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ORIGINAL ARTICLE

Scoliosis in Duchenne's muscular dystrophy: a changing trend in surgical management

A historical surgical outcome study comparing sublaminar, hybrid and pedicle screw instrumentation systems

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Abstract A non-randomised retrospective study to compare the results of surgical correction of scoliosis in Duchenne's muscular dystrophy (DMD) patients using three different instrumentation systems—Sublaminar instrumentation system (Group A), a hybrid of sublaminar and pedicle screw systems (Group B) and pedicle screw system alone (Group C). Between 1993 and 2003, 43 patients with DMD underwent posterior spinal fusion and instrumentation. Group A ($n = 19$) had sublaminar instrumentation system, Group B ($n = 13$) had a hybrid construct and Group C ($n = 11$) was treated with pedicle system. The mean blood loss in Group A was 4.1 l, 3.2 l in Group B and 2.5 l in Group C. Average operating times in Group A, B and C were 300, 274 and 234 min, respectively. Mean pre-operative, post-operative and final Cobb angle in Group A was $50.05 \pm 15.46°$, $15.68 \pm 11.23°$ and $21.57 \pm 11.63°$, Group B was $17.76 \pm 8.50°$, $3.61 \pm 2.53°$ and $6.69 \pm 4.19°$ and Group C was $25.81 \pm 9.94°$, $5.45 \pm 3.88°$, $8.90 \pm 5.82°$, respectively. Flexibility index or the potential correction calculated from bending radiographs were 60 ± 6.33, 70 ± 4.65 and $67 \pm 6.79\%$ for Group A, Group B and Group C respectively. The percentage correction achieved was $72.5 \pm 14.5\%$ in Group A, $82 \pm 6\%$ in Group B and $82 \pm 8\%$ in Group C. The difference between percentage correction achieved and the flexibility index was 12.45 ± 8.22, 12.05 ± 1.3 and $15.00 \pm 1.21\%$ in Group A, B and C, respectively The percentage loss of correction in Cobb angles at final follow-up in Group A, B and C was 12.5 ± 3.5, $16.5 \pm 1.$ and $12.5 \pm 2.5\%$, respectively. Complications seen in Group A were three cases of wound infection and two cases of implant failure; Group B had a single case of implant failure and Group C had one patient with wound infection and one case with a partial screw pull out. Early surgery and smaller curve corrections appears to be the current trend in the management of scoliosis in DMD. This has been possible due to early curve detection and surgery thus having the advantage of less post-operative respiratory complications and stay in paediatric intensive care. Also, early surgery avoids development of pelvic deformity and extension of instrumentation to the pelvis thereby reducing blood loss. This trend reflects the advent of newer and safer instrumentation systems, advanced techniques in anaesthesia and cord monitoring. Sublaminar instrumentation system group had increased operating times and blood loss compared to both the hybrid and pedicle screw instrumentation systems due to increased bleeding from epidural vessels and pelvic instrumentation. Overall, the three instrumentation constructs appear to provide and maintain an optimal degree of correction at medium to long term follow up but the advantages of lesser blood loss and surgical time without the need for pelvic fixation seem to swing the verdict in favour of the pedicle screw system.

Keywords Duchenne's muscular dystrophy · Scoliosis · Sublaminar instrumentation · Hybrid instrumentation · Pedicle screw system

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Introduction

Around 75–90% of patients with Duchenne muscular dystrophy (DMD) who are non-ambulatory seem to develop scoliosis [1]. Surgical correction of the scoliotic deformity is undertaken to retard the rate of deformity progression and to improve quality of life by achieving a better sitting balance [2]. An increase in deformity is associated with an increase in pain and is associated with impairment of cardiac and/or pulmonary function. There has been much debate about the indications and timing of surgery in DMD but there is now a general concurrence that spinal deformity correction should be undertaken prior to onset of cardio-respiratory compromise [3, 4]. There have been suggestions that surgery should be undertaken if the Cobb angle is between 20 and 50° [5–7]. Early operation has been shown to improve outcome in long term follow up studies [3, 4, 8, 9].

Surgical techniques described so far in scoliosis correction in DMD patients range from halo casts with traction wires and buttons [10], Harrington rods [11], Luque's segmental spinal fixation [12] to more recent techniques using pedicle screws and hooks. The older techniques have fallen out of favour in recent years chiefly due to the biomechanical advantages offered by the newer constructs such as a stronger corrective force and increased strength thereby providing a prolonged maintenance of correction.

