



CONGENITAL CHEST WALL DEFORMITIES

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SUMMARY:

Congenital chest wall deformities inhibit patients due to esthetically unpleasant appearance even in the absence of functional cardiac or pulmonary deficit. In most cases the need for correction is based on evident social adaptive difficulties and impairment of worth living sense.

Thoracic scoliosis is seen either with congenital chest wall deformities de novo, or after costal cartilage harvesting, minimal invasive or open correction of chest wall deformities. Anterior chest wall deformities can be put under 5 main categories: Pectus Excavatum, Pectus Carinatum, Poland Syndrome, sternal defects, thoracic deformities seen in diffuse skeletal disorders.

Conservative treatment methods such as using corset are manipulated for starting treatment in childhood and specifically in pectus carinatum. Nuss technique or modifications that are in invasive (Ravitch chondroplasty) or minimal invasive techniques are carried out on elderly people and under the circumstances when conservative methods are unsuccessful. Also vacuum waist technique is one of the treatment methods used in selected cases or for the treatment of the people who do not accept the operation.

In this collection; clinical findings of congenital chest deformities, its classification, embryology, pathophysiology and diagnostic techniques that are mainly used are stated. Also, in company with changing literature; operation indications and traditional surgical correction treatment methods with less invasive new methods are reviewed.

Keywords: Anterior chest Wall deformity, Orthoses, Ravitch chondroplasty, Nuss method.

Level of Evidence: Level V, Review Article

INTRODUCTION

Congenital chest wall deformities can be seen with various anomalies of musculoskeletal system or in a way that it affected isolated costa, cartilage, sternum in various forms. In most of these deformities, there are not certain functional defects in intrathoracic organs and cardiopulmonary and gastrointestinal pathologies that are life-threatening can rarely accompany with it. In almost 6 % of the cases; Marfan Syndrome, Ehlers Danlos Syndrome, connective tissue diseases such as osteogenesis imperfecta and homocystinuria, congenital heart diseases, Down syndrome, scoliosis with the percentage of 26-30% are seen as accompanying diseases⁽¹⁸⁻²⁵⁾. To stage the congenital chest deformities, there is not any classification that is accepted as universal⁽²¹⁾. Those diseases, especially

the patients of Marfan Syndrome, and Ehlers Danlos Syndrome have got scoliosis, thoracolumbar kyphosis and the other spinal deformities⁽²¹⁾. In the Down Syndrome, atlaantoaksial instability is frequently accompanied.

Classically, anterior chest wall deformities can be put under 5 main topics⁽²²⁾:

1. Pectus Excavatum
2. Pectus Carinatum
3. Poland Syndrome
4. Sternal Defects
5. Thoracic deformities in diffuse skeletal disorders;

The deformity ends up with asphyxiating thoracic dystrophy (Jeune Syndrome), spondylothoracic dysplasia (Jarcho-Levin syndrome), cerebrocostomandibular syndrome

along with congenital chest wall deformities and they are deformities that can accompany with different degrees of costa aplesia or hypoplasia ⁽¹⁷⁾.

PECTUS EXCAVATUS

It can also be named as "Funnel chest" and "trichterbrust", "koilosternia", "chone-chondrosternon".

Generally sternum's lower half or two thirds of it get effected. The most distinct area of the deformity is generally at a little above of xiphoid-sternum mergence level. **(Figure-1)**. It is mostly defined 86% at birth or in the first year of life. It almost occurs 2-4 times more in boys than girls. It is the most observed anterior chest deformity with the rate of 1/1000 ^(17,21).

In literature, it is stated that the very first surgical correction intervention attempt was made by Meyers (1911) and Sauerbruch (1913) ⁽²¹⁾. While etiology remains not fully known, it is accepted that the overgrowth of cartilage costa due to the pathogenesis is the key fact of the formation of pectus deformities. Also, it is stated that 37 % of patients with genetic factors have family history ⁽¹⁷⁾. 15% of them have scoliosis and 11 % of these patients have a scoliosis history in their families. The incidence of congenital heart disease is 1.5 % ⁽¹⁷⁾. Patients with Marfan and Prune-Belly syndrome have the incidence of pectus excavatum a lot ⁽²⁷⁾.

Kuhn et al. categorized excavatum types of deformity into 4 in terms of their morphological appearance ⁽¹⁷⁾.

