Surgical Correction of Postradiation Spinal Deformity

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Background: Radiation to the juxtaspinal area in children with malignant tumors induces the appearance of a postradiation spinal deformity (PRSD) with drastic progression during the growth spurt, so surgical correction is mandatory. Methods of surgical treatment depend on the age of the patient, and the type and size of the spinal deformity.

Methods: A long-term retrospective survey of 6 patients receiving surgical management of kyphoscoliosis was conducted. The original tumors were 3 Wilms' tumors, 2 neuroblastomas, and 1 lymphoma. The mean length of time for tumor excision and subsequent radiation was 2.2 years. The total radiation dose averaged 3566 rad. The mean age at initial presentation was 6.1 years and that at spinal correction was 11.8 years. Single posterior surgery was performed in 3 cases, while the other 3 required anteroposterior correction due to severe deformity and scar contracture.

Results: The curves of PRSD were concave toward the side irradiated, and the kyphotic component was more severely involved than was the scoliotic component. Four patients had favorable correction without curve progression. However, in the other 2 younger patients, due to thinness of their back, rigid angulation of the spine, poor bony stock, and medical comorbidity, spinal instrumentation was rather difficult. Postoperative pseudarthrosis and subsequent rod protrusion occurred with progressive kyphosis.

Conclusions: PRSDs consist of uncommon developmental vertebral anomalies, of which curved patterns occur in any combination, but most severely involve the presence of collapsing kyphosis and soft tissue contracture. Surgical correction may be less effective, especially if the children are skinny and have low bone stocks.

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Key words: **postradiation spinal deformity, kyphoscoliosis, Wilms' tumor, neuroblastoma.**

Skeletal sequelae associated with radiation therapy for childhood tumors include scoliosis, kyphosis, hypoplasia of the ilium, rib deformity, leg length discrepancy, osteocartilaginous exostoses, and secondary sarcomas.[1,2] Irradiation has a direct inhibitory effect on the growing epiphyses. Longitudinal growth of the vertebrae by enchondral ossification occurs at the cartilaginous endplates, so irradiation may produce a spinal deformity.[3-6] In 1950, Arkin et al.[7] reported the first clinical case of scoliosis in an

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8-year-old child irradiated for Wilms' tumor. Neuhauser et al.[8] described the roentgenographic findings in this spinal deformity, including transverse growth arrest lines of increased density as "os within os", irregular scalloping of the endplates with loss of vertebral height, stunted axial growth with small-sized vertebrae, and a gross contoured "Morquio" appearance. Soft tissue scarring, skin hyperpigmentation, and underdevelopment of the paraspinal muscles were also noted.[4-6] Growth disturbances in postradiation spinal deformities (PRSDs) are directly related to the radiation dosage and inversely related to the age at irradiation.[4-6,9] As children approach the adolescent growth acceleration, the spinal curvature may show drastic changes due to the rigid structures and primary defects in the vertebral growth potential.[1,5,6,10]

Due to such unacceptable progression of spinal deformities in some of these children, early surgical correction is mandatory to balance the growth of the spine. However, no one operative procedure is appropriate for all types and sizes of deformities. This report is a long-term retrospective study of 6 children with severe spinal deformities who received local radiation in the vicinity of the spine for childhood tumors. They were all followed-up until skeletal maturation. The particular characteristics of the spinal deformities, the effectiveness of different types of spinal surgical corrections and their long-term results are reviewed.

