

REVIEW

Pectus Excavatum: Consensus and Controversies in Clinical Practice



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ABSTRACT

BACKGROUND Pectus excavatum is the most common congenital anterior chest wall deformity. Currently, a wide variety of diagnostic protocols and criteria for corrective surgery are being used. Their use is predominantly based on local preferences and experience. To date, no guideline is available, introducing heterogeneity of care as observed in current daily practice. The aim of this study was to evaluate consensus and controversies regarding the diagnostic protocol, indications for surgical correction, and postoperative evaluation of pectus excavatum.

METHODS The study consisted of 3 consecutive survey rounds evaluating agreement on different statements regarding pectus excavatum care. Consensus was achieved if at least 70% of participants provided a concurring opinion.

RESULTS All 3 rounds were completed by 57 participants (18% response rate). Consensus was achieved on 18 of 62 statements (29%). Regarding the diagnostic protocol, participants agreed to routinely include conventional photography. In the presence of cardiac impairment, electrocardiography and echocardiography were indicated. Upon suspicion of pulmonary impairment, spirometry was recommended. In addition, consensus was reached on the indications for corrective surgery, including symptomatic pectus excavatum and progression. Participants moreover agreed that a plain chest radiograph must be acquired directly after surgery, whereas conventional photography and physical examination should both be part of routine postoperative follow-up.

CONCLUSIONS Through a multiround survey, international consensus was formed on multiple topics to aid standardization of pectus excavatum care.

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Pectus excavatum is the most common type of anterior chest wall deformity.¹ It is characterized by a depression of the sternum and adjacent costal cartilage. Patients often present with physical symptoms, such as exercise intolerance, but also psychosocial symptoms resulting from body image disturbances and related poor self-esteem.^{2,3} The established treatment of choice for pectus excavatum is surgical repair by the Nuss procedure.⁴ Other options include the conventional Ravitch procedure⁵ and nonsurgical treatment by the so-called vacuum bell.⁶

Notwithstanding, before considering any treatment, an indication must be established.

Different studies have elaborated on indications and objective criteria for the Nuss procedure.⁷⁻¹⁰ These include a Haller index of ≥ 3.25 , cardiac and/or pulmonary compression on cross-sectional imaging or

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impaired function test results, and failed previous repairs.⁷⁻⁹ Other institutions have adopted and adapted these criteria, adding criteria such as body image disturbances.¹⁰ The major drawback of these indications and criteria is that they were subjectively drafted by a selected group of experts and lack scientific basis. For example, the Haller index criterion was based on 33 patients with presumed pectus excavatum who underwent surgical correction as judged by presumed experts.¹¹ In conjunction with the fact that the diagnosis of pectus excavatum is associated with considerable intraobserver and interobserver variability, the scientific basis for use of the Haller index is even further blurred.¹²

There is not only a lack of agreement on indications for surgical corrections of pectus excavatum but also no consensus on the diagnostic protocol that should be used to evaluate the eligibility for surgery. In doing so, differences in diagnostic approach can ultimately result in treatment proposal variations. Standardization is therefore urgently needed. In the absence of conclusive scientific evidence, this can only be based on expert consensus. The objective of this international prospective study was to evaluate consensus and controversies on the diagnostic protocol, indications for surgical correction, and postoperative evaluation of pectus excavatum.

MATERIAL AND METHODS

The research was conducted as an international 3-round Delphi survey. The report was written in line with the Guidance on Conducting and REporting DELphi Studies (CREDES) reporting guideline.¹³ Because no patients participated in this study, ethical approval was not required.

PARTICIPANTS. Potentially eligible participants were identified as those affiliated to the Chest Wall International Group, which is an international group of experts who are involved in the treatment of chest wall disorders, including pectus excavatum. Potential participants were contacted by email. Those contacted were also asked to distribute the survey among peers (resident experience level or higher) who were directly involved in the treatment of pectus excavatum patients. No sample size estimation was performed, and no minimum number of participants was set. Only participants who completed all 3 rounds were included in final analysis.