- a. Cup shaped (localized deep depression)
- b. Saucer shaped (diffuse superficial depression)
- c. Grand canyon (depression that is in the form of asymmetrical long pipe)

d. Currarino-Silverman Deformity (pouter pigeon deformity, mixed carinatum / excavatum horns of steer)

For typing and rating pectus excavatum deformity, Welch (1980), Oelsnitz (1981), Hümmer (1984), and Haller (1987) published their studies. One of these authors, Welch, described the rating called "Welch index" by estimating the distance between T3 and T9 vertebra corpus and sternum. Today, Haller index is the most used one.

Surgical Indications:

If a case have two or more criterions below, surgical correction is suggested;

- 1) Pectus excavatum that is progressive or has presence of symptom
- 2) Restrictive lung disease existence that is measured with pulmonary function tests
- 3) Cardiac pressure, pulmonary atelectasis and Haller BT index that is >3.25 **(Figure-2)**
- 4) Cardiac malformations (mitral valve prolapsus or the existence of arrhythmia)
- 5) Nuss pectus excavatum deformity that occurs after an inadequate correction operation.

In many patients, primary indication occurs due to cosmetic and psychosocial reasons. The views of family, child and the environment towards the deformity should be evaluated well. It must be taken into consideration that the deterioration will increase at later ages.



Figure-1.-a,b,c Three cases with difereent degrees of pectus excavatum deformity (Courtesy of Levent Cansever, MD)

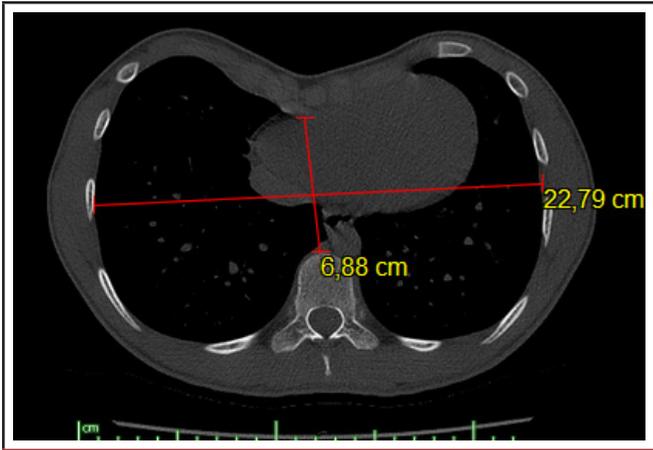


Figure-2. Haller index: maximum latero-lateral distance/shortest anteroposterior distance.

PECTUS CARINATUM

It is the protrusion deformity of chest wall, also called as “pigeon breast” and “chicken breast” and identified by Brodtkin for the first time (**Figure-3.a-b**). This is the second most often seen deformity among chest wall defects (frequency of 5 % in all deformities) and most of them become clear in middle childhood period. It has a genetic predisposition and approximately ¼ of the patients have family history of chest wall deformity; congenital heart disease, marfan syndrome, scoliosis (15%), kyphoses and musculoskeletal defects can be seen ^(17,27).

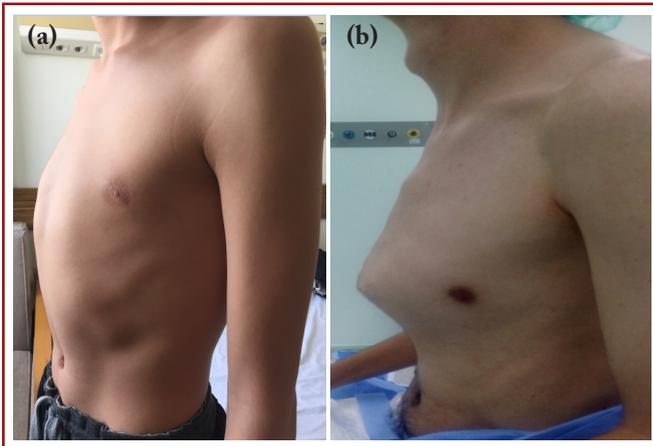


Figure-3.a,b. Two cases with pectus carinatum deformity

Pectus carinatum consists of single-sided or bivious involvement of costa cartilages and deforming spectrums that is on sternum’s upper or lower eminentia. Complex deformity can also be seen. It may occur due to cartilage depression at one side, cartilage protrusion at other side and rotation of sternum.

The most seen clinic appearance of this deformity is the protrusion of sternum object and the symmetric protrusion of

lower costal cartilages. Asymmetrical deformity which occurs due to unilateral protrusion of costa cartilages appears less frequently and mixed type deformity is seen less often.

Also, upper or kondromanubrium deformitie can occur although it is unusual. Here, manubrium prominence and upper costal cartilages are held and the relative depression of sternum object is seen.

