

Thorax deformity, joint hypermobility, and anxiety disorders

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ABSTRACT

Objective: To evaluate the association between thorax deformities, panic disorder, and joint hypermobility.

Method: The study includes 52 males diagnosed with thorax deformity, and 40 healthy male controls without thorax deformity, in Tatvan Bitlis and Isparta, Turkey. The study was carried out from 2004 to 2006. The teleradiographic and thoracic lateral images of the subjects were evaluated to obtain the Beighton scores; subjects' psychiatric conditions were evaluated using the Structured Clinical Interview for DSM-IV Axis I Disorders (SCID-1), and the Hamilton Anxiety Scale (HAM-A) was applied in order to determine the anxiety levels. Both the subjects and controls were compared in sociodemographic, anxiety levels, and joint mobility levels. In addition, males with joint hypermobility and thorax deformity were compared to the group with thorax deformity without joint hypermobility.

Results: A significant difference in HAM-A scores was found between the groups with thorax deformity and without. In addition, 21 subjects with thorax deformity met the joint hypermobility criteria in the group with thorax deformity, and 7 subjects without thorax deformity met the joint hypermobility criteria in the group without thorax deformity, according to Beighton scoring. The Beighton scores of the subjects with thorax deformity were significantly different from those of the group without deformity. Additionally, anxiety scores of the males with thorax deformity and joint hypermobility were found higher than males with thorax deformity without joint hypermobility.

Conclusions: Anxiety disorders, particularly panic disorder, have a significantly higher distribution in male subjects with thorax deformity compared to the healthy control group. In addition, the anxiety level of males with thorax deformity and joint hypermobility is higher than males with thorax deformity without joint hypermobility.

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Several studies have suggested an association between mitral valve prolapse (MVP) and/or joint hypermobility and anxiety disorders, mainly panic disorder,¹⁻⁸ and the contradictory results obtained have made this field controversial and well suited to further research. Several studies have highlighted the relation between thorax deformities and MVP.⁹⁻¹¹ However, we were unable to find any study concerning the association between thorax deformities and panic disorder. The association between thoracic skeletal abnormality and MVP may be a manifestation of a single connective tissue defect during embryonic development of the bony thoracic cage and the atrioventricular valves.¹² Pectus excavatum appears soon after birth and is occasionally associated with kyphosis, scoliosis, or MVP. This may be familial or associated with Marfan, or Ehlers-Danlos syndromes, or with isolated hyper flexibility of the joints. According to some reports, thoracic deformity may be the cause of psychosocial problems due to esthetic worries. This malformation affects an individual's psychosocial development, causing embarrassment, feelings of stigma, social anxiety, and even depression.¹³ In the same way, pectus deformities and atypical costal anomalies are congenital thoracic wall defects that can cause a marked cosmetic defect with psychological trauma. The presence of anxiety disorder has been reported to be 3 times higher in patients with joint hypermobility and MVP, compared with patients who were non-concomitant with MVP. Joint hypermobility syndrome is a genetic condition characterized

by increased active and passive motions, and whose prevalence ranges from 10-15%. The syndrome is more common in women. It has been suggested that MVP and joint hypermobility syndrome may share a common psychopathological mechanism with collagenous tissue diseases. In addition, a significant correlation has been observed between joint hypermobility syndrome and panic disorder in some patients with rheumatological disorder.^{14,15}

In an earlier retrospective study, we considered the psychiatric symptoms of males with pectus excavatum using the Brief Symptom Inventory.¹⁶ Scores on the subscale of phobic anxiety, interpersonal sensitivity, and paranoia, and the General Symptom Index revealed a significant difference compared with the healthy control group.¹⁷ We also determined a close relation between joint hypermobility and anxiety disorders in a prospective study performed on subjects with joint hypermobility, and a control group of subjects without joint hypermobility syndrome.¹⁸ In the light of that research, we performed a prospective study on the relationship between thorax deformity, joint hypermobility, and anxiety disorders.

Methods. The study included 60 male subjects with thorax deformity and 40 males (control) without thorax deformity selected from 600 males during routine general medical examinations in Tatvan Bitlis and Isparta, Turkey, from 2004 to 2006. The subjects with thorax deformity and the healthy control subjects enrolled in the study had not previously been referred to any hospital for any reason. Six subjects with cardiac disorder and 2 subjects with a history of substance abuse were excluded from the 60 subjects with thorax deformity. The teleradiographic and thoracic lateral images of the study subjects were evaluated, and their thoracic indices were calculated using the thoracic anterior posterior diameters in comparison to their lateral diameters. Beighton scoring was used for the assessment of joint hypermobility syndrome by an orthopedic specialist. The Beighton system includes 5 criteria describing 9 maneuvers to explore the degree of joint mobility. The scores range from 0-9, and depend on the degree of movement. Joint hypermobility syndrome is considered positive in subjects with a score of 5 or more out of 9.^{19,20} Subjects exhibiting any evidence of MVP at physical examination, and echocardiographic evaluation were examined by an expert cardiologist. Those subjects with cardiological problems were then excluded from the study. During psychiatric evaluation, the control group and the group with thorax deformity were mixed and administered the Structured Clinical Interview for DSM-IV® Axis I Disorders (SCID-I), Clinician Version, and Hamilton Anxiety Scale

