

1 **Title: Deep venous thrombosis and abortion: an unusual clinical manifestation of severe form of**
2 **pectus excavatum**

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17 ABSTRACT

18 Pectus excavatum is a chest wall malformation with a strong psychological and aesthetic impact.

19 Rarely pectus excavatum patients can show respiratory or cardiac symptoms occurring mainly during

20 physical exertion. We report a case of a 34-year-old pregnant woman with a severe degree of pectus

21 excavatum who developed serious cardiovascular disease resulting in spontaneous twin abortion at the

22 twenty-first week of gestation. Cardiovascular disease was resolved after open surgical correction of

23 pectus excavatum. This case shows how a tardive diagnosis and a delayed surgical approach for pectus
24 excavatum can lead to severe consequences.

25

26 INTRODUCTION

27

28 Pectus Excavatum is the most common deformity of the chest wall (1). Generally, pectus
29 excavatum has only a strong psychological and aesthetic impact, especially in youth and adolescence,
30 showing alterations of their emotional and behavioral sphere, manifesting a real discomfort with their
31 body and aesthetic appearance. In rare cases pectus excavatum can determine the appearance of
32 cardio-respiratory symptoms occurring mainly during physical activity such as arrhythmias, dyspnea,
33 reduced cardiopulmonary fitness, compromised exercise capacity, and fatigue (2). In this article, we
34 describe a case of a 34 years pregnant woman with a severe degree of pectus excavatum determining
35 heart compression and resulting in a spontaneous twin abortion at the twenty-first week of gestation
36 due to a venous thrombosis.

37

38 CASE

39 In December 2019, a 34-year-old woman was referred to our Thoracic Surgery Unit with a severe
40 pectus excavatum (Fig.1A, B, C). According to Chin classification (3), computed tomography (CT) of the
41 chest revealed a type I symmetric PE involving from 4th ribs to 7th ribs with a Haller index of 14.06,
42 compression of inferior vena cava (IVC) and right atrium with thrombotic processes affecting the
43 inferior vena cava up to the superficial and deep femoral system (Fig.2A, B, C, D).

44 The patient did not refer any psychological impairment during her life, but only exertional
45 dyspnea that has been always attributed to asthma, even if not under pharmacological treatment. On

46 March 2019 she experienced a spontaneous twin abortion at the 21st week of gestation, probably due
47 to deep vein thrombosis; no autoimmune disorders (negative anticardiolipin and homocysteine
48 antibodies) or coagulopathies were detected. Therefore, she started therapy with direct oral
49 anticoagulants without obtaining a complete resolution of the thrombotic process, as demonstrated by
50 the echo-color Doppler examination of the lower limbs performed after 6 months. Preoperative
51 transthoracic echocardiogram showed normal ejection fraction (EF: 68%) and the presence of mild
52 mitral and pulmonary valve insufficiency, with systolic retroflexion of the anterior flap of the mitral; in
53 subcostal view the inferior vena cava flow appears reduced and turbulent.

54 Due to the severity of the pectus excavatum, the mini-invasive approach was excluded (4) and
55 she underwent sternochondroplasty by Ravitch's approach through a submammary incision; after the
56 bilateral removal of the costal cartilages from IV to VII ribs we performed a transverse osteotomy in the
57 sternal corpus and placed a steel bar below the sternum to elevate it (Fig. 3A, B, C). Operation time was
58 110 minutes. After three days, postoperative CT showed reduction of compressive effects on the heart,
59 in particular on the right atrium and inferior vena cava, both regularly opacified by the contrast. The
60 antero-posterior diameter of the chest, measured at the deepest level, increased from 19.86 mm
61 (preoperative) to 52.64 mm (postoperative) with a Haller index of 5.08. The patient was discharged on
62 the fifth postoperative day; a 6-months chest CT showed a further improvement both of Haller index
63 (4.73) and the antero-posterior diameter (59.34 mm) (Fig. 4 A, B); the thrombosis was no longer evident
64 at the level of the femoral vein and the patient has discontinued the anticoagulant therapy. Even
65 spirometry performed after surgery showed no signs of obstructive pulmonary disease, demonstrating
66 how the diagnosis of asthmatic disease was inexact. Postoperative echocardiography showed a
67 reduction of mitral and pulmonary valve insufficiency with improvement of cardiac function and the
68 inferior vena cava flow was clearly visualized.