An etiology of pectus carinatum is unknown. It occurs three times higher in boys than girls. Unlike pectus excavatum, it generally appears in childhood and adolescence. Pectus carinatum is seen only in 3 patients in labour. Almost half of the patients face it at the beginning of puberty.

26 % of the patients who suffer from pectus carinatum have a family history having chest wall deformity. 15 % of the patients also carry scoliosis and 12% of them have family history including some level of scoliosis. The patients having scoliosis or quite severe deformity can be suspected of having Marfan syndrome.

Pectus Carinatum is put under 4 types called Kondroglandial symmetrical type, Kondroglandial asymmetrical type, mixed type including Carinatum and excavatum together and Kondromanubrial type (Pouter Pigeon, Currarino-Silverman syndrome). Kondrogladiolar type is the most seen type with the percentage of 89 % ⁽²²⁾. It is like the middle of sternum (gladiolus), the lower part of it and the forward protrusion of costal cartilages in that area ⁽²⁰⁾. With this deformity type, lateral depression (small burn, Harrison’s sulcus) typically exists in costas ⁽¹⁷⁾.

MIXED CARINATUM AND EXCAVATUM

On one side carinatum deformity is observed, on the other side excavatum deformity or depression together with sternal rotation is noticed.

CHONDROMANUBRIAL

It is the most rarely seen form. This form is typically named as pigeon chest deformity and in this form, while protrusion bears on upper sternum corpus part and upper 2nd and the 3rd cartilage costas, the lower part of sternum purports as depressed. Cardiac anomalies are seen more in this form.

POLAND SYNDROME

In 1841, this syndrome took part in literature the publication of Alfred Poland, a medical student in Guy’s Hospital studying on an old convict George ELT, 27. He realized that cadaver does not have major and minor muscles in his cartilage and that there is an anatomic deformity characterised with syndactylia. Since 1962, patients who have this illness have been called as Poland Syndrome.

Poland syndrome is characterized with mamma retention, the full absence of papilla (amastia) or athelia hypoplasia, hypoplasia of subcutaneous tissue, subcutaneous oil and axillary hair development together with scantiness of pilosity on the front chest wall, lack of costa, the absence of costosternal piece of pectoralis major muscle or its hypoplasia, the absence of pectoralis minor muscle and costal cartilage, the hypoplasias or aplasias of serratus, external oblique, latissimus dorsi, infraspinatus and supraspinatus and upper extremity anomalies (syndactylia, brachydactyly or ectromelia) that accompany on the same route. It occurs 2-3 times more in men than women. Although its incidence is reported differently on varied series, it's average is between 1/30.000-1/32.000. It is seen twice as much on the right side than on the left side ^(3,10,19,20).

Although there is not exact conclusion about its etiology, there are two main hypotheses on certain periods of pregnancy. The first one among these is mesodermal plateau damage that is thought to appear in 3-4 weeks of pregnancy or developmental disability ⁽²⁴⁾, or the other one, which is the cut of blood stream in subclavian and vertebral systems during the 6-7 weeks of pregnancy ⁽²⁾. Besides familial facts have rarely been reported in literature, "late dominant germ cell mutation" is also a claim that is being discussed ^(3,5,6).

STERNAL DEFECTS

These defects are less commonly encountered compared to pectus type deformities. Especially the situations where the heart is located outside the thorax are considered lethal ⁽⁸⁾.

1. Basic Cleft Sternum:

The heart is in correct position however, there is a defect in sternum joint located at the frontal thoracic wall. Usually the surrounding tissue and pericardium carries wild type traits. The pulsations from the heart are clearly visible at the sternum defect area. There is a complete or partial separation in the sternum. In the cases of crying or valsalva maneuver, the deformities appear more visible ^(12,27).

2. Ectopia Cordis:

This case is quite rare (5-7 % per million organisms). The most common types are thoracic and throracoabdominal.

Cervical Ectopia Cordis:

Can only be distinguished from Thoracic Ectopia Cordis by the superior positioning size of the heart. Cervico-facial anomalies are common. This case is lethal.

Thoracic Ectopia Cordis:

First case was reported by Stenson (1671) ⁽²⁰⁾. In this situation, the newborns have the contractile heart located outside

without the surrounding tissue. Usually bears additional cardiac malformations. In addition, probands may also show abdominal wall defects such as omphalocell, diastasis recti and eventration.