(HAM-A) by a psychiatrist. A diagnosis distribution of the group with SCID-I was then drawn up, and the anxiety levels of all subjects were determined using HAM-A.^{21,22} Informed consent was given by all subjects before participating in the study, which was approved by the local ethical committee.

Statistical test was carried out using Statistical Package for Social Sciences version 10.0 to evaluate the data. Parametric data were expressed as mean± SD, and categorical data were expressed as percentages. The Mann-Whitney U test was used to compare independent parametric data, and the chi-square test was used for categorical data.

Results. The group with thorax deformity consisted of 52 male subjects, of whom 18.1% were married. Mean age was 21.9±1.3 years, and the mean duration of the subjects' education was 9.4±3 years. The control group included 40 males who had similar sociodemographic characteristics without thorax deformity (**Table 1**). There was a significant difference between the groups with and without thorax deformity in terms of HAM-A scores ($p=0.038$) (**Table 2**). Additionally, the Beighton scores of the subjects with deformity were significantly different to those of the group without deformity ($p=0.008$). Using the Beighton scoring system, 21 subjects with thorax deformity met the joint hypermobility criteria in the group with thorax deformity, while 7 subjects without thorax deformity met the joint hypermobility criteria in the group without thorax deformity. The thoracic index scores, which were obtained by dividing the thorax anterior posterior diameters into the lateral diameters, did not differ significantly between the study group and the control group ($p=0.285$). **Table 3** shows that there was a significant difference between the groups having a Beighton score of 5 points or more ($n=21$) and without ($n=31$) joint hypermobility in terms of HAM-A scores ($p=0.004$). No significant difference was determined between the 2 groups in terms of thoracic index ($p=0.370$). It is noteworthy that joint hypermobility was more frequent in the group with thorax deformity than in the control group ($p=0.04$).

Discussion. Evaluation of the subjects' sociodemographic characteristics revealed no significant differences in terms of age or length of education. The sampling was appropriate for the purpose of the study. This study shows a significant difference between the group with thorax deformity and the control group in terms of HAM-A scores. Based on these data, it appears that anxiety disorders were more common in the group with thorax deformity, and the level of anxiety was also higher in this group compared to the group without thorax deformity, including the group

Table 1 - The sociodemographic characteristics and distribution of psychiatric disorders of the subjects with deformity and the control.

Characteristics	With thorax deformity (n=52)	Control (n=40)	P-value
Age	21.9±1.3	22.7±3.0	0.143
Duration of education	9.4±3.0	9.53±4.3	0.879
Married (%)	18.1	11.1	0.654
Psychiatric Diagnosis	With thorax deformity	Without	
	Frequency n (%)		
Panic disorder	19 (36.5)	4 (10)	
Not otherwise specified anxiety disorder	3 (5.8)	5 (12.5)	
Adjustment	4 (7.7)	0 (0)	
Generalized anxiety disorder	2 (3.8)	0 (0)	
Normal	24 (46.2)	31 (77.5)	
Total	52 (100)	40 (100)	

Table 2 - Comparison between the thorax deformity and control groups.

Parameters	With thorax deformity n=52	Control n=40	P-value
	(Mean±SD)		
HAM-A*	19.1±7.2	15.6±9.2	0.038
Beighton score	3.05±3.0	1.50±2.4	0.008
Thoracic index	1.3±0.08	1.3±0.14	0.285
Number of heart beats	76.6±12.14	77.47±11.4	0.731
*HAM-A - total Hamilton anxiety scale score			

Table 3 - Comparison between the groups with and without joint hypermobility.

