.1016/j.jamcollsurg.2013.06.019
- Jaroszewski DE, Farina JM, Gotway MB, Stearns JD, Peterson MA, Pulivarthi V, Bostoros P, Abdelrazek AS, Gotimukul A, Majdalany DS, et al. Cardiopulmonary outcomes after Nuss procedure. *J Am Heart Assoc.* 2022;10:e022149. doi: 10.1161/JAHA.121.022149
- Chao C, Jaroszewski DE, Kumar PN, Ewais MM, Appleton CP, Mookadam F, Gotway MB, Naqvi TZ. Surgical repair of pectus excavatum relieves right heart chamber compression and improves cardiac output in adult patients—an intraoperative transesophageal echocardiographic study. *Am J Surg.* 2015;210:1118. doi: 10.1016/j.amjsu rg.2015.07.006
- Ewais MM, Chaparala S, Uhl R, Jaroszewski DE. Outcomes in adult pectus excavatum patients undergoing Nuss repair. *Patient Relat Outcome Meas*. 2018;9:65–90. doi: 10.2147/PROM.S117771

- Nuss D, Kelly RE Jr. Indications and technique of Nuss procedure for pectus excavatum. *Thorac Surg Clin.* 2010;20:583. doi: 10.1016/j.thors urg.2010.07.002
- Rodriguez-Granillo GA, Raggio IM, Deviggiano A, Bellia-Munzon G, Capunay C, Nazar M, Martinez JL, Carrascosa P, Martinez-Ferro M. Impact of pectus excavatum on cardiac morphology and function according to the site of maximum compression: effect of physical exertion and respiratory cycle. *Eur Heart J Cardiovasc Imaging*. 2020;21:77. doi: 10.1093/ehjci/jez061
- Kelly RE, Goretsky MJ, Obermeyer R, Kuhn MA, Redlinger R, Haney TS, Moskowitz A, Nuss D. Twenty-one years of experience with minimally invasive repair of pectus excavatum by the Nuss procedure in 1215 patients. *Ann Surg.* 2010;252:1072. doi: 10.1097/SLA.0b013e3181effdce
- Brungardt JG, Chizek PW, Schropp KP. Adult pectus excavatum repair: national outcomes of the Nuss and Ravitch procedures. *J Thorac Dis.* 2021;13:1396. doi: 10.21037/jtd-20-2422
- Haller JA Jr, Loughlin GM. Cardiorespiratory function is significantly improved following corrective surgery for severe pectus excavatum. Proposed treatment guidelines. J Cardiovasc Surg (Torino). 2000;41:125.
- 15. Udholm S, Maagaard M, Pilegaard H, Hjortdal V. Cardiac function in adults following minimally invasive repair of pectus excavatum.

Interact Cardiovasc Thorac Surg. 2016;22:525. doi: 10.1093/icvts/ ivw007

- Töpper A, Polleichtner S, Zagrosek A, Prothmann M, Traber J, Schwenke C, von Knobelsdorff-Brenkenhoff F, Schaarschmidt K, Schulz-Menger J. Impact of surgical correction of pectus excavatum on cardiac function: insights on the right ventricle. A cardiovascular magnetic resonance study. *Interact Cardiovasc Thorac Surg.* 2016;22:38. doi: 10.1093/icvts/ivv286
- Neviere R, Montaigne D, Benhamed L, Catto M, Edme JL, Matran R, Wurtz A. Cardiopulmonary response following surgical repair of pectus excavatum in adult patients. *Eur J Cardiothorac Surg.* 2011;40:e77. doi: 10.1016/j.ejcts.2011.03.045
- Zuidema WP, Oosterhuis JWA, Zijp GW, van der Heide SM, van der Steeg AFW, van Heurn LWE. Early consequences of pectus excavatum surgery on self-esteem and general quality of life. *World J Surg.* 2018;42:2502. doi: 10.1007/s00268-018-4526-9
- Luo L, Xu B, Wang X, Tan B, Zhao J. Intervention of the Nuss procedure on the mental health of pectus excavatum patients. *Ann Thorac Cardiovasc Surg.* 2017;23:175. doi: 10.5761/atcs.oa.17-00014
- Eisingger RS, Islam S. Caring for people with untreated pectus excavatum: an international online survey. *Chest.* 2020;157:590. doi: 10.1016/j. chest.2019.10.034