The senior author's (SHM) management of this condition has evolved over a period of time from the use of early sublaminar instrumentation systems to hybrid constructs and more recently pedicle screw fixation systems. An analysis of the results of these three different instrumentation systems in order to compare the radiological parameters, intraoperative factors and complications is presented.

Materials and methods

Between 1993 and 2003, 43 patients with scoliotic deformity and DMD underwent posterior spinal fusion and instrumentation. All patients were operated by the same surgeon (SHM).

The senior author's preferred method of treatment was Sublaminar instrumentation from T2 to sacrum between 1993 and 1998, Hybrid construct (T2-L5) between 1998–2001 and 2001 onwards, pedicle screw system alone (again T2-L5). Accordingly, patients were divided into three groups for comparison—Group A ($n = 19$) had sublaminar instrumentation system, Group B ($n = 13$) had a hybrid construct and Group C ($n = 11$) treated with pedicle screw system. Preoperative surgical planning included plain radiographs of the whole spine and bending films to assess flexibility of curve. Intraoperative neurological monitoring—somatosensory evoked potential and motor evoked potential was used in majority of cases.

Standard surgical technique was followed in each case. In Group A, all patients underwent sublaminar instrumentation and spinal fusion from T2-Sacrum. Two double stranded short closed loop sublaminar wires used in conjunction with the simple instrumentation designed by the senior author were used at each level (SHM). These double stranded looped wires each measuring 1 mm are available in three different lengths for different areas of the spine. The technique of wire passage and the modified instrumentation are detailed elsewhere [13]. For Group A fixation to the pelvis was performed using an L-shaped pelvic fixation jig. The jig was placed between the posterior iliac blades at the level of L5/S1 facet joints and two holes are drilled in the iliac blades to accommodate the short limbs of the L-shaped rods. All patients in Group A had pelvic fixation. In Group B, all patients had fixation from T2 to L5 using Luque rods and stainless steel sublaminar wires (again in the form of two 1 mm short double strand wire loops as mentioned above) in the thoracic spine and pedicle screws in majority of the lumbar vertebrae. The Luque rods were connected proximally using H shaped bars developed by the senior author (SHM) to provided added stability, control rotational torque and make provision for some growth [14]. In Group C, all patients had pedicle screw fixation with pedicle screws at all levels from T2-L5.The initial five cases had symmetric pedicle screws placed at alternate levels. The final six cases in Group C had symmetrical pedicle screw placement at every single level from T2 to L5. Autologous bone graft and standard technique of facet excision and decortication were followed in all cases (Fig. 1).

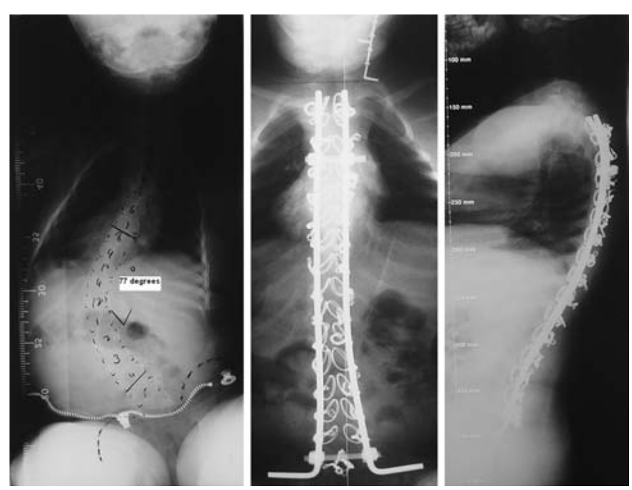

Fig. 1 Plain radiographs showing pre- and post-operative radiographs of the whole spine. Good correction was achieved following Sublaminar Instrumentation in a DMD patient with Scoliosis—Cobb angle measuring 77°. Instrumentation has been extended to the pelvis

Most authors have suggested that pelvic fixation is not necessary if the pelvic obliquity is less than 10° and if curves are smaller (<40°). It was also suggested that in these cases with smaller curves (<40°) and low pelvic obliquity angles (<10°) distal fixation to L5 is sufficient [7, 15]. However, a study conducted by Alman and Kim did not concur with the above [16]. They noticed curve and pelvic obliquity progression despite following the above criteria. They also suggested that the reason for failure maybe the use of sublaminar wiring in the lumbar spine. The same authors also opined that perhaps alternate forms of fixation in the lumbar spine may prevent curve and pelvic obliquity progression [16]. A study from our centre has shown that pedicle screws in the lumbar spine offer a strong platform sufficient to balance the rest of the spine and provide a strong corrective force to achieve and maintain both a good curve as well as pelvic obliquity correction [17]. Hence, Group B and Group C patients had distal fixation only up to L5 and sacro-pelvic fixation was not performed.

