

Prevalence of Pectus Excavatum, Pectus Carinatum and the Most Common Occurring Simultaneous Diseases in About 9,000 Greek Male Soldiers

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Received: March 27, 2017; **Published:** April 05, 2017

Abstract

Objective: To determine the prevalence of anterior chest wall deformities in young Greek male soldiers and to record the accompanying diseases.

Methods: About 9,000 young male soldiers were included in our study. They were aged from 18 to 24 years. They all underwent a thorough clinical examination and a chest X-ray. By soldiers with clinically diagnosed pectus excavatum (PE) and pectus carinatum (PC) further examinations were performed including electrocardiogram (ECG), echocardiography, computed tomography (CT), magnetic resonance tomography (MRI), and lung function tests. All soldiers with PE and PC were additionally examined by a specialist cardiologist, pulmonologist and thoracic surgeon.

Results: Anterior chest wall deformities were detected in 0.22% of the examined soldiers. The ratio of PE versus PC was 5.6:1. In some cases the following diseases coexisted with PE: scoliosis in 23.5%, mitral valve prolapse in 17.6%, ophthalmological defects in 17.6%, right bundle branch block in 5.88%, Marfan syndrome in 5.88%. 47% of soldiers with PE also had first grade family members with PE. All soldiers were asymptomatic.

Conclusion: The prevalence of PE and PC in the examined group was significantly lower compared to the existing data of the current literature (1%).

Keywords: Pectus Excavatum; Pectus Carinatum; Anterior Chest Wall Deformities; Marfan Syndrome; Mitral Valve Prolapse; Soldiers; Greece

Introduction

The anatomical anomalies of the anterior thoracic wall consist an important part of the thoracic nosology. The mainly concerned clinical entities are the pectus excavatum (PE) and pectus carinatum (PC).

PE is defined as the posterior incursion of the sternum and of the adjacent costal cartilages. This is the most common deformity of the anterior chest wall reaching a percentage of 85%. The depression degree of the sternum may vary from person to person. Some patients have a mild grade PE and by others the sternum may almost be attached to the patient's spine [1-3]. There are many hypotheses concerning the development of PE. The today's leading theory focuses on a metabolic defect of the sternocostal cartilage, resulting in a biomechanical weakness and consequently an overgrowth of the sternocostal cartilage [2]. Historically cases of PE have been reported since antiquity. For example, PE was found in mummies of ancient Egypt [4]. However, they were first described as a clinical entity in the 16th century [5]. It has been recently revealed that the first imaging of PE was drawn by Leonardo Da Vinci in 1510 [6].

The diagnostic approach of PE can be done using routine radiology examinations. The first radiological approach is the conventional chest x-ray. PE can be clearly detected in the profile view chest x-ray (Figure 1b). Computer tomography (CT) has general highly contributed to the diagnosis and classification of the diseases of the thorax cavity. The Thorax-CT in these cases may offer a better anatomic definition of the anomaly, a thorough examination of the anatomy of the intrathoracic organs and the calculation of the Haller index (HI), which can be used to set the indication for surgical correction [7]. The surgical treatment has been firstly described by Ravitch. Through the years his technic has been further developed and evolved and many variations have been described [2,8]. A minimally invasive procedure which has been invented and described by D. Nuss in 1987 and is being today widely used. The Nuss procedure includes sliding one or more hollow metal bars behind sternum under thoracoscopic guidance [2] (Figure 1c).

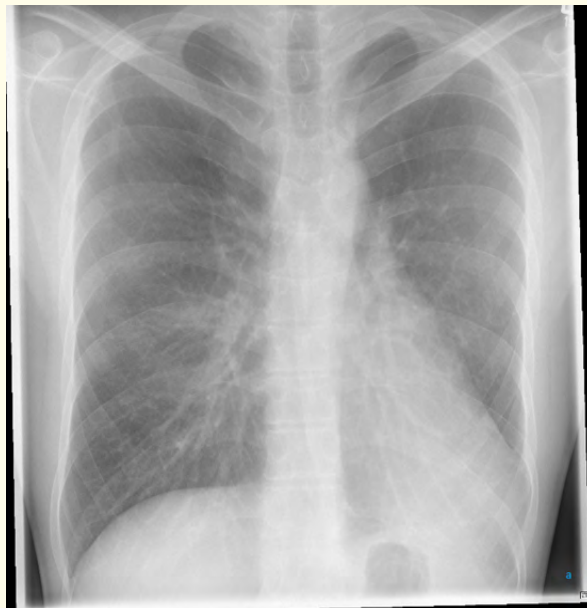


Figure 1a.