SURVEY. The present survey consisted of 3 consecutive rounds. Members N.J., J.D., and E.v.P. of the steering group created the initial survey based on a literature review, which was pilot tested by Y.J., K.H., Y.V., and E.d.L. The survey was adapted accordingly. N.J. and E.v.P. reviewed all entries, deleting duplicates and incomplete responses.

The survey was constructed in the online survey tool Qualtrics (<https://www.qualtrics.com/>) and was distributed by email. Reminders were sent every 2 weeks by email. For round 1, 2 reminders were sent. After round 1, participants were invited for subsequent rounds through the Qualtrics distribution program, allowing no introduction of new participants. For rounds 2 and 3, reminders were sent until at least 70% of the participants of the previous round completed the survey. Data were collected and analyzed after each round. The participants did not meet face to face during the survey period to reduce bias caused by opinions of influential individuals.

The first survey round consisted of 20 questions, addressing the participant's characteristics (eg, the type of institution, affiliated department, professional position, and years of experience) and also addressing aspects of perioperative pectus excavatum care (eg, the use of diagnostic tools and [contra]indications for surgery). The survey of the first round is provided in [Supplemental Table 1](#). Questions on the perioperative aspects were exploratory questions (eg, "What preoperative diagnostic tools do you use routinely?" and "What do you consider the youngest age on which a pectus excavatum correction may be performed?"). Most questions were formulated as multiple-choice questions, with the possibility to add a response option. The remaining questions had an open-ended format.

If in round 1, >90% of the participants selected or proposed a specific response option, then it was agreed upon that consensus had been achieved, and thus disregarded in the following rounds. This was done to avoid participation fatigue.¹⁴ All others, for which no consensus was achieved after the first round, were used as a basis for subsequent rounds. Questions addressing the participants' expertise, which is a given, were obviously only incorporated in round 1 and used to assess the validity of the study.

In the first round, participants were encouraged not only to contribute possible alternative response options to the formulated questions but also to provide additional topics on which consensus was urged. When a response option or topic was proposed by >10% of participants, it was explored in detail in the second and third round. This percentage was chosen arbitrarily.

For rounds 2 and 3, after the comments from the participants on the exploratory questions and answer options in round 1 were analyzed and incorporated, a definite panel of statements was created. The number of statements and topics was thus dependent on the results from the previous round. Statements were categorized into topics, and all statements within a topic had the same question stem (eg, "[x] should be included in routine preoperative evaluation"). In the absence of an established definition for symptomatic pectus

excavatum, no definition was provided for the statement concerning symptomatic pectus excavatum as an indication for surgery. All physical symptoms related to pectus excavatum as judged by the participant were considered. Questions regarding the participant's characteristics were, as mentioned before, not repeated.

The response options for rounds 2 and 3 were presented in the format of a 9-point Likert scale.¹⁴ This scale was selected over a binary answer option to be able to weigh answers and to adhere to predefined criteria for consensus. Disagreement with a statement was defined as a rating between 1 and 3 on the 9-point Likert scale, whereas agreement was defined as a rating between 7 and 9. A rating between 4 and 6 indicated that the participant did neither agree nor disagree with that statement. Consensus was a priori defined as at least 70% of participants agreeing or disagreeing with a certain statement.

The statements presented in the third round were identical to the second. However, during this round, the participants were provided with the overall group response, allowing them to alter their response with this additional insight.

STATISTICAL ANALYSIS. Continuous data are presented as mean and SD or as median and interquartile range in the presence of skewness. Nominal and ordinal data are depicted as frequencies and percentages. Data were analyzed using IBM SPSS Statistics for Macintosh 26.0 (IBM Corp, 2019 release).