Thoracoabdominal Ectopia Cordis (Cantrell's Pentalogy):

Sternum is incised and the heart is outside the thorax in the front. This anomaly was first reported by Wilson (1798) and the pentalogy was described by Cantrell (1958). This anomaly consists of distal sternal cleft, ventral omphalocell, absence of anterior diaphragm, absence of pericardium facing the diaphragm side, cardiac abnormalities (VSD, Fallot tetralogy, ventricular diverticule etc.). Invasive methods after prenatal ultrasound scans could not decrease the mortality rates in these cases. In addition to the lethal effects of all the defects listed, pulmonary hyperplasia can also lead to increased lethality ⁽²²⁾. The initial operative methods should consist of targeting the tissue defects in the abdominal cavity and the heart. So as to prevent infections and mediastinitis, the primer excision and tissue coverage of the omphalocell should be preferred. The compensation of the abdominal wall can be achieved by mobilizing the wall with flebs or supporting of the wall with prosthetic materials. Repair of this locus is easier than Thoracic Ectopia Cordis. Moreover, repair of the cardiac defects should be performed before the heart is supported by prosthetic materials.

OTHER DEFORMITIES

Can progress as parcial development or agenesis in more than one costa. Bifurcations and fusion abnormalities are common in costas. These abnormalities usually do not cause functional or esthetical defects. In advanced cases, pressure can form around cifoeskoliose and intrathoracic organs.

Thoracic Deformities in Diffuse Skeletal Defects

Asphyctic Thoracic Dystrophy (Jeune Disorder):

This is an otozomal recessive disease with no shown chromosomal abnormalities. There is a bell-shaped rigid thorax and a remarkable abdominal structure. The transverse and the antero-posterior diameter of the thorax is shorter. The ribcage is short and wide, and horizontally positioned. All bones are shorter and wider. Extremities are also shorter and pelvis is narrower.

Spondylothoracic Dysplasia (Jarcho-Levin Syndrome):

Characterized by its autosomal recessively transmitted defects like multiple vertabrate, rib abnormalities and breathe obstruction. Since vertabratae are short and the presence of fusion in the posterior ribs in the thoracic vertebrae. Rigid congenital kyphoscoliosis is seen. Pulmonary infections are common. Post-natal lethalties are also common.

Cerebralcostomandibular syndrome:

It was reported in 1966 for the first time. The underlying reasons and genetic causes are not well known. There are three primary observations. Mental retardation, gaps between ribs and micrognathia prognosis. 40% of the probands fail to survive due to breathing defects.

TREATMENT MODALITIES IN PECTUS DEFORMITIES

CONSERVATIVE TREATMENT

1-Dynamic compression orthoses (Argentina brace)

Treatment with dynamic compressive orthoses (DTC – dynamic thorax compressor) was first described in 1979 for pectus carinatum (9,13). Treatment with DTC orthoses, which have screws on the sides instead of velcro or fasteners and allow for the gradual compression of protruding areas, was described by Haje et al. not only for pectus carinatum, but also for pectus excavatum, starting in 1992 (4). In 2006, the authors synthesized the description of the method by publishing the term “dynamic remodeling (DR) method” to designate the use of DTC orthoses during exercises that promote increased intrathoracic pressure. The overall percentage of children and adolescents with (25) and (15) improvement was 60.6 %.

Especially, these corsets are suitable for patients with pectus carinatum deformities (Figure-4.a,b) These are also recommended for patients for whom nucs pectus carinatum or operation options are not available. In a prospective recent case study with 114 dynamic pectus carinatum cases using a compression corset, $8,80 \pm 3,94$ month usage led to 64 % success and 15 % fail rates with 21 % of the patients with necessity of continuity. In addition, asymmetric and elderly patients who fail to use the corset regularly showed no progression of elimination of the deformities (9).

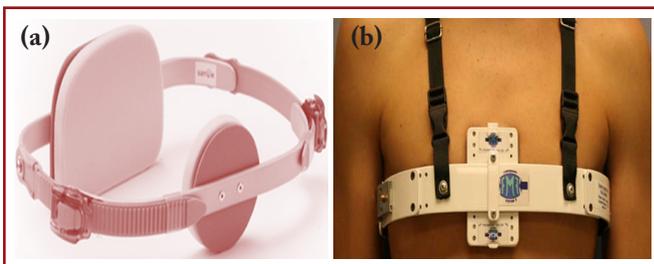


Figure-4.a,b. Dynamic compression orthoses

2- Vacuum Bell Treatment:

It was first described by Eckart Klobe. This method is a major non-invasive approach where symmetrical and light operational methods fail in cases with pectus excavatum. It consists of a system with a vacuum and is applied on the chest and is adjustable to fit (Figure-5).