METHODS

From January 1985 to July 2000, data on 6 consecutive patients with PRSD receiving surgical correction for kyphoscoliosis were collected to review their medical records and spinal radiographs; observations continued for a mean of 10.8 years (range, 5 - 13.8 years) until they had reached skeletal maturity. There were 3 boys and 3 girls. The original tumors included Wilms' tumors in 3 cases, neuroblastomas in 2 cases, and a malignant lymphoma of the spine in 1 case. Locations of the primary tumors were the retroperitoneum in 4 cases, and the mediastinum and thoracic spine in 1 case each. They all had previous histories of multimodal treatments for malignant childhood tumors at the authors' institutions. These comprised excision of the primary tumor with subsequent chemotherapy and local radiation to the vicinity of the thoracolumbar spine.[11,12] One patient with a malignant lymphoma of the thoracic spine had open biopsy only, and the other 5 patients received en bloc excision of the tumor mass. The average age at primary tumor excision was 2.2 years (range, 8 months - 4.8 years). 6 MV supervoltage radiation with the portal crossing the midline was given 1 month after surgery in all cases. The average irradiation dose was 3566 rad, ranging from 2400 and 3000 rad in 1 case each, to 4000 rad in the other 4 cases. The average age at initial presentation of PRSD was 6.1 years (range, 1.5 - 8 years). The trunk became crooked

Table 1. Profile of Patients with Postradiation Spinal Deformities

Case	Gender	Original tumor			Radiation dose	Age at presentation	Brace	Age at spinal surgery
		Age at diagnosis	Location	Stage/Surgery				
1	Female	1 yr 4 mon	R	Wilms' tumor stage II, right nephrectomy	3000	6 yr 3 mon	yes	14 yr
2	Female	1 yr 6 mon	R	Wilms' tumor stage II, left nephrectomy	2400	8 yr	yes	16 yr 5 mon
3	Male	8 mon	R	Wilms' tumor stage III, right nephrectomy	4000	1 yr 5 mon	yes	8 yr 2 mon
4	Female	3 yr 5 mon	M	Neuroblastoma stage IV, right thoracotomy	4000	5 yr 2 mon	yes	6 yr 4 mon
5	Male	4 yr 10 mon	R	Neuroblastoma stage III, left laparotomy	4000	8 yr	yes	14 yr
6*	Male	8 mon	T	Malignant lymphoma, laminectomy	4000	8 yr	yes	16 yr 5 mon

Abbreviations: R: retroperitoneum; M: mediastinum; T: thoracic spine.
* Irradiation-induced myelitis.

with resulting soft tissue fibrosis and underdevelopment of the vertebral column in these growing children. All patients were advised to wear a brace for a certain period of time before definite surgical correction of the spinal deformities. However, the progression of curvature could not be controlled by bracing in all of them. Surgical intervention for the correction of spinal deformities was indicated, because the curvature was rigid and rapidly progressive either in the coronal or the sagittal plane or both during the adolescent growth spurt. The age at spinal surgery averaged 11.8 (range, 5.3 to 16.4) years (Table 1).

The curvature patterns varied, with 1 being pure scoliosis, 1 kyphosis only, and the other 4 kyphoscoliosis. The severity of the kyphosis was measured in all patients using the modified Cobb method[13] from the most sagittally tilted vertebra at either end of the deformity as seen on lateral spinal radiographs taken with the patient in an erect posture immediately before surgery, after surgery, at 1 year, and at the

