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HISTORICAL ASPECTS OF PECTUS EXCAVATUM

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ABSTRACT

Pectus excavatum (PE), also known as "funnel chest," is the most common congenital malformation of the chest wall, with historical references dating back to ancient Egypt. This article explores the historical evolution of PE, tracing its recognition, terminology, and medical interpretations over time. Early descriptions emerged in the 16th century, with significant developments in understanding its etiology and treatment occurring in the 19th and 20th centuries. Various hypotheses have been proposed regarding its causes, ranging from genetic predisposition to diaphragmatic contracture and skeletal abnormalities. The surgical treatment of PE has evolved from early attempts at costal cartilage resection to modern minimally invasive techniques, particularly the Nuss procedure. This review highlights key historical milestones, influential figures in the field, and the progressive refinement of surgical interventions that have shaped the current approach to managing pectus excavatum.

KEYWORDS: Pectus excavatum, Historical review, Thoracic surgery.

INTRODUCTION

The excavated chest (pectus excavatum) (PE), also known as "funnel chest," is the most common congenital malformation of the chest wall. In English medical literature, the Latin term "pectus excavatum" is primarily used for this malformation, which German authors often translate as "trichterbrust" [1]. In 1934, Snyder named this condition "koilosternia," derived from the Greek words "koilos" (hollow) and "sternon" (chest), and the Greek translation "chone-chondrosternon" was proposed in 1938 by A. Ochsner et al. Other names for the malformation include thorax en enton noir, pectus infundibuliforme, cobbler's chest, and Cobbler's chest [2, 3, 4, 5]. Brodkin H.A. (1948) suggested the name congenital chondrosternal depression [6]. Brown A.L. (1939) referred to pectus excavatum as a chest deformity characterized by a funnel-shaped deformation of the sternum, with its apex located approximately at the level of the junction of the xiphoid process with the body [7].

The first image of the pectus excavatum deformity, dating back to approximately 2400 B.C., comes from ancient Egypt and depicts the relief of a man with an anterior chest wall deformity [8].

Some authors, upon reviewing CT scans of ancient Egyptian mummies, have identified cases of pectus excavatum (PE) previously reported by medical historians. This confirms the suspicion of a notable absence of descriptions of PE from antiquity, even though the malformation was quite common during that time [9].

In Europe, the oldest preserved skeletal remains with pectus excavatum were discovered among the 48 skeletons found in Hungary in graves dating from the 10th to the 16th centuries [10].

Individuals with pectus excavatum can also be seen in paintings by Leonardo da Vinci (1452-1519) and Jusepe De Ribera (1591-1652) [11, 12].

The first descriptions of this anomaly appeared only in the 16th century. Schenck J. (1530-1598) was the first to gather data from the literature and provide a description of this anomaly, which was identified by Bauhinus J. and is considered the earliest cited report. In 1594, Bauhinus J. published a classic work in which he described the clinical manifestations of a 7-year-old child who had difficulty breathing and paroxysmal coughing due to severe pectus excavatum, with a risk of suffocation. He believed the condition was caused by a short diaphragm [7, 13].

In 1863, von Luschka reported the case of a 24-year-old man with a 6 cm deep depression in the anterior chest wall. In 1870, Eggel published the first case report of a patient with a funnel-shaped chest depression, which he called "miraculum naturae," assuming that the cause of the deformity was a weakness and abnormal flexibility of the sternum, resulting from nutritional disorders or developmental insufficiency [14].

Coulson W. (1820) was the first to highlight the genetic predisposition of this deformity after identifying three brothers with pectus excavatum from the same family. Later, Williams C.T. (1872) described a 17-year-old patient born with pectus excavatum, whose father and brother also suffered from the same condition [15]. Subsequently, the familial predisposition to this deformity was demonstrated by Lester and Sainsbury, who believed the defect was usually recessive, although in one family it manifested as a dominant trait [18].

In 1860, Woillez E. suggested that the cause of the deformity of the anterior chest wall was a contracture of the central diaphragmatic tendon, which had been previously mentioned by Bauhinus J. in 1609. Bauhinus had documented observations of diaphragmatic traction in 1596 [16].