The initial seven patients operated in Group A did not have auto transfusion using cell saver technique. The remaining 12 patients in Group A and all patients in Group B and C had intra-operative auto transfusion by cell saver techniques. All patients had haemoglobin levels monitored in recovery and in the post-operative period. Transfusion was performed in keeping with the departmental guidelines. Post-operative care included close monitoring on the paediatric intensive care unit for at least 48 h. Mean follow up of patients was 5.6 years in Group A, 4 years in Group B and 4.1 years in Group C.

Case notes were reviewed and parameters such as intraoperative blood loss, surgical time and complications were recorded. Pre-operative erect and bending radiographs were studied and flexibility index of the curves were then calculated as bending films Cobb angle subtracted from erect film Cobb angle (EFCA) divided by EFCA. Pelvic obliquity angles and Cobb angle measurements on post-operative radiographs were used to estimate the degree of correction achieved in the immediate post-operative period and during subsequent follow up.

Results

All 43 consecutive DMD patients who underwent corrective surgery for scoliosis were considered in the study. Group A comprised of 19 patients treated by sublaminar instrumentation system, Group B comprised of 13 patients treated with Hybrid construct while Group C had 11 patients treated with pedicle screw system.

The average age of patients at the time of operation in Group A was 14.6 ± 1.8 years, Group B 11.4 ± 1.84 years and Group C 11.9 ± 2.69 years, respectively. The mean surgical time in Group A, Group B and Group C were 300 ± 51.96, 271.42 ± 24.78 and 234.54 ± 35.31 min, respectively. The average blood loss in Groups A, B and C were 4.53 ± 1.02, 3.52 ± 1.39 and 2.5 ± 1.04 l respectively. The results are summarized in Table 1.

Preoperative Cobb angle measured 50.05 ± 15.46° in the sublaminar instrumentation group, 17.76 ± 8.50° in the hybrid construct group and 25.81 ± 9.94° in the pedicle screw group. Flexibility index or the potential correction calculated from bending radiographs were 60 ± 6.33, 70 ± 4.65 and 67 ± 6.79% for Group A, Group B and Group C respectively. Post-operative mean Cobb angle was 15.68 ± 11.23°, 3.61 ± 2.53° and 5.45 ± 3.88° in Groups A, B and C respectively. Correction achieved in Group A was −34.36 ± 5.57°, 14.15 ± 8.10° in Group B and 20.36 ± 9.16° in Group C. At final follow-up, mean Cobb angle measured in each of the groups were—Group A: 21.57 ± 11.63°, Group B 6.69 ± 4.19° and Group C 8.90 ± 5.82°. The data is represented in Fig. 2.

A good correction was achieved in the immediate post-operative period in Group A (−34.36 ± 5.57°, $P < 0.001$), Group B (−14.15 ± 8.10°, $P = 0.001$) and Group C (−20.36 + ±9.16°, $P = 0.003$). Loss of correction at final follow up was 5.78 ± 4.21° in Group A, 3.07 ± 2.92° in Group B and 2.63 ± 1.74° in Group C. The percentage correction achieved in the immediate post-operative period was 72.5 ± 14.5% (Group A), 82 ± 6% (Group B) and 82 ± 8% (Group C). When percentage correction was subtracted from the Flexibility index, the correction achieved in Group A, Group B and Group C were

Table 1 Comparison of preoperative characteristics and surgical outcome

Mean ± SD	Group A sublaminar instrumentation $n = 19$	Group B hybrid construct $n = 13$	Group C pedicle screw $n = 11$
Age at operation	14.6 ± 1.8	11.4 ± 1.84	11.9 ± 2.69
Follow up (years)	5.6	4	4.1
Blood loss (l)	4.1 ± 1.79	3.2 ± 1.08	2.5 ± 1.04
Surgical time (min)	300 ± 40.9	284 ± 35.4	234 ± 35.3
Complications	Three infection, two implant failure	One implant failure	One infection, one partial screw pullout

