Spinal Deformity Correction in Duchenne Muscular Dystrophy (DMD): Comparing the Outcome of Two Instrumentation Techniques

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Study Design: A retrospective matched cohort study.

Purpose: To compare the results of combined Luque rod–sublaminar wiring (thoracic) and pedicle (lower lumbar) instrumentation (SLW) versus those with pedicle screw fixation (PS) for scoliosis correction in Duchenne muscular dystrophy (DMD).

Overview of Literature: PS fixation is gaining popularity. Two instrumentation systems were not compared before in a matched cohort of patients.

Methods: Two groups of patients with DMD were matched according to the age at surgery, magnitude of deformity and vital capacity. Indications for surgery included loss of sitting balance, rapid decline of vital capacity and curve progression. In group 1 (22 patients) SLW fixation was used from T2/3 to pelvis or sacrum. In group 2 (18 patients) PS fixation was used from T2/3 to L5. Five patients had all level segmental PS fixations. Minimum follow-up was 2 years (range, 2 to 13 years). Radiographs, lung function tests and subjective/objective assessment were performed at standardized intervals.

Results: Mean Cobb angle in group 1 improved from 45.3° (range, 26 to 75°) to 17.7° (range, 0 to 37°) and mean pelvic obliquity improved from 14.5° (range, 8 to 28°) to 5.6° (range, 0 to 15°). Mean Cobb angle in group 2 improved from 42.8° (range, 28 to 80°) to 7.3° (range, 0 to 20°) and mean pelvic obliquity improved from 11.2° (range, 7 to 30°) to 2.0° (range, 0 to 5°) ($p < 0.05$). Mean operating time and blood loss were less in group 2 ($p < 0.05$). In group 1, the infection rate and instrumentation failure was higher, and subjective/objective outcomes showed no significant difference between the groups.

Conclusions: PS fixation had superior correction and controlled pelvic obliquity without the need for pelvic fixation.

Key Words: Muscular dystrophy, Duchenne, Spinal deformity, Scoliosis, Pedicle Instrumentation, Sub laminar wiring

Introduction

Progressive scoliosis develops in most patients with Duchenne muscular dystrophy (DMD) due to paralysis of the extensor muscles [1]. The progression can be rapid leading to serious deterioration of unsupported sitting ability and deterioration of the respiratory function already compromised by the disease [2,3]. Spinal stabilization is

required to prevent these disabilities. Spinal instrumentation in DMD has seen a sea of change over the last five decades [4-14].

The trend in the 1980s was to extend the fixation to the pelvis or sacrum. However, the prolonged operating time and increased blood loss persuaded many to stop the distal instrumentation at L5 level [10,15-17]. Most current literature deals with the instrumentation of the spine using hooks and wires or hybrid constructs with pedicle screws in lum-

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bar region. Although, the use of pedicle screw constructs in thoracic spine for idiopathic scoliosis was established in late 90s [18-20], it was not until recently, Hahn et al. [21] reported for the first time the outcome of patients following use of only pedicle instrumentation in DMD.

The present study compares the clinical and radiographic results of combined Luque rod-sublaminar wiring (thoracic) and pedicle screws (lower lumbar) (SLW) versus only pedicle screw fixation (PS) in two consecutive patient cohorts with scoliosis in DMD.

Materials and Methods

The data for two-matched consecutive cohorts were collected retrospectively at a single institution (University Hospital, Nottingham). These patients were treated within a span of 17 years from 1990-2007. Group 1 consisted of the historic cohort of 22 patients treated between 1990 and 2000 with Luque rod-sublaminar wiring with pelvic fixation with either Galveston or L-rod technique (SLW group). Group 2 consisted of the cohort of 18 patients who were treated between 2000 and 2007 with pedicle screw con-

structs (PS group). The senior authors (SHM and JKW) were involved in treating patients from both groups.

The groups were matched according to three criteria: similar age at surgery, similar magnitude of spinal curvatures (all type 1 and 2) [22], and vital capacity. Indications for surgery were loss of sitting balance, presence of scoliosis with or without evidence of curve progression and diminishing vital capacity. Both groups had similar operative exposures and upper thoracic fixation levels.

In group 1, the correction was achieved using Luque rods fixed with sublaminar wires in the thoracic and lumbar spine (Fig. 1). The Galveston technique was used in 6 cases and the L-rod configuration in 16 cases for pelvic fixation.