Parameters	Thorax deformity and joint hypermobility n=21	Thorax deformity without joint hypermobility n=31	P-value
	(Mean±SD)		
HAM-A*	21.26±5.8	16.42±8.6	0.004†
Thoracic Index	1.30±0.16	1.27±0.1	0.370
Number of heart beats	76.6±12.14	77.47±11.4	0.731
*HAM-A - total Hamilton anxiety scale score, † - significant results			

with no psychiatric diagnosis. In a retrospective study comparing a group with thorax deformity with a control group without thorax deformity, we reported that psychiatric symptoms might be more frequent in the thorax deformity group compared to the normal population.¹⁷ There was also a significant difference between the group with thorax deformity and the control group in terms of Beighton scores in this study. We were unable to find any other studies supporting this result. Furthermore, we determined a significant difference in HAM-A scores between the groups with and without joint hypermobility. This result confirms the reports in a study by Bulbena et al²³ and Martin-Santos et al,²⁴ that joint laxity is highly prevalent in patients with panic disorder.

It is noted that the distribution of the areas at 5 and above the cut-off point for hypermobility in the Beighton scores was significantly higher in the group with thorax deformity compared to the no deformity group. Several studies have indicated the concomitance of joint hypermobility, MVP, and anxiety disorders.^{1,8} However, to our knowledge, there are no studies in the literature concerning the prevalence of joint hypermobility in subjects with thorax deformity. Therefore, our findings show that joint hypermobility has a higher distribution in subjects with thorax deformity compared to the normal population. A duplication of a portion of chromosome 15q has been found to be strongly associated with phobic disorders.²⁵ Gratacos et al²⁶ studied the molecular basis of the co-occurrence of panic and phobic disorders with joint laxity. We also determined a close relation between joint hypermobility syndrome and anxiety disorders in a controlled study.¹⁸ An interstitial duplication of human chromosome 15q24-26 (known as DUP25), which is significantly associated with panic, or agoraphobia, or social phobia, or joint laxity in families, and with panic disorder in nonfamilial cases, has been identified.²⁶

One group of studies highlights the concomitance of mitral valve prolapse and thorax deformity, and indicates that a common physiopathological and genetic mechanism may play a role between thorax deformity and MVP. The results obtained from our study suggested that there might also be a similar relationship in the concomitance of thorax deformity and panic disorder. Concomitance of vascular diseases, asternic body type and anxiety disorders have already been reported by several authors.²⁷ We observed that thorax deformity, joint hypermobility, and MVP may function with a common physiopathological mechanism, and several studies indicating the concomitance of MVP and joint hypermobility in disorders such as Marfan syndrome, which may present with anxiety disorders and depression, and Ehler Danlos syndrome, and defining the association of each clinical presentation (MVP and joint hypermobility) with anxiety disorders suggests

that anxiety disorders may also be frequently involved in the close relationship between thorax deformities of collagenous tissue disorders. This hypothesis has been supported by the fact that joint hypermobility has a higher distribution in the thorax deformity group compared to the control group. It has also been indicated that thorax deformity may be subject to depressive findings developing secondary to esthetic concerns.²⁸ However, it is not sufficient to account for the presentation of anxiety disorders solely in terms of esthetic concerns. The striking concomitance of joint hypermobility, thorax deformity, and MVP with anxiety disorders suggests the presence of a common physiopathological mechanism.

In conclusion, it seems that anxiety disorders, and particularly panic disorder, have significantly higher distributions in subjects with thorax deformity and/or joint hypermobility compared to the healthy population, in addition to the information supplied in the literature. The association of thorax deformity and joint hypermobility may increase the anxiety level compared to group with only thorax deformity. The small sample size strongly limits the usefulness of our data, however, the design could be expanded and replicated. Including only male subjects was another limitation of the study. Although, it is difficult to generalize the results of this study unless it is supported with wide-sampled and genetic based studies, the results of this study may be a remarkable step for future studies in this field.

References

1. Arkonaç O, Gültekin N, Özer S, Uner S, Toker F. [Mitral valve prolapsuslu olgularda psikiyatrik bozukluklar]. *Düsiimen Adam* 1991; 4: 36-40. Turkish
2. Hamada T, Koshino Y, Misawa T, Isaki K, Gejyo F. Mitral valve prolapse and autonomic function in panic disorder. *Acta Psychiatr Scand* 1998; 97: 139-143.
3. Tamam L, Ozpoyraz N, San M, Bozkurt A. Association between idiopathic mitral valve prolapse and panic disorder. *Croat Med J* 2000; 41: 410-416.
4. Chan FL, Chen WW, Wong PH, Chow JS. Skeletal abnormalities in mitral-valve prolapse. *Clin Radiol* 1983; 34: 207-213.
5. Katerndahl DA. Panic and prolapse: Meta-analysis. *J Nerv Mental Dis* 1993; 181: 539-544.
6. Toren P, Eldar S, Cendorf D, Wolmer L, Weizman R, Zubadi R, et al. The prevalence of mitral valve prolapse in children with anxiety disorders. *J Psychiatr Res* 1999; 33: 357-361.
7. Koshino Y, Murata T, Omori M, Isaki K. Prevalence of mitral valve prolapse in patients with anxiety disorder. *Clin Psychiatry* 1990; 32: 963-970.
8. Kocabasoglu N. [Panic disorder agoraphobia and other comorbid entities]. *Yeni Symposium* 2002; 40: 68-75. Turkish
9. Kumar UK, Sahasranam KV. Mitral valve prolapse syndrome and associated thoracic skeletal abnormalities. *J Assoc Physicians India* 1991; 39: 536-539.
10. Peh WC, Teo WS, Kwok RK, Quek S. Thoracic skeletal abnormalities in young men with mitral valve prolapse. *Ann Acad Med Singapore* 1985; 14: 676-681.