In the first treatment session 15 minutes of application is sufficient. The patient can continue applying the treatment at home under supervision from a physician. 1-2 hours of application a day was reported sufficient for young and adult patients.

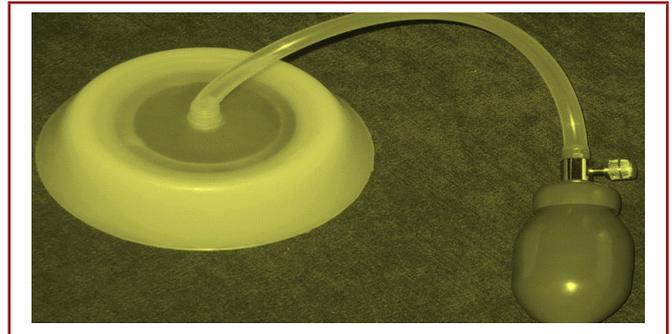


Figure-5. Vacuum bell system

SURGICAL APPROACH

Open Surgery

Operational surgery on Pectus Excavatum, was carried out by Meyer (1911) and Sauerbruch (1913) for the first time.

In 1949, the modified method derived from this technique was defined by Ravitch.

1. Excision of deformed rib cartillages with their perikondria
2. Separation of xifoides from sternum
3. Separation of intercostel (rib) bands from sternum
4. Transverse sternal osteotomie (Figure 6 a-e)

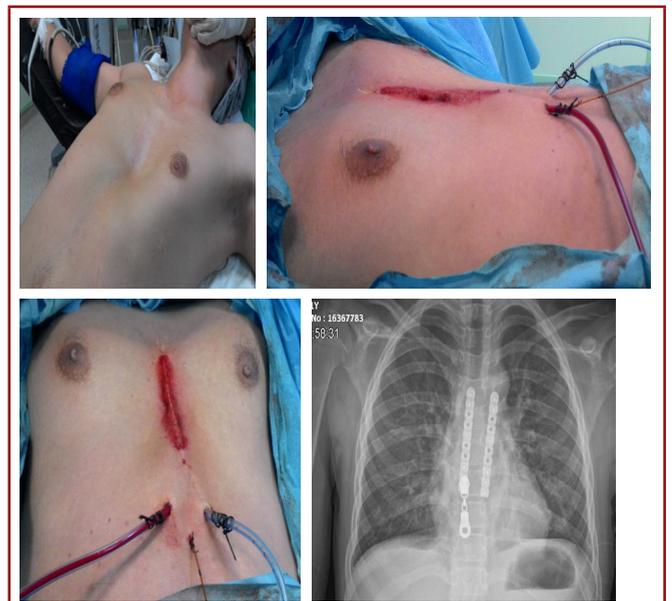


Figure-6. a-d. Operative view of a case who had Ravitch operation due to pectus excavatum. (Courtesy of Ali Cevat Kutluk, MD)

This technique allows swapping the kirschner string with sternum. In 1957 and 1958 respectively, Baronofsky and Welch indicated another technique. In this technique, the main steps are protection of pericondrial sheaths on rib cartillages, dorsal intercostal bands, sternal osteotomie, fixation of the sternum to anterior structures by silk stiches. This technique was reported to have high success rates.

In addition, in 1957, a tripod fixation method was described by Haller. This strategy follows the posterior sternal osteotomie, subpericardial resectioning of the lower cartillage deformities, posterolateral oblique division of normal secondary and tertiary costal cartillages. In 45 cases, the success rate was reported as 100%.

Sternal turnover technique was also reported first in Japan. Sternum was twisted 180 degrees as a free-graft and reattached to costal cartillages. This radical approach was considered limited due to its potential for high complications in children with pectal excavatum. Sternal necrosis is also one of the rare complications.

Operational Repair of Pectus Carinatum

First repair was carried out four decades ago. In 1952, Ravitch reported the repair of condromanubrial bud. He managed this repair by resecting the costal cartillages followed by carrying out double osteotomie in the sternum.

Modern techniques were first applied in 1963. This method consisted of subpericondrial resection of the costal cartillages and resection of lower sternum and the strengthening of the rest of the sternum with the rectus muscle. Afterwards, in 1973, a more elaborate method was developed based on subpericondrial resection of the budding costal cartillages and protection of the whole sternum.

In today's medical approaches, a transverse osteotomie by frontal cortex of the sternum and with replacement of the sternum from anterior to posterior benefiting from the posterior cortex fracture and correction of the frontal protrusion.