The pre-operative characteristics of the entire study group and the surgical outcome parameters studied

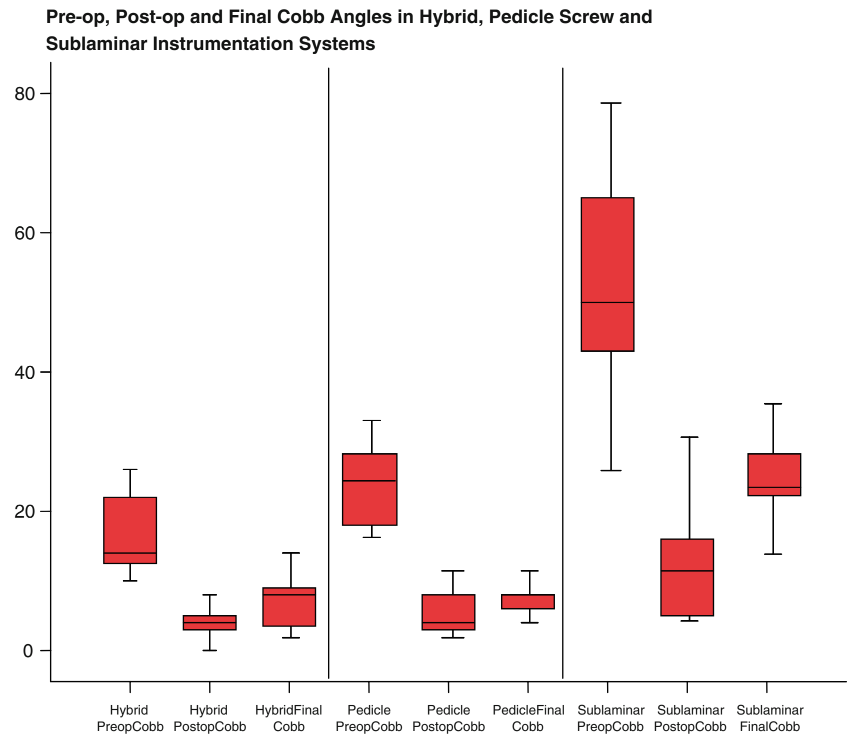

Fig. 2 A representative *box-plot* showing the mean pre-op, post-operative and final Cobb angles in each of the three groups. The *line in the box* represents the mean and the limit lines represent 95% confidence limits

Table 2 Mean pre-operative, post-operative and final follow-up Cobb angles in Hybrid, pedicle screw and sublaminar instrumentation groups

	Sublaminar	Hybrid	Pedicle screw
Pre op Cobb angle (mean ± SD)	50.05 ± 15.46	17.76 ± 8.50	25.81 ± 9.94
Flexibility index (mean ± SD) (%)	60 ± 6.33	70 ± 4.65	67 ± 6.79
Post-operative Cobb angle (mean ± SD)	15.68 ± 11.23	3.61 ± 2.53	5.45 ± 3.88
Correction achieved (mean ± SD)	−34.36 ± 5.57	−14.15 ± 8.10	−20.36 ± 9.16
% Correction achieved (%)	72.5 ± 14.5	82 ± 6	82 ± 8
Final Cobb angle (mean ± SD)	21.57 ± 11.63	6.69 ± 4.19	8.90 ± 5.82
% Correction at final follow up (%)	60 ± 11	65.5 ± 7.5	69.5 ± 10.5
Loss of correction (mean ± SD)	5.78 ± 4.21	3.07 ± 2.92	2.63 ± 1.74
% Loss of correction (mean ± SD) (%)	12.5 ± 3.5	16.5 ± 1.5	12.5 ± 2.5

The mean pre-operative, post-operative and final follow-up Cobb angles in the three different Groups A, B and C. The flexibility index, absolute correction and the percentage correction achieved. The absolute loss of correction and the percentage loss of correction are also represented here

12.45 ± 8.22, 12.05 ± 1.3 and 15.00 ± 1.21%, respectively. At final follow-up, the correction maintained was 60 ± 11% (Group A), 65.5 ± 7.5 % (Group B) and 69.5 ± 10.5% (Group C). The findings are summarised in Table 2.

