Original Article

Preoperative imaging of clinically relevant intrathoracic abnormalities in pectus excavatum patients

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Background: Preoperative radiological imaging in pectus excavatum sometimes coincidentally yields additional intrathoracic abnormalities. In the context of a larger research project investigating replacement of CT scans by 3D-surface scanning as routine preoperative work-up for pectus excavatum, this study aims to quantify the incidence of clinically relevant intrathoracic abnormalities found incidentally using conventional CT in pectus excavatum patients.

Methods: A single-center retrospective cohort study was conducted including pectus excavatum patients, receiving CT between 2012 and 2021 as part of their preoperative evaluation. Radiology reports were reviewed for additional intrathoracic abnormalities and scored into three subclasses: non-clinically relevant, potentially clinically relevant or clinically relevant findings. Also, two-view plain chest radiographs reports, if available, were evaluated for those patients with a clinically relevant finding. Subgroup analysis was performed to compare adolescents and adults.

Results: In total, 382 patients were included, of whom 117 were adolescents. Although in 41 patients (11%) an additional intrathoracic abnormality was found, only two patients (0.5%) presented with a clinically relevant abnormality requiring additional diagnostics, postponing surgical correction. In only one of the two patients, plain chest radiographs were available, which did not show the abnormality. Subgroup analyses revealed no differences in (potentially) clinically relevant abnormalities between adolescents and adults.

Conclusions: The prevalence of clinically relevant intrathoracic abnormalities in pectus excavatum patients was low, supporting the notion that CT and plain radiographs can be safely replaced by 3D-surface scanning in the preoperative work-up for pectus excavatum repair.

Keywords: Pectus excavatum; funnel chest; diagnostic imaging

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Introduction

Pectus excavatum is the most common congenital chest wall deformity, characterized by dorsal deflection of the sternum and adjacent cartilage (1). It is typically diagnosed in early childhood. The deformity is estimated to occur in 1:300 live births (2) and affects predominantly the male sex, with a ratio ranging from 2:1 to 9:1 (3). Pectus excavatum can be accompanied by additional skeletal abnormalities such as scoliosis, and the outward projection of the ribs adjacent to the depression (4,5). If indicated, pectus excavatum can be surgically corrected via the open Ravitch procedure (6) or the minimal invasive Nuss procedure (7).

Prior to treatment, patients are screened to determine the morphology and severity of their chest wall deformity [using objective indices such as the Haller index (8)] and to assess the presence of cardiopulmonary compression. Conventionally, this is done through computed tomography (CT), whereby pectus excavatum patients are exposed to a considerable amount of radiation (7,9). Currently, there is a lack of consensus on the diagnostic workup for pectus excavatum patients, and some experts still advocate for CT, as it provides a comprehensive view including all the aforementioned aspects (10). In addition, this imaging technique coincidentally detects additional intrathoracic abnormalities. Unfortunately, alternative imaging modalities associated with no or limited exposure to radiation have several disadvantages. For example, magnetic resonance imaging (MRI) is not suitable for claustrophobic patients, and is also more prone to motion artefacts, which often occurs when imaging pediatric patients (11-13). Plain chest radiographs on the other hand provide no information on the presence of cardiopulmonary compression, and literature on their ability to detect additional clinically relevant intrathoracic findings in pectus excavatum patients is limited (14). Echography is an acceptable alternative to detect cardiac compression; however, it provides no information on the deformity required for surgical planning (15). Information on the sensitivity and specificity of echography for the detection of different types of intrathoracic abnormalities is scarce. A novel promising, radiation-free imaging technique is three-dimensional (3D) surface imaging. This technique accurately assesses the morphology and severity of the deformity (13,16). Automatic quantification of morphological features is not limited to the Haller index (17). Furthermore, it provides information on cardiopulmonary compression as cardiac compression prediction models have been developed for and tested in pectus excavatum patients (18). However, this technique cannot be used to detect possible additional intrathoracic abnormalities. Therefore, the question is whether routine screening of pectus excavatum patients for intrathoracic abnormalities is warranted, especially because this patient group is predominately of young age.