Surgery options in Poland Syndrome

In the cases of missing ribs and paradoxal movements of the ribcage, approaches improvement of the skeletal system and support of the ribcage are commonly used. Methods involving repair of the missing ribs, correction of assymetries in the sternum by transverse osteotomie, improvement of stabilization by by marlex mesh besides costa grefts or repair of the chest wall by marlex mesh sandwich method are among the strongest approaches ⁽¹⁹⁾.

in order to solve esthetic problems, autogenous tissue transfer or patient-specific silicone implants are usually recommended. Lately, the methods involving injection of fat as main or side approaches are becoming more and more common ^(6,16).

Implants provide easy and fast options that are also most commonly used. When implants are decided to be used, a pattern is prepared on top of the patient by plaster or a 3-d computer model is created.

Poland syndrome and its side deformities define the patient's functional and esthetic requirements. The priority is to protect the heart and the lungs by increasing stability of the ribcage. Therefore, patient-specific solutions should be evaluated and after considering patient's opinion and requirements, unique methods should be applied supporting the symmetric and secure body shape ^(3,6).



Figure-7.a-d. A case who had Ravitch operation due to pectus carinatum. (Courtesy of Selcuk Kose, MD)

Minimal Invasive Surgery

1-Nuss Method (minimal Invasion in Pectus Excavatum)

A technique was reported with resection of sternum retrosternal barring of the costa cartillages or elevation without division (nuss method) (Figure-8.a-d).

This method was improved after years and an almost perfect version was created (Figure-9.a-c).

In a study carried out by Nuss ⁽²⁶⁾ between 1987-2008 consisting of 1015 cases showed that the most common complication is the spontaneously recessing pneumothroax. misplacement of the bar and infection are also common risks however, the incidence was reported as less than 1 %. The most dangerous complication is the rare cardiac rupture. recovery phase is between 4-5 days. Pain control, pulmonary function physiotherapy and educational support should always be considered in the cases. The epidural catheter to control the pain is critical and can be kept for 2-4 days depending on the situation. For maintenance

of the situation, oral or parenteral analgesic administration is recommended.

After discharging of the patient, a 6-week restriction plan is applied on physical activities and exercise including an initial 2-3 week ban for work/school activities.

The bar on the pectus is usually removed under general anesthesia after 2-4 years without any reported complications so far. Patients are sent home after 2 hours post-operation. Reoccurrence rate is below 5% after all these implications are carried out.