The first who described pectus excavatum as a medical condition was Eggel (1870), and early treatment for it was outlined by Ebstein W. in 1882. This treatment included breathing exercises, fresh air, sleeping on the side, and other measures [5, 17].

Hagmann described the deformity in 1880, and in 1882, Wilhelm Ebstein reported five cases of this condition and proposed the term "trichterbrust" (funnel chest) [19]. According to Brown A.L. (1939), the modern name and interest in this thoracic deformity are due to the comprehensive presentation made by Ebstein W. in 1882 [7].

The etiology of pectus excavatum has long been a point of controversy among scholars. Ebstein W., citing Woillez E. (1860), considered the malformation to be the result of contracture of the central tendon, similar to how Dupuytren's contracture affects the fingers [6, 13]. Brown A.L. (1939) noted that Eggel, in 1870, among the many theories regarding the possible etiology of pectus excavatum,

supported the idea that the comparative elasticity and relatively insecure fixation of the lower ribs could allow the diaphragm to pull the lower part of the sternum inward during inspiration [7].

In 1873, Flesch proposed that the cause of funnel chest was excessive growth of the ribs, an idea supported by Hagmann (1888) and later by Sweet (1944) [14, 18].

Langer and Zuckerkandel (1880) believed that the deformity of the chest was due to insufficient development during the intrauterine period. They suggested that excessive intrauterine pressure causes the fetus's mandible to compress the sternum [14].

Muller C. and Bien G., observing that during normal embryonic development the sternum goes through a stage of depression, proposed the hypothesis that pectus excavatum results from the sternum remaining in this embryonic position. This hypothesis was invalidated by the discovery of normal-length sternums in patients with pectus excavatum [13].

In 1929, Kelley S.W. reported the occurrence of funnel chest in patients with rickets, providing supporting photographs for confirmation. This opinion was not shared by Brown A.L., who, along with Lester Ch. and Brodkin, attributed the cause of pectus excavatum to the shortening of the central tendon of the diaphragm and the action of the substernal ligament. In 1986, Ravitch M. noted that rickets and scurvy, previously considered causal factors of pectus excavatum, likely have no connection to the condition, though no plausible explanation was provided [20, 21].

In 1950, Lester Ch., in a paper published in the Journal of Thoracic Surgery, stated that the deformity of pectus excavatum is caused by a shortened central tendon of the diaphragm, which flattens the dome of the relaxed muscle. When the muscle contracts during inspiration, the lower end of the sternum, to which the diaphragm attaches, is pulled backward, eventually causing a funnel-shaped deformity of the chest wall with the apex at the level of the xiphoid process [22].

The concept of a restrictive retrosternal ligament or a shortened internal tendon of the diaphragm was challenged by Kenneth J. Welch (1958). Gross R.E. (1953) argued that while the substernal ligament appears to be an important factor, it is doubtful that it is the underlying cause of the external depression of the anterior chest wall [20].

In 1899, Tietze A. described the first surgical correction of pectus excavatum in Wroclaw. In the early 20th century, several surgical correction techniques for pectus excavatum were proposed, initiated by Meyer L. (1911), Klapp R. (1912), and Zahradnicek J. (1925) [23].

The surgical correction procedure for pectus excavatum practiced by Meyer L. (1911), described by some authors as the first attempt at surgical correction of the deformity through resection of the costal cartilages, involved the excision of the second and third costal cartilages on the right side. This procedure yielded unsatisfactory results [5, 24, 25].

In 1913, Sauerbruch F., a pioneer in thoracic surgery who developed a negative pressure chamber for chest operations, addressed pectus excavatum by excising the costal cartilages on the left side, from the fifth to the tenth, along with a quarter of the adjacent sternum. This surgery represented progress in the correction of pectus excavatum. In 1920, the surgical technique was modified to include bilateral resection of the costal cartilages and sternal osteotomy with external traction for six weeks [26].

In 1925, Zahradnicek J. treated a 16-year-old boy with pectus excavatum by using two metal wires inserted through the sternum to maintain traction [17].