1. Technique

Pedicle screws are first placed in the lumbar spine. Posterior superior iliac spine is identified and the outer wall of ilium is dissected in a subperiosteal plane to reach the greater sciatic notch. This position acts as a reference for insertion of the rod on either side. The rods are contoured and prebent to realign the deformed spine. The basic princi-

Fig. 1. A 11-year-old boy, wheelchair bound for 8 months with Duchenne muscular dystrophy. (A) Preoperative antero-posterior radiograph right sided showing 60° curve. (B, C) Two year post-operative antero-posterior and lateral radiographs showing sublaminar wiring instrumentation with Luque rods and distal fixation to pelvis with L-rod configuration.

ple determining the sequence depended on where the initial correcting load needed to be applied. Once the rods are fixed to the lumbar pedicle screws (as the foundation) a cantilever corrective load (that is, one that has support at one end) to the rod or rods was applied, their unattached ends are brought to the anchorage at the opposite end. A strong foundation is defined as strong enough to accept corrective loads and to resist deforming loads without dislodgment of the anchors or plastic deformation of the rods. The thoracic foundations consisted of instrumentation spanning at least three consecutive vertebrae. The instrumentation sequence was initiated on the convex side of the thoracolumbar curve and then added on the concave side, with the cephalad rod connected side to side to the intrailiac post (Galveston) anchorage or L rod configuration. Two short closed loop sublaminar wires were used to fix the rods to the spine.

In group 2 fixation of the spine was carried out with pedicle screws in the thoracic as well as in the lumbar spine (Fig. 2) and to L5 vertebrae in all the patients. Pedicle screws with a diameter of 5mm and 6mm were used in the thoracic and the lumbar spine respectively. Anatomical landmarks were used to insert the screws. In 5/18 patients all level segmental pedicle screw construct was used and the rest had alternate segmental pedicle screw constructs in which every other vertebra in the main lumbar region and every second or third vertebra in the upper thoracic region were instrumented. The average number of pedicle screws used was 24 (range, 18 to 34) per patient. Following instru-

mentation all had autologous bone graft and supplemental femoral head allograft for fusion following standard facet excision and decortication. All patients had intra-operative neurophysiologic monitoring (SSEPs and SMEPs). Post-operatively, all the patients were managed in a pediatric intensive care unit until they no longer required any ventilatory support.

The following clinical data were recorded: forced vital capacity (FVC) and forced expiratory volume in the 1st second estimated immediately preoperatively and at final follow-up, electrocardiogram findings, echocardiogram results, weight, age at surgery, estimated blood loss, operation ("skin to skin") time, complications and the duration of hospitalization following surgery.

Antero-posterior and lateral radiographs of the whole spine in sitting were taken in all patients preoperatively and during the follow-up. The Cobb angle of the scoliosis and the pelvic obliquity were measured from the radiographs preoperatively, 6 months postoperatively and at the final follow-up. An independent assessor assessed the radiographs and measurements were made independently of the surgeons involved. Functional assessment was performed using the EK scale [23]. The EK scale comprises of ten categories (EK1-10) which was developed to determine the need for and impact of intervention in the non-ambulatory stages of DMD. The questionnaire incorporates 10 items which deal with the patients ability to use wheelchair, ability to transfer from wheelchair, ability to stand, ability to balance in the wheelchair, ability to move the arms, ability

Fig. 2. A 13 year-old-boy wheelchair bound for 2 years with Duchenne muscular dystrophy. (A, B) Preoperative antero-posterior and lateral radiographs showing left sided scoliosis with 40° curvature. (C, D) Two year post-operative antero-posterior and lateral radiographs showing instrumentation with pedicle screws.

to use the hands and arms for eating, ability to turn in bed, ability to cough, ability to speak and physical well being. A range of scores can be possible from 0-30. Lower the score better the function.

2. Statistical analysis

Pre and post-operative and final follow-up radiographs were used to analyse the Cobb angle and pelvic tilt. Post-operative corrections were analysed using paired *t*-test. Similarly correction percentage at initial post-operative radiographs and final follow-up radiographs was compared using paired *t*-test ($p < 0.05$ was considered to identify significant differences). We determined the duration of operation, mean blood loss and mean post-operative hospital stay to see if these factors were associated with better outcome with chi square test.

