Pediatric scoliosis and the lung

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ABSTRACT

Objective: To determine the pattern of scoliosis in the pediatric age group and to determine the course of the disease and the result of surgical intervention.

Methods: A retrospective review of all charts of patients referred to the Pulmonary Clinic of the King Faisal Specialist Hospital, Riyadh, Kingdom of Saudi Arabia (KSA), for scoliosis and preoperative evaluation from the period November 1992 to October 2001. Demographic and clinical data, preoperative and postoperative pulmonary function test (PFT) and effect of surgery on the lung were presented.

Results: A total of 66 patients with scoliosis were followed. The age at diagnosis was 8.5 ± 4.4 years. Thirty-four (52%) males and 32 (48%) females. Fifty-seven (86%) were alive and 9 (14%) died. A period of follow up was 5.3 ± 3.7 years. Idiopathic scoliosis was found in 20 patients (30%), congenital anomalies associated with scoliosis constituted 35 (53%) of the total scoliosis referral and secondary type due to trauma or malignancy formed 11 (17%). Mean Cobb's angle of 70 ± 30 degrees. The most common curvatures were: thoracic 31 (47%), lumbar 12 (18%) and thoracolumbar 30 (45%). Thirteen patients (20%) had 2 or 3 types of curvatures simultaneously. Baseline PFT showed moderate restrictive lung disease in approximately two thirds of the patient population. Scoliosis surgery was carried out in 34 (52%) of the patients. Posterior approach was more commonly used in 30 (88%) of the patients. Pneumonia and atelectasis were the most common complication encountered in 15 (44%) of the patients. Recurrence of scoliosis after operation recurred in 12 (35%) of the population. Comparisons between both groups have shown that patients with congenital anomalies and scoliosis were diagnosed earlier, had significant postoperative complications and higher mortality compared to those with idiopathic scoliosis ($p<0.05$). Changes in PFT for patients who underwent surgery for scoliosis with Cobb's angle >40 degrees showed less deterioration compared to those with angle <40 degrees ($p=0.004$), due to progressive deterioration without surgery.

Conclusion: Scoliosis is a common problem in KSA. Patients need to be referred early for proper intervention before it becomes a severe magnitude and difficult to be fixed and to prevent permanent complications. Patients with congenital anomalies and scoliosis had poor prognosis compared to those with idiopathic scoliosis.

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Scoliosis develops as a result of an underlying pathological process, which leads to a lateral curvature of the spine. This in turn causes a rotation of the vertebral bodies and distortion of all attached structure that leads to the characteristic prominence of the rib cage on the convex side and anterior flattening and widening of the interspaces. As a consequence of such distortion, the patient may suffer functional pulmonary disabilities.[1] The most common type of scoliosis is the idiopathic form, which consists of 85% of all scoliosis population.[1,2] It is divided into 3 forms: infantile (<3 years), juvenile (3-9 years) and adolescent (10 years old to skeletal maturity).[2] The second type is congenital form (5%), which is formed due to failure of formation or segmentation of the vertebra.[3] The third one is the secondary type due to cerebral palsy,[4] vertebral trauma[5] or spinal cord tumor as neuroblastoma.[1] The most common detectable pulmonary function abnormality is a restrictive pattern when the angle is >50-60°.[2,6-9] It is usually progressive if not detected and treated early before the adolescent growth spurt to reduce late complication.[4] Cobb's angle