Hence, this paper is set up to provide evidence on the prevalence of clinically relevant intrathoracic abnormalities secondary to pectus excavatum in patients of all ages seeking surgical correction.

Methods

This retrospective cohort study was conducted at the Department of Surgery, Division of General Thoracic Surgery of Zuyderland Medical Centre in Heerlen, The Netherlands, a tertiary referral center for chest wall disorders. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Medical Ethics Committee Zuyderland & Zuyd (ID: METCZ20220037; approval date: April 13th, 2022), and individual consent for this retrospective analysis was waived.

All patients who were referred for pectus excavatum and received CT as part of their diagnostic work-up, were eligible for inclusion. Patients imaged between 2012 and 2021 were considered, with no age restrictions. Patients who received prior thoracic surgery were excluded.

Variables and data analysis

The following patient characteristics were extracted from the electronic patient files: (I) sex; (II) age. Patients aged 10 to 19 years were considered adolescents, and patients aged 19 years or older were considered adults (19).

Radiology reports of obtained CTs were reviewed by two researchers (NJ, IM). Intrathoracic abnormalities were derived from the reports and scored according to the classification as introduced by Rattan *et al.* (14):

- ❖ Class I abnormality: incidental and of no significance;
- ❖ Class II abnormality: potentially clinically significant;
- ❖ Class III abnormality: affected decision to perform surgery (also postponed surgery).

The class 3 abnormalities were divided into two subsections:

- ❖ IIIa: surgery is postponed allowing diagnostic work-up;

- IIIb: surgery is postponed allowing treatment of the abnormality, or the surgical procedure was cancelled.

When a class III abnormality was described in the CT radiology report, the two-view plain chest radiographs report, if available, was reviewed to evaluate whether the specific abnormality was also detected by plain chest radiography. In the presence of an abnormality, its medical consequences were also derived from the electronic patient record.

Subgroup analysis on the number of CT scans with an additional intrathoracic abnormality in adolescents compared to adults was performed. In this analysis, the presence of multiple abnormalities in one individual patient was considered as one CT scan with an intrathoracic abnormality.

The prevalence of the different types of abnormalities among adolescents compared to adults was also assessed. If one patient had multiple abnormalities across different classes (e.g., one class I abnormality and one class II abnormality), this was valued as such. However, each patient was only scored once within each class.

Statistical analysis

Nominal variables were depicted as absolute numbers and percentages. Continuous data was presented as mean and standard deviation (SD) or as median and interquartile range (IQR) in the presence of skewness. The Fisher exact test was used for subgroup analysis and a P value ≤0.05 was considered statistically significant. Data were analyzed using SPSS statistics (IBM Corp. IBM SPSS statistics for MacOS, Version 27.0, Armonk, NY, USA).

Results

A total of 382 patients were enrolled, based on previously stated selection criteria. Median patient age of the entire cohort was 23 years (IQR, 18–36 years), with 117 (31%) patients within the adolescent group and 265 (69%) in the adult group. Median patient age was 16 years (IQR, 14–17 years) in the adolescent group and 29 years (IQR, 23–44 years) in the adult group. Most patients were male (n=302, 79%). In 341 patients (89%), no abnormalities other than the pre-identified pectus excavatum were reported. In the remaining 41 patients (11%), a total of 47 abnormalities were reported (*Table 1*). Class I abnormalities were most frequently observed, comprising 36 (77%) reported abnormalities in 31 (8%) patients. Nine (19%) class II abnormalities were reported in 8 (2%) patients. Of note, the single patient suffering from a thoracic aorta aneurysm was already diagnosed with the aneurysm and enrolled in a follow-up program at another institution. In 2 (0.5%) patients a class IIIa abnormality was identified, including a thymic lymphoma and pericardial effusion. Both abnormalities did not necessitate direct treatment and entered follow-up. Successful surgical correction of the deformity by the Nuss procedure was established in both patients. For the patient with a thymic lymphoma, preoperative plain chest radiographs were available. However, the abnormality was not visible on the plain chest radiographs. In the presented cohort, screening of 191 patients was required for the