Figure 1b.

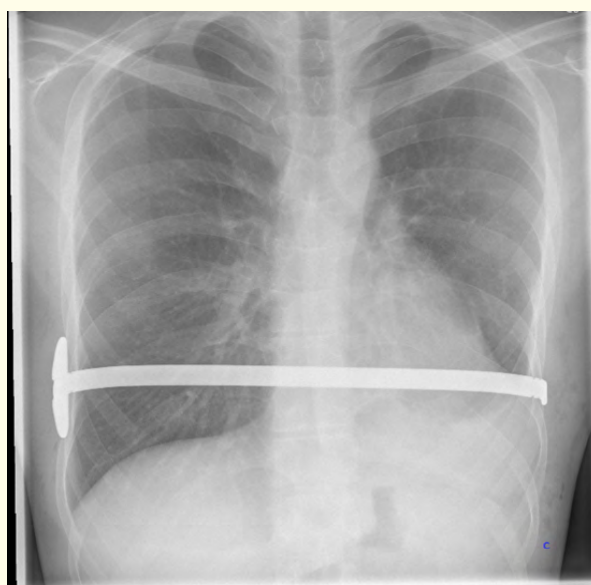


Figure 1c.

Figure 1: Patient with PE. In the posterior anterior chest x-ray a small degree of scoliosis to the right side can be detected. (Figure 1a). In the lateral chest x-ray of the same patient the posterior depression of the inferior sternum can be seen (Figure 1b). The chest x-ray after the correction of the PE deformity with the Nuss procedure (Figure 1c).

PC is on the contrary characterized by the protrusion of the anterior chest wall. It is possibly caused by the overgrowth of the costal cartilage [7,8]. It is also called pigeon chest because of the chest morphology. The deformity consists of the 15% of anterior chest wall anomalies. The treatment is primarily not surgical and is accomplished through compressive orthotic braces. This conservative approach has shown successful results. However, in some severe cases a surgical treatment may also be considered [1,2,7,8].

Despite the evolution of the surgical procedures there is still a lack of information in the literature concerning prevalence of these entities. The purpose of the current study is to describe the prevalence of PE and PC in young Greek male soldiers. To our knowledge there has never been any other similar study in Greece before. In addition, through research in the PubMed library using relevant keywords (pectus excavatum, pectus carinatum, anterior chest wall deformities, chest wall deformities, thorax deformities) separately or in combination, we were able to confirm the lack of many reports about this issue. Therefore, in the present study the prevalence of PE and PC was investigated in a population of about 9,000 Greek male soldiers and the accompanying diseases have also been recorded and evaluated.

Material and Methods

In our study 8996 male soldiers took part. They were all drafted in the Greek army from August 2010 to November 2013. They came from all parts of Greece. They were all men aged from 18 to 24 years. According to the World Health Organization (WHO) and UNESCO people aged from 10 or 15 to 24 years can be characterized as young [9,10]. The military service in Greece is compulsory for all males older than 18 years. It is therefore believed that our population fulfills the criteria to be characteristic as young men of this age and may be considered as a characteristic sample of the Greek male population of this age.

The diagnostic approach of PE and PC requires a thorough investigation. The diagnosis and evaluation of these diseases requires carrying out several examinations. However, the performance of all available examinations is not always necessary. Generally, chest x-rays, Thorax-CT, the magnetic resonance imaging (MRI), electrocardiography (ECG), echocardiography, lung function tests and cardiopulmonary exercise testing are most of tests which may be used to diagnose and evaluate PE and PC [1]. In the present study, the following diagnostic procedure was performed. On the first day of recruitment all soldiers were clinically examined by general practitioners. Chest X-Rays were performed by all soldiers and were also checked and evaluated from radiology specialists. Soldiers with diagnosed chest wall deformities were later referred to the central military hospital for further examination. There they were all examined by pulmonologists, cardiologists and thoracic surgeons. The further examinations performed included ECG, echocardiography and lung function tests. Thorax-CT was also used when necessary. The chest deformities were then classified as mild, moderate or severe [7,11]. After a thorough study of the soldiers' medical history and the newly acquired data, we concluded to the results described below.

