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ORIGINAL ARTICLE

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Late-onset spinal deformities in children treated by laminectomy and radiation therapy for malignant tumours

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Abstract This is a retrospective study of 76 children who had had malignant tumours treated with laminectomy or laminoplasty and/or radiation therapy affecting the spine. Spinal tumours in children are extremely rare. However, their treatment can result in progressive spinal deformity. Radiation therapy affecting the growing spine can lead to asymmetric vertebral growth, causing kyphosis and/or scoliosis. These spinal deformities pose one of the most challenging problems for the spine surgeon. The aim of this article is to describe late-onset post-laminectomy/post-radiation spinal deformities and to evaluate the results of their treatment. Seventy-six children, with a mean age of 4 years and 7 months (range, 2 months to 16 years), underwent surgical removal of malignant tumours, between 1961 and 1995. Sixty-seven of them developed post-laminectomy/post-radiation spinal deformity. Conservative treatment consisted of bracing and corrective plaster casts. In 46 cases the deformity was treated surgically. A distraction plaster cast was used as preoperative preparation in the more severe and rigid curves, with or without neurological impairment. Surgery consisted of combined anterior and posterior fusion in 39 cases and posterior fusion in seven cases. Posterior instrumentation was used in 38 cases. The mean follow-up period was 6 years and 7 months (range, 9 months to 20 years and 2 months). Nine children did not develop deformity following the primary tumour treatment. One of them underwent laminectomy with posterolateral fusion and eight had laminoplasty combined with external immobilisation. Forty-six children developed iatrogenic kyphosis and underwent surgical correction from a mean of 75° pre-correction to a mean of 32°. The mean scoliotic angle correction was 66° preoperatively to 34° postoperatively. At follow-up, the mean correction loss was 7° in the sagittal plane and 5° in the coronal plane. Preoperative distraction plaster cast treatment resulted in a correction of 39% in kyphosis and of 58% in scoliosis, and in a partial or complete recovery of neurological deficits in all but one patient. In severe and rigid curves that develop following treatment of paediatric spinal tumours, preoperative application of a distraction plaster cast can reduce deformity and facilitate surgical correction. Furthermore, in the case of pure bony compression of the spinal cord due to the apical vertebra of the deformity, treatment with the distraction plaster can result in recovery from the neurological impairment. The prevention of post-laminectomy/post-radiation spine deformities is emphasised. Rigid external immobilisation for a period of 4 months in the cervical spine and of 6 months in the thoracic spine is recommended after both laminoplasty and laminectomy with posterolateral fusion.

Keywords Laminectomy · Laminoplasty · Radiation therapy · Spinal deformity · Spine fusion · Distraction plaster

Introduction

Spinal deformity following laminectomy or radiation therapy in children has been well described by a number of authors. Advances in paediatric oncology have resulted in an increasing survival rate and in some cases, complete cure, although at times with very considerable sequel and the emergence of secondary pathology. Tachdjian reported spinal deformities in 26% of infants and children who had undergone laminectomy [21]. Others have reported incidences of 95–100% [1, 8, 9, 18]. The surgical treatment of spinal tumours is often supplemented with radiotherapy. The first case report of scoliosis following radiation therapy was published by Arkin et al. in 1950 [2]. Two years later, Neuhauser described the late effects of radiotherapy on growing vertebrae [12]. The growth potential of the irradiated vertebral growth cartilage decreases; the vertebral endplates become irregular, and the vertebral height decreases. If a vertebra is affected asymmetrically by the radiation, anterior or lateral wedging of the vertebral body develops, causing kyphosis and/or scoliosis. The fibrosis and contracture of the irradiated tissue may accelerate the progression of the deformity [5]. More severe deformities are to be expected if a spine previously treated with laminectomy is irradiated [10, 13].

Preventing iatrogenic deformity in paediatric spinal tumours may be achieved by protecting the spine from radiation, ensuring symmetric radiation of the vertebrae in order to diminish the asymmetric growth disturbance [5]; performing posterolateral fusion in the same stage as laminectomy [1, 3, 8, 14, 15]; or performing laminoplasty [16, 18] if the posterior elements are not involved by the tumour.

