Turkish Journal of Medical Sciences

http://journals.tubitak.gov.tr/medical/

Research Article

Turk J Med Sci
(2018) 48: 1200-1206

doi:10.3906/sag-1807-180

The prevalence of chest wall deformity in Turkish children

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Received: 19.07.2018 • **Accepted/Published Online:** 09.09.2018 • **Final Version:** 12.12.2018

Background/aim: This study aimed to identify children who have chest wall deformity, the prevalence of deformity, and the factors affecting the psychological and physical disorders caused by the deformity.

Materials and methods: The study was conducted among 14,108 girls and boys aged 11–14 years, who were 5th–8th grade secondary school students in Ankara Province between October 2014 and March 2015.

Results: Of the 14,108 students in our study, the mean age of the children was 12.53 ± 1.11 years (median 12.54, 11–14 years) and chest wall deformity was detected in 199 (1.41%) students. Male/female and pectus carinatum/pectus excavatum ratios were 2.16 and 1.59, respectively. According to multivariate logistic regression analysis, physical disturbance was found to be statistically significantly higher among children in age group 11 [adjusted OR (95% CI) =16.01 (1.89–135.61), P < 0.011] and in children who were aware of the deformity [adjusted OR (95% CI) = 0.31 (0.13–0.71), P < 0.006], and psychological disturbance was found to be statistically significantly higher in girls [adjusted OR (95% CI) = 15.44 (1.68–141.59), P < 0.015] and in those with a presence of family history [adjusted OR (95% CI) = 18.66 (1.92–181.60), P < 0.012].

Conclusion: In this study conducted in a large population, chest wall deformities were found to be more prevalent in boys (0.96%) and pectus carinatum was found as the most common deformity type in our country, contrary to the literature.

Key words: Chest wall deformity, pectus, prevalence, Turkish children

1. Introduction

Pectus excavatum (PE) is the most common chest deformity overall and the most common in males (1,2). Pectus carinatum (PC) is seen less than PE in the general population (3). Poland syndrome is another chest wall deformity seen at a ratio of 1/30,000 (4,5).

In this study, it was aimed to identify children who have chest wall deformity, the prevalence of deformity, the factors affecting the psychological and physical disorders caused by the deformity, and early diagnosis of these psychological and physical disorders and to take the required actions in the district of Mamak in Ankara Province.

2. Materials and methods

Approval was obtained from the National Education Directorate of Ankara Province (Date/Number: 14.11.2014/5317227) and the Clinical Research Ethics Committee of Ankara Numune Research and Training Hospital (Date/Number: 01.10.2014/E-14-306). The study was conducted with 14,108 girls and boys aged between 11 and 14 years, who were 5th–8th grade secondary school students in the Mamak district of Ankara Province between October 2014 and March 2015. Seventeen schools were randomly selected among the secondary schools in Mamak.

Written informed consent was obtained from the families one day before visiting the school. The students whose families did not agree to participate were not involved in the study. There were two thoracic surgeons for the children's examination. Male and female students were examined separately in single cabins with top clothes completely removed and their anterior and posterior chest walls were inspected. A questionnaire form and a physical examination form were completed for the children who were found to have chest wall deformities. The first part of the self-filled questionnaire form included names of the students, whether they knew about chest wall deformity (CWD), whether they were aware of the CWD, family history, the most common physical disturbance they

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experienced (chest pain, dyspnea, exertional dyspnea, palpitation, or no disturbance), and whether they felt any psychological disturbance due to this deformity. In the physical examination, height and weight were measured, presence of murmur was examined on cardiac auscultation, and scoliosis was examined by inspection. For the students who were detected to have PE deformity, deformity severity was measured by anthropometric index calculation by the proportion of sternal deformity depth to the largest diameter of chest wall. The cut-off value of the anthropometric index was accepted as 0.12. Deformity type and degree (mild, moderate, severe) and subtype of PC (chondromanubrial, chondrogladial symmetrical, chondrogladial asymmetrical, and mixed type) were detected by inspection (6). In the inspection of the children, if we could not detect PC and PE clearly in the first physical examination, we accepted it as mild. If we could detect PC and PE clearly in the first physical examination, then we accepted it as moderate or severe. Subjective deformity degree detection during the inspection was done by the same physician for all students. We did not perform chest X-rays during the study. Therefore, the surgical correction criterion was that the anthropometric index value for PE be more than 0.12. The surgical correction criterion was moderate or severe scoring on the physical examination for PC.

