Research Journal of Biological Sciences 2 (7): 765-772, 2007
ISSN: 1815-8846

Surgical Treatment of Congenital Scoliosis

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Abstract: Congenital scoliosis is a multifactorial spine deformity with some other coexisting anomalies that can be prevented and possibly relatively corrected by means of individualized operative treatment. To assess different aspects of congenital scoliosis in order to propose optimal preventive as well as corrective operational individualized techniques to reach the optimal outcome. Of 105 consecutive scoliotic patients presenting to Tabriz Shohada Orthopedics Hospital since 2003 to 2005, 35 cases were congenital scoliosis and they form the basis of this study. All cases were thoroughly and meticulously examined and Para-clinically assessed for the spinal deformity, co-existing anomalies, operative strategies, curative out comes and the resulted complications. Fifteen male and 20 female were studied through this survey, with the age range of 2-17 years. The average follow-up was 21 months. Associated kyphosis was seen in 19 (54.28%) patients. Amongst 5 different patterns of curve presentation Hemivertebrae was found to be the commonest type of vertebral anomaly (31.42%). General abnormalities were seen in 25 (73.5%) patients. Genitourinary anomalies were seen in 3 (8.57%) patients. Cardiovascular anomalies were seen in 5 patients (14.28%) with Mitral valve prolapse being the commonest anomaly. In coronal plane the greatest angle before operation was 110° and the least one 50° with mean degree of 72.22 and the mean correction degree of 68.09% in coronal plane. We found one case of hypokyphosis; however, the associated hyperkyphosis was corrected during the operation.

Key words: Congenital, scoliosis, treatment, multifactorial spine, hypokyphosis

INTRODUCTION

Congenital Scoliosis (CS) is a lateral curvature of the spine caused by congenital anomalies of vertebral development that result in an imbalance of the longitudinal growth of the spine (Samdani and Storm, 2007; Birch and Roach, 2002; Giampietro *et al*., 2003). An incidence of approximately 0.5 to 1/1,000 births has been observed for CS (Giampietro *et al*., 2003).

The etiology of congenital scoliosis is largely unknown (Maisenbacher *et al*., 2005). An autosomal recessive form of CS observed in male and female siblings of Iranian ancestry has been described (Giampietro *et al*., 2003). Congenital scoliosis results from defects formed during the embryologic development of the spinal vertebrae (Hedequist and Emans, 2007; Tian *et al*., 2006; Fatyga *et al*., 2006; Barney and Freeman, 2003; Newton and Wenger, 2001).

CS is classified by orthopedists as a failure of segmentation (partial or completely fused vertebrae), failure of formation (such as hemivertebrae) and mixed defects. Each of these may cause development of a spinal curve based on asymmetric growth. The severity of the curve is related to the type of defect and whether or not the primary problem is accompanied by any compensatory developmental changes. The progression and ultimate prognosis are dependent upon the specific vertebral anomaly and anatomic location (Giampietro *et al*., 2003; Hedequist and Emans, 2007).

Because CS arises from significant developmental disruptions, involvement of other organ systems is common (Giampietro *et al*., 2003). Patient evaluation focuses on the history and physical examination, followed by appropriate imaging modalities (Hedequist and Emans, 2007). Once CS is identified on clinical exam, x-ray studies and evaluation by a pediatric orthopedic surgeon are indicated. Computerized tomography or magnetic resonance imaging (Suh *et al*., 2001) scans may be required for further delineation of underlying vertebral and spinal cord anomalies. An evaluation for associated cardiac and renal anomalies should be performed (Giampietro *et al*., 2003).

The hallmark of surgical treatment is early intervention before the development of large curvatures. Approximately 50-75% of patients with CS ultimately require surgical correction because of curve progression.

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The primary goal of treatment of congenital scoliosis is to prevent the development of a severe deformity. Do not wait until a severe deformity has developed and then attempt to perform a major and dangerous corrective procedure. For patients with a marked spinal growth imbalance, no treatment is perfect. The best result that can be achieved is spinal growth that is balanced on the convexity. In these circumstances, the optimum result is a short relatively straight spine rather than the severely crooked spine that would have developed without treatment (Giampietro *et al*., 2003; Hedequist and Emans, 2007).

Our aim from this survey, was assessment of different aspects of congenital scoliosis in order to propose optimal preventive as well as corrective operational individualized techniques to reach the optimal outcome, as well as evaluation of the results and consequential impediments with other concomitant studies, so as to prove the accuracy of the essay.