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is the term used for measurement of the magnitude of the curvature that is determined with x-ray of the spine.[2] The most common detectable pulmonary function abnormality is a restrictive pattern when the angle is >50-60º.[2] Anomalies in congenital scoliosis occur at sites other than the spine in 30-60% of children.[8,10-13] The most common sites are: the genito-urinary tract, the cardiac system, and the spinal cord.[10-13] Many studies have shown that congenital scoliosis have been associated with vatar association and Klipple-Feil syndrome. Intraspinal anomalies such as tethered cord, diastomyelia, syringomyelia and low conus medullaris were described in 18-38% of neurologically normal patients.[3] Genito-urinary defect occurred in 20-40% of patients with vertebral anomalies. Restrictive lung disease may develop in curve magnitude >60º. Idiopathic scoliosis is usually progressive if not detected and treated early before the adolescent growth spurt to reduce late complication. Bracing can be used in juvenile and adolescent types if Cobb's angle <50º, but surgery is indicated if angle >50º.[14-17] Harrington's and Luque rods are the most common rods that are used to fix the curvature.[15] The most common postoperative complication is those related to the lung, which includes: atelectasis, pneumonia, pulmonary edema, respiratory failure, inability to clear secretion and reduction of lung volumes, which may improve 2 years after surgery.[15,16] Other complications that are related to the surgical procedures are: wound infection, neurological impairment and detachment of the fixation rod.[15] In this report, we carried out a retrospective chart review to determine the demographic and clinical data on patients with scoliosis. Preoperative, postoperative pulmonary function test (PFT) and effect of surgery on the lung are also presented. A comparison between patients with idiopathic and those who have congenital anomalies in combination with scoliosis is discussed.

Methods. A retrospective review of all charts of patients referred to Pulmonary Clinic of the King Faisal Specialist Hospital and Research Centre, Riyadh, KSA, for scoliosis and preoperative evaluation from the period November 1992 to October 2001. Demographic and clinical data was collected. Type of scoliosis, pattern of curvatures and the magnitude of the Cobb's angle, pulmonary function test with preoperative and postoperative values were determined. Use of brace, type of surgery, type of instrument used, and blood gas status was determined. Postoperative complications were identified. Idiopathic scoliosis is defined as scoliosis with no apparent cause and no associated anomalies. Secondary scoliosis is a result from causes such as trauma, poliomyelitis and cerebral palsy. Congenital scoliosis is defined as scoliosis secondary to defects in vertebral formation or segmentation or other congenital anomalies or syndromes that are associated with spinal deformity that is detected at birth, such as myelomeningocele, muscular dystrophy, congenital myopathy, vatar association, Klippel Feil syndrome, limb anomalies such as arthrogryposis multiplex congenita.

Statistical analysis. The data was entered into an EXCEL spreadsheet. They were then transferred into JMP statistical package (version 3.2, SAS Institute Inc.) where the statistical analyses were performed. For continuous variables, histograms were reviewed and means, SD and median were calculated. The following comparisons using the student t-test were made for all variables by status (alive and dead) and by sex. Additionally, the group with congenital scoliosis and congenital anomalies were compared with idiopathic scoliosis by status and then separately by sex. Chi square was calculated for all nominal variables by status (alive and dead) and by sex. Results were presented at a level of significance of $p<0.05$

Results: A total of 66 patients with scoliosis were followed. Thirty-four (52%) males and 32 (48%) females. Fifty-seven (86%) were alive and 9 (14%) have died. Age when scoliosis was noted by parents was 4.6 ± 3.7 years. The age at diagnosis in our center was 8.5 ± 4.4 years. Period of follow up was 5.3 ± 3.7 years. Idiopathic scoliosis was found in 20 patients (30%) (**Figures 1a & 1b**), scoliosis associated with congenital anomalies consisted of 35 patients (53%) (**Figure 2**), and 11 patient (17%) with secondary scoliosis (**Table 1**) due to cerebral palsy, malignancy **(Figures 3a & 3b)**, trauma, or poliomyelitis. Mean Cobb's angle of 70 ± 30 degrees. The most common curvatures were: cervical curvature formed 2 (3%), thoracic 31(47%), lumbar 12 (18%) and thoracolumbar 30 (45%). Thirteen patients (20%) had 2 or 3 types of curvatures simultaneously. Sixty-two (94%) were progressive in nature while waiting for surgery. The brace was used in 25 (38%) in the preoperative period while 14 (21%) was used in the postoperative period (**Table 2**). Baseline blood gas was within normal limits. Baseline PFT was carried out in 41 patients (62%) of the patient population, which showed a restrictive pattern in 33 (52%) of the patient population, combined obstructive and restrictive patterns in 4 (10%) (**Table 2**), normal pattern in 4 (10%). Mild restrictive pattern in one (3%) of the patient population, moderate in 14 (35%), severe in 21 (52%) and normal in 4 (10%). Scoliosis surgery was carried out in 34 (52%) of patients (**Table 2**) (**Figure 1b & Figure 4**). Posterior approach was more commonly used in 30 (88%) of the patients. Harrington rod was used in 22 (65% [**Figure 4**]), whereas Luque rod was used in 7 (21%) (**Figure 1b**). Pneumonia and atelectasis were the most common complication encountered in 15 (44%) of the patients (**Table 3**). There were 10 patients who had 3 or more complications simultaneously. Recurrence of scoliosis after operation recurred in 12 (18%) of the population (**Table 3**). Nine patients have died (14%) (**Table 4**). This included patients who did not go for surgery and progressed quickly and developed respiratory and

