



## THE DEVELOPMENT OF SCOLIOSIS AND KYPHOSIS IN PATIENTS WHO HAVE UNDERGONE STERNOTOMY OR THORACOTOMY DUE TO CONGENITAL HEART DISEASE (CHD)

### KONJENİTAL KALP HASTALIĞI NEDENİYLE STERNOTOMİ VEYA TORAKOTOMİ YAPILMIŞ HASTALARDA SKOLYOZ VE KİFOZ GELİŞİMİ

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#### SUMMARY:

**Objectives:** To evaluate the prevalence of scoliosis and kyphosis development in patients who have been treated surgically with medial sternotomy or left thoracotomy due to congenital heart anomalies.

**Patients and Methods:** 170 patients treated with surgery due to congenital heart diseases (CHDs) between 1987 and 2009 were included in this study. 102 of the patients were female (60%) and 68 were male (40%). The CHDs were grouped as cyanotic (149 patients, 87.6%) and acyanotic (21 patients, 12.4%). The number of the patients who were operated on before the age of 5 was 131 (77%), and the number over the age of 5 was 39 (23%). Sternotomy was applied to 158 patients, and the other 12 received left thoracotomy. The measurements were made using the Cobb technique, using standing PA and lateral chest X-rays taken during routine follow-up. The kyphosis angle was measured separately on the lateral thoracic X-rays at two levels, one between T2 and T5, and the other between T5 and T12.

**Results:** We detected scoliosis in 25.2% of the patients (43 out of 170). While the Cobb measurements were between 10 to 20 degrees in 86% of the cases, the rest (14%) were over 20 degrees. It was found that there was no statistical significance with regards to the scoliosis progression between the sternotomy and thoracotomy patients ( $p=0.161$ ). The average kyphosis angle was found to be  $32.92^\circ$  in the sternotomy patients, and  $29.67^\circ$  in the thoracotomy patients. The average T2-5 kyphosis was  $14.5^\circ$  and the average T5-12 kyphosis was  $16.2^\circ$ . There were no statistically significant differences between the thoracotomy and sternotomy patients with regard to scoliosis progression ( $p=0.532$  and  $p=0.107$ ). The T2-5 kyphosis angles of the scoliosis patients were found to be considerably lower when compared to patients without scoliosis. The scoliosis prevalence rate was found to be increased by 8.42-fold in children operated on under the age of 5 with respect to those operated on over the age of 5. There were no statistically significant differences in whether the heart disease was cyanotic or acyanotic with respect to scoliosis or kyphosis progression ( $p=0.362$ ,  $p=0.586$ ,  $p=0.184$ ).

**Conclusion:** The scoliosis prevalence rate is found to be increased in patients treated with either sternotomy or thoracotomy due to CHD, and the T2-5 kyphosis is also found to be increased compared to the normal population. These spinal deformities are more commonly observed in children operated on under the age of 5.

**Key Words:** Scoliosis, kyphosis, etiology, congenital heart disease

**Level of Evidence:** Retrospective clinical study, Level III

#### ÖZET:

**Amaç:** Konjenital kalp anomalisi nedeniyle medyan sternotomi veya torakotomi ile cerrahi tedavi uygulanmış hastaların uzun dönem takiplerinde skolyoz ve kifoz prevalansını araştırmak.

**Hastalar ve Metod:** 1987 ile 2009 yılları arasında konjenital kalp hastalığı nedeniyle ameliyat edilmiş 170 hasta çalışmaya dâhil edilmiştir. Hastaların 102'si kız, 68'i erkektir. Hastaların mevcut kalp hastalıkları siyanotik ve asiyantotik olarak iki gruba ayrılmıştır, buna göre siyanotik olanlar 149 (% 88), asiyantotik olanlar ise 21 kişidir (%12). Ameliyat oldukları dönemde 5 yaşın altında olanlar 131 (% 77), 5 yaş üzerinde olanlar ise 39 kişidir (% 23). Hastaların 158'ine (%93) sternotomi, 12'sine (% 7) ise sol torakotomi uygulanmıştır. Hastaların kontrole geldiği tarihte ayakta çekilen toraks PA ve lateral grafileri üzerinden Cobb yöntemiyle ölçümler yapılmıştır. Lateral toraks grafilerinde T2-T5 kifozu ve T5-T12 kifozu ayrı ayrı ölçülmüştür.