11. Tamura K, Fukuda Y, Ishizaki M, Masuda Y, Yamanaka N, Ferrans VJ. Abnormalities in elastic fibers and other connective-tissue components of floppy mitral valve. *Am Heart J* 1995; 129: 1149-1158.
12. Udoshi MB, Shah A, Fisher VJ, Dolgin M. Incidence of mitral valve prolapse in subjects with thoracic skeletal abnormalities--a prospective study. *Am Heart J* 1979; 97: 303-311.
13. Smith KA. Pectus excavatum. More than meets the eye. *Orthop Nurs* 2004; 23: 190-194.
14. Grahame R, Edwards JC, Pitcher D, Gabell A, Harvey W. A clinical and echocardiographic study of patients with the hypermobility syndrome. *Ann Rheum Dis* 1981; 40: 541-546.
15. Pitcher D, Grahame R. Mitral valve prolapse and joint hypermobility: evidence for a systemic connective tissue abnormality? *Ann Rheum Dis* 1982; 41: 352-354
16. Sahin NH, Durak A. [Kısa semptom envanteri (Brief Symptom Inventory-BSI): Türk gençleri için uyarlaması]. *Türk Psikoloji Dergisi* 1994; 31: 44-56. Turkish
17. Gulsun M, Tonbul M, Gokce O, Evrensel A, Yıldız M. Göğüs Deformiteli Olgularda Psikiyatrik Semptom Dağılımlarının Arastırılması: Retrospektif Bir Arastırma. Türkiye Psikiyatri Derneği 9. Bahar Sempozyumu, Belek-Antalya, 2005.
18. Gulsun M, Doruk A, Uzun O. [Anxiety disorders in a nonclinical male sample with joint hypermobility syndrome]. *Yeni Symposium* 2006; 44: 165-168. Turkish
19. Beighton P, Solomon L, Soskolne CL. Articular mobility in an African Population. *Ann Rheum Dis* 1973; 32: 413-418.
20. Beighton PH, Grahame R, Bird H, editors. *Hypermobility of Joints*. 2nd ed. London (UK): Springer-Verlag; 1989.
21. Çorapçıoğlu A, Aydemir Ö, Yıldız M, Esen-Danacı A, Köroğlu E. DSM-IV Eksen I Bozuklukları (SCID-I) için Yapılandırılmış Klinik Görüşme, Klinik Versiyon. Ankara (TR): Hekimler Yayın Birliği; 1999. Turkish
22. Hamilton M. The assessment of anxiety states by rating. *Br J Med Psychol* 1959; 32: 50-55.
23. Bulbena A, Duró JC, Porta M, Martín-Santos R, Mateo A, Molina L, Vallescar R, et al. Anxiety disorders in the joint hypermobility syndrome. *Psychiatry Res* 1993; 46: 59-68.
24. Martín-Santos R, Bulbena A, Porta M, Gago J, Molina L, Duró JC. Association between joint hypermobility syndrome and panic disorder. *Am J Psychiatry* 1998; 155: 1578-1583.
25. Flint J. Psychiatric genetics: a frightful chromosome. *Current Biology* 2001; 11: 907-909.
26. Gratacos M, Nadal M, Martín-Santos R, Pujana MA, Gago J, Peral B, et al. A polymorphic genomic duplication on human chromosome 15 is a susceptibility factor for panic and phobic disorders. *Cell* 2001; 106: 367-379.
27. Vertogradova OP, Voitsekhn VE, Krasnov VN, Sinitsin VN, Suvorov AK. [Age and features of depressions during the second half of life]. *Zh Nevropatol Psikhiatr Im S S Korsakova* 1986; 86: 1380-1384. Russian
28. Lawson ML, Cash TF, Akers R, Vasser E, Burke B, Tabangin M, et al. A pilot study of the impact of surgical repair on disease-specific quality of life among patients with pectus excavatum. *J Pediatr Surg* 2003; 38: 916-918.

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