cardiac impairment 6 (9.5%), patients with limb anomalies one (1.5%), and patients with congenital anomalies or syndromes 3 (4.5%) (**Table 4**).

Idiopathic scoliosis (Table 2). Of the 20 patients with idiopathic scoliosis, 10 (50%) were males and 10 (50%) were females (**Table 2**). Age at diagnosis was 10 ± 4 years, average of 1.8-16 years. Nineteen (95%) are alive and one (5%) died. Eighteen (90%) patients had a progressive pattern. Cervical curvature was found in one (5%) of the patient population with a Cobb's angle of 90^0, thoracic curvature in 13 (65%) with an angle of 71 ± 29^0, lumbar curvature in 4 (20%) with an angle of 63 ± 25^0 and thoracolumbar in 6 (30%) with an angle of 79 ± 35^0 of the idiopathic population. Nine (45%) of the patients had combined curvatures of 2 or 3 types in the same patient (**Table 2**). Of the 6 (30%) patients who were unable to do PFT, 3 were below 5 years and remained clinically stable and the other 3 had progressive deterioration of their curve and one of them died before any surgical intervention. Surgery with instrumentation was carried out in 11 (55%) at the age of 11 ± 3 years (**Figure 1b**). Posterior approach for surgery was the main type applied in 10 (50%). Harrington rod was used in 9 (46%). The main complication of surgery was pneumonia and atelectasis in 4 (20%) (**Table 2**). Recurrence of scoliosis after surgery occurred in 5 (25%) (**Table 2**). Comparison between PFT that was carried out in preoperative and postoperative period showed stabilization of all values with no apparent deterioration.

Congenital scoliosis (Table 1 & 2). Of the 35 patients with congenital scoliosis, 30 (85%) had scoliosis associated with congenital anomalies (**Figure 2**) and genetic diseases (**Figure 4**), and 5 (15%) with an isolated congenital scoliosis of the spine due to a defect in vertebral formation, development or vertebral segmentation with no apparent associated anomalies (**Table 1**). Sixteen (46%) males and 19 (54%) females. Twenty-seven (77%) are alive and 8 (23%) died (**Table 2**). The most common associated congenital anomalies and genetic diseases are: 9 muscular dystrophy, 5 myelomeningocele, 5 with isolated congenital scoliosis and no congenital anomalies and the rest are different syndromes that constitute 1-2 cases each (**Table 1**). The age when scoliosis noted by parents was 4 ± 3.4 years. The age when referred to a tertiary care center for proper management was 7.4 ± 4.3 years (**Table 2**). The patients were followed for 5.8 ± 4 years. Thirty-three (94%) patients had a progressive pattern. Cervical curvature was found in one (3%) of the patient population with a Cobb's angle of 70º, thoracic curvature in 12 (34%) with an angle of 66 ± 26, lumbar curvature in 7 (20%) with an angle of 78 ± 42 and thoracolumbar in 19 (54%) with an angle of 59 ± 24 (**Table 4**). There were 6 (17%) of patients that had 2 or 3 types of curvatures simultaneously. Of 27 patients who were able to do baseline PFT, the result of PFT test showed 23 (86%) with restrictive pattern (**Table 2**). Surgery with instrumentation was carried out in 20 (57%) at an age of

