Cardiopulmonary Function in Relation to Pectus Excavatum

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Abstract

Pectus excavatum (PE) is the most common, congenital deformity of the anterior chest wall and represents around 90 % of all anomalies of the anterior chest wall. PE has been very well investigated over the years with a vast amount of studies being produced, still, no consensus has yet been reached on the direct impact of PE on cardiopulmonary function.

Keywords

Cardiopulmonary function • Pectus Excavatum • Chest wall deformity • Congenital deformity • Anterior chest wall

Pectus excavatum (PE) is the most common, congenital deformity of the anterior chest wall and represents around 90 % of all anomalies of the anterior chest wall. PE has been very well investigated over the years with a vast amount of studies being produced, still, no consensus has yet been reached on the direct impact of PE on cardiopulmonary function.

The question of a possible impact on cardiopulmonary function was raised as early as in the 1920s, where the German thoracic surgeon Dr Sauerbruch [1], reported on a young adult who was suffering from PE and complained about increasing dyspnoea and being exhausted easily. The patient underwent surgery and some years following the surgery, he reported back with clear subjective improvements and was now able to work for hours a day without being exhausted as easily.

Many other cases like that of Dr Sauerbruch have since been presented, where patients preoperatively present with symptoms of varying degree, with more than 60 % presenting with exercise intolerance, lack of endurance and shortness of breath [2]. Through the last 20 years, after the minimal technique by dr. Nuss was introduced

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[3]; a great amount of patients in their early teens have undergone corrective surgery with cosmetically great results, and therefore the question has never been more interesting than now. In several papers, where patients and parents describe the changes of the patients' lives following surgery, it is well-documented that it is not just the appearance that changes, but also their physical exercise capacity. Likewise, any preoperative symptoms that may have been described are reported as nearly disappeared or decreased following surgery. Accordingly, many studies have focused their investigation on this change happening with the patient undergoing surgery.

The psychosocial change reported following surgery has recently been examined thoroughly by a research group from our institution at Aarhus University hospital in Denmark [4]. With this study, Jacobsen et al. have published results on a large patient-group of 172 children who were all undergoing the minimally invasive Nuss procedure. They were asked about their health-related quality of life following corrective surgery for PE, and the same type of questionnaire was also handed out to a healthy control-group of 387 age-matched schoolchildren. All the participants were between the ages of 8–20 years. Uniquely for the patients, they were also asked to fill out the Nuss Assessment Questionnaire, which retrospectively investigated possible changes in perceived physical and psychosocial aspects following the Nuss operation.

The Nuss Assessment Questionnaire found the patients to report of an increased self-esteem and body-concept postoperatively. The other questionnaire, which looked at differences between the patient and the control group, found that the perceived health related quality of life was higher in the patients compared to controls. Furthermore, the patients even reported of a higher physical functioning compared to the controls. Other studies have found similar results. In North America, Kelly et al. published an article on patients reporting of markedly improved body image and perceived ability for physical activity following corrective surgery [5]. And as recent as October 2014, Kuru et al. [6] found comparable results with that of Jacobsen et al. In short, there can be no doubt of the positive impact that surgery has on most of the patients' body image and self-esteem.

In light of the abovementioned findings, the interesting subject is clearly whether these reported changes following surgery can be attributed solely to the changed body image and bettered self-esteem – or if the surgery also has a physiological impact on the patients' physical capacity? When considering studies based on resting cardiac function in patients suffering from PE, no noticeable impact on the cardiopulmonary function has yet been established [7]. But during exercise, different reports have noted a measurable, decreased stamina among patients [8, 9].

With a study from 2005, Rowland and colleagues [8] examined patients with PE preoperatively, and matched them with age-matched healthy controls. All participants were in their early teens and went through upright bicycle exercise tests. As an important detail, the authors also considered the habitual exercise levels of the participants by handing out questionnaires. The exercise tests showed a decreased cardiac index (cardiac output compared to the individual's body surface area) reached in the patient group during maximal exercise. This could not be attributed to a difference in habitual exercise level between the two groups. Similar results have been noted in other, small-scale studies. However, common for these pre-operative studies are that they lack follow-up testing after the funnel chest has been corrected.

A couple of studies have looked at the patients after follow-up and find there is a sustained significant increase in cardiac performance. Neviere et al. followed 70 adult patients, age 18–62 years, after correction by a modified Ravitch procedure for 12 months and found that the maximal O_2 uptake measured during exercise increased significantly from 77 ± 2 % to $87_{\pm}2$ %, $p<0.01$ [10]. Another group, O'Keefe and colleagues looked at cardiopulmonary exercise function in young adults following surgery after the Nuss procedure [11]. Sixty-seven patients at age 14 years were included prior to surgery. Through increased bicycle exercise testing, their exercise O_2-pulse (a surrogate for stroke volume during exercise) were measured and

compared to post-operative measurements. Following bar-removal, O'Keefe noted a significant increase in O_2-pulse during exercise. They also investigated cardiac index before and after surgery, although without significant difference. It should be emphasized though that this parameter was measured at rest and the findings are thus in line with those of former studies.