**Sonuçlar:** Çalışmamızda 170 hastanın % 25.2'sinde (43 hasta) skolyoz saptanmıştır. Bunlardan % 86'sında 100 ile 200 arasında eğrilik, % 14'ünde 200 ve üzerinde eğrilik saptanmıştır. Sternotomi yapılan hastalarda skolyoz gelişme oranı ile torakotomi yapılanlar arasında istatistiksel olarak bir fark bulunmamıştır ( $p=0.161$ ). Hastaların ortalama kifoz açıları sternotomi yapılanlarda  $32.920$  iken, torakotomi yapılanlarda  $29.670$  saptanmıştır. T2-T5 kifozu ortalaması  $14.50$  iken T5-T12 kifozu  $16.20$ 'dir. Kifoz gelişimi açısından torakotomi yapılan hastalarla sternotomi yapılanlar arasında anlamlı fark bulunmamıştır ( $p=0.532$  ve  $p=0.107$ ). Skolyoz gelişen hastaların T2-T5 kifozu skolyozu olmayanlara göre anlamlı derecede düşük bulunmuştur. 5 yaş altında ameliyat edilenlerde skolyoz görülme oranı, 5 yaşından büyük olanlara göre 8.42 kat daha fazla saptanmıştır. Kalp hastalığının siyanotik olmasının skolyoz ve kifoz gelişimi açısından anlamlı bir farkı yoktur ( $p=0.362$ ,  $p=0.586$ ,  $p=0.184$ ).

**Sonuç:** Konjenital kalp hastalığı nedeniyle sternotomi ve torakotomi yapılan hastalarda skolyoz gelişme sıklığı artmış olup, T2-T5 kifozu da normal popülasyona göre daha fazla bulunmuştur. Daha küçük yaşta ameliyat olanlarda omurga deformiteleri daha sık ortaya çıkmaktadır.

**Anahtar Kelimeler:** Skolyoz, kifoz, etiyoloji, konjenital kalp hastalığı

**Kanıt Düzeyi:** Retrospektif klinik çalışma, Düzey III

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## INTRODUCTION:

The prevalence of congenital heart disease (CHD) in newborns is 1%. CHD is defined as a risk factor for thoracic scoliosis, but the underlying reasons are unknown<sup>10</sup>.

In open heart surgery, the major surgical approaches are sternotomy, thoracotomy, or both. Scoliosis is also detected after non-cardiac sternotomy and thoracotomy procedures<sup>3,5,7</sup>. It is believed that not the cardiac disease itself, but the surgery, resulting in damage to the anterior chest wall, has a role in scoliosis development. There have been some studies on the development of scoliosis in children who have undergone thoracotomy and sternotomy. Most CHD patients receive median sternotomy, and the scoliosis prevalence is found to be higher in this group<sup>1</sup>. If the surgical approach is thoracotomy, the incidence of scoliosis is higher regardless of the CHD.

The aim of this study is to investigate the effects of cyanotic and acyanotic states of CHD and the age at which the surgery takes place for patients undergoing sternotomy or thoracotomy for CHD on scoliosis and kyphosis development in the mid-phase.

## PATIENTS AND METHOD:

173 patients who received surgery for CHD between 1987 and 2009 were included in the study. Three patients were diagnosed with scoliosis according to the PA chest X-rays at the time of surgery and excluded from the study. 170 patients were included. 102 of the patients were female and 68 of them were male. The cardiac diseases of the patients were categorized as either cyanotic or acyanotic, and

87.6% (149 patients) were cyanotic while 12.4% (21 patients) were acyanotic. 77% (n=131) of the patients at the time of surgery were under the age of 5, and 23% (n=39) were older than 5 years of age. 93% of (n=158) the patients had sternotomy, and 7% (n=12) had thoracotomy.

Cobb angle measurements were made using thoracic PA and lateral X-rays taken in the follow-up appointments. In the lateral thoracic X-rays, the T2–5 and T5–12 kyphosis were measured separately.

## Statistical Analysis:

In this study, all statistical analyses were performed using the NCSS (Number Cruncher Statistical System) 2007 package program. In the evaluation of data, descriptive statistics (average, standard deviation, median, inter-quartile range) were employed. For the comparison of two groups, the Mann Whitney U-Test was used. For the comparison of qualitative data, the chi-square test was employed. Results were considered significant with a p-value < 0.05.