MATERIALS AND METHODS

A total of 105 consecutive scoliotic patients presenting to Tabriz Shohada Orthopedics Teaching Referral Hospital since 2003 to 2005, 35 cases were congenital scoliosis. We performed this prospective study on these 35 patients with congenital scoliosis.

Throughout all steps of this study taking a full detailed history and performing a full physical examination was mandatory because associated anomalies of many organs are common. Maternal perinatal history, Family history and Developmental milestones were fully explored. A detailed neurological examination was performed. The genitalia were examined for maturity, epispadias, hypospadias and the presence of undescended testicles. Extremities, Spine and Static were comprehensively examined while the patient was fully disrobed and erect. The hand was examined for clubhand, thenar hypoplasia, or other more subtle anomalies. The feet studied for clubfeet, cavus or varus deformities, vertical tali, clawing of the toes, or other signs of motor weakness. A cardiovascular evaluation was performed in all of the patients by both clinical examination and echocardiography to exclude cardiac anomalies.

Standard Posteroanterior (PA) radiography including the iliac crest distally and most of the cervical spine proximally and lateral views of the entire spine were used for the initial evaluation.

Standing films are customary, as they show the characteristics of the curve under gravity with its compensation and torso-pelvic relationships. Supine coned-down views of the anomalous region are especially

Fig. 1: Determination of vertebral maturity

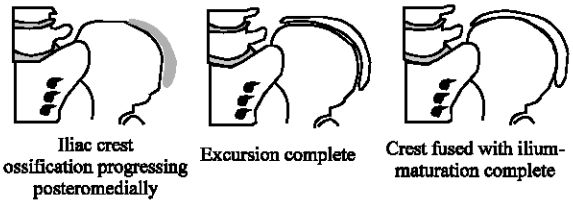

Fig. 2: Determination of skeletal maturity

helpful to visualize defects and their patterns; additionally we utilized 40-inch distance supine films in some uncertain cases, along side with 72-inch distance routine standing ones. Right and left bending films were obtained in order of bracing and surgery so as to depict the rigidity or flexibility of curves and their adjacent motion segments in this study.

The congenital curves were classified as a failure of segmentation or a failure of formation. The roentgenograms were thoroughly examined for any evidence of widening of pedicles or midline bony defects that may indicate an underlying cord anomaly. Roentgenogram of the hallmark bones was made to determine skeletal age which is considered in addition to other signs of maturation, including vertebral ring apophyses, Risser sign (iliac apophyses excursion) and physical signs of maturity such as breast development and pubic hair (Fig. 1 and 2).

All curvatures were carefully measured with the Cobb technique, including compensatory or secondary curves in seemingly normal parts of spine. Theses measurements included each end of the anomalous area and each end of the entire curve generally considered for treatment.

MRI and CT scan studies were reserved and leaved for neurological abnormalities such as diastematomyelia according to neurosurgens' indications.

The scoliotic curves magnitude must be measured from the standing AP view. We applied the most commonly used method (the Cobb method of measurement recommended by Terminology Committee of the Scoliosis Research Society). To use the Cobb method, one must first decide which vertebrae are the end-vertebrae of the curve. These end-vertebrae are the vertebrae at the upper and lower limits of the curve which

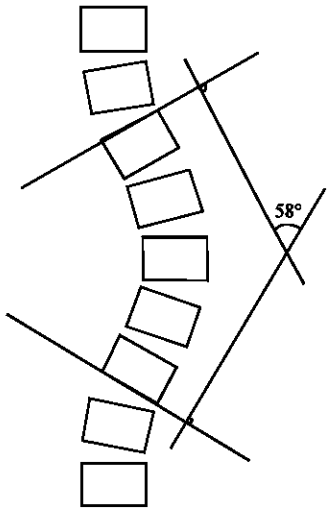

Fig. 3: Cobb method for measurement of scoliosis

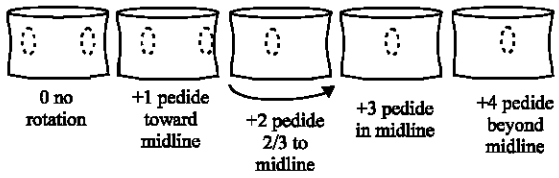

Fig. 4: Nash and Moe measurement of rotational component of scoliosis

tilt most severely toward the concavity of the curve. Once these vertebrae have been selected, one then draws a line along the upper endplate of the upper body and along the lower endplate of the lower body as shown below. The angle of interest is simply the angle between these two lines (Fig. 3) (Gupta *et al*., 2007).