Results

A total number of 8996 soldiers 18 to 24 years old were examined. The prevalence of chest wall abnormalities (posterior and anterior deformities) in the soldiers of our study was 1.11% (100 persons). 0.22% had anterior chest wall deformities (PE and PC). PE was detected in 17 soldiers (0.18%) and PC in 3 soldiers (0.03%). The ratio of PE versus PC was 5.6:1. The anterior chest wall deformities correspond to 20% of all detected chest wall deformities.

From the diseases that accompanied PE the most common was scoliosis. 4 soldiers had scoliosis simultaneously to PE (23.5%). 3 soldiers were detected with PE and MP (17.6%). From these cases 1 soldier had severe PE and MP. One soldier was diagnosed with simultaneous PE, pulmonary valve insufficiency and MP.

3 soldiers with PE (17.6%) were also diagnosed with eye disorders. One of them showed color vision deficiency, one myopia and one strabismus. A soldier with PE also reported history of bronchial asthma (5.88%). A PE soldier showed right bundle branch block (RBBB) in the ECG, had however no other clinical findings (5.88%). Another soldier with a mild PE presented Marfan syndrome (MS) (5.88%) (Figure 2). 8 PE soldiers (47%) reported a relevant family history, having a parent with PE.



Figure 2: Soldier with MS and mild PE.

PC was detected in 3 soldiers. The PC cases reached the 3% of all chest wall defects (anterior and posterior) and the 15% of all anterior chest wall deformities. A PC soldier presented a simultaneous MP (33.3%) and one other soldier PC and scoliosis (33.3%).

From the clinical examination of the rest soldiers (8976 persons) 80 soldiers were diagnosed with deformities of the posterior chest wall. 71 of them had scoliosis and 9 of them had kyphosis.

All soldiers with anterior chest wall deformities were asymptomatic.

Discussion

The prevalence of thoracic deformities in the general population varies. The chest wall defects have been described to appear to about 1% of the population [1,2]. Regarding PE in a study in mummies from ancient Egypt the prevalence was at 1.32% [4]. In the US in 10,000 births 23 children are expected to have PE [3]. A study from Brazil concerning school students has shown that deformities of anterior chest wall range from 1.95% to 4.9% [11]. Zou., *et al.* reported that PE among school students in China aged 7 - 14 years was 0.583%. [5]. In school students of the same age in Turkey the prevalence of PE was 3.2%, while in Iran in the same school population was 1.03% [3,12]. As far as the present study is concerned, the chest wall defects also approach the 1% of the literature. The PE prevalence was however in our study lower compared to other studies. On the other hand, our findings are similar to the findings of post-mortem studies and to the afterbirth US findings [3,11].

PE is much more common than the PC. In our study the PE:PC rate was 5.6: 1. This rate was equivalent to other studies (2.2:1 - 5:1) [11]. Our study showed that the prevalence of PE among Greek male soldiers was 0.18% and that of PC 0.03%. The findings in two other studies among students in Brazil (PE 0.95% and PC 0.04%) and in postsurgical PE results were also similar [11].

The role of radiology in the diagnostic approach of PE and PC is crucial and it may involve the use of chest x-rays, CT and MRI. The Haller index (HI) is a very useful tool which has been used to assess the severity of incursion of the sternum into the mediastinum. It is calculated from the ratio: maximal transverse diameter/narrowest anterior posterior length of chest in CT. Clearly described guidelines concerning the treatment of PE do not exist. However, if the HI is greater than 3.25 it is considered as a pathological finding. If HI is less than 2.56 then it is considered a normal finding. A HI between 2.56 and 3.25 is referred as a mild grade PE and no surgical intervention is required. A HI < 1.98 is indicative of PC. However, a rough radiologic diagnosis or a verification of the deformity can be made with normal chest x-rays [7,13]. In the present study neither a HI calculation nor a CT was performed by all patients. Only soldiers who had a severe PE or were selected for surgery underwent a thorough radiological examination.