RESULTS

A cumulative number of 313 potentially eligible participants were identified. Of these, 11 were excluded due to invalid contact details, and 217 provided no response or an incomplete response to the survey, leaving 85 valid responses (27% response rate). Respectively, 62 and 57 valid responses were received to the second and third rounds, corresponding with a response rate of 20% and 18% (Supplemental Figure).

PARTICIPANT CHARACTERISTICS. Of the 57 participants who completed all 3 rounds, most (70%) worked in an academic hospital and were attendings (58%) in the field of pediatric surgery (45%) and thoracic surgery (44%). All continents (except Antarctica) were represented in the sample, and most participants were located in Europe (60%) and North America (19%).

Most participants (70%) had >10 years of experience treating patients with pectus excavatum. Up to 39% of participants treated >200 pectus excavatum patients during their ongoing career. A relatively small group (21%) treated <50 pectus excavatum patients successfully. The number of new patients diagnosed in the participant's institution each year was spread more

evenly. Further specifications on the characteristics of participants are summarized in Table 1 and the Figure.

SURVEY ANALYSIS. In round 1, based on the comments of 11% of participants, the answer option "physical examination" was added to the single question: "Which methods do you use to determine the improvement of correction postoperatively?." In addition, consensus was already achieved for 1 question: 92% of participants agreed that a plain chest radiograph should be routinely acquired directly after corrective surgery. No new topics were proposed to be included in the following rounds.

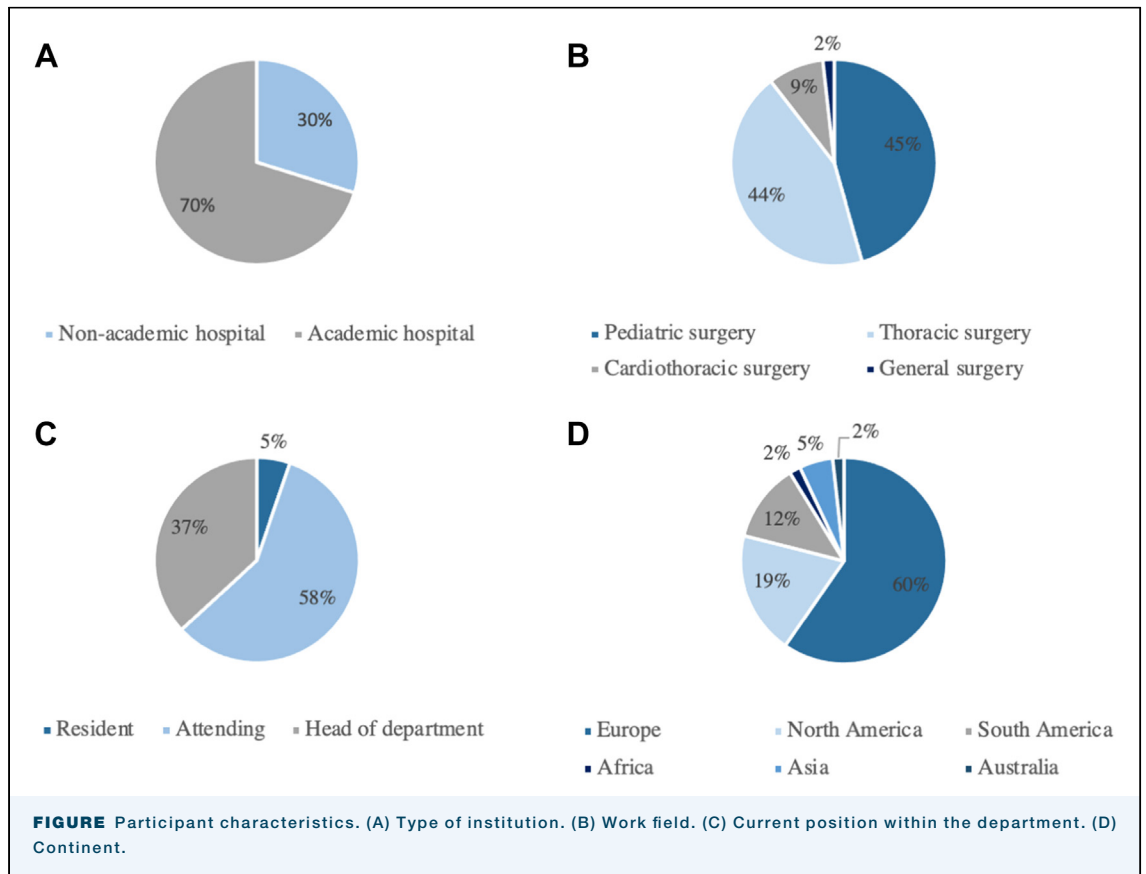
The second and third survey rounds consisted of 62 statements spread across 10 topics. During round 3, the level of (dis)agreement further increased across almost all statements that met the criteria of consensus in the second round. In addition, consensus was achieved on an additional 4 statements. Statements for which consensus was achieved are described in Table 2, and all statements are summarized in Supplemental Table 2 and Supplemental Table 3 for the sake of clarity and readability.

