Acta Orthopaedica Scandinavica

ISSN: 0001-6470 (Print) (Online) Journal homepage: https://www.tandfonline.com/loi/iort19

One-stage surgical correction of congenital thoracic lordosis--report of 2 cases

Abdullah Gögüs, Ufuk Talu & Azmi Hamzaoglu

To cite this article: Abdullah Gögüs, Ufuk Talu & Azmi Hamzaoglu (2001) One-stage surgical correction of congenital thoracic lordosis--report of 2 cases, Acta Orthopaedica Scandinavica, 72:4, 413-418, DOI: 10.1080/000164701753542096

To link to this article: https://doi.org/10.1080/000164701753542096

Case reports

One-stage surgical correction of congenital thoracic lordosis—report of 2 cases

Abdullah Gögüs[1], Ufuk Talu[2] and Azmi Hamzaoglu[2]

[1]Kadir Has University, School of Medicine, Dept. of Orthopedics and Traumatology, [2]Istanbul University, Medical Faculty of Istanbul, Dept. of Orthopedics and Traumatology, Correspondence: Dr. A. Gögüs, Ataköy 5. Kisim D 10/6, TR-34750 Istanbul, Turkey
E-mail: agogus@hotmail.com
Submitted 00-10-10. Accepted 01-01-06

Case 1

The patient's deformity, first noticed by her parents when she was 1 year of age, was treated with braces and physiotherapy for several years during which time it progressed. She was seen by the senior author (AH) in 1990 at the age of 14 years. Standing radiograms revealed thoracic lordosis between the 4th and 12th thoracic vertebrae of –92°, a sterno-vertebral distance of 6 cm and multiple congenital anomalies in the lumbar vertebrae, but essentially no frontal plane deformity (Figure 1). She was short of breath, especially during physical activities. Her pulmonary function tests showed a vital capacity of 660 mL (one third of normal). Mitral valve prolapse, hypertrophy of the right ventricle due to thoracic deformity, and increased pulmonary artery resistance as well as incomplete right bundle branch block on ECG were diagnosed. The patient had no neurological deficit. CT and MRI scans were not done.

The surgical technique that we used combined the principles set forth by Bradford et al. (1983) for severe idiopathic lordoscoliosis of anterior diskectomies and anterior closing wedges, multiple bilateral rib osteotomies and sublaminar wiring over a kyphotically-contoured stiff rod with the concept of osteotomies of laminar synostosis and osteotomies of fused facet joints using Winter and Leonard's method (1990) (Figure 2). Internal rib osteotomies or resections were performed via the thoracotomy on the convex side and the concave rib osteotomies or resections were done via a posterior approach. She was operated on between T2 and L1 levels. As hardware, we used two 6.4 mm Luque rods and sublaminar wires. Wake-up test was normal and total duration of the operation was 8.5 hours. The blood loss was 1.3 L. The postoperative course was difficult. She needed ventilatory support for 17 days and had a secondary suture for her wound, which finally healed uneventfully. External support was not used. 1 year later, we did a Galveston spino-pelvic fixation between L2 and the ilium mainly for her congenital lumbar scoliotic curve and, to a lesser extent, for her pelvic obliquity of 2 cm (Figure 1). At the 10-year follow-up, she had no complaints apart from her short stature. The thoracic kyphosis angle measured –7° (85° of correction), pelvic obliquity 3 cm and trunk shift 2.6 cm (Figure 1). Her pulmonary function tests 2 and 10 years after the index procedure revealed a vital capacity of 1.1 L (half of normal) and 1.3 L, respectively.

Case 2

An 8-year-old girl with progressive spinal deformity, but no dyspnea was first seen by the senior author (AH) in August 1997. Her physical examination showed thoracic lordosis with slight right thoracic scoliosis. Standing radiograms revealed a low thoracic lordosis between the 4th and 12th thoracic vertebrae of –30° and right convex scoliosis of 27°. CT scans of the thoracic vertebrae showed fusion of the facet joints of T6-T10 on the left, of T7-T11 on the right side and fusion of the spinous processes between T5-T6 and T7-T11. Spina bifida between T7-T10 was also clearly seen on the CT scans (Figure 3). On MRI scans, no intramedullary anomaly was detected. Pulmonary function tests showed a vital capacity of 720 mL (three-fourths of normal). No cardiac or renal anomaly was found. Neurologically she was intact.