Table 1 Overview of reported abnormalities

Class	Description of abnormality	n
I	Lung nodule(s)	17
	Small area of ground-glass opacity	3
	Small area of avascular lung parenchym	1
	Small area of pleural thickening	2
	Subpleural adhesions	1
	Pleural effusion	1
	Atelectasis	3
	Emphysema	1
	Thickened bronchovascular bundle	2
	Air trapping lung	1
	Coronary artery calcification	2
	Unilateral diaphragmatic paralysis	1
	Subtotal	36
II	Small lung infiltrate	6
	Pericardial cyst	1
	Ectopic thymus tissue	1
	Aneurysm thoracic aorta	1
	Subtotal	9
III	Pericardial effusion	1
	Thymus lymphoma	1
	Subtotal	2
Total		47

Table 2 Abnormalities among adolescents compared to adults

Abnormality	Adolescent, n [%]	Adult, n [%]	P value
Abnormality			0.05
Yes	7 [6]	34 [13]	
No	110 [94]	231 [87]	
Class I			0.03
Yes	4 [3]	27 [10]	
No	113 [97]	238 [90]	
Class II			>0.99
Yes	2 [2]	6 [2]	
No	115 [98]	259 [98]	
Class III			0.52
Yes	1 [1]	1 [0.4]	
No	116 [99]	264 [99.6]	
Total	117 [100]	265 [100]	

detection of one class III abnormality.

Subgroup analysis revealed that an abnormality was found in 13% (34/265) of the adults, while an abnormality was reported in 6% (7/117) of the adolescents (*Table 2*). This difference in prevalence between both groups was statistically significant (P=0.05).

In addition, class I abnormalities were more frequently found in adults (10%, 27/265) compared to adolescents (3%, 4/117, P=0.03). For class II and III abnormalities, no significant differences were observed between both age groups (*Table 2*).

Discussion

Pectus excavatum patients are preoperatively screened to evaluate the severity and morphology of the deformity, and to assess the presence of cardiopulmonary compression. Currently, there is no consensus on the diagnostic protocol with some experts advocating CT as the primary imaging technique as it provides a comprehensive view including all aforementioned aspects (12). Coincidentally, additional intrathoracic abnormalities are found using this imaging technique. In the search for a diagnostic protocol that prevents patients from being exposed to unnecessary radiation, with 3D surface imaging as a promising radiation-free alternative to CT (13,16,17), the question arises on the place of routine screening for intrathoracic abnormalities in preoperative pectus excavatum work-up. The objective of this study was therefore to determine the prevalence of additional clinically relevant intrathoracic abnormalities in pectus excavatum patients of all ages seeking surgical correction.

In our retrospective cohort of 382 patients, consisting of all patients who embarked on preoperative assessment for pectus excavatum repair, 41 patients (11%) had one or multiple additional intrathoracic findings, of whom only two (0.5%) were classified as a clinically relevant abnormality. Hence, 191 patients needed to be screened to detect one clinically relevant abnormality. A decade ago, Rattan *et al.* (14) conducted a retrospective study among 209 pectus excavatum patients and accidental findings which affected the decision to perform or postpone surgery, were observed in only 1% of patients. As opposed to Rattan *et al.*, who only included children and young adults (mean 13.1 years, 3.4 SD), patients of all ages were included in the current study. The scientific starting point created by Rattan *et al.* potentially constituted a skewed image when extrapolated to the entire pectus excavatum population as the number of additional findings is generally known to increase with age (20). However, although class I abnormalities were more frequently observed in adults (median age of 29 years, IQR, 23–44 years) by the present series, the presence of (possible) clinically relevant findings was comparable among both age groups.