## RESULTS:

25.2% (43 patients) of the 170 patients included in the study had scoliosis. 86% (37 patients) of those had 10–20° of curvature and 14% (6 patients) had ≤20° of curvature (Table-1).

The age values at the time of operation of the scoliosis (+) group were lower than that of the scoliosis (-) group, with statistical significance (p=0.002). No statistically significant distribution was observed between the gender distributions of the scoliosis (+) and scoliosis (-) groups (p=0.084).

No statistically significant differences between the distribution according to the type of surgery were found between the scoliosis (+) and scoliosis (-) groups (p=0.161). Surgery at <5 years of age was found to be significantly associated with the scoliosis (+) group, with n=42 (95.3%), compared to the scoliosis (-) group with n=90 (70.9%) (p=0.001).

The scoliosis prevalence in the <5 age group was found to be 8.42 times higher than the >5 age group. There were no statistically significant differences between the scoliosis (+) and scoliosis (-) groups according to the presence of cyanosis (p=0.482) (Table-2).

When the relationship between scoliosis and kyphosis were investigated, the T2-5 kyphosis values were found to be significantly lower in the scoliosis (+) group than in the scoliosis (-) group (p=0.0001).

There were no statistically significant differences between the T5-12 kyphosis values of the scoliosis (+) and scoliosis (-) groups (p=0.152) (Table-3) (Figures-1, 2, and 3).

The test age values of the sternotomy group were significantly higher than the thoracotomy group (p=0.039). The operation age values of the sternotomy group were found to be significantly higher than the thoracotomy group (p=0.017).

**Table-1.** Comparison of epidemiological properties of patients with and without scoliosis.

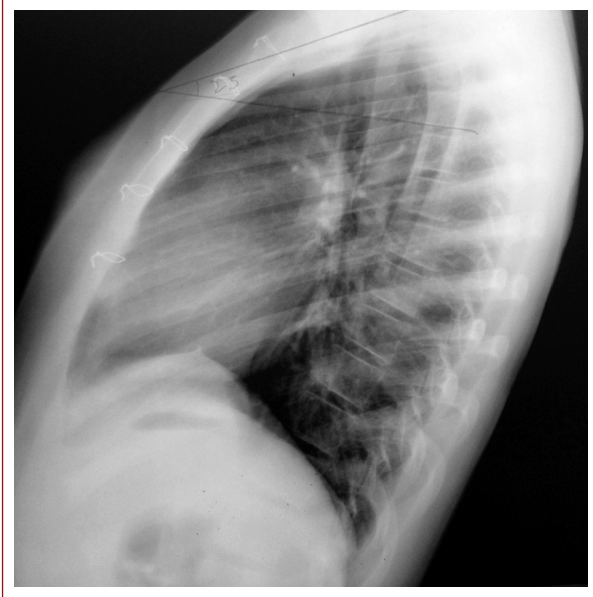
|                    |                             | Scoliosis (-) n:127            | Scoliosis (+) n:43          | MW                   | p            |
|--------------------|-----------------------------|--------------------------------|-----------------------------|----------------------|--------------|
| <b>Control age</b> | <b>Avrg±SD Median (IQR)</b> | 8.63±4.97<br>7.34 (5.91-10.51) | 6.97±4.8<br>6.26 (3-9.59)   | 2096                 | <b>0.023</b> |
|                    | <b>Operation Age</b>        | 3.99±4.51<br>3.04 (0.85-5.92)  | 1.8±1.93<br>1.10(0.42-2.88) | 1862                 | <b>0.002</b> |
| <b>Gender</b>      | <b>Female</b>               | 81 63.80%                      | 21 48.80%                   | x <sup>2</sup> :2.98 |              |
|                    | <b>Male</b>                 | 46 36.20%                      | 22 51.20%                   | p=0.084              |              |