A genetic predisposition of PE has not been verified yet. However, a positive family history is present in about 30% of patients with PE. In our study a 47% of PE positive family history was recorded among the PE cases. Similar were the results in Brazil. A percentage of 44 - 65% of positive PE family history was found in that study [11]. Pinar Kuru, *et al.* also reported equivalent results in school students in Turkey. Almost half of the students with PE had a positive family history [14]. However, the etiology is not clear and a direct genetic relevance has not been defined. From the clinical view the hypothesis that the pathophysiology may have a genetic background and that PE can be inherited either dominantly or recessively is strong, as about 40% of the patients have affected family members with similar congenital deformities. Some genes that can be involved in the development of PE have been identified. For example, mutations in the sulfotransferase gene GAL3ST4 can be a potential cause of PE [14]. In addition, experimental data from mouse models have shown that Gpr126 can be another genetic cause for the pathogenesis of adolescent idiopathic scoliosis and PE [15]. What is more a genetic connection is thought to exist in the development of PC too. This hypothesis is based on the fact that PC is detected in genetic inherited diseases such as Marfan disease, Noonan syndrome, Morquio syndrome, osteogenesis imperfecta, mitral valve prolapse, and homocystinuria [7].

PE is considered to be connected in a great degree with scoliosis. It seems that scoliosis appears more frequently in patients with PE than in the general population. Scoliosis is detected in 0.5-3% of PE cases [17]. In Brazil scoliosis was detected in 11.5% of students with PE [11]. In our study 23.5% soldiers with PE also had scoliosis. A reason why our percentage of coexistence of PE and scoliosis is higher than in the other studies may be due to the fact that our population consisted only from male persons. Similarly, Hong, *et al.* reported a high percentage of patients with PE and scoliosis (22.58%) in a population where more males took part [16]. In addition, Kuru, *et al.* also reported a high percentage in Turkey [14]. Despite the fact that the exact mechanism that connects the two diseases is not clearly known it can be easily understood that a deformity of the sternum would also affect the shape of the spine. In fact, the more asymmetric the PE is, the greater the deformity of the vertebral column would be [17-19]. Scoliosis also appears to be more often in the right side of the body. That fact is even more often if the 6th to 10th thoracic vertebrae are involved. Wang, *et al.* reported a theory according to which the sternum deformation pushes the heart to the left. For this reason, counterforces push the spine to the right [19]. Scoliosis has also been described in cases with PC. In 1989 Waters reported that 14% of patients with PC and scoliosis had required a therapeutic intervention [20]. In our study one soldier was detected with simultaneous PC and scoliosis. The common mechanism of the pathogenesis of the two deformities is possibly the overgrowth of the costal cartilages that push the sternum forward and consequently the spine latterly.

The effect and the consequences of the PE in the cardiovascular function remain a debatable issue. It is nowadays mostly believed that the cardiovascular function is not seriously affected if no other syndrome, such as Marfan syndrome, coexists [21]. However, the posterior sternum depression makes the anterior posterior thorax diameter smaller and as result presses and pushes the heart away [22]. This direct mechanical compression to the heart can lead to stroke volume and cardiac output reduction in cases of a severe deformity [1,21,22]. During the clinical physical examination, a systolic cardiac murmur can be sometimes heard and a MP can be diagnosed [1]. In our study, all soldiers with PE and PC underwent a clinical examination, ECG, chest x-ray and echocardiography. In many cases the anatomical morphology of the thoracic cavity can make the performance of the echocardiography even more difficult. In some patients, appropriate echocardiographical approach can be obtained through the subcostal window. In addition, there is always the alternative of transesophageal

echocardiography (TEE). The possibility of cardiac MRI also exists and may be very useful in some selected cases. The MRI has also the advantage that it cannot be affected from the thoracic deformities (Figure 3).