In 1939, Ochsner A. and DeBakey M. reported a successfully treated case of pectus excavatum, with correction of the deformity maintained one-year post-operation and clinical symptom improvement. At the same time, they presented an extensive literature review at the American Association for Thoracic Surgery, concluding that treatment of this thoracic deformity could be both conservative and surgical [27]. Conservative methods included breathing exercises, postural gymnastics, and orthopedic measures. The authors classified surgical procedures into three groups: 1) chondro-sternal resections, 2) T-sternotomy with or without division and traction of the costal cartilage, and 3) sternal mobilization with division or resection of costal-cartilaginous structures [19].

In 1939, Brown A.L. performed a surgical correction technique for pectus excavatum in infants, based on the removal of ligaments connecting the sternum to the diaphragm [7]. Hausmann (1955) recommended performing this procedure starting from the age of 2 years [28].

With various minor modifications, the surgical technique proposed by Brown A.L. forms the basis of contemporary treatment for this deformity [29].

Ochsner J.L. and Ochsner A. (1966) believed that the ideal age for surgical correction of pectus excavatum was quite controversial [3]. Some authors advocated for performing the surgery before the child reached 2 years of age, while others reported good outcomes when the operation was performed between 2 and 6 years of age. There were also opinions suggesting that surgical intervention should be carried out during adolescence. Haller J.A. et al. (1970) considered the age range of 6 to 10 years optimal for correction, noting that patients under 3 years and those older than 20 years had lower chances of achieving excellent results [30].

Sweet R.H. (1943), applying the principles outlined by Brown A.L., reported two cases of successfully operated pectus excavatum and proposed a few minor modifications to Brown A.L.'s surgical technique. According to Sweet, these modifications were worth considering [31].

In 1949, Ravitch M.M. proposed an approach that, excluding the need for external traction, included radical resection of all costal cartilages, complete detachment of the sternum from the thoracic wall, and a wedge-shaped sternal osteotomy. The shortened costal cartilages were reattached to the correctly positioned sternum with non-absorbable sutures. Ravitch believed that the younger the patient, the greater the likelihood of achieving a completely normal chest contour, with the surgery indicated for removing a cosmetic defect that represents a social handicap. Early surgery was considered important for correcting progressive skeletal deformity and improving cardiac or pulmonary symptoms, serving as prophylaxis against the progression of clinical symptoms [32].

Later, Ravitch M.M. (1965), having performed approximately 164 surgeries for pectus excavatum in children and adults, reaffirmed the basic principles of the operation: subperichondrial excision of all deformed cartilages, complete release of the sternum by dividing the xiphoid and intercostal bundles, transverse osteotomy of the sternal bone, and absence of external fixation. Based on accumulated experience, Ravitch M.M. proposed a modification involving an oblique division of the lowest normal cartilage for sternal osteotomy, with a space higher than the previous form, always performed on the posterior surface of the sternum. Occasionally, the author used internal fixation or support, especially in adolescents or adults with long sternal structures [33].

Lester Ch. (1950) believed that near-complete correction of pectus excavatum could be achieved in cases where surgical intervention occurred before the age of 5. The proposed technique involved

making an incision over the lower end of the sternum to expose the xiphoid and costal arch, followed by detachment of the xiphoid from the sternum through an incision at the xiphosternal junction and entering the anterior mediastinum by dividing the substernal ligament. The correction of the deformity relied on the elasticity of the chest and growth [22].

Humphreys G. and Jaretzki A. (1980) considered that surgical correction of pectus excavatum is rarely indicated before the age of 3 years, with the best results being achieved in children between the ages of 3 and 6 years, although satisfactory outcomes can be obtained at any age [34].

The sternal eversion procedure or sternal turnover, first described by Lexer E. (1927) and continued by Nissen R. (1944), was a completely new concept in the correction of pectus excavatum. It was supported in 1954 by Jean and Robert Judet, then by Jung A. (1956) [35], and later by Wada J. (1970) [36]. This invasive technique involves transverse division of the sternum and costal cartilages, total mobilization of the chest wall with axial rotation of 180 degrees of the bone and muscle graft (sternal turnover), and reattachment of the sternum and costal cartilages through suturing. This procedure was not widely adopted due to major complications in the event of infection and sternal necrosis [21, 37].