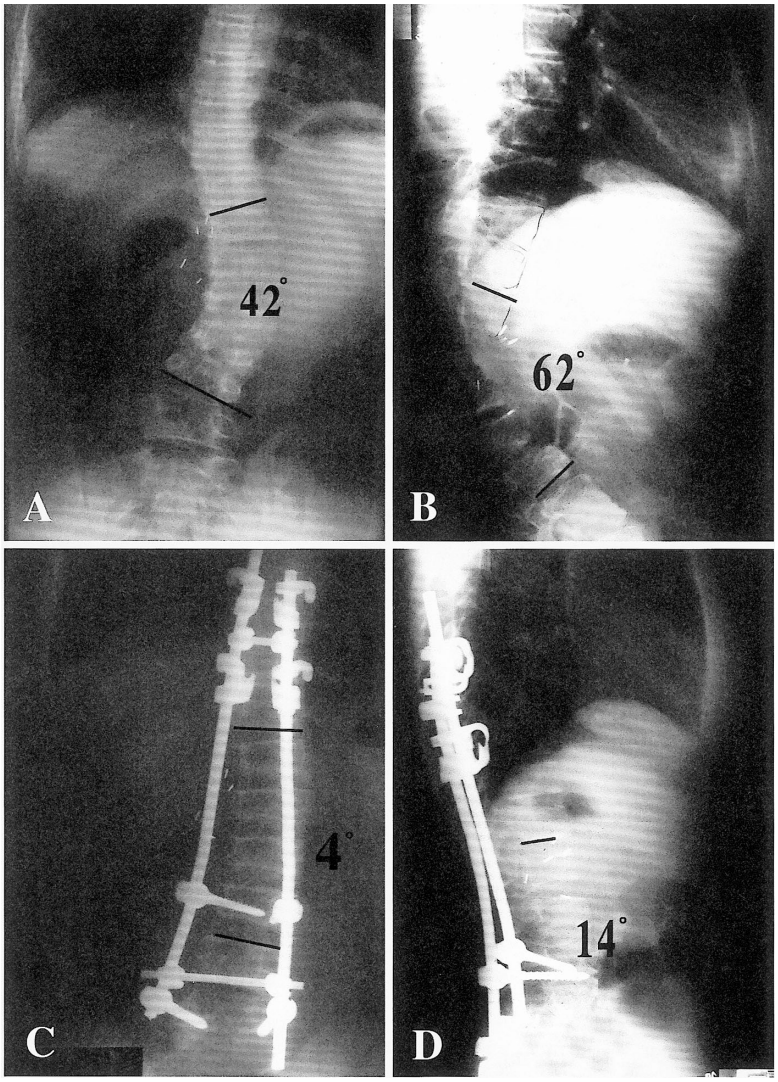

Fig. 1 Two adolescent girls (Cases 1 and 2) sustained a PRSD after treatment for a stage II Wilms' tumor. Posterior spinal fusion and instrumentation were performed. (A) Preoperatively, the scoliotic angle from the T12 to L3 levels was 42°, and (B) the kyphotic angle was 62°. (C) Postoperatively, the scoliotic angle was 4°, and (D) the kyphotic angle was 14°. Seven years later, normal alignment in the coronal and sagittal planes had been maintained.

final follow-up. The scoliosis was also measured using the Cobb method[13] on anteroposterior spinal radiographs. Care was taken to measure all radiographs from exactly the same anatomic points with reference to the preoperative radiographs. Skeletal maturity was determined radiographically when there was complete ossification and fusion of the iliac apophyses. All patients showed curvature which was concave toward the side irradiated. Kyphosis was always the major component of the PRSD, and the apex of deformity occurred mostly in the area of the thoracolumbar junction. Surgical procedures were individualized, according to the age at presentation of the spinal deformity, the location of the primary tumor, the size of the compromised vertebral column, the soft tissue condition of the trunk, and the magnitude of the spinal deformity. None of the patients had an occult intraspinal anomaly or spinal cord compression at the apex of the kyphoscoliosis. The surgery was considered a failure if at final follow-up, the kyphosis was 10° or greater than it was immediately after surgery, or if there were any neurologic deficits.

Posterior Cotrel-Dubousset instrumentation and spinal fusion were used in 2 adolescent cases (cases 1 and 2) (Fig. 1). Combined anterior and posterior spinal release, instrumentation, and fusion were performed in 3 cases (cases 3, 5, and 6) due to severe rigidity and large curvature of the kyphoscoliosis. One patient (case 4) was inaccessible for anterior release surgery due to severe respiratory distress after the initial tumor surgery and a right pneumonectomy. A posterior Luque rod with segmental wiring fixation was applied in 2 of the 3 patients after anterior spinal release, with (case 5) and without (case 6) a Zielke anterior implant. Pediatric Cotrel-Dubousset implants were used for posterior surgical correction in 2 juvenile cases (cases 3 and 4). Implant-related problems were complicated due to the thinness of the back, the rigidity of the spine, and the poor bony stock. These factors affected the outcomes of the surgical correction. The bone graft chips used for spinal arthrodesis were taken from the iliac bone of patients. Before 1988, however, large allogenic bank bone and bone substitutes were not available at the authors' institutions.