The clinical relevance of the identified cases in this study was the necessity to perform a more elaborate diagnostic workup. However, no patient needed direct treatment for the condition which was incidentally detected and in no patient the decision to surgically treat the pectus excavatum was changed.

For only one of two cases with a class III abnormality, preoperative plain chest radiographs were available, which did not show the secondary finding. This in contrast to Rattan *et al.* (14), who demonstrated that there was no additional value for CT compared to two-view plain chest radiographs in identifying the found abnormalities in their series. However, as the decision to perform surgery was not affected, the finding in the current study does not support the necessity of acquiring a CT scan in the preoperative assessment for pectus excavatum repair. Furthermore, CT is associated with exposure to ionizing radiation which is considered a carcinogen. The acquisition of a single chest CT results in a mean radiation exposure of 7.0 mSv is (range, 4.0–18.0) (21). Especially children and adolescents are at risk of developing a radiation-related pathology due to their relatively long life-time risk (22-24). A study by Miglioretti *et al.* (25) on the development of solid cancer and leukemia among pediatric patients (<15 years) who received a chest CT, demonstrated a life-time attributable risk of 18 per 10,000 (0.2%) patients. The number needed to harm based on that study would be 556. In the study by Rattan *et al.* (14) 105 (209/2) patients needed to be screened to detect one abnormality. Though, Rattan *et al.* took all sorts of accidental findings into account, all detected clinically relevant findings were intrathoracic abnormalities. In the current study 191 (382/2) patients had to be screened to detect one clinically relevant intrathoracic abnormality.

The present study only reports on the necessity of CT in identifying additional intrathoracic abnormalities. Part of surgeons demand CT for their surgical planning in more severe or morphologic aberrant cases of pectus excavatum. In that case, a low-dose CT scan can be used in order to decrease radiation exposure as only costal structures need to be visualized. However, this does not apply for all surgeons and/or centers, and based on the presented data it is not possible to differentiate which patients benefit from receiving a CT.

This paper suggests that routine acquisition of a CT scan only to evaluate the presence of additional intrathoracic abnormalities is not justified in the light of the hazardous effects of radiation exposure, especially in a young population.

Limitations

In the present study, both CT scans and plain chest radiographs were reviewed by different radiologists. This could affect results due to interobserver bias. On the other hand, this study represents a real-world situation and the conclusion is therefore better applicable in other settings. Another limitation of the study is that not all patients routinely received a chest CT scan as part of their diagnostic work-up. This predominantly affected patients under the age of 18, as for those patients a CT was only performed upon discretion of the surgeon, compared to patients 18 years and older who got a CT done per protocol. This may have led to an overestimation of the actual number of additional findings as the number of findings increases with age. However, as the prevalence of (possible) clinically relevant abnormalities was comparable for both age groups, this effect seems to be limited in the present study.

The study is further limited by its retrospective design. However, based on the relatively low prevalence of intrathoracic findings, it is not feasible to include enough patients to achieve sufficient power in a prospective non-inferiority study. Therefore, real-world evidence studies, such as the current exhibit the best available evidence.

Conclusions

Prevalence of clinically relevant additional intrathoracic abnormalities detected using computed tomography in pectus excavatum patients of all ages is 0.5% with an associated number of 191 patients needed to screen to detect one abnormality. Furthermore, the detected abnormalities only led to additional diagnostics and postponement of the surgery and did not affect the decision to surgically correct the deformity. Among patients who both received a chest computed tomography scan and plain chest radiographs, the former could not be substituted by the latter to detect the reported clinically relevant abnormalities. However, as the found intrathoracic abnormalities did not affect the decision to perform surgery, this study suggests that there is no place for routine screening for intrathoracic abnormalities in the preoperative work-up for pectus excavatum repair.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-22-1366/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Medical Ethics Committee Zuyderland & Zuyd (No. METCZ20220037; approval date: April 13th, 2022) and individual consent for this retrospective analysis was waived.

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