If a post-laminectomy/post-radiotherapy deformity develops, it can result in a severe and disabling deformity that sometimes threatens spinal cord function. The relation between neurological deficits and spinal deformity is complex. If a deficit appears or deteriorates, this can stem either from the recurrence of a spinal tumour, or from the anterior bony compression of the cord at the level of the apex of the kyphosis. In addition, a paralytic deformity may develop at a distance from the initial neurological lesion [6]. This can result in further deterioration of spinal stability, which is already affected by the deformity at the level of the laminectomy.

Post-laminectomy and post-radiation spinal deformities pose one of the most challenging problems to the spine surgeon. The aim of this paper is to present our strategy and the results of the surgical and non-surgical management of iatrogenic spinal deformities in children.

Materials and methods

This is a retrospective study based on the analysis of the charts of the 76 patients who had been treated with laminectomy/laminoplasty, with or without adjuvant radiation therapy for spinal or extraspinal malignant tumours between 1961 and 1995. Ten children underwent laminoplasty. Two other patients had laminectomy followed by posterior fusion without instrumentation. Sixty-seven of 76 children developed deformities following laminectomy (or laminoplasty with improper immobilisation) and/or radiation therapy (Table 1). The initial pathology was intraspinal tumour (such as a spinal cord tumour) in 30 cases, juxtaspinal tumour (such as a neuroblastoma) in seven cases, extraspinal

Table 1 Patients developing spinal deformities after various types of primary tumour treatment. Since the patients were (small) children, the spine was not instrumented when the posterior fusions were carried out

Type of primary treatment	No. of patients	No. of secondary deformities
Laminectomy only	21	21
Irradiation only	21	21
Laminectomy and irradiation	22	22
Laminectomy + non-instrumented posterolateral fusion + external immobilisation	1	0
Laminectomy + non-instrumented posterolateral fusion without external immobilisation	1	1
Laminoplasty + external immobilisation	8	0
Laminoplasty without external immobilisation	2	2
Total	76	67

tumour (e.g., a tumour farther from the spine, such as a Wilms tumour) in 12 cases and a combined intradural and extradural (dumbbell) tumour in 18 cases. The mean age at laminectomy/radiotherapy was 4 years, 7 months (range, 2 months to 16 years). The male:female ratio was 31:36. The laminectomy was cervical in seven cases, cervicothoracic in nine, thoracic in 20, thoracolumbar in nine and lumbar in one case. On average, six laminae were resected. In seven cases, at least one total facet resection was performed. Forty-four patients initially presented with neurological symptoms. All but three of these were ameliorated after laminectomy and tumour resection. The mean radiation dose after laminectomy was 4,500 rads. Patients who had had extraspinal tumours received an average dose of 3,500 rads.

The post-laminectomy/post-radiotherapy spinal deformity was diagnosed on average 2 years, 9 months after the initial treatment (range, 1 week to 12 years, 5 months). Pure kyphosis was detected in 36 cases, cervical spondylolisthesis with focal kyphosis in two cases, swan-neck deformity with upper thoracic hyperkyphosis in four cases, kyphosis with mild scoliosis in 15 cases, and angular kyphosis with progressive rotational dislocation in four cases. A further two cases were in the stage of pre-rotational dislocation (e.g., S-shaped, combined scoliosis with increased intervertebral rotation in the junctional zone). Scoliosis with hyperrotatory paradoxic kyphosis [4] was found in four cases.

In 63 of the above 67 cases, conservative therapy was started when the child was first seen in our institution. Four patients were operated on immediately after a period of preoperative traction and/or Stagnara's plaster-cast distraction [20] (Fig. 1). Of the 63 conservatively treated patients, 42 were operated on later, after an average period of 6 years and 7 months of conservative treatment.