Body mass index (BMI) (kg/m^2) was calculated using weight and height. BMI percentiles according to age and sex were calculated using childhood and adolescence (2–20 years) percentile curves of the Centers for Disease Control (7,8). Students were accepted as thin if BMI was below 5%, normal if BMI was between 5% and 84%, overweight if BMI was between 85% and 94%, and obese if BMI was ≥95%.

The students who were detected to have CWD were referred to the hospital for further investigation.

2.1. Statistical analysis

The sample size was calculated before starting the research. Chest deformity prevalence was found as 1.03% in the study of Rajabi-Mashhadi et al. (9). Based on this prevalence rate, with 80% power, a ±20% deviation, and 0.05 significance level, the minimum sample size was calculated as 14,000 children. These 14,000 children were distributed proportionally according to age and sex (R3.0.1 open source program). Mean unit size was calculated as 800 students considering the number of children at schools; 17 schools were found to be enough and 3 schools were determined as replacements. All of the students aged between 11 and 14 years were analyzed.

Statistical analysis was done using SPSS 18.0 for Windows (SPSS Inc., Chicago, IL, USA). Categorical data were tested with Fisher's exact test, Pearson's chi-square test, and Yate's corrected chi-square test. A logistic regression model was used for analysis of the variables that were found to be significant in univariate analysis. $P < 0.05$ was accepted as statistically significant.

The crude prevalence rate for our study population and adjusted prevalence rate for the standard Turkish population for the age group of 11–14 years were calculated by the following formulae:

Crude prevalence rate = [count/population] × 100,000

Adjusted prevalence rate x-y =∑yi-x[(counts/popi) × 100,000 × [stdmili/∑yj-xstdmilj]]

3. Results

Of the 14108 students enrolled in our study, 6828 (48.4%) were girls and 7280 (51.6%) were boys. Mean age of the children was 12.53 ± 1.11 (median: 12.54, 11–14) years. There were 1621 (23.7%) girls and 1726 (23.7%) boys in age group 11; there were 1669 (24.4%) girls and 1808 (24.8%) boys in age group 12; there were 1869 (27.4%) girls and 1912 (25.2%) boys in age group 13; there were 1669 (24.4%) girls and 1834 (25.2%) boys in age group 14. There was not a significant difference between age groups with regard to sex ($P = 0.452$). CWD was detected in 199 (1.41%) students. Of them, 63 (31.7%) were girls and 136 (68%) were boys. The prevalence of deformity was 0.45% (n = 63) for girls and 1.96% (n = 136) for boys ($P < 0.0001$) (Table 1). Male/female ratio was 2.16 in CWD cases. Mean age of the students in whom deformity was detected was 12.78 ± 1.98 (median: 13, 11–14) years. Deformity was most frequent in age group 14 with 68 (34.2%) students and demographic characteristics of the patients are shown in Table 1. Crude and population-adjusted prevalence rates by age group and sex for the 11–14 age group are shown in Table 2. Crude prevalence rate was 5.63%, and adjusted prevalence rate was 1.42% for the 11–14 age group population. Among all students, there were 121 (0.86%) cases of PC, 76 (0.54%) of PE, 1 (0.007%) of Poland syndrome, and 1 (0.007%) of rib anomaly. The PC/PE ratio was 1.59. Chondrogladial symmetrical type was the most common PC subtype with 49 cases (40.5%). On inspection, deformity was mild in 123 (61.8%), moderate in 69 (34.7%), and severe in 7 (3.5%). In 76 students with PE, deformity was mild in 42 (55.3%), moderate in 30 (39.5%), and severe in 4 (5.3%). The anthropometric index was 0.12 or above for 22 (28.9%) and below 0.12 for 54 (71.1%) of 76 students with PE. Twenty-two children who had PE and 76 children who had PC needed correction of the chest wall abnormality. Scoliosis was detected on inspection in 10 (5.03%) of the children who were detected to have CWD (5.03%). The CWD degree of scoliosis cases was mild in 1 (10%), moderate in 8 (80%), and severe in 1 (10%). Of the children with CWD, 48 (24.1%) stated that they knew about the deformity and 67 (33.7%) stated that they were aware of the deformity. Family history was positive for CWD in 15 (7.6%), while 136 (68.3%) stated

Table 1. Deformity prevalences by some sociodemographic characteristics.