To determining the vertebral rotation, we used the Nash and Moe method. In this method, if the pedicles are equidistant from the sides of the vertebral bodies, no vertebral rotation is present (0 rotation). The grades progress up to grade +4 rotation in which the pedicle is past the center of the vertebral body. After the measurement of curve angle, we estimated the degree of rotation of the vertebra at the apex of the curve by looking at the relation of the pedicles to midline according the mentioned method (Fig. 4).

Treatment methods: The method of selected surgery depends on the age of the patient, the site and type of vertebral anomaly, the size of the curvature and the presence of other congenital anomalies. The most common technique, used in 19 cases was combined anterior and posterior fusion with the following details.

Anterior approach: After pre-operation preparations and induction of anesthesia, patient was placed in the lateral decubitus position with the convex side of the curve elevated.

A curvilinear incision along the rib that is one level higher than the most proximal level to be fused was made which results in thoracotomy.

Consequently, appropriate rib was excised for future-coming grafting and fusion along side effective exposure. Thereafter diskectomy and subsequent anterior disk space fusion was made.

Posterior approach: At prone position of subject after preparing and draping a straight incision is made upon the fusion area in midline and with an electrocautery and muscle separation, diskectomy was done. Thereafter, the proper curve area was decorticated and the bone graft was applied.

- One case underwent hemivertebrae excision because of special condition of him.

All the patients were meticulously examined for the possible future-coming complications. The intra-operative 'wake-up' test to ensure voluntary lower limb movement has been supplanted by electrophysiological monitoring of the spinal cord using Somatosensory Evoked Potentials (SSEP) in some patients that needed curve correction and instrumentation. But our theaters were not equipped by SSEP so we used "wake-up" test for evaluating the unforeseen neurological complications during operation.

RESULTS

Across the years of 2003 till 2005 upon 105 consecutive patients with scoliosis that had been scheduled for operation during a 2 year period, 35 cases were congenital scoliosis and they form the basis of this study. Fifteen male (42.86%) and 20 female (57.14%) were studied through this survey. Congenital scoliosis was more frequent in girls (male: Female ratio 1:1.33).

The median age at the time of medical intervention for congenital scoliosis was 10 years (range 2 years to 17 years). The average follow-up was 21 months (6 to 21 months). Associated kyphosis was seen in 19 (54.28%) patients. There were 5 different patterns of curve presentation amongst these subjects which can be seen in Table 1.

Right-sided and left-sided curves were seen with a frequency of 4.6:1 (28 right-sided versus 7 left sided). The

Table 1: Pattern of the curves among congenital scoliosis

Type of curve	Number (%)
Thoracolumbar	22 (62.87%)
Thoracic	10 (28.57%)
Lumbar	2 (5.71%)
Cervico Thoracic	1 (2.85%)
Lumbosacral	0 (0%)
Total	35 (100%)

Table 2: Vertebral anomalies

Type of vertebral anomaly	Number of patients
Hemivertebrae	11
Unsegmented bar with hemivertebrae	9
Unilateral unsegmented bar	2
Wedge vertebrae	6
Block vertebrae	1
Mixed anomalies	6
Total	35

vertebral anomalies seen are shown in Table 2. Hemivertebrae (31.42%) was found to be the commonest type of vertebral anomaly.

General abnormalities were seen in 25 (73.5%) patients in different combinations. The incidence of various general abnormalities noted is shown in Table 3.

Of all the patients, 2 had neurological deficits at the time of observation and both had associated kyphosis. Furthermore, they had bilateral weakness of toe extensors with absent ankle jerk.

Intraspinal anomalies were seen in 5 (11.42%) patients with diastematomyelia being the commonest anomaly seen in 3 patients. In all of 3 patients with diastematomyelia, it was the thoracolumbar region. In these patients the diastematomyelia was at the site of spinal deformity. The excessive spinal spore was lucratively resected during the operation by a veteran consultant neurosurgeon in all mentioned subjects. Lumbar canal stenosis was not seen.

Genitourinary anomalies were seen in 3 (8.57%) patients. None of them had clinical features suggestive of genitourinary anomalies, but were incidental findings detected on screening; left kidney agenesis was found during the sonography along in one case. Another study revealed left kidney duplication along side with a mild left kidney hydronephrosis in the latter subject. Cardiovascular anomalies were seen in 5 patients (14.28%) (Table 4).

For the curve measurement we used Cobb method in both sagittal and coronal plates not only before operation but also post operatively to assess the outcomes along side with carrying out follow up courses.