Results

1. Group 1

DMD was diagnosed at a mean age of 4.5 years (range,

1.5 to 9 years). The patients became wheelchair bound at a mean age of 9.6 years (range, 6 to 12.5 years) and operated at a mean age of 12.5 years (range, 9 to 16 years). At the time of surgery, the FVC was 57% (range, 34 to 72%). The scoliosis was thoracic in 2 cases, thoracolumbar in 14 cases and lumbar in 6 cases. The mean body mass index (BMI) was 22.5 kg/m². The mean follow-up in this group was 4.5 years (range, 3 to 9 years). The mean Cobb angle was 45.3° (range, 26 to 75°) and the mean pelvic obliquity was 14.5° (range, 8 to 28°) at the time of surgery. Immediately after surgery, the mean Cobb angle measured 16° (range, 0 to 35°) and at final follow-up was 17.7° (range, 0 to 37°) (Fig. 3A). The immediate postoperative pelvic obliquity measured 5.7° (range, 0 to 15°) and at final follow-up was 5.6° (range, 0 to 15°). The mean operative time was 260 minutes (range, 180 to 380 minutes), mean estimated blood loss was 3.4 l (range, 2 to 10.0 l), mean intensive care stay was 56 hours (range, 48 to 80 hours) and mean hospital stay was 8.5 days (range, 6 to 16 days). Two patients had excessive intra-operative bleeding treated with blood transfusion. Five patients had infection (four superficial and one deep). Three patients required prolonged ventilatory support but didn't require any tracheotomy. Two patients had implant failure

Fig. 3. (A) The distribution of Cobb angle at the time of surgery, immediately after surgery, and at final follow-up in individual cases in group 1 (SLW). (B) The distribution of Cobb angle at the time of surgery, immediately after surgery, and at final follow-up in individual cases in group 2 (PS). SLW: Luque rod-sublaminar wiring (thoracic) and pedicle (lower lumbar) instrumentation, PS: Pedicle instrumentation, Preop: Pre-operative, Postop: Post-operative, FU: Follow-up.

(one had loosening of the implant and the other had proximal junctional kyphosis). Pelvic obliquity was maintained at or below 10° in all but two patients.

2. Group 2

The mean age of diagnosis in this group was 5 years (range, 4 to 12 years). The patients became wheelchair bound at a mean age of 9.8 years (range, 6 to 13 years) and operated at a mean age of 11.8 years (range, 9 to 15 years). At the time of surgery, the FVC was 53% (range, 34 to 78%). Fifteen patients had C shaped curve with equal distribution of sidedness. The curves were mainly thoracic in 3, thoracolumbar in 13 and lumbar in 2 patients. The mean follow up in this group was 2.3 years (range, 2 to 6 years). The mean BMI was 25.4 kg/m². The mean Cobb angle was 42.8° (range, 28 to 80°) and the mean pelvic obliquity was 11.6° (range, 0 to 20°) at the time of surgery. Immediately after surgery, the mean Cobb angle measured 4.5° (range, 0 to 15°) and at the final follow-up was 7.3° (range, 0 to 20°) (Fig. 3B). The immediate postoperative pelvic obliquity measured 2.6° (range, 0 to 6°) and at final follow-up was 2° (range, 0 to 5°). The mean operative time was 216 minutes (range, 180 to 300 minutes), mean estimated blood loss was 2.0 l (range, 1 to 3.7 l), mean intensive care stay was 44 hours (range, 36 to 72 hours) and mean hospital stay was 7 days (range, 6 to 14 days). Two patients had infection treat-

ed with drainage, irrigation and antibiotics. One patient had broken rod and loose screw, which was revised.

Comparing the two groups it was observed that although both groups had surgery for similar curve magnitudes, the mean curve magnitude at final follow up was significantly higher in group 1 ($p < 0.01$) (Table 1). The percentage of curve correction and pelvic obliquity correction was significantly higher in the group 2 ($p < 0.01$). Intra-operative instrumentation was more time consuming in group 1 ($p < 0.01$). Intra-operative blood loss was significantly higher in group 1 ($p < 0.01$). The functional outcome as measured by EK scale was similar in both groups. The mean EK score was 8.95 (range, 2 to 23) in group 1 as compared to 8.72 (range, 3 to 19) in group 2 ($p = 0.8$).