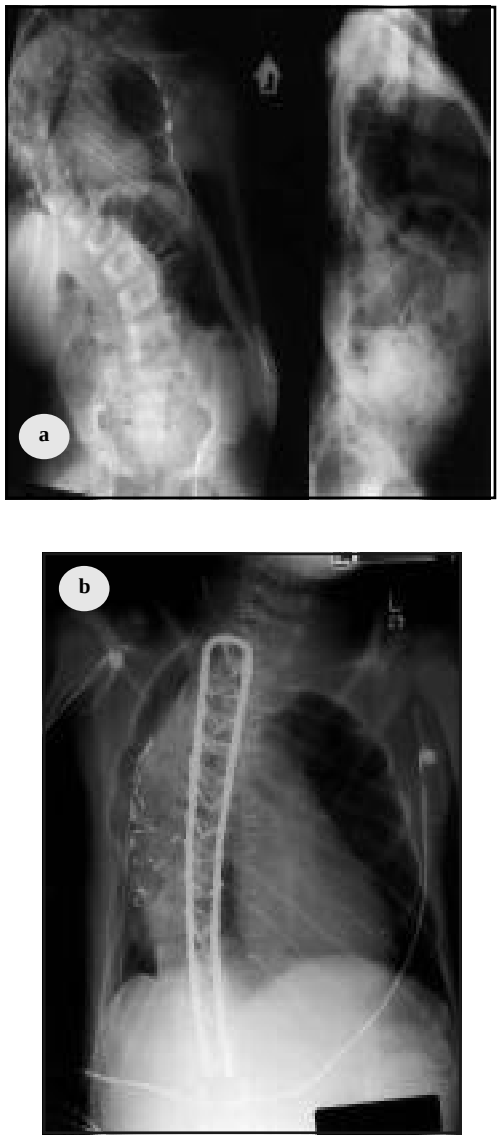

Figure 1 - X-ray showing the **a)** thoracolumbar spine (T-L) anteroposterior and lateral of patient with idiopathic scoliosis showing marked S-shaped scoliosis with a main curve in mid thoracic area, convexity to the right, with an angle of approximately 100 degrees. There is smaller L convex cervical and lumbar curve. There is lack of normal kyphosis and lordosis within T-L spines in the lateral position as seen by a straight spine. **b)** Patient's anteroposterior and lateral with idiopathic scoliosis (same patient as in Figure 1a) showing status post spinal fusion, with endotracheal tube and internal jugular line in the proper place. Marked straightening of the severe scoliosis. T - thoraco, L - lumbar

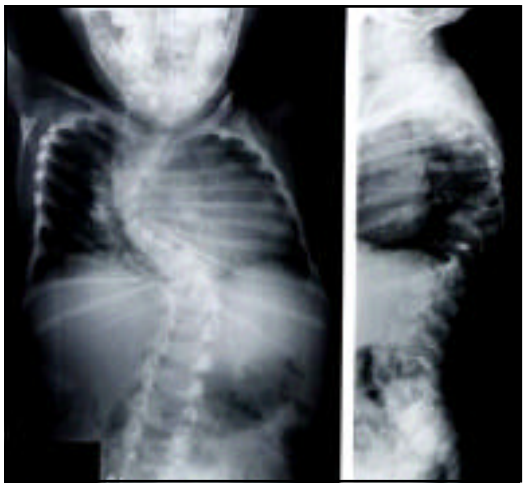

Figure 2 - X-ray of thoracolumbar spine anteroposterior and lateral of a patient with multiple congenital anomalies: showing severe kyphoscoliosis. Right convex curve in the mid thoracic region and left convex curve in the lower thoracic region. Spina bifida occulta in the lower lumbar region but normal spinal cord. In mid thoracic region, some asymmetric under segmentation at the apex of the right convex curve suggesting congenital type of scoliosis. It measures 80 degrees between T2-T10 with a left curve measures 76 degrees between T11-L3. There are rib deformities showing posterior tapering, quite slender. T - thoraco, L - lumbar

Table 1 - Diseases associated with scoliosis (N=66).