In summary, 46 patients were treated surgically for iatrogenic spinal deformities (Table 2). The mean age at the time of the surgery was 13 years, 7 months. The sex ratio was 25 males to 21 females. In general, preoperative traction was applied for 1–3 weeks. In stiff and angular deformities, a Stagnara's plaster cast with halo traction was applied and progressive distraction was performed (seven cases, Table 3). Combined anterior and posterior fusion was carried out in 39 cases. Anterior tibial strut grafting was applied in 35 cases. These grafts were applied in a palisade fashion and always in the concavity if there was an associated scoliosis. Anterior instrumentation was not used in any patients. Posterior-only fusion was performed in seven cases. Posterior instrumentation was used in 38 cases (CD instrumentation in 34 cases, Harrington instrumentation in three cases and Harrington rods with Wisconsin hooks in one case). All of the non-instrumented spines were immobilised with a rigid external support (a plaster cast, or a halo cast) for an average period of 6 months, but this external immobilisation was also applied in 18 instrumented cases where the quality of the metal–bone junction was considered to be poor.

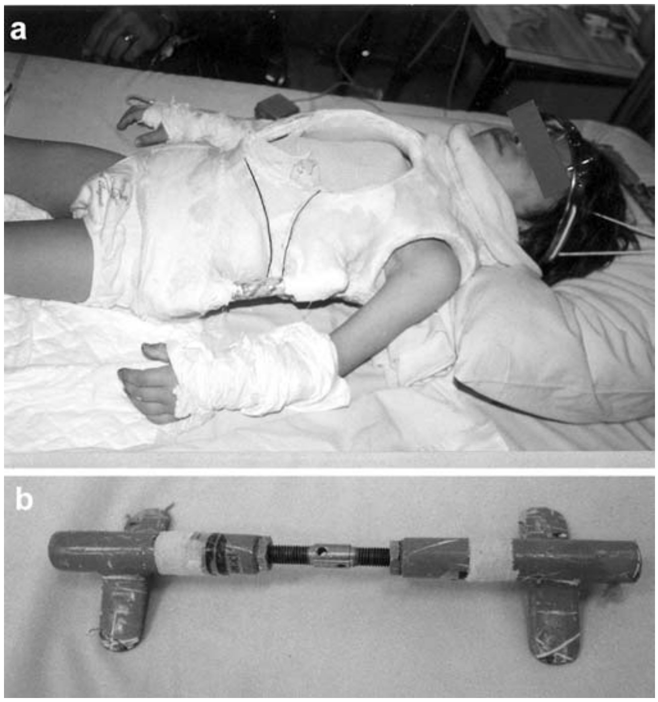

Fig. 1

The mean follow-up period was 6 years, 7 months (range, 9 months to 20 years, 2 months) after the surgical correction of the spinal deformity. Physical examination of the patients and standard anteroposterior (AP) and lateral radiographs were taken at the last follow-up examination. The curve magnitudes were measured using the Cobb method and the average values are given with standard deviation (SD). Statistical analyses were performed with Student's *t*-test. Statistical significance was set at the level $P < 0.05$.

Results

Ten children underwent laminoplasty. Two of them did not have the usual postoperative external immobilisation for 4 months on the cervical spine or 6 months on the thoracic spine, and both developed deformities. Two patients had laminectomy with immediate posterior fusion without instrumentation. One of these was left without external immobilisation and developed deformity.

In general, the spinal deformities were detected within 5 months following cervical laminectomy, 15 months following thoracic laminectomy and 77 months after radiation therapy. Twenty-one patients had had only

Table 2 Distribution of conservative and operative management for iatrogenic spinal deformities

	No. of patients
Patients with post-laminectomy and/ or post-irradiation deformity	67
Conservative treatment only	21
Initial treatment: non-surgical	63
Immediate surgery	4
Surgery after bracing	42
Total of surgeries	46

conservative treatment. Six of them had died and four of them are still treated with bracing. Forty-six patients underwent surgical correction for post-laminectomy/ post-radiation spinal deformities (Table 4). Seven patients presenting with severe deformities with or without neurological lesions were preoperatively treated with Stagnara's distraction plaster and halo traction. The magnitude of kyphosis decreased from 111° (SD, 31.9°) to 68° (SD, 34.0°) ($P=0.00$) by the end of the distraction. Three of seven patients had no neurological problems before, and the distraction casting did not result in any neurological deficit. One patient with deep tendon reflex alteration and mild muscle clonus recovered completely after the distraction. Another patient with altered deep tendon reflex response and positive bilateral Babinski sign remained unchanged after casting. One patient with severe paraparesis and hypoesthesia and pyramidal signs recovered almost completely, and only a mild muscle weakness was detectable after distraction. In one patient with spastic paraplegia and presence of dumbbell tumour, some diminution of muscle strength of the lower limbs was observed by the end of the distraction (Table 3).