RESULTS

The scoliotic angles according to the Cobb method before surgery were 38.4° (range, 6°-53°). This was corrected to 15.4° after surgery, but had increased to 27.4° (range, 3°-60°) at the final follow-up. The kyphotic angles before surgery measured 59.2° (range, 40°-70°). This was corrected to 38° after surgery but had increased to 56.8° (range, 14°-88°) at the final follow-up. The curvature patterns of spinal deformation were related to the portion of the vertebrae irradiated, with concavity toward the side irradiated and kyphosis being more severe than scoliosis. Usually, the initial changes of the vertebral anatomy occurred 9 months to 4 years after irradia-

Table 2. Surgical Results in Patients with Postradiation Spinal Deformities

Case	Levels of deformity		Cobb angles preop	postop	final	Surgical approach (implant)	Percentage of correction loss	Complication	Follow-up
1	T12-L3	S	42°	4°	4°	P (CDI)	S: -	screw breakage	7 yr 6 mon
		K	62°	14°	14°		K: -		
2	T11-L3	S	53°	22°	25°	P (CDI)	S: 13%		5 yr
3	T9-L3	S	40°	20°	20°	A+P (PCDI)	S: -	*implant impingement;	11 yr 11 mon
		K	70°	55°	88°		K: 60%	pseudarthrosis	
4	T4-L2	S	51°	30°	60°	P (HRRPCDI)	S: 100%	*implant impingement;	11 yr 8 mon
		K	54°	40°	86°	K: 115%	pseudarthrosis		
5	T10-L3	S	6°	1°	3°	A+P (VDS+LR)	S:		13 yr 8 mon
		K	40°	30°	40°		K: 33%		
6	T2-T8	K	70°	45°	50°	A+P (LR)	K: 10%	wire protrusion	13 yr 10 mon

Abbreviations: S: scoliosis; K: kyphosis; A: anterior release and fusion; P: posterior fusion; CDI: Cotrel-Dubousset instrumentation; PCDI: pediatric Cotrel-Dubousset instrumentation; HR: Harrington rod; VDS: ventral derotation system; LR: Luque rod.
* Failed correction and curve progression.

tion. Brace treatment was less effective for progressive kyphosis than for scoliosis. Spinal fusion with instrumentation was undertaken at different ages in these patients, if significant curve progression was documented. Single posterior surgery was performed in 3 patients, while the other 3 needed combined anterior release and posterior surgical correction. There were no neurological complications nor deep wound infection noted after surgery (Table 2). However, implant impingement beneath the skin of the apical prominence with superficial wound infection was noted in 3 cases (cases 3, 4, and 6) 1 year postoperatively, which was attributed to hypoplasia of the paraspinal muscles and atrophy of the soft tissues. Subsequent loss of correction with progressive kyphosis was noted in 2 (cases 3 and 4) of them, due to hook dislodgment and rod impingement during the follow-up period (Fig. 2). The protruding wire was removed in the other patient (case 6) with no sequelae. The condition of his spastic paraplegia after