Following corrective and preventive results were reached as a result of individualized operations:

- In coronal plane the greatest angle before operation was 110° and the least one 50° by mean of 72.22 degrees.

Table 3: General abnormalities

Abnormalities	Number of patients
Cardiovascular anomalies	5
Ligamentous laxity	0
Hypertrichosis	2
Club foot	1
Diastematomyelia	3
Spinabiphida	1
Defective dental occlusion	0
Low Lumbar hairy patches	2
Multiple congenital contractures	0
Hydromyelia	1
Sacral agenesis with deformity of right lower	0
Lumbar hyper pigmentation	2
Morquio's syndrome (Mucopolysaccharidoses-IV)	0
Klippel fiel syndrome with Sprengel's shoulder	1
Torticollis with Sprengel's shoulder	1
Amelia	0
Pectus carinatum or excavatum	0
Umbilical, inguinal hernia	2

Table 4: Cardiovascular anomalies

Cardiovascular anomaly	Number of patients
Mitral valve prolapse	2
Atrial septal defect	1
Rheumatic heart disease	0
Mitral regurgitation	2
Pulmonary regurgitation	0
Valves stenosis	0
Total	5

Mean correction degree was 68.09% in coronal plane but data from last visits of patient indicates 1° to 4° loss of correction which is not considerable. This data depicts the successful prevention of curve progression in the mentioned plate.

- Sagittal plane also evaluated for curative outcomes:

We did not find any case of hypokyphosis; however, the associated hyperkyphosis was corrected during the operation. Mean Sagittal curvature magnitude pre-operation was 16° over the normal rang which is corrected to between 20°-60° post-operationally.

- Apical vertebra rotation was measured by Nash and Moe Method and the mean correction rate was 46% of one grade which represents that correction in this parameter is not considerable.
- Achieved mean correction rate for Apical vertebra translation from plumb line was 3cm (rang 1.5 to 4cm) by percentage of 47%; this indicates a substantial correction.

Pre-operation sitting and Standing height in comparison with early postoperative same heights showed just 2cm enhancement through following period. It reveals the better results of prevention rather than correction.

- Blood loss for each patient was documented from anesthesiologist's records which were averaged 2.98 unites. For each subject it shows the sum of unites during different steps.

DISCUSSION

Scoliosis occurs relatively frequently in the general population and its frequency depends upon the magnitude of the curve being described. Scoliosis of greater than 25 degrees has been reported in about 1.5/1000 persons in the United States (Yawn *et al*., 1999; Greiner, 2002). An incidence of approximately 0.5 to 1/1,000 births has been observed for congenital scoliosis (Giampietro *et al*., 2003). There is not any comprehensive screening system regarding scoliosis, particularly congenital. Hence, the morsel of information we possess is a result of routine and random referrers to spine specialists. However, maternal health service providers especially at the first frontiers of health care, namely, initial examiners of newborn ought to pay much more attention to the spine dismorphology and related clues such as hairy patches and sacral dimpling.

An statistics review upon the 105 consequent scoliotic patients who were underwent operation across the years of 2003 till 2005 of this study revealed that 35 subjects had congenital spinal deformity (33.33%) which is just over the text based data and similar studies for mentioned just like Mohanty and Kumar (2000) studies upon patterns of presentations (Barney and Freeman, 2003; Mohanty and Kumar, 2000).

Very similar to Campbell's orthopedics text-book statistics, 15 male and 20 female were studied through this survey. Concomitant with the above facts, congenital scoliosis was more frequent in girls (male: Female ratio 1:1.33) (Barney and Freeman, 2003).

Congenital scoliosis, the most common deformity of the immature spine, is not simply an abnormal lateral curvature of the vertebral column. A rotational deformity of variable magnitude is nearly always present and distortion in the sagittal plane may occur (Mohanty and Kumar, 2000).