69

70 DISCUSSION

71 Aesthetic discomfort is the main indication for pectus excavatum correction. The pectus
72 excavatum related complications are known until decades and clinical presentation can be various.
73 Hemodynamic complications, arrhythmias, decrease of pulmonary function and dyspnea are rare but
74 possible and they can be considered in decision-making process (4). In our clinical case, however, the
75 patient had never experienced cardiovascular disorders before pregnancy and she never had shown
76 aesthetic or psychological impairment due to the congenital malformation. The mechanism underlying
77 our clinical situation is probably due to physiological modifications occurring during twin gestation and
78 pectus excavatum: the overload on the compressed heart modifies the blood flow balance, determining
79 the IVC thrombosis. The prothrombotic state, induced by pregnancy, typically results as a deep iliac and
80 femoral system thrombosis, and pulmonary embolism is a common and dangerous complication;
81 however, the right atrium and inferior vena cava thrombosis is a rare condition and in this case the
82 pectus excavatum works as a mechanical risk factor. We suppose, on the basis of the preoperative
83 imaging and her medical history, that IVC thrombosis is strong related with the abortion, but the real
84 mechanism is unclear. At the time of spontaneous abortion, the deep iliac thrombosis was
85 demonstrated by echocolor-doppler and the echocardiography shows indirect sign of IVC thrombosis.
86 The patient was referred to our department after 6-month of anticoagulant therapy and CT-scan shows
87 minimal residual thrombosis at atrial-IVC junction; for these reasons we can speculate, but not
88 demonstrate by imaging the direct correlation between thrombosis and abortion.

89 For the first time we present the case of IVC compression due pectus excavatum in pregnant
90 woman who presents abortion and the relationship is not described before in literature. White et al (5)
91 described the case of pectus excavatum with compression of IVC and recurrent syncope in 22 years-old

92 woman; they propose that the compression of the sternum on right atrium and the reduction in upright
93 cardiac output caused the disorder. Iannucci et al in 2015 described the case of lower extremity edema
94 in a child due to pectus excavatum, after 3 months of surgical correction the patient presents complete
95 resolution of her edema and leg pain (6).

96 Although the mini-invasive approach is actually the first surgical choice (7,8), in this case the
97 severity of malformation and the impressive heart compression have suggested a traditional surgical
98 approach. We obtained a progressive increase of the antero-posterior diameter of the chest allowing
99 the decompression of the caval-atrial junction, a correct venous return to the heart and consequently
100 to the systemic circulation with an improvement in the overall cardio-circulatory performance. The
101 patient has no more exertional dyspnea and doesn't show edema in the lower limbs with an
102 improvement of quality of life.

103 In conclusion this particular case shows as a delayed diagnosis of a severe form of pectus
104 excavatum can lead to sudden symptomatic and unfortunately dramatic manifestations. Corrective
105 surgery has a high success rate, but the timing and surgical approach should be determined on case-
106 by-case.

107 Compliance with ethical standard: Investigations is in accordance with the Helsinki Declaration
108 of 1964 and all subsequent revisions.

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133 Figures

134 **Fig.1** Preoperative view of pectus excavatum **a)** frontal plane **b)** sagittal plane **c)** transverse plane.

135 **Fig.2 a)** Preoperative sagittal CT scan showing the deep depression of the sternal corpus **b)** Preoperative
136 axial CT scan with Haller Index **c)** Preoperative coronal CT scan showing thrombosis in the deep femoral
137 system **d)** Preoperative coronal CT scan showing thrombosis in the inferior cava system.

138 **Fig.3** Intraoperative view **a)** Bilateral removal of the costal cartilages (white lines) **b)** Steel bar placed
139 under the sternum **c)** Submammary incision at the end of surgery.

140 **Fig.4 a)** Postoperative axial CT scan at 3th day showing improvement of antero-posterior diameter **b)**

141 Postoperative axial CT scan at 6 months showing further improvement of antero-posterior diameter.