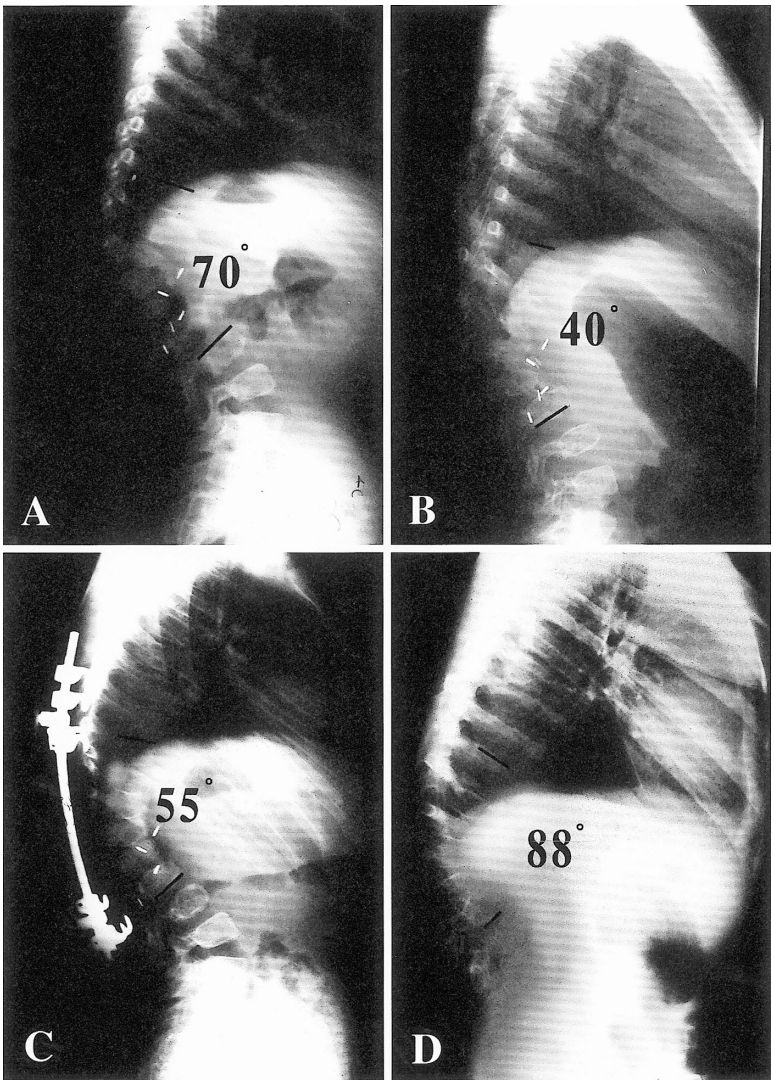

Fig. 2 An 8-year-old boy sustained a PRSD after treatment for a stage III Wilms' tumor. Combined anteroposterior spinal surgery was performed. (A) Preoperatively, the thoracolumbar kyphosis was 70°. (B) Kyphosis was 40° after anterior release and a rib graft. (C) After posterior pediatric instrumentation 1 year later, kyphosis was 55° with a low hook pullout. (D) Twelve years postoperatively, a collapsing kyphosis was 88°.

local irradiation remained the same.

In the 2 juvenile patients (cases 3 and 4) with spinal fixation using pediatric Cotrel-Dubousset implants, the postoperative courses were rather complicated. The implants had to be removed because of implant exposure and superimposed wound infection. Pseudarthrosis was noted at their thoracolumbar junctions. Attempted repair of the nonunion site and immobilization by underarm plaster jacket failed due to an insufficient amount of autogenous iliac bone chips. In addition, a lack of anterior vertebral columnar support and imbalanced spinal growth