PREOPERATIVE EVALUATION. In round 2, consensus was reached on the topic of which tests should be included in routine preoperative evaluation. Participants agreed (81%) that conventional photography (ie, visually documenting the deformity through simple photography) should be included in routine workup.

Consensus was also reached on the topics concerning which tests should be added in the presence of cardiac or pulmonary complaints. In the case of cardiac complaints, echocardiography and cardiopulmonary exercise

TABLE 1 Participant Characteristics

Variables	Participants n (%)
Years of experience treating pectus excavatum	
≤3 years	2 (4)
4-6 years	4 (7)
7-9 years	11 (19)
≥10 years	40 (70)
Number of new patients diagnosed with pectus excavatum per year	
≤25 cases	9 (16)
26-50 cases	19 (33)
51-75 cases	13 (23)
76-100 cases	4 (7)
≥101 cases	12 (21)
Total patients successfully treated	
≤50	12 (21)
51-100	14 (25)
101-150	7 (12)
151-200	2 (4)
≥201	22 (39)
Total	57 (100)



testing should be performed based on expert consensus (77% and 72% agreement, respectively). In the presence of pulmonary complaints, respondents agreed that spirometry should be the designated diagnostic method (74% agreement). After round 3 was conducted, participants now agreed (79% vs 67% in the second round) that

TABLE 2 Statements on Which Consensus Was Achieved (Level III Evidence)	
Statement	Participants
TEE should be included in routine preoperative screening	74% disagreement
Conventional photography should be included in preoperative screening	83% agreement
Spirometry should be added on when patients present with pulmonary complaints	79% agreement
Echocardiography should be added on when patients present with cardiac complaints	79% agreement
ECG should be added on when patients present with cardiac complaints	79% agreement
Cosmetic discomfort or body image disturbances should be considered an indication for pectus excavatum repair	84% agreement
Cardiac compression on imaging should be considered an indication for pectus excavatum repair	75% agreement
Abnormal cardiac function tests should be considered an indication for pectus excavatum repair	88% agreement
Abnormal pulmonary function tests should be considered an indication for pectus excavatum repair	74% agreement
Progression of deformity should be considered an indication for pectus excavatum repair	84% agreement
Symptomatic pectus excavatum (physical) should be considered an indication for pectus excavatum repair	97% agreement
There should be a follow-up program for patients who do not meet the initial requirements for repair	88% agreement
The youngest age for pectus excavatum correction in standard cases should be 12 years old (special cases may be an exception to this)	75% agreement
CT scan should be included in routine postoperative evaluation	72% disagreement
MRI should be included in routine postoperative evaluation	83% disagreement
TEE should be included in routine postoperative evaluation	81% disagreement
Conventional photography should be included in routine postoperative evaluation	86% agreement
Physical examination should be included in routine postoperative evaluation	98% agreement

CT, computed tomography; ECG, electrocardiogram; MRI, magnetic resonance imaging; TEE, transesophageal echocardiography.

an electrocardiogram (ECG) should be part of the preoperative evaluation in case of cardiac symptoms, whereas there was no room for transesophageal echocardiography (TEE) during the routine preoperative evaluation (74% vs 67% in the second round). Round 3 did lead to withdrawal of 1 statement that consensus was agreed upon during the second round, namely, the necessity of cardiopulmonary exercise testing in preoperative evaluation in patients with cardiac complaints (68% agreement vs 72% in the second round).