Subsequent modifications to the surgical technique for correcting pectus excavatum focused on internal stabilization of the sternum. To this end, Dorner R. et al. (1950) utilized homologous ribs [27], an idea supported by Dailey J. (1952), who used a substernal osteoperiostal rib graft for the repair of pectus excavatum [38].

The modified Ravitch M.M. operation, which did not include external traction, had a higher rate of recurrences. To prevent sternal regression, Wallgren and Sulamaa developed, in 1956, an internal support technique, using a curved steel bar placed subcutaneously across the chest and pushed through the caudal end of the sternum, supporting the sternum against the ribs on both sides. Later, Adkins and Blades placed the bar posterior to the sternum [24]. Over the next 40 years, this reconstructive procedure for pectus excavatum became the preferred technique for patients of all ages [39].

The use of a short support bar together with costal cartilage resection has been widely adopted as the standard repair for addressing this deformity [21].

Welch K.J. (1958) advocates a less radical approach compared to the technique proposed by Ravitch M.M., achieving good results in 75 patients without sectioning all intercostal bundles and the rectus abdominis muscle [40].

The method described by Robiscek F. (1963) involved subperiosteal excision of the cartilages, transverse displacement of the sternum, and excision of the forward-displaced lower segment of the sternum. The rectus abdominis muscle and xiphoid process were then sutured to the newly formed lower part of the sternum. In cases of asymmetric deformities, the author used bilateral cartilage resection and double osteotomy of the sternum [17].

Stanger P. and colleagues (1968) found that reoperations following procedures involving rib cartilage resection were quite challenging due to extensive scarring and the loss of normal anatomical structure, with resected cartilages regenerating into a rigid, plate-like structure. They advocated for the development of a new surgical technique to address recurrent cases of pectus excavatum [41].

Murray P. and Long G.A. (1965) addressed the treatment of pectus excavatum depression by implanting silicone pouches, which restored only the external contour of the chest, while the shape of the thoracic

wall remained unchanged [42]. Despite its drawbacks, the method has been supported over the years by several authors [43, 44].

Some authors were opposed to the resection of costal cartilages in the surgical correction of pectus excavatum in very young patients, considering the development of asphyxiating chondrodystrophy in young patients a complication of this early surgery. Based on experimental studies on rabbits that developed the condition after cartilage resection during the growth phase, they advocated for the development of alternative techniques that avoid the removal of costal cartilage and a reevaluation of the optimal age for repairing this malformation [45].

Professor Haller J.A., in the paper "Thoracic Wall Constriction After Overly Extensive and Early Operations for Pectus Excavatum," highlighted the risk of "acquired asphyxiating chondrodystrophy." Following a review of the literature, most surgeons abandoned open repair of pectus excavatum in young children, preferring to wait until after puberty. Additionally, the amount of cartilage resected was reduced, with the procedure being referred to as the "modified Ravitch procedure" [46].

Considering the flexibility and malleability of the thoracic wall for a considerable period, as noted by Kelly S. in 1929 and reaffirmed by Haller in 1986, Nuss D. concluded in the mid-1980s that there was no need to excise these thoracic structures and that an alternative approach should be found. In 1987, Nuss D. corrected a severe case of pectus excavatum in a 4-year-old child by placing a bar under the sternum without cartilage resection and without sternal osteotomy, achieving excellent correction [24].

In 1998, Nuss D. and colleagues reported the results of their 10-year experience (1987-1996) in treating 148 patients with pectus excavatum using this method [47]. Subsequently, the technique underwent several enhancements, including the addition of stabilizers, thoracoscopic assistance, and sternum elevation [48, 49].

Although there are conservative options [50], surgical treatment remains the only effective method for advanced deformities of the thoracic wall, with the minimally invasive Nuss technique being considered the most popular method for correcting pectus excavatum [51, 52].

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