The mean preoperative kyphosis Cobb angle was 76° (SD, 22.9°), which was reduced to 33° (SD, 20.1°) ($P=0.00$) after the surgery. Whether the kyphosis was sharp (five or fewer vertebrae involved) or long (nine or more vertebrae involved) did not significantly change the final correction rate. The sharp Cobb angle kyphotic deformities were corrected from an average 77° down to 33°, and the long Cobb angle kyphotic curves were corrected from 76° down to 37°. When the kyphosis was accompanied by scoliosis, the preoperative extent of the scoliosis was 45° (SD, 19.8°), which decreased to 25° (SD, 20.2°) ($P=0.00$). The mean correction loss was 7° in the sagittal plane and 3° in the coronal plane after a mean follow-up period of 6 years and 7 months. In five cases, the scoliosis presented the main indication for surgery. In these cases the coronal curve decreased from 66° (SD, 13.6°) to 34° (SD, 5.6°) ($P=0.00$). The average loss of correction was 5°. Six of 21 conservatively treated patients died and a further six patients were lost for the follow-up. In five patients the conservative treatment has

Table 3 Data on patients prepared with distraction plaster (DTR: deep tendon reflex)

Patients	Scoliosis (°)			Kyphosis (°)			Neurological status		
	Pre-traction	Post-traction	Post-surgery	Pre-traction	Post-traction	Post-surgery	Pre-traction	Post-traction	Post-surgery
1	-	-	-	82	35	38	Normal	Normal	Normal
2	121	88	80	144	118	108	Normal	Normal	Normal
3	93	67	37	140	80	44	Spastic paraplegia	Decreased muscular strength in the lower limbs	Idem
4	57	18	4	102	42	26	Paraparesis, hypo-aesthesia, DTR alterations, positive Babinski sign	Almost complete recovery, minimal muscle weakness	Complete recovery
5	60	20	13	128	92	51	Normal	Normal	Normal
6	38	15	13	67	40	18	DTR alterations, muscle clonus	Complete recovery	Complete recovery
7	92	41	33	26	8	7	DTR alterations, bilateral positive Babinski signs	Idem	Complete recovery
Average	77	32	20	111	68	48	–	–	–

Table 4 Kyphosis angles in iatrogenic spinal deformities

	($n=46$)	Laminectomy only ($n=11$)	Laminectomy and radiotherapy ($n=17$)	Radiotherapy only ($n=18$)
Preop	76° (SD ± 22.9)	68° SD ± 23.9)	79° (SD ± 32.9)	70° (SD ± 12.5)
Postop	33° (SD ± 20.1)	38° (SD ± 19.1)	30° (SD ± 21.6)	28° (SD ± 23.0)
Follow-up	40° (SD ± 24.3)	46° (SD ± 22.7)	35° (SD ± 23.2)	35° (SD ± 27.7)

already finished. Their cervicothoracic/thoracic kyphosis improved by means of 7° (40° before treatment and 33° at follow-up).

Complications

Neither neurological nor acute septic complications were observed after the correction and fusion. Complete removal of the instrumentation was necessary in five cases: a prominent implant causing pain in one case and late deep infection in four cases (9%). All four of these children belonged to the irradiated group. The infection developed 4.4 years postoperatively (range, 2.5–6.2 years). Junctional kyphosis under the fusion was detected in six cases. It measured 9° on average, and involved one segment in five cases and two segments in one case.

Discussion

Spinal deformities following radiation therapy and laminectomy are more likely to develop in children than in adults [1, 9, 10, 11, 13, 14, 15, 19]. This is due to the greater proportion of cartilage, the higher viscoelasticity of the immature tissues and the growth potential of the spine [15]. Removal of the laminae increases compressive forces on the anterior aspect of the vertebral bodies and increased instability is observed after removal of both facets at one level [8]. The biomechanical studies by Robert et al. [17]. revealed a considerably increased pressure in the nucleus pulposus of the involved discs after laminectomy in the infantile spine, whereas in adults the analogous increase is much less important.