Pelvic obliquity was measured on antero-posterior radiographs as the angle formed by a line joining the two highest points on the iliac crests and a perpendicular to the line passing through the tips of the L4 and L5 spinous processes. The mean pre-operative pelvic obliquity was 16 ± 12° in Group A. The pelvic obliquity in Group A patients was 8.5 ± 4° immediate post-operative and at final follow-up was found to measure 10.6 ± 10°. On the other hand, the pre-operative pelvic obliquity in the Group B and Group C was 8.2 ± 7° and 9.4 ± 8.2°, respectively. The mean post-operative pelvic obliquity in Group B was 4.4 ± 3.8° and in Group C was 5.6 ± 5.4°. At final follow-up, the pelvic obliquity in Group B and C was 5.9 ± 4.4° and 6.4 ± 4.7°, respectively. Growth as measured by migration of the H-rod was found to be a mean of 1.8 cm (range 0–2.2 cm) in Group A and 2.0 cm (range 0–2.5 cm) in Group B.

Complications noted in Group A were two implant failures and three cases of infection. Only one of the implants failed in Group B. In Group C there was one case of wound infection and one case with partial screw pull out.

Discussion

The main goal of the present study was to assess the outcome in view of the changing trends in the surgical treatment of scoliosis in DMD patients. To the best of our knowledge, there is no available literature, comparing the outcome of these three different types of instrumentation systems used in the correction of scoliosis in DMD.

Surgical treatment of scoliosis in DMD is mainly performed to restore the balance of the spinal column in both coronal and sagittal planes and to improve table top activities. Surgery has also been advocated to improve lung compliance; however this is controversial [18–20]. Surgical treatment can halt progression of deformity in patients with DMD, however, their compromised general condition, quality and requirements of life, and associated medical co-morbidities should be taken into account prior to embarking on such major surgical procedures. Instrumentation techniques have therefore evolved over the years to achieve these goals by decreasing surgical time and blood loss with minimum neurovascular complications.

Early attempts at treatment of neuromuscular scoliosis were in the form of body cast, halo casts and halo cast with buttons and traction wires. With the advent of Harrington instrumentation significant improvement in curve correction, retardation of curve progression and decrease in post-operative recumbence period was seen [10]. Initial single rod- long segment fixation techniques were superseded by segmental fixations in order achieve a better correction, avoid post-operative immobilization and reduce loss of correction over time [12].

In a critical appraisal of segmental sublaminar instrumentation in spinal deformities of varying aetiologies, Herndon et al. noticed this technique had a higher risk of spinal cord injury, implant failure and loss of correction. In order to avoid these complications; they advocated the use of supplementary anterior arthrodesis preceding posterior instrumented fusion. They also recommended the use of allografts (to improve fusion rates) and external support in the form of a post-operative body orthosis [21]. However anterior surgery in DMD is not advised due to the already severely compromised cardiorespiratory compromise. In order to reduce the risk of cord injury as a result of passing sublaminar wires in the spinal canal, spinous process wiring was proposed, however, this constituted a weaker construct.

The quest for a robust and safe instrumentation technique resulted in the advent of pedicle screws and hook systems [22]. Pedicle screws are penetrating anchors which are superior to gripping fixation obtained by laminar wires and cables. It provides a greater pull out strength and hence a biomechanically stronger deformity correction force [23, 24]. Some authors tried a combination of these two techniques and compared the results with pedicle screw systems and found similar results in terms of the junctional change, lowest instrumented vertebra, operative time and post-operative clinical outcome. However the pedicle screw system was still found to be superior in achieving a better major curve correction and lesser neurological complications [25].The risk can be minimized by better understanding of the anatomy and biomechanics involved in pedicle screw fixation [24, 26, 27]. The advantage of the hybrid system is that it can avoid the placement of pedicle screws in the upper thoracic spine, which is technically demanding and is fraught with the risk of neurological and vascular complications.

The trend in our study showed that the mean age at time of surgery in the sublaminar instrumentation group (Group A) was 14.6 ± 1.8 years. These cases were all performed in the earlier phase of the study period. On the other hand Group B and Group C patients underwent surgery at 11.4 ± 1.84 and 11.9 ± 2.69 years respectively. Also the pre-operative Cobb angle in Group A was 50.05 ± 15.46° as opposed to 17.76 ± 8.5° and 25.81 ± 9.94° in Group B and Group C respectively. This could reflect a changing trend in our practice as a result of early detection of deformity, rapid referrals and joint clinics with paediatric neurologists. Early surgery was also found to have the benefit of less post-operative respiratory complications and stay in paediatric intensive care. The other advantages of early surgery are that it avoids development of pelvic deformity and extension of instrumentation to the pelvis thereby reducing blood loss. This trend also reflects the advent of newer and safer instrumentation systems, advanced techniques in anaesthesia and cord monitoring.