Diseases	**n**
Idiopathic	20
Secondary N=11 (17%)	
Trauma	3
Malignancy	3
Polio myelitis	3
Cerebral palsy	2
Congenital scoliosis N=35 (53%)	
Isolated congenital scoliosis	5
Klipple-Feil syndrome	1
Muscular dystrophy	9
Congenital myopathy	1
Myelomeningocele	5
Rigid spine syndrome	1
Absent right lung	1
Ehlers Danlos syndrome	1
Congenital cardiac defect	1
Growth hormone deficiency	1
Arnold chiari malformation	2
Spina bifida	2
Marfan's syndrome	1
Cerebral dysgenesis	1
Neurofibromatosis	2
Vatar association	1

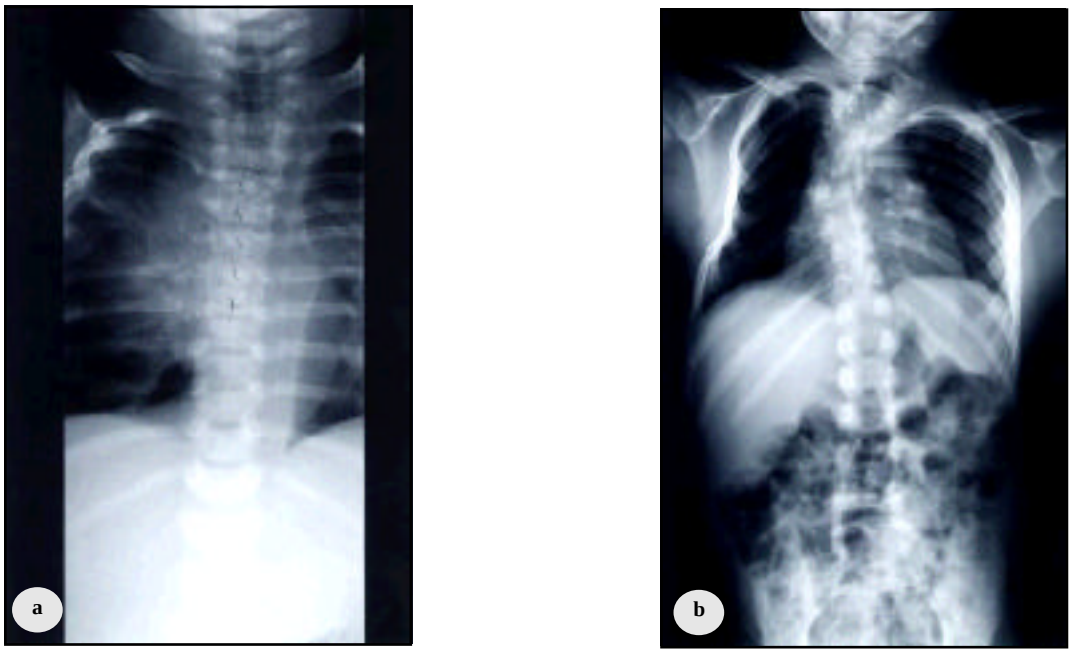

Figure 3 - Chest x-ray of **a)** anteroposterior of a patient with neuroblastoma in the posterior mediastinum displacing the carina anteriorly and eroding and splaying 3, 4, 5, 6 ribs. The right pedicles of T_3-T_6 eroded with loss of definition of the lateral margin of the vertebral bodies. Noted also diffuse calcification throughout the soft tissue mass. Widening of the para vertebral soft tissue on the left from T_3 and down to T_{11}. **b)** Thoracolumbar spine anteroposterior of patient in (Figure 3a). Post-mass resection showing severe kyphoscoliosis. The kyphosis angle measures 66 degrees and the angle of scoliosis with right convexity measures 35 degrees. T - thoraco

Table 2 - Comparisons between idiopathic scoliosis and patients with scoliosis and congenital anomalies group (N=66).