INDICATIONS FOR CORRECTIVE SURGERY. Among the potential indications for corrective surgery, participants agreed on the following indications during the second round: physically symptomatic pectus excavatum (88% agreement), progression of the deformity (74% agreement), abnormal cardiac function test results (84% agreement), and cosmetic discomfort or other body image disturbances (79% agreement). Of note, no consensus was reached for both the Haller index and correction index.

Most participants (75% agreement) agreed that 12 years is the youngest age at which pectus excavatum correction should be performed in straightforward cases. No consensus was obtained regarding contraindications for surgical correction. The necessity of a follow-up program for patients who do not meet the initial criteria for surgery was endorsed by 77% of participants, achieving consensus.

After round 3, both cardiac compression on imaging (ie, computed tomography [CT] or magnetic resonance imaging [MRI]; 75% vs 68% in the second round) and abnormal pulmonary function test results (74% vs 68% in the second round) were added to the indications for corrective surgery.

POSTOPERATIVE EVALUATION. Regarding the topic postoperative evaluation, based on the responses in the second round, both physical examination and conventional photography should be included in postoperative follow-up (agreement of 74% and 77%, respectively) besides performing a plain chest radiograph directly postoperatively, on which consensus already had been reached in round 1. Participants agreed that there was no room for diagnostic methods such as CT, MRI, and TEE for routine postoperative evaluation (83%, 88% and 79% agreement, respectively). Round 3 did not lead to withdrawal or the addition of any statement regarding postoperative evaluation.

POST HOC SENSITIVITY ANALYSIS. For round 3, a post hoc sensitivity analysis was performed among participants who had successfully treated >50 pectus excavatum patients (Supplemental Tables 4, 5). The statement on the necessity of TEE in routine postoperative evaluation just fell short of consensus (66%), whereas 81% of the entire participant group did agree that TEE should not be routinely performed. However, consensus was

achieved on 2 additional statements when selecting this participant group. Participants (73% vs 68%) agreed that a cardiopulmonary exercise test should be preoperatively acquired in the case of cardiac complaints. Regarding the postoperative evaluation, participants agreed that body plethysmography (73% vs 68%) should not be routinely performed.

COMMENT

By conducting an international multiround survey, we intended to evaluate consensus and controversies regarding the diagnostic protocol, indications for surgical correction, and postoperative evaluation of pectus excavatum. We compiled 62 statements on which participants were asked to provide their opinion. After the third and final round, consensus was reached on 18 of these 62 (29%) statements.

This study addresses the standard diagnostics as well as additional ones that are advised upon indication of cardiac and/or pulmonary impairment. Next, indications for corrective surgery and what steps should be initiated when patients do not meet these requirements are included. It moreover involves part of the postoperative evaluation and follow-up. Although adhering to the consensus as evaluated in this study in a large proportion of standard cases is recommended, we recognize that experts may deviate for substantiated reasons. Despite such a deliberate deviation, other parts of the study may still be applied.