It was found that the correction achieved with all the three instrumentation systems was significant (Table 3). It would appear that the percentage correction achieved with both the hybrid instrumentation system and the pedicle screw system was higher than correction achieved with sublaminar instrumentation system (Table 2). Since the pre-operative curves were higher in Group A patients, curve flexibility index is necessary in order to compare the percentage correction achieved in these three groups. Hence, a more realistic comparison is obtained when the immediate post-operative correction is subtracted from the flexibility index. This showed a comparable correction was achieved with all three types of instrumentation systems, with the pedicle screw system group showing a marginally better mean correction (Group A:Group B:Group C = 12.45 ± 8.22%:12.05 ± 1.3%:15.00 ± 1.21%, respectively). At

Table 3 Comparison of the correction achieved in each group—Wilcoxon signed rank test (immediate post-operative Cobb angle compared to pre-operative Cobb angle)

	Sublaminar system	Hybrid system	Pedicle screw system
Z	−3.824 (a)	−3.180 (a)	−2.936 (a)
P	<0.001	0.001	0.003

The results of Wilcoxon signed rank sum test used to compare the immediate post-operative Cobb angle to pre-op Cobb angle in each of the three groups. Note that a significant correction was achieved in all the three groups

final follow up, pedicle screw system also appeared to retain correction in a marginally better fashion than the hybrid group and the sublaminar group (percentage correction at final follow-up Group A:Group B:Group C = 69.5 ± 10.5%:65.5 ± 7.5%:60% ± 11%). When the loss of correction between immediate post-operative and final follow up was compared in these three groups, the sublaminar system appeared to have lost nearly the same degree of correction as the pedicle screw system (12.5 ± 3.5 versus 12.5 ± 2.5%) and both these systems were marginally superior to the hybrid system (16.5 ± 1.5%) (Figs. 3, 4).

The results from the study need to be carefully interpreted. Percentage curve correction is a useful traditional technique to compare groups with varying curve magnitudes. However, in our study, the curves in Groups B (17.76 ± 8.50) and Group C (25.81 ± 9.94), were of a small magnitude. Percent correction when curves are so small can be misleading and there is a risk of over-interpreting the results. In addition to this, the standard deviations overlay significantly and there does not appear to be any real difference between any of the systems given the small size of the curves to begin with in the correction. Group A, which was chronologically performed earlier and patients from this group had curves with larger magnitudes compared to Groups B and C. By considering flexibility index, the potential corrections possible in the different groups to the actual correction achieved with different instrumentation systems can be compared in a fairly reliable fashion.

Patients with neuromuscular scoliosis have a higher incidence of intraoperative blood loss (up to seven times greater risk) during scoliosis surgery than other patients. One of the possible reasons for this is that they require more extensive surgery with fixation of multiple vertebral body segments [28]. It has also been postulated that the lack of dystrophin associated with DMD can impair vasoconstrictive properties leading to increased blood loss [29]. We noted that intra operative blood loss and operating time was maximal with the Sublaminar system (4.1 l) chiefly due to bleeding from epidural vessels (at the time of laminotomy and wire passage) and from pelvic instrumentation. A previous study from our centre showed that

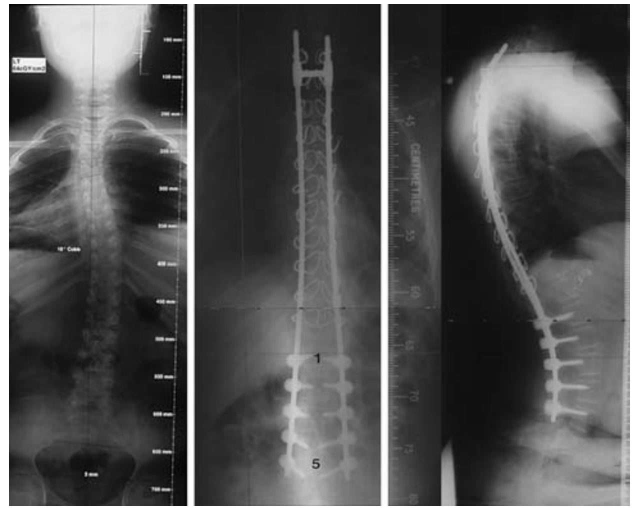

Fig. 3 Plain radiographs showing pre-operative and post-operative radiographs of a patient with DMD and scoliosis corrected using the Hybrid instrumentation system. Notice that pedicle screws have been used in the lumbar spine and sublaminar wiring in the thoracic spine