Variable	Total N=66	Idiopathic N=20	Congenital N=35	*p* value*
Male	**34**	10	16	
Female	**32**	10	19	
Age at diagnosis	**8.5 ± 44 years**	10 ± 4 years	7.4 ± 4.3 years	0.03
Alive	**57**	19	27	
Died	**9**	1	8	
Age when scoliosis appeared	**4.6 ± 3.7 years**	4 ± 4 years	4 ± 3.4 years	
Age at follow up	**14 ± 3**	14 ± 3 years	13 ± 4 years	
Progressive curvature	**62**	18	33	
Non progressive	**4**	2	2	
Period of follow up	**5.3 ± 3.7 years**	4 ± 4 years	5.8 ± 4 years	
Brace preoperative	**25**	4	17	0.03
Brace postoperative	**14**	3	8	
Type of curvature				
Cervical	**2**	1	1	
Thoracic curvature	**31**	13	12	0.03
Lumbar curvature	**12**	4	7	
Thoracolumbar curvature	**30**	6	19	
Pulmonary function test				
Restrictive pattern	**33**	10	23	0.0005
Obstructive and restrictive patterns	**4**	1	2	0.0005
Normal PFT	**4**	0	2	0.0005
Mild restrictive pattern	**1**	0	1	0.001
Moderate restrictive pattern	**14**	7	7	0.001
Severe restrictive pattern	**21**	2	16	0.001
Surgery	**34**	11	20	
No surgery	**32**	9	15	
Age at surgery	**10.8 ± 3.3 years**	11 ± 3 years	11 ± 4 years	
Posterior approach	**30**	10	17	
Combined approach	**4**	1	3	
Type of rods				
Luque rod	**7**	0	7	
Harrington rod	**22**	9	11	
Universal spinal system	**2**	1	1	
Mosley fixation rod	**4**	1	3	
Complications				
Pneumonia	**15**	4	11	0.0001
Sepsis	**1**	0	1	0.0001
Infection of the site	**2**	0	2	0.0001
Seizure	**1**	0	1	0.0001
Respiratory arrest	**1**	0	1	0.0001
Detachment of wire	**4**	1	3	0.0001
Ventilation/ home O_2	**3**	1	2	0.0001
Recurrence of scoliosis	**12**	5	7	0.0001

*p values not mentioned as they are > 0.05, PFT - pulmonary function test

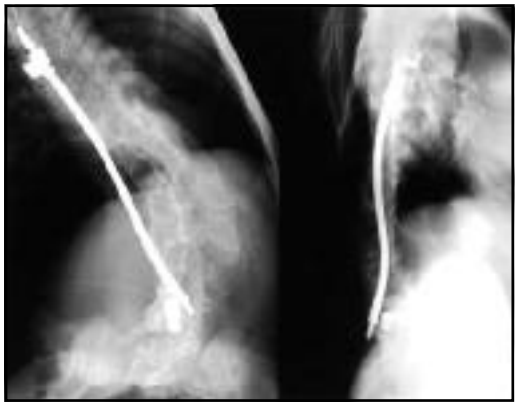

Figure 4 - X-ray of thoracolumbar spine showing a detachment of the wire in a patient with rigid spine syndrome.

Table 3 - Complication of surgery (N=34).*

Complications	n	(%)
Pneumonia or atelectasis	15	(44)
Sepsis	1	(3)
Infection of the site	2	(6)
Seizure	1	(3)
Respiratory arrest	1	(3)
Detachment of instrument or wire	4	(12)
Prolonged ventilation or home oxygen	3	(8)
Recurrence of scoliosis	12	(35)

*10 patients had 2 or 3 complications at the same time.