PREOPERATIVE EVALUATION. One of the largest pectus excavatum series published to date concerns the study of Kelly and colleagues,¹⁵ including 1215 patients. Their standard preoperative evaluation included a thoracic CT, and in part of the patients, a spirometry, ECG, or echocardiogram. The first mentioned was primarily obtained to calculate the Haller index, for which a threshold of 3.2 was applied as indication for corrective surgery. In contrast, our results show that there is no room for the Haller index as indication for surgery. This is also reflected by the fact that no consensus was obtained on the use of CT and plain chest radiographs during the preoperative evaluation.¹⁶

In the current study, a clear distinction was made among patients experiencing physical symptoms. For patients with pulmonary complaints, spirometry is advised, whereas an echocardiogram and ECG are advised in the presence of cardiac complaints. Nonetheless, we acknowledge that for symptoms such as dyspnea, it can be hard to differentiate between an underlying cardiac or pulmonary problem based on anamnesis. Therefore, a comprehensive evaluation by spirometry, ECG, and echocardiography may be advocated in the presence of cardiac or pulmonary complaints given the established relationship of the cardiopulmonary system.

This contrasts with a portion of the surgeons who advocate that the pulmonary implications of pectus excavatum are negligible. The minimal improvement of pulmonary indicators, which are already within the limits of expected values, is imputed to enhanced breathing mechanisms after removal of the bar as airway or pulmonary disease is absent in most patients.^{17,18} They refer to the pulmonary overcapacity of the human body and attribute symptoms such as exertional dyspnea to the inability to linearly increase cardiac output due to external compression causing right-sided diastolic dysfunction. This is strengthened by worsening of the pulmonary indicators during the in situ period of the Nuss bar, caused by increased rigidity of the chest wall, whereas experienced exertional dyspnea during exercise does not worsen during this period.¹⁹

It is interesting to note that no consensus was achieved on the necessity of cardiopulmonary exercise testing in the preoperative evaluation by the overall participant group. Solely cardiac function tests (ECG and echocardiography) at rest were recommended, whereas many patients only experience cardiac complaints during exercise due to the inability to increase cardiac output, as mentioned before. This is also reflected by available literature, where an abnormal cardiac function may only come to light during exercise. Also, post hoc sensitivity analysis revealed that criteria for consensus on the necessity of cardiopulmonary exercise testing were met when selecting participants who successfully treated >50 pectus excavatum patients.

INDICATIONS FOR CORRECTIVE SURGERY. In the same study by Kelly and colleagues,¹⁵ a set of criteria and indications for surgical correction were proposed. Patients had to be symptomatic, present with a severe pectus excavatum, and had to fulfill 2 or more objective criteria to qualify for surgical correction. These included the presence of cardiac and/or pulmonary compression on CT or echocardiography, a Haller index of ≥ 3.2 , restrictive lung disease, mitral valve prolapse, or arrhythmia. They added 2 additional criteria, namely, significant body image disturbances and failed previous repair.

In contrast, the supported view of participants in the current study did not identify pulmonary compression or failed previous repair as indication for surgery just like the Haller index, which was already previously mentioned, whereas progression of the deformity was added to the list of indications. Moreover, only 1 requirement was deemed a sufficient reason for corrective surgery by participants.

It is noteworthy that cardiac compression on imaging was identified as a criterion for corrective surgery, especially since historically it has often been evaluated by CT or cardiac MRI, neither of which has a place in preoperative evaluation based on current expert

consensus. Nevertheless, signs of cardiac compression are also often visible on an echocardiogram, which does form part of the suggested preoperative evaluation in case of cardiac complaints. In addition, the criterion holds in case of deliberate deviation and use of additional alternative diagnostic methods.

SURVEY CHARACTERISTICS. A multiround survey is an effective tool to create consensus by allowing multiple experts from a multitude of different countries and medical centers to remotely submit their opinions on a specific subject. The anonymity, as applied in this study, allows for a nonconfrontational manner of data collection. Furthermore, it allows the investigator to retain control over the feedback provided by the participants and value each participant's feedback equally, which may reduce bias. These 2 aspects allow both dominant and submissive participants to submit their opinions without being weighed.