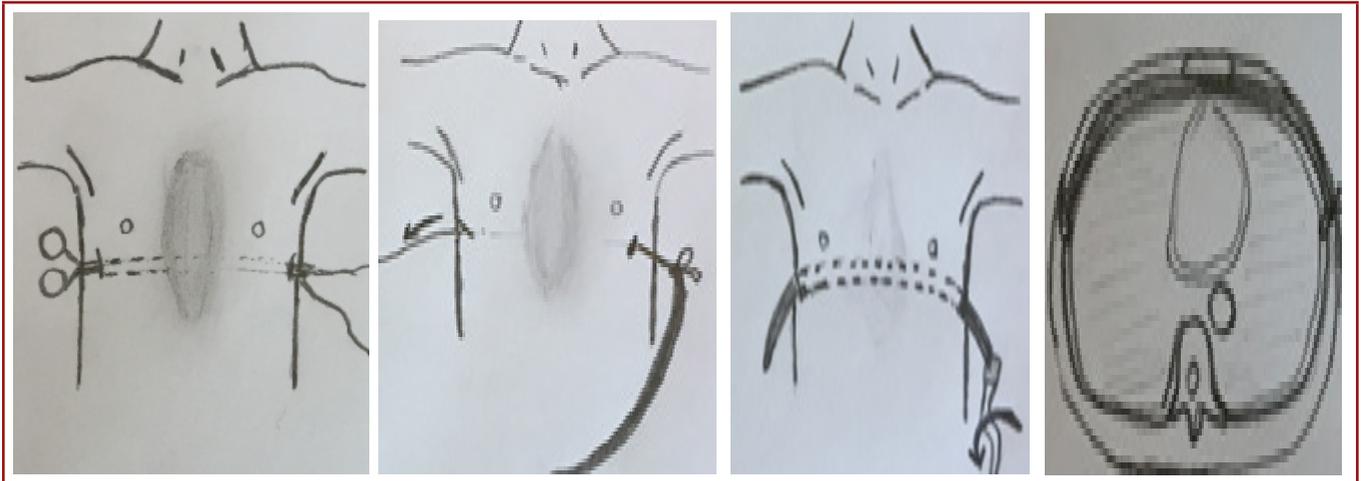


Figure-8.a-d. The steps of Nuss operation

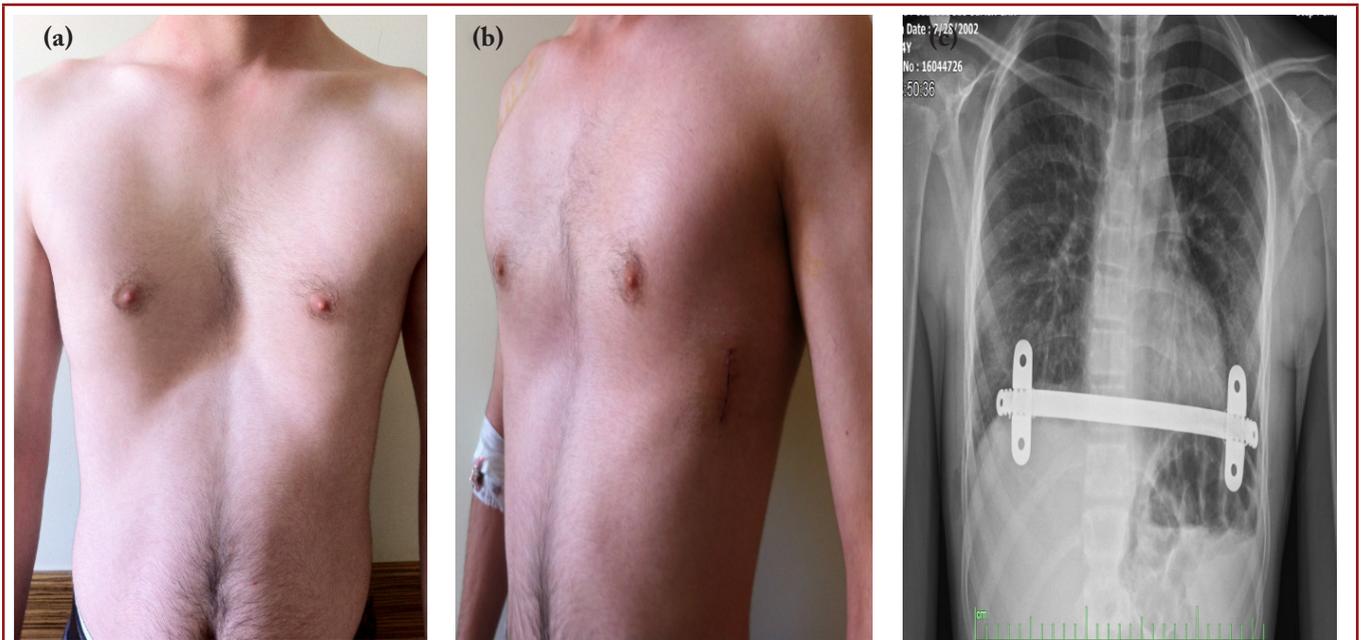


Figure-9.a-c. Preoperative view of a case with pectus excavatum (a). Postoperative radiograph and view after Nuss operation (b-c) (Courtesy of Levent Cansever, MD)

2- ABRAMSON METHOD (Minimal Invasive Surgery on the Pectus Caritanum)

Dr. H. Abramson is the first scientist to perform a modified version of the Nuss method on 40 pectus caritanum patients and present the results in scientific congresses ⁽¹⁾. (Figure-10.a-c).

In this technique, quite similar to Nuss method, success rate is high in pectus caritanum patients as mentioned. In this method, the tunnel formed with the bars is used to pressure the sternum where the budding is most visible and on both sides, attached to the ribs by steel strings using screws. The bar remains attached for approximately 2-3 years and removed under general anesthesia together with the screws ⁽¹³⁾.