While post-laminectomy deformities in children appear relatively early (in our series, 5 months after cervical/cervicothoracic laminectomy and 15 months after thoracic laminectomy), deformities after radiation therapy may only manifest many years later (77 months after radiation therapy in this series), with a frequency of 10–100% [15]. Patients must be followed up until the end of adolescence, because post-radiation deformities can develop and/or deteriorate in the period of rapid growth during puberty.

Immobilisation following laminectomy does not prevent deformity [3, 13]. However, we consider that bracing may retard the progression of the deformity. Therefore, we recommend that all children treated with laminectomy be provided with a proper external support from the first postoperative day. Simultaneous laminectomy and posterolateral fusion (without internal fixation) are recommended by some authors [1, 3, 8, 14, 15], but it is also mandatory to have a postoperative external immobilisation of the spine in a correct posture [6, 8]. Laminoplasty may also be a solution if the tumour does not involve the posterior vertebral elements, but a necessary condition for success is the adequate fixation of the replaced block of laminae with intraosseous sutures and immobilisation of the spine with a cast or halo cast in the postoperative period. In our series, two of the ten patients who had undergone laminoplasty did not participate in postoperative immobilisation. Both patients returned with kyphotic deformities, and during the posterior correction fusion we observed the displacement and non-union of the block of laminae in both cases.

In most cases the patient usually requires surgical treatment. Preoperative traction for 1–3 weeks is applied in every case. The indications for a Stagnara distraction plaster [20] are severe and rigid deformities with or without neurological impairment. In our series, we utilised the distraction plaster in six cases with angular kyphosis and in one case with kyphoscoliosis. The goal of this treatment is multiple. The distraction plaster can diminish the deformity considerably, increase the lung capacity and allow a better correction on the operating table. Less force may be required to maintain the correction obtained with the distraction plaster, through the viscoelastic property of the spine. After preparation with a distraction plaster, the forces acting on the anterior strut grafts and on the posterior instrumentation can be lower, as can the risk of complications. If neurological signs are present, due to the compression of the spinal cord by the bony structures at the apex of the deformity, progressive distraction may resolve both the compression and the neural signs, particularly if these lesions have appeared only recently. When the cases are more chronic or secondary to tumour recurrence, no neurological improvement is to be expected. If there is long-standing purely mechanical bony compression of the spinal cord, the chances of recovery are low, but some improvement is not excluded. Better results can be expected when the neurological problem is of short duration and caused by the posterior parts of the vertebral bodies at the apex of the kyphosis. If a tumoral origin of the neural symptoms is excluded, but the traction or the distraction cast does not solve the neurologic symptoms, anterior decompression of the spinal cord (corpectomy) is indicated. In our series, seven patients were prepared with a distraction cast, which ensured an average correction of 39% of the kyphosis and complete recovery in two of the three patients with neural impairments.

With the exception of moderate (less than 70°), non-angular and flexible kyphotic deformities, combined anterior and posterior fusions were carried out with posterior internal fixation. The mechanical advantages of anterior strut grafting are well-known [3, 10]. There is, however, a concern for graft dislodgement, which can be prevented with proper surgical technique. Each graft is shaped to be pointed, so that it can be impacted into the adjacent vertebral bodies. This is facilitated with the manoeuvre done by the assistant, who pushes the back of the patent at the apex of the kyphosis. The angle of the kyphosis opens, and the grafts are placed and impacted. As the assistant stops pushing the patient's back, the deformity tends to aggravate and the grafts are impacted more strongly. The closure of the periosteum around the strut graft is usually impossible, but the suture of the parietal pleura is feasible and also helps prevent graft dislodgement. In our series, we did not observe any complication with the tibial strut graft (neither tibial fracture nor strut fracture).