11 ± 4 years. Posterior approach for surgery was the main type applied in 17 (85%). Harrington rod was used in 11 (55%) (**Table 2**). There were 2 patients who had 2 types of rods inserted. The main complication of surgery was pneumonia and atelectasis in 11 patients (55%) (**Table 2**). Recurrence of scoliosis after surgery occurred in 7 (35%) (**Table 2**). Four (20%) of patients, who had surgery, developed 2 or 3 complications simultaneously at the same period of time. Recurrence of scoliosis after surgery occurred in 7 patients (35%). Comparison between PFTs, which were carried out in preoperative and postoperative period showed stabilization of all values with no apparent deterioration. Eight of the patients (23%) who did not have surgery died from the progression of their scoliosis and the severity of their congenital anomalies. The remaining 7 patients (20%) are still alive, but continued to have progressive scoliosis.

Comparison of idiopathic and congenital scoliosis. Comparisons between both groups have shown that patients with congenital anomalies and scoliosis were diagnosed earlier (p=0.03), had more thoracolumbar curvature, had more restrictive lung disease, had significant postoperative complications (p=0.0001), and higher mortality (p=0.001) compared to those with idiopathic scoliosis (**Table 2**). Comparisons between the same groups in reference to baseline blood gases showed no significant difference in all values between both types (p>0.05). Changes in PFT for patients who underwent surgery for scoliosis with Cobb's angle >40º, showed less deterioration in forced vital capacity (FVC) and forced expiratory volume in one second (FEV_1) compared to those with angle <40º, due to progressive deterioration without surgery (p<0.05) (**Table 5**).

Discussion. Scoliosis is a common disease in the pediatric age group.[1] The diagnosis of idiopathic scoliosis is missed as most of scoliosis <50º are a symptomatic, and it only becomes apparent to parents when it causes back pain or cosmetic abnormality.[1,2,6,7] Congenital scoliosis is usually discovered at birth, specially if associated with other congenital anomalies.[3,8-13] School screening of scoliosis is not practiced in KSA, therefore, most of the idiopathic scoliosis cases are discovered late and patients may have already developed a complication or progressive curvature before they are referred to a tertiary care center. Our hospital is the major tertiary care center in this country for scoliosis surgery and genetic diseases. For this reason, the bulk of the referrals are those of scoliosis in association with congenital anomalies or genetic diseases (53%) compared to the idiopathic type (30%). This is different from what is reported in the literature as 85% and 5%.[1-3,7,8] This may not reflect the real incidence in the general population in this country, since most of the mild curvatures are followed in primary care centers. Adolescent idiopathic scoliosis is not represented properly in our pediatric population (30% of the total scoliosis population compared to 85% from the literature)[1,2,7] since most of the cases were referred to an adult pulmonologist for evaluation and not to a pediatric pulmonologist as the age limit in our center is <14 years. Most of the idiopathic scoliosis cases were referred late, for approximately 4-5 years after being noticed by parents and further delayed by 2 years before proper surgical procedure was carried out. The magnitude of the curve is of a severe type at diagnosis 60-90º due to delay in referral. Most of the complications in our study are related to the lung. A similar percentage of our patients who had the brace in the preoperative period was 38%, which is similar to 36% that was cited in the literature.[2,6,7,14,16,17] Most of the curves were progressive in nature (90%) due to delay in

Table 4 - Mortality data (N=66).*

Variable	Alive N=57 (86%) n	(%)	Died N=9 (14%) n	(%)
Male	30	(45)	4	(6)
Female	27	(41)	5	(8)
Surgery	31	(46)	3	(4.5)
No surgery	26	(40)	6	(9.5)
Limb deformity†	4	(6)	1	(1.5)
Cardiac	2	(3)	0	0
Congenital anomalies/syndromes‡	21	(32)	3	(4.5)
Poliomyelitis/trauma	11	(17)	0	0
Idiopathic scoliosis	19	(29)	1	(1.5)