	CWD (+)		CWD (-)		Total		P
	N	%	N	%	n	%	
Sex Girls Boys Total	 63 136 199	 31.6 68.4 100.0	6765 7144 13,909	 48.6 51.4 100.0	 6828 7280 14,108	 48.4 51.6 100.0	0.0001
Age groups 11 12 13 14 Total	 30 51 50 68 199	 15.1 25.6 25.1 34.2 100.0	 3317 3426 3731 3435 13,909	 23.84 24.63 26.82 24.70 100.0	 3347 3477 3781 3503 14,108	 23.73 24.64 26.80 24.83 100.0	0.003

Table 2. Crude and adjusted prevalence rates by age group and sex for 11–14 age group in the Turkish population.*

Population (11–14 years age group)	Crude prevalence rate (%)	Adjusted prevalence rate (%)
Total male population	7.42	1.87
Total female population	3.70	0.94
Total population	5.63	1.42

*Population data were obtained from the Turkish Statistical Institute for 11–14 age group for girls and boys of the Turkish population.

that they did not know about the family history of CWD. CWD was reported for the father in 7 cases (46.7%), mother in 3 (20.0%), sibling in 2 (13.3%), grandfather in 2 (13.3%), and cousin in 1 (6.7%). The most commonly reported CWD-related physical disturbance was dyspnea in 20 (10.1%), chest pain in 10 (5%), exertional dyspnea in 6 (3%), and palpitation in 6 (3%) of the children. Nine (4.52%) children with CWD reported psychological disturbance due to CWD. This prevalence was found as 11.1% in girls and 1.5% in boys and the difference was found to be statistically significant ($P = 0.005$).

Prevalence of awareness of deformity was found to be statistically significantly higher in the children with a family history of CWD ($P = 0.001$).

Univariate analysis was performed for the presence of physical disturbance with respect to sex, age groups, deformity degree, deformity awareness, family history, having information about CWD, and BMI percentile curves. The significant risk factors for the presence of physical disturbance were found as being in the age group of 11 years ($P < 0.015$), having a mild deformity ($P < 0.004$), having a moderate deformity ($P < 0.017$), and being aware of the deformity ($P < 0.001$). A logistic regression model was established to understand the effective factors for the physical disturbance. According to multivariate logistic regression analysis, physical disturbance was found to be statistically significantly higher in children in the 11 year age group [adjusted OR (95% CI) = 16.01 (1.89–135.61), $P < 0.011$] and in children who were aware of the deformity [adjusted OR (95% CI) = 0.31 (0.13–0.71), $P < 0.006$] (Table 3).

Univariate analysis was performed for the presence of psychological disturbance with respect to sex, age groups, deformity degree, deformity awareness, family history, having information about CWD, and deformity type. Psychological disturbance was found to be statistically significantly higher among girls ($P < 0.002$), the moderate deformity group ($P < 0.035$), children who were aware of the deformity ($P < 0.0003$), children with a family history ($P < 0.0001$), and children who knew about CWD ($P < 0.0001$). According to multivariate logistic regression analysis, psychological disturbance was found to be statistically significantly higher in girls [adjusted OR (95% CI) = 15.44 (1.68–141.59), $P < 0.015$] and in the presence of family history [adjusted OR (95% CI) = 18.66 (1.92–181.60), $P < 0.012$] (Table 4).

Table 4. Univariate and multivariate logistic regression analysis between psychological disturbance and sex, age groups, deformity degree, deformity awareness, family history, having knowledge about CWD deformity, and deformity type in children with CWD.