Discussion

The scoliosis correction in DMD is challenging because of the primary muscular pathology and the frequently associated reduced pulmonary reserve. Several instrumentation techniques has evolved over the last five decades since Harrington [6] first reported the use of rods and hooks in spinal fusion for DMD. This was discarded in early 70s due to multiple complications encountered in various series [6,9,22]. Luque [8] introduced sublaminar wiring in early 80s for spinal stabilization in deformities which was undertaken in many centres with variable success. The authors

Table 1. A comparison of the key mean data from the two groups

Data	Group 1 (SLW)	Group 2 (PS)	<i>p</i> -value
No. of patients	22	18	-
Mean age at diagnosis (yr)	4.5 (1.5-9)	5 (4-12)	-
Mean age at surgery (yr)	12.5 (9-16)	11.8 (9-15)	-
Mean preoperative FVC (% pred. value)	57 (34-72)	53 (34-78)	-
Mean preoperative Cobb angle (range, °)	45.3 (26-75)	42.8 (28-80)	-
Mean preoperative Pelvic tilt angle (range, °)	14.5 (8-28)	11.2 (0-20)	-
Flexibility Index (range)	62.8 (54-70)	65.5 (57-72)	-
Mean body mass index (kg/m ²)	22.5	25.4	< 0.01 ^{a)}
Mean final Cobb angle (range, °)	17.7 (0-37)	7.3 (0-20)	< 0.01 ^{b)}
Mean final pelvic tilt angle (range, °)	5.6 (0-15)	2 (0-5)	< 0.01 ^{b)}
Mean % Cobb angle correction (range)	62 (51-70)	83 (77-89)	< 0.01 ^{b)}
Mean % pelvic tilt correction (range)	63 (45-81)	83 (73-97)	< 0.01 ^{b)}
Mean operative time in min (range)	260 (180-380)	216 (180-300)	< 0.01 ^{a)}
Mean blood loss in l (range)	3.4 (2-10)	2 (1-3.7)	< 0.01 ^{a)}
Mean intensive care stay (hr)	56 (48-80)	44 (36-72)	< 0.05 ^{a)}
Mean hospital stay in days (range)	8.5 (6-16)	7 (6-14)	-

SLW: Luque rod-sublaminar wiring (thoracic) and pedicle (lower lumbar) instrumentation, PS: Pedicle screw fixation, FVC: Forced vital capacity.

^{a)} χ^2 test, ^{b)}Paired *t*-test.

reported a mean correction of deformity on the frontal plane equal to 50%, and only the severest cases required postoperative immobilization. The advantages of surgery that they described include increased comfort and sitting tolerance [1,4,5,8,10-12]. More recently, Cervellati et al. [24] recommend the use of Luque rods and sublaminar wiring since it ensures a satisfactory, stable and durable correction of the deformity with good restoration of trunk balance.

In 1984, Allen and Ferguson [25] proposed a modified system of the segmental spinal instrumentation, known as the Galveston technique. This modification provided better fixation and correction of pelvic obliquity. However, they pointed out that pelvic fixation is a technically demanding procedure that increases operative time and risk. The fixation to pelvis was the norm for better sitting stability. Sussman [12] suggested that instrumentation and fusion to L5 might be adequate in DMD. Fusion to the fifth lumbar vertebra has been recommended [15] in cases in which pelvic obliquity (measured as the angle between a line tangential to the superior margins of both iliac crests and a line perpendicular to one through the spinous processes of L4 and L5), is less than 10°, and the Cobb angle is less than 40°. Sengupta et al. [17] recommended L5 fixation for young patients with smaller curves and minimal pelvic obliquity. Bony fusion to the pelvis is difficult to achieve because of the severe osteopenia in patients with DMD. Increased blood loss is another intra-operative complication which is dreaded by surgeons and anesthetist. In the current series i.e., SLW group had significantly increased operating time as well as blood loss which confirm the observation of others [26]. This was due to bleeding from epidural vessels during the subperiosteal dissection for passage of the sublaminar wire.

Since the introduction of segmental pedicle fixation in idiopathic scoliosis in mid 90s, many surgeons are using pedicle fixation to good use in DMD patients [19,20,26]. Hahn et al. [21] reported the first series of twenty patients treated with pedicle instrumentation from T3/T4 to the pelvis. Pelvic fixation was done with iliac screws similar to Galveston technique [14]. The mean total blood loss was 3.7 l, intensive care unit stay was 77 hours and hospital stay was 19 days. The mean Cobb angle correction was 77% from 44° to 10°, pelvic obliquity improved 65% from 14° to 3°. In our series, lower mean blood loss of 2 l was recorded, intensive care unit stay was maximum of 48 hours and mean hospital stay was 7 days in the PS group. The radiographic results in this series of pedicle instrumentation

were comparable to the series presented by Hahn et al. [21]. This series had a mean Cobb correction of 83% and pelvic obliquity correction of 83%. The pelvic obliquity correction achieved was much higher as compared to others. It is an interesting difference since the pedicle instrumentation group didn't have any pelvic fixation as described for every patient in the series presented by Hahn et al. [21]. It is presumed that the reason for such a difference may be due to the fact that their series had patients who had higher pelvic obliquity (at least five patients had more than 20° pelvic obliquity).