In most cases, posterior fixation was done in the second stage. Usually, it is not necessary to do it first, because the post-laminectomy/post-radiation spinal deformities are the consequence of a chronic instability of the spine, as opposed to an acute instability that would require a posterior fixation first. Furthermore, after the anterior release and strut grafting, more correction can be achieved. Posterior instrumentation serves more for fixation than correction. This is because of the poor bony quality of the affected part of the spine that is seen frequently in these tumour patients. If one applies extra force to correct the deformity from the posterior, one risks pullout of the hooks and breakage of the laminae. But if the anterior release precedes the posterior instrumentation and fusion, it is enough just to fix the achieved position. Utilising the increased visco-elastic feature of the immature spine, continuous halo traction for 1 week between the two stages can reduce the force required to maintain the achieved correction.

If accompanying scoliosis is present at the level of the kyphosis, the anterior approach is always made from the concavity of the scoliosis. In hyperrotatory paradoxic kyphosis [4, 7], the approach for anterior release is on the convex side of the scoliosis. There is no strut grafting, but anterior fusion with the use of either bone chips or rib-inlay grafting is performed [6].

Posterior fusion was carried out in only four cases and was satisfactory in improvement and stabilisation of the kyphosis. (In two of four cases cervical posterior fusion and external immobilisation was applied. In the rest, thoracic hyperkyphosis was corrected with posterior fusion and internal fixation.) In cases involving non-angular, less severe and flexible thoracic hyperkyphosis (< 70°), posterior-only fusion with internal fixation proves to be sufficient [13, 15].

No differences were found between the correction rates in short and long kyphotic deformities. The length of the kyphosis does not seem to be a predictive factor for the outcome of the surgery. This could be rather the Cobb angle and the rigidity of the deformity.

The complication rate in our series was higher than usual in spinal deformities. In 9% of the cases the implants had to be removed because of septic conditions. All these four patients had received radiation after the tumour resection. Septic complication was not observed in the laminectomy-only group. The compromised soft tissue due to radiation therapy may be a hotbed for infection.

Although many case reports are available on iatrogenic spinal deformities, they are mostly confined to the cervical spine. In the recent study by Otsuka [13], nine of the 12 patients with thoracic or lumbar post-laminectomy or post-radiation kyphosis were treated with combined anterior and posterior fusion with posterior instrumentation, and three patients with posterior-only fusion with instrumentation. The average preoperative kyphosis was corrected from 84° to 35°, with an average loss of correction of 4° at 5 years, 6 months after surgery, which is similar to our present series of 46 patients.

Conclusions

After laminectomy and/or radiation therapy, children should be closely observed, because it is highly possible that they will develop spinal deformities. Postoperative immobilisation does not prevent a post-laminectomy spinal deformity, but it can be conducive to lesser deformities with fewer difficulties in the final treatment.

Orthotic treatment is indicated as soon as a deformity develops. If the deformity progresses, a corrective plaster cast may be effective in slowing the progression. The goal of non-surgical treatment is to slow the progression and to defer the spinal fusion until optimal development and growth of the spine and lungs have been achieved.

In severe deformities, and particularly those involving a purely bony compression of the spinal cord, a distraction plaster cast of Stagnara is recommended. This can result in improvement of the neurological symptoms, especially if the neurological compromise has appeared recently. Furthermore, a distraction cast combined with respiratory management can appreciably improve the vital capacity and respiratory function of the patient.

With the exception of moderate, regular and flexible thoracic hyperkyphosis, anterior and posterior fusion with posterior instrumentation is the recommended procedure. Anterior strut grafting is recommended in all kyphotic cases except hyperrotatory paradoxic kyphosis.

If no internal fixation is applied, postoperative external immobilisation (a plaster cast or a halo plaster) for 6–12 months is mandatory. Postoperative external immobilisation is also recommended, even with posterior instrumentation, if the instrumentation is not felt to be optimal, with severe and rigid curves and dysplastic bone, or after radiation therapy at an early age.

The prevention of post-laminectomy/post-radiation spinal deformities is of primary importance. Symmetrical radiation prevents vertebral wedging. Isolated laminectomy should be avoided. If possible, laminoplasty is the procedure of choice to explore the spinal canal in the treatment of tumours. After laminoplasty or laminectomy combined with posterolateral fusion, rigid external immobilisation for a period of 4 months in the cervical spine and of 6 months in the thoracic spine is mandatory.

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