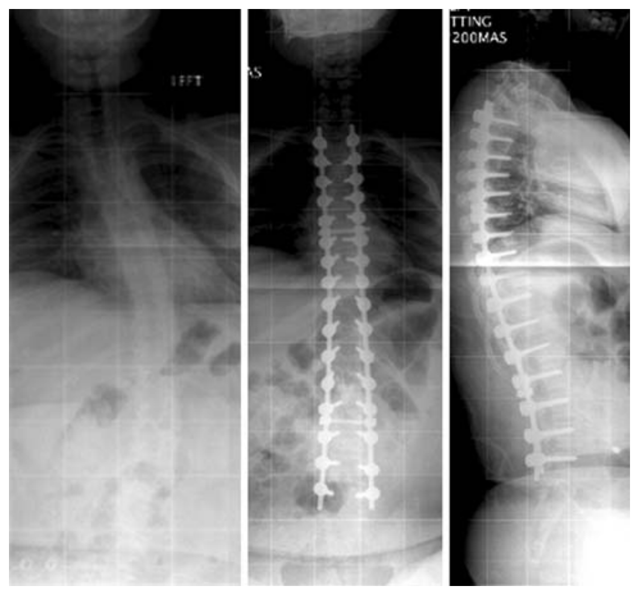

Fig. 4 Plain radiographs showing pre-operative and post-operative radiographs in a DMD patient with scoliosis. Pedicle screw instrumentation has been used to achieve a good correction in this case. Notice the placement of pedicle screws at all levels and that instrumentation has been restricted to L5

lumbar fixation to L5 was adequate if the surgery was performed in patients at a young age with smaller curves and minimal pelvic obliquity [17].

Conclusion

Early surgery and smaller curve corrections appear to be the changing trend in the management of scoliosis in DMD

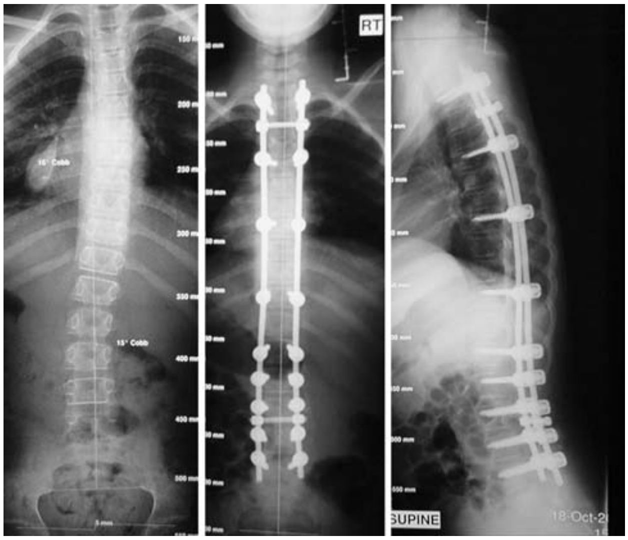

Fig. 5 Plain radiographs showing another example of a DMD patient with scoliosis treated with pedicle screw instrumentation

(Fig. 5). This has been possible due to early curve detection and surgery thus having the advantage of less post-operative respiratory complications and stay in paediatric intensive care. Also, early surgery avoids development of pelvic deformity and extension of instrumentation to the pelvis thereby reducing blood loss. This trend reflects the advent of newer and safer instrumentation systems, advanced techniques in anaesthesia and cord monitoring. All three instrumentation constructs appear to provide a good immediate post-operative correction. Sublaminar instrumentation system group had increased operating times and blood loss compared to both the hybrid and pedicle screw instrumentation systems. With both the hybrid and pedicle screw instrumentation systems medium to long term results comparable to the sublaminar group were achieved even without pelvic fixation. The pedicle screw system had the technical difficulty of screw placement in the upper thoracic spine which was obviated by the hybrid system. Overall, the three instrumentation constructs appear to provide and maintain an optimal degree of correction at medium to long term follow up but the advantages of lowest blood loss and least surgical time without the need for pelvic fixation seem to swing the verdict in favour of the pedicle screw system.

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