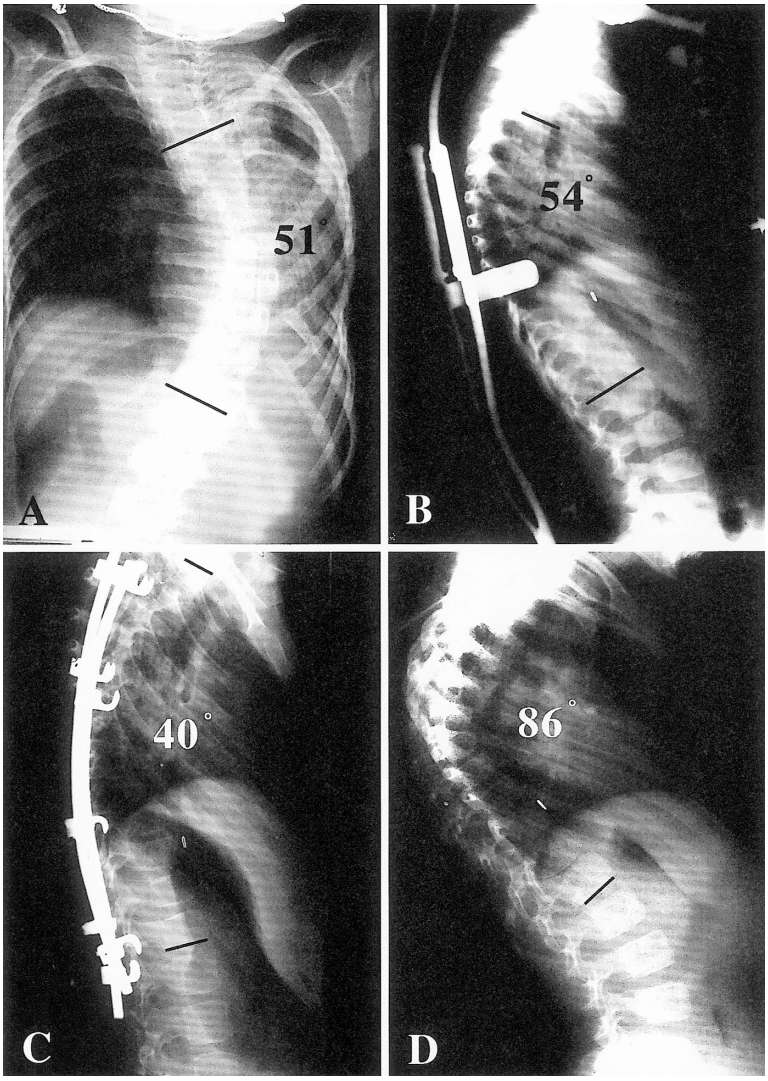

Fig. 3 A 6-year-old girl sustained a PRSD after treatment for a stage IV mediastinal neuroblastoma with spinal invasion. (A) Scoliosis progressed to 51°, and (B) kyphosis to 54° even with a brace support. Subcutaneous Harrington rodding failed, and the previous right pneumonectomy inducing respiratory distress made anterior surgery impossible. (C) Posterior pediatric instrumentation attained a kyphotic angle of 40°. Rod impingement, pseudarthrosis, and superimposed wound infection were found 1 year postoperatively. The implant was removed, and cast immobilization was continued, but kyphosis progressed to 55° with eventual bony union. (D) During the adolescent growth spurt at 12 years of follow-up, angular kyphosis had progressed to 86° with no neurological sequelae.

eventually induced collapsing kyphosis (Fig. 3). Medical comorbidity, such as frequent urinary tract infections (case 3), or poor pulmonary function with a forced vital capacity of 23% of normal control and occasional asthmatic attacks (case 4), prohibited further reconstruction of the spine. Progression of the kyphosis at the thoracolumbar junction was noted in 2 cases, one from 55° to 88° (case 3), and the other from 40° to 86° (case 4) at final follow-up. The remaining 4 cases maintained normal alignment in both the coronal and sagittal planes without further progression. There were no pseudarthroses nor implant problems among them.

DISCUSSION

Unlike idiopathic scoliosis, PRSD is caused by initial tumor surgery and irradiation, and is characterized by considerable structural alteration in the vertebral column and the paraspinal soft tissues. Due to the radiosensitivity of the growing epiphyseal cartilage, local radiation leads to varying degrees of disorganization in the proliferative zone and vascular growth buds.[3,9] These changes frequently impair the longitudinal growth anteriorly or anterolaterally to the transverse axis of vertebral rotation in the sagittal plane, and eventually develop into a rigid spinal curvature together with a prominent kyphotic component.[14] The dose-effect relationship is particularly steep between a cumulative dose of 2000 and 3000 rad, manifesting as irregular scalloping of the vertebral endplates with diminished axial height and small vertebral bodies.[4,8,15] A dose of 5000 rad is the critical level for bone necrosis and myelopathy due to vascular compromise of the spinal cord.[10] Axial malalignment of the spine only occurs many years later, when the remaining normal healthy portion of the vertebrae grows and aggravates the deformed structures. The severity of the spinal deformity is also influenced by the young age at irradiation especially before 2 years old, and by the later bowstring effect of soft tissue contracture after the initial tumor surgery, as well as rib and iliac hypoplasia.[3,4,9] All patients in our series were under 5 years of age at the time of radiation therapy, and 4 of them were even less than 2 years old. The objective of surgery is to prevent further PRSD progression and achieve correction by balancing the growth of the spine. The different approaches to spine-instrumented fusion depend upon the age of the patient, the size of the vertebral anomaly, the condition of the paraspinal soft tissues, and the rigidity and magnitude of the spinal curvature.