**Table-2.** Distribution of the patients according to age at surgery and cyanotic status of the disease.

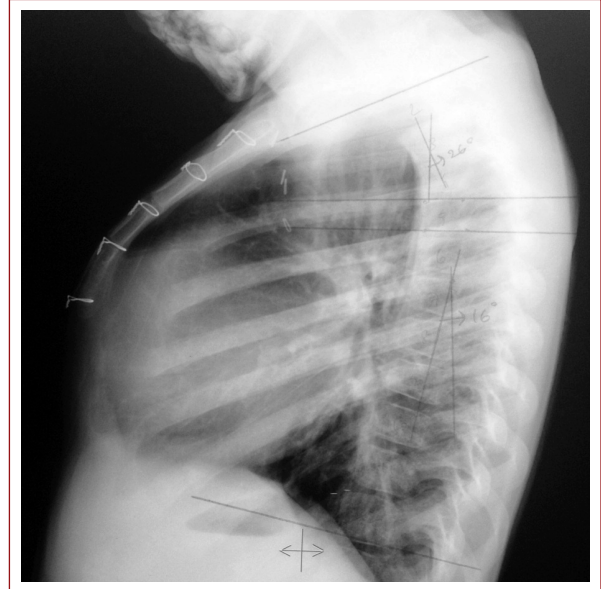
|                          |                          | Scoliosis (-) n:127 |        | Scoliosis (+) n:43 |        | OR 95% GA             |             |
|--------------------------|--------------------------|---------------------|--------|--------------------|--------|-----------------------|-------------|
| <b>Type of operation</b> | <b>Sternotomy group</b>  | 116                 | 91.30% | 42                 | 97.70% | x <sup>2</sup> :1.96  | 0.251       |
|                          | <b>Thoracotomy group</b> | 11                  | 8.70%  | 1                  | 2.30%  | p=0.161               | 0.031-2.01  |
| <b>Age of operation</b>  | <b>&lt;5 age</b>         | 90                  | 70.90% | 41                 | 95.30% | x <sup>2</sup> :10.89 | <b>8.42</b> |
|                          | <b>&gt;5 age</b>         | 37                  | 29.10% | 2                  | 4.70%  | <b>p=0.001</b>        | 1.94-36.67  |
| <b>Cyanosis</b>          | <b>Cyanosis (-)</b>      | 110                 | 86.60% | 39                 | 90.70% | x <sup>2</sup> :0.49  | 0.66        |
|                          | <b>Cyanosis (+)</b>      | 17                  | 13.40% | 4                  | 9.30%  | p=0.482               | 0.21-2.09   |

**Table-3.** Scoliosis and kyphosis relationship in patients.

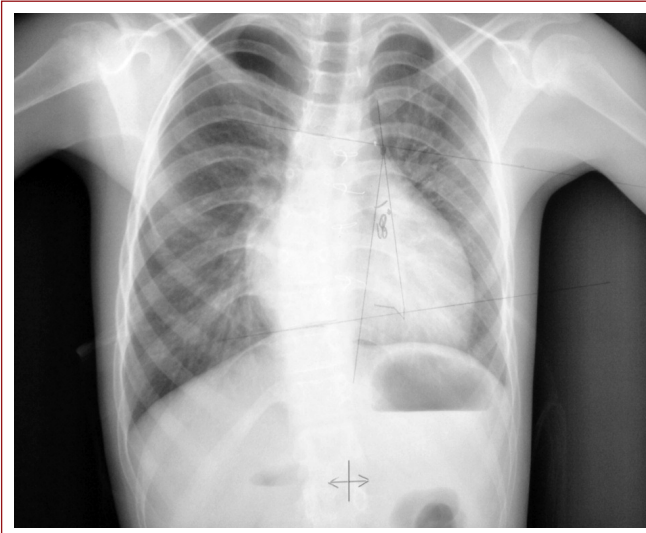
|                 |                      | Scoliosis (-) n:127 degree | Scoliosis (+) n:43 degree | MW   | p      |
|-----------------|----------------------|----------------------------|---------------------------|------|--------|
| t2-t5 Kyphosis  | Avrg±SD Median (IQR) | 15.9±6.58<br>15 (11-20)    | 12.47±5.7<br>10 (10-15)   | 1682 | 0.0001 |
| t5-t12 Kyphosis | Avrg±SD Median (IQR) | 17.44±5.03<br>18 (14-20)   | 18.3±5.95<br>20 (15-21)   | 2223 | 0.152  |



**Figure-1.** Lateral thoracic X-ray showing increased T2-5 kyphosis (25°) of a patient who underwent sternotomy taken at the year 7 follow-up.



**Figure-3.** Lateral thoracic X-ray showing increased T2-5 kyphosis (26°) and decreased T5-12 kyphosis (16°) of a patient who underwent sternotomy taken at the year 9 follow-up.