Risk factor	Psychological disturbance (+)		Psychological disturbance (-)		Crude OR (95% CI)	P	Adjusted OR (95% CI)	P
	n	%	n	%				
Sex								
Female	7	11.1	56	88.9	8.38 (1.69–41.57)	0.002	15.44 (1.68–141.59)	0.015
*Male	2	1.5	134	98.5	-	-	-	-
Deformity degree								
*Mild	2	1.6	121	98.4	-		-	-
Moderate	6	8.7	63	91.3	0.17 (0.03–0.88)	0.035	0.66 (0.08–5.8)	0.551
Severe	1	14.3	6	85.7	0.09 (0.01–1.25)	0.074	0.31 (0.01–14.88)	0.704
Deformity awareness								
Yes	8	11.9	59	88.1	17.76 (2.17–145.20)	0.0003	0.47 (0.01–17.55)	0.681
*No	1	0.8	131	99.2	-	-		
Family history								
Yes	4	26.7	11	73.3	13.02 (3.06–55.43)	0.0001	18.66 (1.92–81.60)	0.012
*No	5	2.7	179	97.3	-	-		-
Having knowledge about CWD								
Yes	7	14.6	41	85.4	12.72 (2.54–63.57)	0.0001	0.14 (0.004–5.29)	0.290
*No	2	1.3	149	98.7		-		-
**Deformity type								
PE	15	19.7	61	80.3	0.89 (0.44–1.83)	0.7683	0.85 (0.13–5.62)	0.866
*PC	26	21.5	95	78.5	-	-	-	-

*Reference group.
**Two patients with Poland syndrome and rib anomaly were eliminated from crude OR and all adjusted OR values were calculated with 197 students.

Of the children with PE, 36.8% (n = 28), and of the children with PC, 31.4% (n = 38) were found to be aware of the deformity. A statistically significant relationship could not be detected between deformity type and awareness of deformity (P = 0.431).

A statistically significant relationship was detected between deformity degree and awareness of deformity (P = 0.0001). Awareness increased as deformity degree increased.

A statistically significant relationship was not detected between deformity degree (P = 0.32) and deformity type (P = 0.550) or BMI according to age percentile curves.

4. Discussion

Congenital chest deformity prevalences vary in the general population and among communities. CWD prevalence was reported as 1% in the general population (10) and varies between 1% and 1.95% in the literature (9,11). Prevalence was reported higher in the white race in the study of Westphal et al. (11). When the literature in Turkey is examined, the prevalence of thoracic deformities was determined by Esme et al. as 0.768%, by Yucesan et al. as 0.76%, and by Soysal et al. as 1.28% (12–14). The population-adjusted prevalence rate was found as 1.42% in Turkish children, which is consistent with the literature.

Table 3. Univariate and multivariate logistic regression analysis between physical disturbance and sex, age groups, deformity degree, deformity awareness, family history, having knowledge about CWD, and BMI percentile curves in patients with CWD.

Risk factor	Physical disturbance (+)		Physical disturbance (-)		Crude OR (95% CI)	P	Adjusted OR (95% CI)	P
	n	%	n	%				
Sex								
Female	12	19	51	81	0.83 (0.39–1.76)	0.6282	1.26 (0.54–2.99)	0.593
Male*	30	22.1	106	77.9				-
Age groups								
11	1	3.3	29	96.7	12.96 (1.65–101.54)	.015	16.01 (1.89–135.61)	0.011
12	12	23.5	39	76.5	1.45 (0.63–3.32)	0.376	1.19 (0.48–2.99)	0.702
13	8	16	42	84	2.35 (0.94–5.85)	0.068	2.57 (0.93–7.11)	0.680
14*	21	30.9	47	69.1	-			-
Deformity degree								
Mild	21	17.1	102	82.9	12.14 (2.21–66.85)	0.004	5.35 (0.69–41.62)	0.109
Moderate	16	23.2	53	76.8	8.28 (1.46–46.83)	0.017	4.39 (0.59–32.53)	0.147
Severe*	5	71.4	2	28.6	-	-	-	-
Deformity awareness								
Yes	23	34.3	44	65.7	3.11 (1.54–6.26)	0.001	0.31 (0.13–0.71)	0.006
No*	19	14.4	113	85.6	-		-	-
Family history								
Yes	5	33.7	10	66.7	1.99 (0.64–6.16)	0.2280		-
No*	37	20.1	147	79.9	-	-	-	-
Having knowledge about CWD								
Yes	14	29.2	34	70.8	1.81 (0.86–3.81)	0.1162	-	-
No*	28	18.5	123	81.5	-			
BMI percentiles								
<5%*	6	20	24	80				
5%–84%	32	19.9	129	80.1	1.01 (0.38–2.67)	0.988	-	-
85%–94%	1	50	1	50	0.25 (0.01–4.60)	0.351		
≥95%	3	50	3	50	0.25 (0.04–1.56)	0.138		