Regarding the minimum number of samples required for creation of a consensus statement, Akins and colleagues²⁰ showed that a panel consisting of only 23 experts can produce a stable conclusion when a Delphi method is applied, under the condition that all experts have a sufficient understanding of the field of interest (ie, knowledge and practical engagement with the issue). Akins and colleagues²⁰ moreover concluded that the Delphi method can be used in practice fields where the population of experts is limited. In addition, they state that small samples also suffice in fields with many available experts and can be applied when the use of a small sample is more practical.²⁰ Overall, sample sizes of 10 to 100 participants are commonly accepted.²¹

The construct of the multiround survey as conducted in the current study is similar to a Delphi survey; however, wide selection criteria for participants were applied. Chest wall disorders, including pectus excavatum, can be classified as a rare entity treated by a limited group of experts worldwide. In this study, 57 of the initially invited 313 participants participated in all 3 rounds. Most of the participants are allied to the Chest Wall International Group, a large scientific association for thoracic wall disorders. The subjective representativeness of our sample is demonstrated by the fact that 73% of experts have >10 years of experience treating pectus excavatum patients and represent a multitude of countries. Also, post hoc sensitivity analysis on participants who treated >50 cases revealed only a slight deviation from the results of the overall participant group.

Conversely, it must be noted that there are no available guidelines on how to objectively value the representativeness of an expert sample. The single study available regarding adequate caseload for those directly involved in treatment of pectus excavatum is the study by De Loos and colleagues²² on the learning curve of the

Nuss procedure for surgical repair. They established that after a 10-procedure proctoring period, the Nuss procedure is a safe procedure to adapt and perform without a complication-based learning curve, while performing at least 1 procedure per 35 days. The number of >50 successfully treated patients during a participant's ongoing career was therefore arbitrarily chosen as a criterion in post hoc analysis in the current study.

Participant fatigue was limited, given that most participants who completed the first round also completed the second and third round (57 of 85).

LIMITATIONS. The current study is limited by its room for interpretation. Even though the survey's statements were carefully formulated by the steering group and adapted based on the comments after round 1, the risk for misinterpretation remains. Especially when working with a diverse expert panel, one must consider the different social and cultural backgrounds that may affect the interpretation of questions. Because there is no face-to-face interaction between participants or between the participants and the observers, there is no room for discussion where these differences in interpretation may come to light and, thus, can go unnoticed. However, participants were encouraged to seek contact in case of any unclarity. In addition, the concern of reliability remains for consensus statements; if 2 samples of experts receive the same set of questions, they may not come to the same consensus.

Further detailed research on the different topics and statements included in this study is recommended. The current study, for example, does not address to which degree a cardiopulmonary function test result should be abnormal, or the nature of the abnormality observed, for it to be valued as an indication for surgical correction of the deformity. Also, consensus was not reached for all topics within and should be further explored. In addition, solid objective scientific reports remain ahead of consensus statements such as the present study. Future studies are needed including assessment of criteria to initiate conservative treatment of pectus excavatum.

INTERPRETATION. Through means of an international multiround survey among those who are directly involved in treatment of pectus excavatum, consensus and controversies regarding the diagnostic protocol, indications for surgical correction, and postoperative evaluation of pectus excavatum were evaluated.

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Karel W. E. Hulsewe reports a consulting or advisory relationship with Johnson & Johnson and being a board member of the Dutch Federatie Medisch Specialisten (Federation of Medical Specialists), a federation of the 33 professional associations for medical specialists in the Netherlands. Yvonne L. J. Vissers reports a consulting or advisory relationship with Johnson & Johnson and being a board member of Nederlandse Vereniging voor Longchirurgie (NVVL). Erik R. de Loos reports consulting or advisory relationships with Johnson & Johnson and Baxter, and reports being a board member of the Dutch Advanced Trauma Life Support Association and Faculty of the European Society for Thoracic Surgery's Chest Wall Course. Frank-Martin Haecker reports being an executive board member and past president of the Chest Wall International Group. Jose Ribas Milanez de Campos reports being an executive board member and current president of Chest Wall International Group. The other authors have no conflicts of interest to disclose.

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