**Figure-2.** Thoracic PA X-ray showing thoracic scoliosis (18°) of a patient who underwent sternotomy taken at the fourth year follow-up.

There were no significant differences between the gender distribution of the sternotomy and thoracotomy patients ( $p=0.903$ ) (Table-4).

There was no statistical significance in the operational age, whether <5 years of age or >5 years of age, between the sternotomy and thoracotomy groups ( $p=0.070$ ).

The scoliosis distribution of the sternotomy and thoracotomy groups was not found to be significant ( $p=0.161$ ). The presence of cyanosis in the thoracotomy group ( $n=4, 33.3\%$ ) was significantly higher than in the sternotomy group ( $n=17, 10.8\%$ ) ( $p=0.022$ ) (Table-5).

There was no significance between the scoliosis values of the sternotomy and thoracotomy groups ( $p=0.223$ ). There was no significance between the T2–5 kyphosis values of the sternotomy and thoracotomy groups ( $p=0.532$ ). There was also no significance between the T5–12 kyphosis values of the sternotomy and thoracotomy groups ( $p=0.107$ ) (Table-6).

No statistically significant differences were observed between the scoliosis values of the cyanosis (+) and cyanosis (-) groups ( $p=0.362$ ). The T2–5 kyphosis values of the cyanosis (+) and cyanosis (-) groups were not significant ( $p=0.586$ ).

There was no statistically significant difference in the T5–12 kyphosis values of the cyanosis (+) and cyanosis (-) groups ( $p=0.184$ ) (Table-7).

## DISCUSSION:

The incidence of adolescent idiopathic scoliosis is 2–3%. The prevalence of scoliosis in the population with congenital heart disease is 2–24%<sup>9,10</sup>. The development of scoliosis in the population with congenital heart disease depends on many factors<sup>4</sup>. An increase in the incidence of scoliosis in children with congenital abnormalities has been shown in the literature<sup>7,10,11</sup>, although a study by Feiz et al. showed no difference<sup>6</sup>.

It has been shown that the development of scoliosis after open heart and chest surgeries may be due to the surgical approach<sup>3,5,7</sup>. Gilsanz et al. showed scoliosis in eight patients who had undergone surgery for esophageal atresia<sup>7</sup>. Durning et al. showed scoliosis in nine of 18 patients who received thoracotomy for a trachea-esophageal fistula<sup>5</sup>.

**Table-4.** Age and gender comparison according to sternotomy and thoracotomy.

|               |                     | Sternotomy group n:158 |        | Thoracotomy group n:12 |        | MW             | p         |
|---------------|---------------------|------------------------|--------|------------------------|--------|----------------|-----------|
| Control Age   | Avg±SD Median (IQR) | 8.38±5.01              |        | 5.97±3.72              |        | 608.5          | 0.039     |
|               |                     | 7.27 (5.63-10.20)      |        | 6.12 (3.27-7.28)       |        |                |           |
| Operation Age | Avg±SD Median (IQR) | 3.59±4.22              |        | 1.33±1.83              |        | 556.5          | 0.017     |
|               |                     | 2.28 (0.51-4.95)       |        | 0.71(0.084-2.88)       |        |                |           |
| Gender        | Female              | 95                     | 60.10% | 7                      | 58.30% | $\chi^2:0.015$ | $p=0.903$ |
|               | Male                | 63                     | 39.90% | 5                      | 41.70% |                |           |

**Table-5.** Patients' operation age, presence of cyanotic heart disease and presence of scoliosis, according to implementation of sternotomy or thoracotomy.

|               |               | Sternotomy group n:158 |        | Thoracotomy group n:12 |         |               |
|---------------|---------------|------------------------|--------|------------------------|---------|---------------|
| Operation age | <5 Age        | 119                    | 75.30% | 12                     | 100.00% | $p=0.070$     |
|               | >5 Age        | 39                     | 24.70% | 0                      | 0.00%   |               |
| Scoliosis     | Scoliosis (-) | 116                    | 73.40% | 11                     | 91.70%  | $\chi^2:1.96$ |
|               | Scoliosis (+) | 42                     | 26.60% | 1                      | 8.30%   | $p=0.161$     |
| Cyanosis      | Cyanosis (-)  | 141                    | 89.20% | 8                      | 66.70%  | $\chi^2:5.24$ |
|               | Cyanosis (+)  | 17                     | 10.80% | 4                      | 33.30%  | $p=0.022$     |