*a total of 9 patients died, †as in arthrogryposis multiplex congenital, ‡as vatar association, Klipple-Feil syndrome

Table 5 - Change in PFT in all patients in relation to Cobb's angle (N=41).*

Value	>40 degrees†	<40degrees†
FVC‡	-5.4	-15.7
FEV_1‡	-6.4	-18.7
FEV1/FVC	-5.8	-3
$MMEF_{25-75\%}$	-6.9	-18.7
PEF	-6.7	-13.7
FRC	-5	-8
RV	5.4	17.5
TLC	-1.5	-11
RV/TLC	-18	15
NIP	3	-15
PEP	14	20

*pulmonary function test (PFT) at follow- up baseline values of PFT. †Values are expressed in percent predicted for age, ‡p values < 0.05, FVC - forced vital capacity, FEV_1- forced expiratory volume in one second, MMEF - maximum mid expiratory flow, PEF - peak expiratory flow, FRC- functional residual capacity, RV-residual volume, TLC - total lung capacity, NIP - negative inspiratory pressure in centimeter of H_2O PEP - positive expiratory pressure in centimeter of H_2O

surgery. Beals et al[10] described 61% of 218 patients had associated anomalies. Most were part of the vatar association[3,8-11] and Klipple-Feil syndrome.[12] In our study 30 of 35 patients (85%) had associated anomalies. Only one case was clearly identified as vatar association and another case with Klipple-Feil syndrome. There were 5 cases with isolated congenital scoliosis and no associated congenital anomalies. Other rare genetic diseases were represented in our report in 18 cases of our population that involved a multi system defect as rigid spine syndrome, neurofibromatosis, cerebral dysgenesis, Marfan's syndrome, lung agenesis and growth hormone deficiency. Ten patients had scoliosis due to genetic disease such as muscular dystrophy and congenital myopathy. Genito-urinary defect described in the literature to occur in 20-40% of patients with vertebral anomalies.[13] In our report, we had one case with kidney abnormalities, but most of the patient with myelomeningocele had reflux due to neurogenic bladder. Mcmaster and Ohtsuka[8] and Winter et al[18] reviewed a combined 585 patients with congenital scoliosis until skeletal maturity. They found that 75% of patients required surgical treatment. In our report 20 (57%) patients required surgical intervention and 17 (49%) patients of both types of scoliosis used the brace preoperatively and only 8 of them used it postoperatively. This study has shown that patients with congenital anomalies in association with scoliosis have been diagnosed much earlier 7.4 ± 4.3 years compared to 10 ± 4 years for idiopathic scoliosis (p=0.03), had significant restrictive lung disease pattern in PFT (p=0.0005), had more significant complications in comparisons with those with idiopathic scoliosis (p<0.05) (**Table 2**). The most interesting aspect of the study is that; patients who had an angel of >40º and went for surgery from both types of scoliosis (congenital and idiopathic) had less deterioration in their PFT specially in relation to FVC and FEV_1 compared to those patients who had an angel of <40º but did not go for surgery (p<0.05) (**Table 5**). The reason is that those patients with an angel of <40º deteriorated very quickly and unable to go for surgery. This confirms the previous report of early surgery to prevent progressive deterioration.[6] A special precaution should be applied to this data due to the small number of patients who were able to do PFT.

This study has shown that scoliosis is a common problem in KSA. Patients need to be referred early for proper intervention before it becomes a severe magnitude, difficult to be fixed and to prevent permanent complications. Further effort is needed to identify patients at risk for respiratory complications before surgery to prevent permanent home ventilation or long-term oxygen supply. Further, studies should be carried out to include all referral of scoliosis including adult and pediatric population to reflect the proper incidence in our population. School screening should be encouraged to detect smaller curvatures and treat them properly to prevent further complications and to prevent high mortality.[6]

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