Wilms' tumors represent the third most common malignant tumor of the abdomen in children, exceeded in frequency by neuroblastomas and lymphomas in Taiwan.[16] Excision of this embryonic renal tumor usually leaves the vertebral column intact, which allows assessment of the effects of irradiation alone on the juxtaspinal structures.[4,5,10,14] Riseborough et al.[5] reported on 81 surviving patients with Wilms' tumor, of whom 59 had spinal deformities. Seven patients underwent spinal fusion with a 30% correction rate, but 3 of them had pseudarthrosis, and 2 had infections. In our 3 cases with Wilms' tumor (cases 1-3), 2 patients had kyphoscoliosis, and 1 had pure scoliosis of the spine. One patient (case 3) was noted to have progression of the kyphotic deformity even after both anterior and posterior fusion and instrumentation. The pediatric implant had to be removed, because a collapsing kyphosis induced hardware impingement on the skin, wound infection, and pseudarthrosis. We attempted to repair the pseudarthrosis by adding 2 further vertebral levels above and below the previous fusion site, but further reconstruction was halted due to the poor general condition of this young child, and this resulted in only a fair outcome. The other 2 children had good correction results, probably because they were older at the time of spinal surgery and possessed sufficient quality of bone stocks for spinal reconstruction.

Neuroblastomas are of neural crest origin, arising from anywhere along the distribution of the sympathetic chain and adrenal gland. More than half of these tumors occur in children younger than 2 years old, and many are diagnosed after the occurrence of metastasis.[6,11] In contrast to Wilms' tumors, neuroblastomas may invade the spinal column and compromise neurological function. Radical tumor resection and irradiation inevitably produce spinal instability and lead to severe kyphoscoliosis.[6,10,11,14,17,18] The most common location of kyphotic deformities is at the thoracolumbar junction with a long radius of curvature extending into the lumbar spine, or a short local kyphos with the apex at the T12-L1 levels.[4,6,14] Lonstein[17] advocated prompt anterior column stabilization to obtain a solid fusion mass to balance over the pelvis. Of our 2 cases with neuroblastomas, 1

(case 4) had severe, progressive kyphoscoliosis of the spine at a very young age. Anterior stabilization surgery could not be applied in the following years, because of repeated asthmatic attacks after initial pneumonectomy. Therefore, posterior instrumentation even with repeated bone grafting ultimately could not maintain the corrected results. Later, hardware impingement occurred with a collapsing kyphosis. The PRSD in the other patient (case 5), who had a favorable general condition, did not progress after anteroposterior fusion and instrumentation, and thus this patient had a successful outcome.

The total amount of bone grafting is an important factor contributing to the success of solid fusion. We suggest that posterior fusion should extend not only to the most sagittally tilted vertebra at either end of the kyphosis, but also to at least 1 additional vertebra above and below.[18] Pseudarthrosis is difficult to detect radiographically in kyphoscoliotic patients. Repeated bone grafting is recommended if kyphosis has progressed by more than 5° by 6 months after the arthrodesis. Also, an anterior strut graft is placed at the apex of the curve and spans a long segment; reconstitution is difficult among small pediatric patients and is prone to fracture.[19] The current 4 cases (cases 1, 2, 5, and 6) had favorable corrective results, presumably because these children were older when they underwent surgery. Their spinal segments were sufficiently large in size to place instrumentation parts, and abundant bone chips could be taken from the ilium for fusion. However, hypoplasia of the ilium and ribs were noted in 2 juvenile cases, indicating that autogenous grafts were not easy to harvest. A bone bank had not yet been set up in our hospital during that period. Pediatric posterior implants are much less effective in stabilizing large rigid curves, and are often prominent beneath the skin over the apex of the kyphosis as well as obscuring areas available for posterior arthrodesis.[19] These factors may partially explain the unfavorable outcomes in some patients.