**Table-6.** Scoliosis and kyphosis relationship in patients according to sternotomy or thoracotomy operation.

|                 |                       | Sternotomy group n:158        | Thoracotomy group n:12           | MW    | p     |
|-----------------|-----------------------|-------------------------------|----------------------------------|-------|-------|
| Scoliosis       | Avrg ±SD Median (IQR) | 3.62±6.30<br>0 (0-10) 0 (0-0) | 1.83±6.35                        | 795   | 0.223 |
| t2-t5 Kyphosis  | Avrg±SD Median (IQR)  | 15.11±6.66<br>14 (10-20)      | 14±4.51 846.5<br>13.5 (10-16.75) | 0.532 |       |
| t5-t12 Kyphosis | Avrg±SD Median (IQR)  | 17.81±5.31<br>18 (14-20)      | 15.67±4.83<br>14.5 (12.5-19.5)   | 685   | 0.107 |

**Table-7.** Kyphosis and scoliosis relationship in the patients according to cyanotic or acyanotic heart disease.

|                 |                      | Cyanosis (-) n:149       | Cyanosis (+) n:21        | MW   | p     |
|-----------------|----------------------|--------------------------|--------------------------|------|-------|
| Scoliosis       | Avrg±SD Median (IQR) | 3.68±6.48<br>0 (0-10)    | 2.19±4.77<br>0 (0-0)     | 1417 | 0.362 |
| t2-t5 Kyphosis  | Avrg±SD Median (IQR) | 14.93±6.67<br>14 (10-20) | 15.71±5.5<br>15 (10-20)  | 1451 | 0.586 |
| t5-t12 Kyphosis | Avrg±SD Median (IQR) | 17.82±5.44<br>18 (14-20) | 16.52±3.83<br>16 (14-20) | 1286 | 0.184 |

A study by Van Biezen et al. showed increased scoliosis development in children who had undergone a thoracotomy procedure for coarctation of aorta<sup>13</sup>. Ruiz-Iban et al. surveyed 128 children with CHD who had undergone a median sternotomy procedure and observed scoliosis in 34.4%<sup>12</sup>. Herrera-Soto et al. surveyed 68 children who had undergone first thoracotomy and then sternotomy for CHD, and found scoliosis in 26%<sup>9</sup>. Another study by Herrera-Soto et al. showed that 28% of 108 children who had undergone sternotomy had scoliosis<sup>8</sup>.

In our study, 25.2% of the 170 patients had scoliosis. 86% of them had curvature between 10° and 20°, and 14% of the patients had more than 20° curvature.

Other studies in the literature have conducted global kyphosis calculations. Increases in the T2–5 kyphosis were a result of decreased T5–12 kyphosis, and evaluated as a compensatory deformity<sup>2</sup>. In our study, it was determined that cessation of the sternal growth did not increase T5–12 kyphosis, in opposition to the predictions of other studies.

Studies have shown the relationship between cyanotic heart disease and increasing spinal deformities, and also cyanosis and scoliosis. Herrera-Soto et al. showed that the development of scoliosis in cyanotic heart diseases was 4.5 times higher, and the curvature was more prominent<sup>9</sup>.

The tight closure of the sternum after osteotomy may result in early obtundation of the growth of the sternum<sup>8</sup>.

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In a study by Herrera-Soto et al., 21% of the patients had increased kyphosis, 9% of them were in the scoliosis patient group and 12% were in the other group<sup>9</sup>. In our study, the kyphosis measurements were conducted in two different regions, T2–5 and T5–12. The T2–5 kyphosis was found to be increased, and the T5–12 kyphosis was decreased, compared to a normal population.

In our study, the development of scoliosis and kyphosis in cyanotic heart disease was not seen to be statistically significant when compared to acyanotic heart disease.

The longitudinal growth of the spine is 2 cm/year aged 0–5 years, 0.9 cm/year aged 5–10 years, and 1.8 cm/year from 10 years of age to puberty<sup>6</sup>. In this study, the age of the surgery and the surveillance periods of the patients were variable, but 95.3% (n=41) of the patients who developed scoliosis were operated on before the age of 5, and only 4.7% (n=2) of the patients were older than 5.

As a result, this study shows that sternotomy or thoracotomy operations pose a risk for spinal deformity development. The most important limitation of this study is a lack of surveillance of all of the patients up to the age of puberty.

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