Alman and Kim [15] questioned the use of pelvic fixation of rods since the pelvic obliquity progressed in 78% of their patients despite pelvic fixation. But they also recommended that if the apex of the curve is caudal to L1, one may use pelvic or sacral fixation. They further suggested the use of hooks or screws in the lumbar region to avoid the failures seen in sublaminar wiring. Use of pedicle screw system for DMD was started in this institute since the turn of the millennium with an idea to avoid the complications observed in the sublaminar wiring. It was observed in our practice that pedicle screw system had better intra-operative control of the correction of the curve in sagittal and coronal plane as compared to the sublaminar wiring. Gayet et al. [27] introduced the flexible spine instrumentation with rods fixed to the lumbar and sacral spine using screws. They achieved good correction with improved spinal growth by 5 cm. Thus, the quest for better instrumentation techniques continued until the pedicle screws were found to be biomechanically superior since they are penetrating anchors as compared to the gripping fixation obtained by wires and cables [28,29].

Kim et al. [30] compared two techniques i.e., pedicle instrumentation and hybrid system of sublaminar wiring and lumbar pedicle screws in idiopathic scoliosis. They found similar results in junctional changes, lowest instrumented vertebra, surgical time and clinical outcome. Although, similar curve magnitudes were observed in both surgical groups in this series, the curve correction (both in coronal and sagittal plane) achieved was much higher in the pedicle instrumentation group [26]. These results obtained in DMD patients points out that pedicle instrumentation has a better deformity correction force and probably maintains the correction even without pelvic or sacral fixation. Good functional outcome was evident in five patients who had high BMI and had pedicle instrumentation at all segments from T2 to L5. Increased body weight owing to steroid

intake in these patients may be benefited by pedicle instrumentation at all levels since they may be at risk of implant failure. This precludes the need for revision surgery in these patients who has little longevity.

In this study, both the groups were matched for mean age at surgery, mean spinal curvature and diminishing vital capacity. The matching study cohorts make it easier to interpret the results. In one series, three methods of instrumentation techniques were compared but none of the groups were matched. The number of subjects in each group was less and authors had to interpret the results of curve correction with caution [26].

The risks involved in pedicle screw instrumentation are minimized by better understanding of the orientation of the pedicles during surgery and intra-operative image guidance. In both the groups early surgery results in less post-operative respiratory complications and therefore, reduced hospital stays. Early surgery in DMD also avoids pelvic instrumentation since pelvic obliquity was usually less than 10° . The percentage curve correction was significantly higher (83% vs. 62%) in the PS group as compared to SLW group. PS system appeared to retain the correction achieved at final follow-up.

Although the radiographic parameters show quite significant change in the PS group as compared to SLW group, the functional outcome remains similar in both groups. This is owing to the fact that both the techniques provide adequate support to the spine for a better sitting balance and may prevent further deterioration of pulmonary function. We recommend that pedicle instrumentation has better inherent capability of maintaining the corrected curve due to its biomechanical superiority.

The strengths of our study include single centre study, matching cohort of patients, availability of pre and post operative data, involvement of an independent assessor and critical analysis of the data. There were few limitations. First limitation, it's comparing a historical surgical outcome study from two different decades. Second limitation, the SLW group had pelvic fixation while the PS group had no pelvic fixation. Third limitation, since curve flexibility index was a prerequisite to compare the percentage correction, immediate post-operative correction was subtracted from the flexibility index in this series for interpreting the results.

Conclusions

Spinal instrumentation with pedicle screws for scoliosis correction in patients with DMD results in a good correction of the spinal and pelvic deformity. Pedicle instrumentation gives a solid foundation, on which the rest of the spine may be held upright, maintain the balance and rotation. Due to the improvement of lumbar curve and pelvic obliquity by pedicle instrumentation, a good sitting balance is restored even without the pelvic or sacral fixation in patients with pelvic obliquity less than 10° .

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