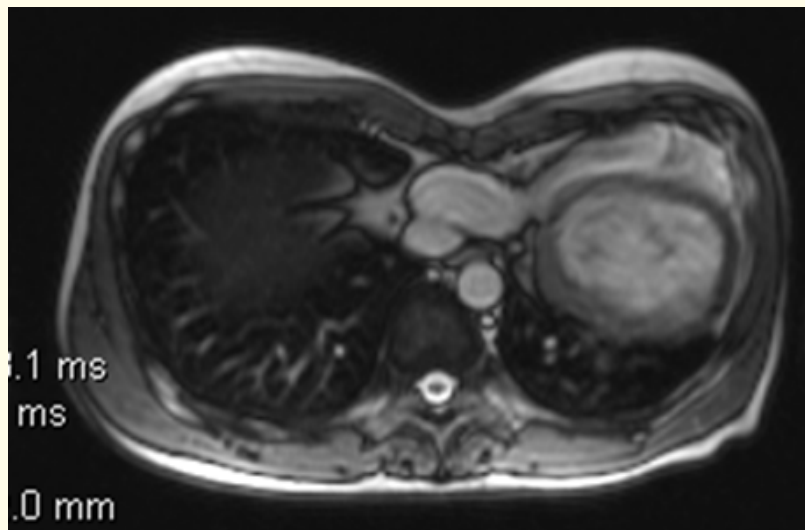


Figure 3: PE in MRT. The heart pushing away to the left due to PE can be seen. The patient 's deformity was corrected with the Nuss technic.

In our study 3 soldiers were presented with PE and Mitral prolapse (MP), consisting the 17.6% of the soldiers with PE. However, the percentage of patients with PE and MP can even reach a percentage of 25%. This percentage seems to be higher as the patient's age rises [1]. In addition, a soldier with PC was detected with undiagnosed MP. As far as the pathogenesis of the MP in PE is concerned it is believed that it may be attributed to the pressure phenomena due to the sternum depression. This hypothesis is also supported by the fact that the MP relents after the PE surgical correction.

An additional cause of the MP in sternum deformities can be the degenerative changes that can occur due to congenital syndromes that correlate and may coexist with anterior chest wall deformities. Such examples of connective tissue disorders are Marfan, Ehlers-Danlos, Poland, MASS (Mitral valve prolapse, not progressive Aortic enlargement, Skeletal and Skin alterations) syndromes [1,22,23]. Especially the Marfan syndrome is detected in 2 - 3 persons among 10,000. In 66% of Marfan patients PE can coexist [23,24]. In our study one soldier with Marfan syndrome and PE was detected among the about 9000 persons examined.

Electrocardiographic changes have been also detected in patients with PE. ECG is usually normal in most patients. However, in some cases ECG changes can be detected. They can vary from bundle branch blocks (BBB), ST segment changes suggestive of cardiac ischemia or even Brugada-type ECG pattern. The oftener detected ECG changes are the right-axis deviation and depression of the ST segment [21,25]. In our case a soldier was detected with right BBB. This soldier as well as all other soldiers included in the study were asymptomatic.

The progressive development of the anterior chest wall deformities and especially the PE can lead to the gradual development of restrictive lung disease [26]. In our study a soldier was detected with PE and bronchial asthma. The description of coexistence of bronchial asthma and PE is old [2,27,28]. Cserhati, *et al.* had already since 1984 reported that bronchial hyperreactivity and lung functional disorder

ders leading to chronic obstruction were significantly more frequent in patients with thorax deformities. Such a relevance also appeared in this study with 7.34% of patients with chest wall deformities having a coexisting bronchial asthma [28]. Fonkalsrud, *et al.* reported that 7% of patients who underwent a surgical correction of a deformity had bronchial asthma [27]. In our case the percentage was lower (5.88% - one soldier). Both static and dynamic exercise pulmonary function tests have been used for the examination of the anterior chest wall deformities [1]. In children, these examinations are usually normal. By aging or gradual progress of the deformities a more serious gradual obstructive and restrictive pattern pulmonary disease can be detected [1,18,21]. However, in only a small percentage of patients a typical restrictive pattern (low FVC with proportionate decrease in the FEV1 and expiratory flow rates) can be found. This pulmonary pathophysiology can be normalized after the surgical deformity correction [1,26]. In our case both static and exercise pulmonary function tests were performed in all soldiers with anterior chest wall deformities. No soldier presented a typical restrictive pattern.

The coexistence of serious ophthalmological diseases with PE is more often in Marfan patients. Mainly lens dislocation, and less commonly signs of early cataracts and/or glaucoma have been detected in these patients [21,28]. In our case the detected ophthalmological problems were randomly diagnosed and had no connection with Marfan syndrome.