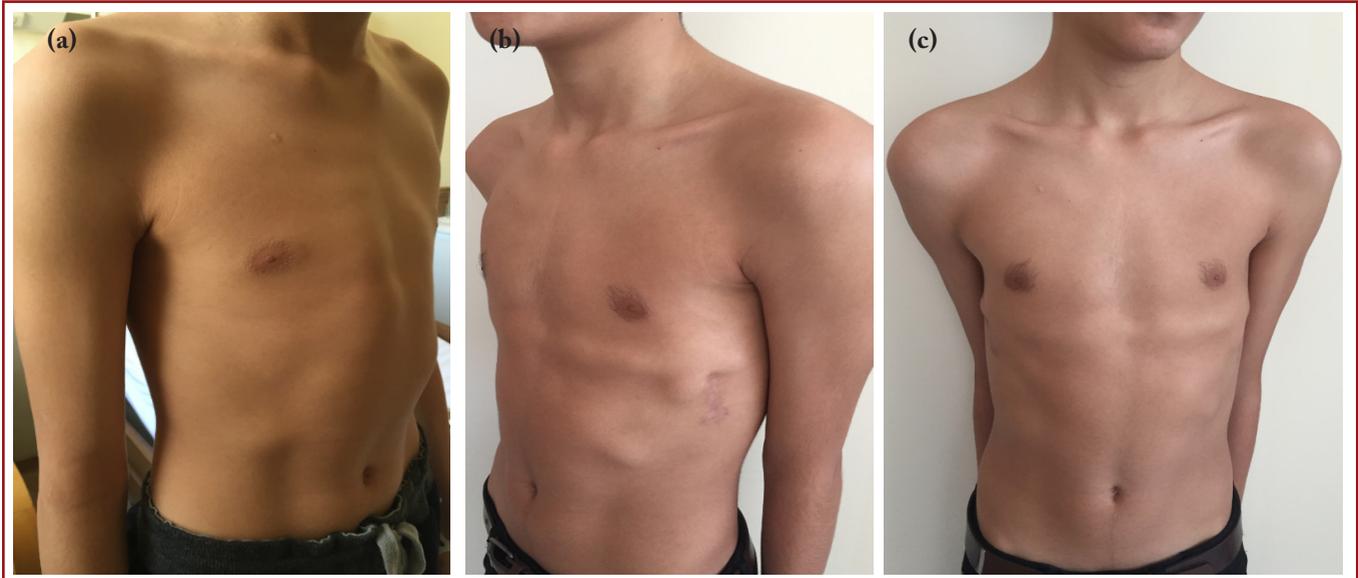


Figure-10.a-c. Preoperative view of a case with pectus carinatum (a). Postoperative view after Abramson operation (b-c) (Courtesy of Levent Cansever, MD)

CONCLUSION

Anterior chest wall deformities, known universally as pectus deformities, are often observed in medical practice. These deformities are usually hidden by patients due to psychological problems, allowing them to remain unknown. As recently as 1990, anterior chest wall surgery was considered to have matured with no new innovations. Suddenly this has become an exciting and dynamic area of surgery with new ideas and innovations being prescribed at conferences and in medical journals almost on a monthly basis. Since the publication of Donald Nuss about the success of the minimally invasive repair of pectus excavatum (MIRPE) in 1998 the demand for correction of all sorts of thoracic wall deformities boomed almost all over the world ⁽⁴⁾.

Scoliosis of the vertebral column is associated with chest wall anomalies. There is however still lack of knowledge about the pathogenesis of thoracic anomalies going along with scoliosis, whether they are expression of a secondary reactive deformation based on compelled biomechanical forces due to heavily distorted and multiaxially shortened thorax or if they are genetically contingent and accompanied with the scoliosis deformity itself. Likewise pectus excavatum deformity has been reported to be associated with thoracic scoliosis in 15-20% of

cases. Moreover thoracic scoliosis can usually be seen after open correction of chest wall deformities but it has been reported even after minimal invasive (Nuss) correction of the deformities⁽²³⁾.

The conservative treatment option for pectus deformities is based on the principles of Nicolas Andry, considered “the father of orthopedics,” and the effects of this treatment can be explained by Julius Wolff’s law of bone remodeling. Therapeutic forces applied regularly on deformed bones and cartilage may produce a gradual remodeling in a beneficial and corrective direction, and this can be observed especially in the anterior chest wall, which is a flexible region ⁽¹⁴⁾.

The MIRPE due to several advantages remains as the ideal therapeutic option in childhood and adolescence, even in selected cases in adulthood. Nevertheless the more rigid and more severe the deformity appears beyond puberty, the hybrid technique MOVARPE seems to represent an alternative method with lesser pain periods, lesser pectus bar implantation period, and a lower rate of common complications, to be noted at first a tilting of the bar ^(7,11).

Nevertheless, many of the techniques described so far should be performed only at specialized centers, which fulfill the requirements of broad experience by sufficient numbers of

cases treated. However, conclusive studies are still lacking due to variable surgical expertise or inhomogeneous patient collectives, thus the proper selection of the most appropriate techniques for a multitude of indications sometimes remains cumbersome.

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