Pseudarthrosis, wound infection, hardware impingement, and loss of correction are the major concerns with surgical correction of PRSDs.[4-6,10,14] King and Stowe[14] reported on 13 patients with Wilms' tumors and neuroblastomas who underwent spinal corrective surgery; pseudarthrosis, due to hypoplasia and avascular changes in the irradiated bones, was found in 7 patients even after repeated fusion attempts. This high incidence of pseudarthrosis was not significantly improved by the addition of posterior instrumentation. Also, the infection rate was 16.6% in patients with Wilms' tumors, and 28.6% in those with neuroblastomas. Lonstein[17] reported pseudarthrosis rates of 25% with anterior fusion alone, 50% with posterior fusion alone, and 9.5% with combined approaches. For kyphotic curvature of greater than 60° in PRSDs, the authors advocate long anterior and posterior release, and fusion with instrumentation to include the normal spinal segments in order to avoid these complications.[19,20] Otherwise, implant impingement and its removal with subsequent loss of correction are inevitable.

Recent advances in the introduction of new pharmaceutical agents for chemotherapy and modern equipment for local radiation have greatly increased the survival of these children after tumor extirpation.[1,12,14,15] The incidence and severity of irradiation-induced skeletal sequelae were significantly lower than those of previous reports.[1] In recent years, we have rarely seen spinal deformities secondary to irradiation in pediatric patients, because of the addition of current chemotherapy without irradiation in children beyond the age of 2 years.[1,2] However, PRSD may still appear in some patients with deficient bone stocks and avascular soft tissues. Kyphosis and soft tissue contracture contribute to curve progression during the adolescent growth spurt. Therefore, all clinicians who care for children with this disease etiology need to be aware of the evolution and prognosis of spinal deformities. Early and careful detection of curve progression and evaluation of the juxtaspinal condition can decrease treatment-related morbidity. Bracing is not effective in situations of progressive kyphosis. Once surgical correction is decided upon, combined front-and-back surgery with long segment fusion and instrumentation should be done to obtain maximal control and correction of the kyphoscoliosis, and to diminish the possibilities of pseudarthrosis and infection.

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腫瘤放射治療引發的脊椎變形手術療法

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背 景： 兒童胸腹腔惡性腫瘤切除術後的放射治療，是會引發胸腰椎變形，尤其在骨骼迅速成長時段，脊椎後側凸彎曲度變化特別明顯，早期手術矯正有其必要。然而手術方式依罹病年紀，受犯椎骨發育大小及彎曲柔軟度而有所差異。

方 法： 過去15年，合計六例腫瘤經放射療法引發脊椎變形者，接受矯正手術，所有病人都追蹤至成年。其原位腫瘤包括三例威姆氏胚性腎肉瘤、兩例神經母細胞瘤，一例惡性淋巴瘤，平均在2.2歲時切除，腫瘤照射量3566 rad.，初始發現脊椎變形是6.1歲，背架矯正無效，接受手術矯正是在11.8歲時，其中三例後位矯正固定術，另三例則須前後位手術處理。

結 果： 椎體變形曲度之凹側與腫瘤手術切除側同，且後凸面厲害程度大於側彎面，是此類病例特色。四例得到滿意矯正結果，另兩例年紀甚輕者，其軀幹瘦薄、脊椎變形角度大且硬、骨量較輕及合併內科疾病，使得矯正固定器擺置不易，導致融骨不良、鋼條突出頂住皮膚而傷口感染、喪失預期矯正效果，使得脊椎後凸畸形明顯。

結 論： 腫瘤放射治療引發脊椎變形的表徵是多樣性的，最嚴重會導致脊椎塌陷、後彎角度大且硬。在瘦小患者，脊椎矯正器組合不易，常無法有滿意結果，合併發生假性關節及術後感染。

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關鍵字： 放射治療引發脊椎變形，後側凸彎，威姆氏胚性腎肉瘤，神經母細胞瘤。

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