The spine is formed during a process called somitogenesis. This formation is a very early event and takes place between 20 and 30 days of gestation, during the first month of pregnancy. Many other organs and tissues are being made during this important time and from the same origin in development, including the heart, kidneys, brain, limbs and other organs. This fact proves the probable co-incidence of concomitant anomalies. Spinal cord anomalies are particularly widespread, occurring in up to 20% of CS cases (Giampietro *et al*., 2003). Common associated abnormalities are found in the nervous, urogenital, gastrointestinal and cardiovascular systems (Loder and Dayioglu, 1990). Additional associations include the anomalies of the ribs and chest wall as well as Sprengel deformity of the shoulder (Tsirikos and McMaster, 2005), Klippel-Feil syndrome (short neck, low posterior hairline, fusion of cervical vertebrae), Goldenhar's syndrome (associated with craniofacial anomalies, including microtia and epibulbar dermoids due to abnormal branchial arch development), incontinentia pigmenti (hyperpigmented whorls and streaks associated with eye, skin, hair, nail, teeth and central nervous system abnormalities), other recognizable syndromes, or the VACTERL association (Vertebral malformations, Anal atresia, Cardiac malformations, TracheoEsopha-geal fistula, Renal and Radial anomalies and Limb defects) (Giampietro *et al*., 2003). Genitourinary abnormalities have been reported to occur in 43% of patients with CS. A 13% incidence of renal and ureteral abnormalities in patients with CS has been reported. Renal ectopia was observed to occur in conjunction with scoliosis with a 10-fold increase in chicks with CS induced by surgical technique (Giampietro *et al*., 2003; Lonstein and Bradford, 1995; Rothman, 1999).

Given the above fact exerted the idea that comprehensive multi organ examination in conjunction with some other paraclinical studies such as sonography as well as echocardiography must be performed. We had 3 patients (8.57%) with genitourinary anomalies which had not potential hazard to the child. Amongst neurological as well as intra-spinal lesions, noted above, we just encountered Sprengel's deformity, Klippel-Feil deformity, diastematomyelia, Spinabiphida, neurofibromatosis with cafe-au-lait spots and hydromyelia with the sum of 12 subjects (34.28%), which is roughly concomitant with the reference information this data emphasis over the fact of conjunctive neuro- anomalies.

CS is classified by orthopedists as a failure of segmentation (partial or completely fused vertebrae), failure of formation (such as hemivertebrae) and mixed defects. The progression and ultimate prognosis are dependent upon the specific vertebral anomaly and anatomic location (Giampietro *et al*., 2003; Hedequist and Emans, 2007). Hemivertebrae will usually result in a curve that exceeds 40 degrees by 10 years, except in instances where two hemivertebrae occupy an adjacent and opposite orientation on the spinal column. Multiple hemivertebrae on the same side are associated with a more rapid progression and usually require treatment before 5 years. A unilateral unsegmented bar has a very high likelihood of resulting in severe progressive scoliosis. An unsegmented bar with a hemivertebrae is associated with

the most severe prognosis, with the possibility of a spinal curve exceeding 50 degrees by 3 years of age (Giampietro *et al*., 2003; Hedequist and Emans, 2007; Barney and Freeman, 2003; Lonstein and Bradford, 1995; Lonstein, 1999; Shahcheraghi and Hobbi, 1999).

In our series, 11 patients (31.42%) had at least one hemivertebrae as the base of deformity which was the most common one before unsegmented bar with hemivertebrae (25.71%).

Knowledge of the natural history of CS is essential because the natural history dictates the prognosis and treatment. The large study of McMaster and Ohtsuka of 216 patients is the best in this regard. They followed the untreated patients for 5 years and found that the rate of deterioration and the ultimate severity of the curve depended on both the type of anomaly and the site at which it occurred. They revealed that only 11% of cases were nonprogressive, whereas 14% were slightly progressive and the remaining 75% progressed significantly. Other natural history studies from Nasaca and Winter confirm theses findings (Barney and Freeman, 2003; Lonstein and Bradford, 1995).

MRI and CT scan are particularly useful in detailing bony canal anatomy and associated spinal cord dysraphism. MRI and CT scan studies were reserved and leaved for neurological abnormalities such as diastematomyelia according to neurosurgens' indications; MRI is advisable in all scoliotic children under the age of 12 years. Neuroanatomical defects, infection or neoplasia may be found. The whole spine from the base of the occiput to the sacrum should be imaged sagittally with selective occipito-cervical, cervicothoracic, thoracolumbar and lumbosacral junction axial views. CT scanning is rarely indicated. It is useful when interpretation of plain radiographs can be difficult, for example in the evaluation of severe curves or where there are multiple congenital defects (Lonstein and Bradford, 1995; Cassar and Einestein, 2002).

The primary goal of treatment of congenital scoliosis in this study was to prevent the development of a severe deformity. So we did not wait until a severe deformity has developed and then attempt to perform a major and dangerous corrective procedure.

We tried to achieve an optimum result in patients with congenital scoliosis, by over emphasizing on 3 keys:

Early diagnosis: If the diagnosis is made early, while the curvature is still small, an opportunity exists for prophylactic surgery to balance the growth of the spine.