*Reference group.

CWD prevalence rates were higher in boys than in girls, again consistent with the literature (9,15).

The PE/PC ratio was reported as 2.2–5/1 in different studies (9,11,15–18). When the literature in Turkey is examined, it is reported that the prevalence of PE deformities is higher than the prevalence of PC (15,19). Although PE prevalence is higher than some PCs, PC was reported higher than PE in Argentinean and African

populations (10). In our study, PC prevalence was found higher than PE (PC/PE = 1.59), similar to the Argentinean and African populations.

Coexistence of CWD and scoliosis is generally accepted knowledge. While Westphal et al. reported this coexistence as 15% among Brazilian students, this rate was found as 5.03% in our study (11). This difference may be due to the lack of concurrent use of inspection and radiological examination in our study. There was no association between scoliosis and deformity degree, which was also reported by Frick et al. (20).

Deformity awareness is higher among children with a positive family history, particularly in closed societies. Awareness increases in children with a positive family history of CWD, as this disease is known within the family.

Ample studies are available in the literature reporting that CWD affects cardiopulmonary functions. Cardiopulmonary functions were reported to improve postoperatively. In particular, FEV1 was reported to improve 3 years after the pectus bar was removed in PE and cardiac symptoms were reported to improve as the heart reached its normal position (21–24). Loff et al. (25) reported that PC did not cause physical disturbances; however, it caused psychological disturbances. Koumbourlis et al. (26) reported that there were no physical symptoms during rest in PE and obstructive pulmonary disease prevalence was high. On the other hand, Coskun et al. (15) reported that restrictive pulmonary disease prevalence was higher in PE cases. More than half of CWD patients in our study did not have physical complaints and there was not a significant impairment in respiratory function tests. Westphal et al. (11) reported chest pain as the most common physical disturbance. However, we found dyspnea as the most common physical disturbance. In our study, the physical disturbance rate was found to be higher in the 11 years age group and among children who were aware of the disease in multivariate logistic regression analysis. This may be because the deformity becomes prominent mostly around the age of 11. Higher reporting of physical disturbance in children who were aware of the disease suggests that the deformity affects the child psychologically as well as physically.

The main reason for correcting CWD is reported as correcting body image rather than correcting physical disturbances and related psychosocial problems (15,27–29). Hadolt et al. (30) reported positive outcomes after Nuss operation in PE cases in the young who did not have self-esteem and who felt bad psychologically. Ji et al. (31) reported that children with PE had more psychosocial problems compared to the general population and being between 12 and 16 years old, deformity severity, and being mocked negatively affected these problems. In our study, psychological disturbances were found higher in females and in the presence of family history in multivariate logistic regression analysis. Increased awareness and increased physical disturbance in children with a family history seems to arise from the reflection of psychological disturbance as a physical disturbance, rather than being due to increased physical disturbance. Girls caring about body image more than boys and being more sensitive than boys has resulted in sex being an important factor that affects the presence of psychological disturbance in CWD. Awareness naturally increased as deformity degree increased and deformity became more evident as its degree increased.

In this study conducted in a large population, CWD was found more prevalent in boys (0.96%) and PC was found as the most common deformity type in our country. Being in the 11 year age group and deformity awareness were found to be effective on the presence of physical disturbance in CWD while family history and being female were found to be effective on psychological disturbance.

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