The clinical symptomatology of the anterior chest wall deformities varies. The clinical symptomatology in children and adolescents is also random. In children, the symptomatology is reported mainly on exertion and thus it is difficult to be documented at rest. Symptoms can appear in adult life as the thoracic wall loses its elasticity. The appearance of symptoms also depends on the severity and the degree of progression of the PE. Especially in PE cases symptoms may occur after mild exercise and can appear as dyspnea or in the soldiers' case difficulty to complete the required physical training due to earlier fatigue compared to other soldiers. The main symptoms which may appear during exercise include the progressive loss of endurance, chest pain, worsening, progressive fatigue, wheezing and generally intolerance of the exercise [1,3,21,26]. In addition, palpitations, tachycardia, frequent upper respiratory infections, fainting/dizziness and chest pain at rest have also been reported. In adults with PE symptoms from the cardiopulmonary system could be directly attributed to PE [1]. The symptomatology to this age group can be similar as in childhood. Symptoms can appear clinically as shortness of breath and exercise intolerance [7]. In our study, all soldiers reported no symptoms. Coskun, *et al.* reported no symptomatology among school students [12]. On the contrary in the city of Manaus in Brazil 65.4% of school students with anterior chest wall deformities reported chest pain, dyspnea or palpitations [11]. For this reason, the symptomatology of the anterior chest wall deformities should always be approached with great attention even if the actual symptoms exist due to the psychological burden of the deformity that may restrict everyday activities [1,2].

In many countries, these deformities are firstly recognized from the general practitioner [1]. In most countries, there is no primary health care PE and PC screening program or a patient's information program or an organized preoperative and postoperative follow-up program. Zou, *et al.* have proposed a PE screening network program based on the experience that was acquired through their study. The organization of an analog program in a country in combination with widespread information could lead to early detection and treatment of abnormalities of the anterior chest wall [5]. As a result, within the framework of this screening program PE and PC surgical candidates could be early identified and postoperatively closely followed-up. Within such a screening program a recurrence of symptoms or of the deficiency could be after the surgical repair early diagnosed and treated [3].

Despite its uniqueness in Greece our study presents generally special features. It consists of a population of young soldiers whereas most other studies concentrated on school students aged from 7 to 14 years. The advantage of these studies is that the surgical correction of the deformity is preferred in these ages. A disadvantage of our study is also that no anthropometric characteristics were included. Moreover, the psychological burden was also not evaluated. The psychological burden because of the deformities in absence of clinical symptoms is an indication for surgical repair [1]. What is more, because of the nature of the study our study neither described the etiology of PE nor explored its risk factors. Restricted by the examination conditions, we were not able to carry out onsite measurement of the HI index for the PE patients.

| PE and commonest diseases | Percentage (%) |
|--------------------------------------|-----------------------|
| Anterior chest wall deformities | 0.22% |
| Ratio of PE:PC | 5.6:1 |
| PE | 0.18% |
| Scoliosis | 23.5% |
| Mitral valve prolapse | 17.6% |
| Accidental ophthalmological findings | 17.6% |
| Right bundle branch block | 5.88% |
| Marfan syndrome | 5.88% |
| Symptomatology | 0% |
| First grade family members with PE | 47% |

Table 1: Percentage of commonest diseases with PE.

| Study origin | Population | Ages | Percentage |
|---------------------|-------------------|-------------|-------------------|
| Ancient Egypt [4] | Mummies | - | 1.32% |
| Brazil [11] | School students | 7 - 14 | 1.275% |
| China [5] | School students | 7 - 14 | 0.583% |
| Turkey [12] | School students | 7 - 14 | 3.2% |
| Iran [3] | School students | 7 - 14 | 1.03% |
| Greece | Soldiers | 18 - 24 | 0.18% |

Table 2: Prevalence of PE in the countries of the world.

Conclusion

Our study is unique in the literature. It describes the prevalence of abnormalities of the anterior chest wall in young Greek male soldiers and directly refers to the young Greek male population. The prevalence of PE was 0.18% and of PC 0.03% in the general population. This rate is lower than that described in the literature. Alongside the percentage of the commonest diseases that accompany these deformities were recorded as a reference basis.

Conflict of Interest

None to declare.

Acknowledgments

All authors contributed equally.

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Volume 3 Issue 3 April 2017

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