Anticipation: The prognosis for deterioration of congenital scoliosis can be anticipated based on the amount of spinal growth remaining, the type and site of the vertebral anomaly and the degree of growth imbalance it produces.

Prevention of deterioration: All patients assessed radiologically at 4 to 6 month intervals and once progression is established, immediate treatment was necessitated to prevent further deterioration.

According to the reference data, 75% of congenital curves are progressive and only 5-10% can be treated with bracing, so surgery remains the fundamental treatment. Furthermore, surgery is the most effective treatment for severe or progressive congenital scoliosis. Many different forms of operative treatment exist for congenital scoliosis and all have their place. No single operative procedure can be applied to all types of deformities (Barney and Freeman, 2003; Newton and Wenger, 2001; Lonstein and Bradford, 1995).

In this study, the method of surgery selected depends on the age of the patient, the site and type of vertebral anomaly, the size of the curvature and the presence of other congenital anomalies. Successful surgical treatment depends on selecting the right procedure and applying it at the right time.

Given the above facts depict the individualized operation technique for each child in the base of 6 described thorough procedures. The most frequent technique was combined anterior and posterior fusion, which was carried out in 19 subjects while, posterior fusion without instrumentation upon 12 patients and the third one was posterior fusion with instrumentation in 4 children. One case underwent hemivertebrae excision because of special condition of him. After the operation all of them have been covered by Body Jacket circular cast. Throughout anterior approach we excised the proper rib for future-coming fusion supply as well as providing a better loom.

According the fact that only a few studies have been published in recent years analyzing different aspects of the congenital scoliosis (Samdani and Storm, 2007; Barney and Freeman, 2003; Newton and Wenger, 2001; Lonstein and Bradford, 1995) we compared the achieved consequences by reference values that are acceptable and concomitant.

Neurological injury is the most dreaded the incidence of serious post-operative neurological deficit, including paraplegia is around 0.5%. The intra-operative 'wake-up' test to ensure voluntary lower limb movement has largely been supplanted by electrophysiological monitoring of the spinal cord using Somatosensory Evoked Potentials (SSEP). In specialist centers, this has a low false negative rate in one multi-centre study only 0.063% of

post-perative neurological deficits went undetected (Mohanty and Kumar, 2000). But our theaters were not equipped by SSEP so we used "wake-up" test for evaluating the unforeseen neurological complications during operation. There was not any theatre in situ neurological complication.

To date studies explaining the spectrum of complication for spine surgeries, particularly scoliosis revealed impediments such as an inadvertent pneumothorax during posterior procedures, as may dural tears. A postoperative ileus after either anterior or posterior procedures and pulmonary atelectasis are both quite common. Cardio-respiratory problems are seen in those with a neuromuscular etiology and excessive bleeding can be troublesome in dystrophic patients (Mohanty and Kumar, 2000; Lonstein and Bradford, 1995).

Blood loss for each patient was documented from anesthesiologist's records which were averaged 2.98 unites. For each subject it shows the sum of unites during different steps. This disclose that theatre in situ complications such as massive bleeding did not encountered during different steps of surgeries; however, we had a seriously bleeding patient who needed 9 unites along side with crystalloid infusions for maintaining the optimum circulation.

The overall deep infection rate following scoliosis surgery is up to 5%. Symptomatic Pseudoarthrosis following surgery is 2.5%. The risk is higher in neuromuscular cases, neurofibromatosis and noninstrumented fusions. Further surgery is indicated if the pseudoarthrosis causes pain or loss of position. Finally, post-operative progression of the deformity is also occasionally seen (Barney and Freeman, 2003; Newton and Wenger, 2001; Mohanty and Kumar, 2000).

CONCLUSION

Our complication profile comprised one case of abdominal pain resulted from ileus of peritoneal manipulation, 3 cases of fever and pneumonia with alveolar consolidation and atelectasis. One case of feverless cough with tachypnea, Wound infection during first week of operation in 3 subjects and hydro or hemo-thorax in 5 patients which had not potential risk for patients.

Given the above facts shows that infections consisting pulmonary along side early wound infections were dominant in this study, whilst we encountered hemo- or hydro-thorax as a result of chest and mediastinum aggressive manipulations in 5 patients. Underlined above, tempts us to speculate that despite the protracted operation time for scoliosis surgeries, further awareness must be embarked on the sterilization dealings and aseptic procedures in all theatres.

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