Another important aspect that should be pointed out with this study by O'Keefe is that there is no comparable control group. Therefore it is impossible to judge whether the improved exercise function found is caused by the impact of surgery or simply by the concurrent growth of the patients. As noted earlier, they were circa 14 years of age at inclusion and on average 3 years older at the time of follow-up.

At our institution in Denmark, we set out to take all factors in to mind – including a comparable control group, investigated during exercise and tested both before and after surgery. The results after 3 years follow-up were published in 2013 [12]. The exercise capacity of teenagers was examined before undergoing the Nuss procedure and compared to a group of healthy, age-matched control subjects. Furthermore, the habitual exercise levels were documented through all 3 years. Prior to surgery, we found teenagers with PE to have a lower maximal cardiac index (CI_{max}) compared to a group of healthy age-matched controls during incremental bicycle exercise [13]. One year following the modified Nuss procedure patients had significantly increased their CI_{max} during exercise, however still scoring significantly lower compared to the age-matched controls [14].

The same study-population was investigated again after the pectus bar-removal in order to determine whether patients would further increase their cardiopulmonary function to a level comparable with the healthy, age-matched control subjects, and in this way continuously taking growth during the investigational period into consideration. Following the bar-removal the cardiac exercise parameters had increased in the patients to such a level, that no difference existed between the two groups anymore. In other words, the patient group had normalized their maximum cardiac index during exercise.

The exercise results from our 3-year follow-up study are illustrated in Fig. 12.1. When looking at the patient group, a significant increase of 21 % was found within this group over the 3 years – a similar increase could not be found in the control subjects. With these results it is thus emphasized that the change found in the patient group could not solely be attributed to the concurrent growth during the investigational period.

The maximum heart rate reached during the exercise tests did not differ between the two groups during the 3 years. And much like Rowland et al. we also considered the possible different habitual exercise habits of the patients and the control subjects. However, at no point during the 3 years did we find any significant difference between the groups. The increased exercise function could

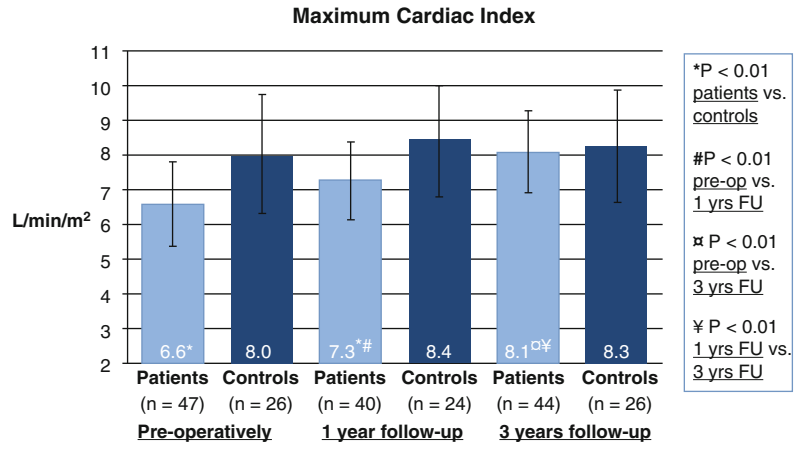

Fig. 12.1 Maximum cardiac index (With permission from Maagaard et al. [12])

thereby not be explained by a higher level of physical activity following the corrective surgery.

The Haller Index was also examined by MRI in both groups throughout the 3 years follow-up study and these measurements showed a significant decrease in the indices in the patient group, with no changes seen in the control group. No difference existed between the groups following bar-removal. But in contrast to the study done by Swanson et al. [15], we did not find any correlation between a decreased cardiac exercise function and a high Haller index. In other words, it was not the severity of the chest wall deformity in our study that determined the level of reduced exercise function.

Echocardiographic studies investigating postoperative results both after the Ravitch and also the Nuss Procedure have shown an increased right ventricular end-diastolic diameter, which might be caused by the decreased pressure from the sternum, causing better filling of the right ventricle. However, these studies are only done at rest [16, 17].

With these results it is illustrated that following corrective surgery, the cardiac exercise function of the patients with PE normalizes compared to a healthy, age-matched control group and also increases the cardiac performance in adults. Surgical correction of PE should be considered in all patients who presents with symptoms of reduced physical performance and not only for patients with cosmetic complaints.

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