For posterior instrumentation, we used pediatric pedicle screws placed at T2 and T12 levels.

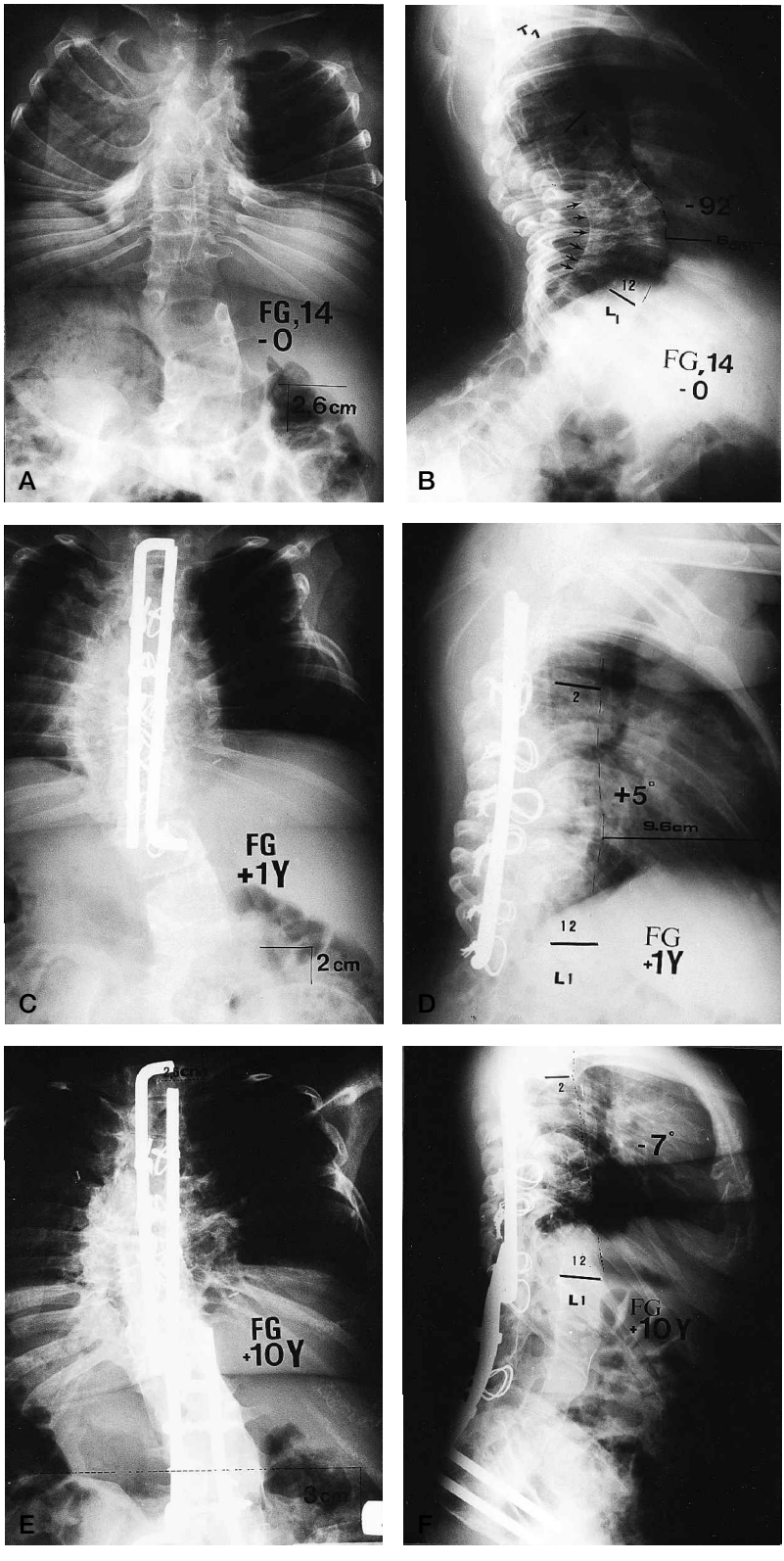


Figure 1. Case 1.

A. Preoperative standing posteroanterior radiograph at age 14 shows no thoracic scoliosis, but pelvic obliquity of 2.6 cm.

B. Initial lateral standing radiograph shows thoracic lordosis of –92° between T4 and T12 and a T10-sternum distance of 6 cms. Black arrows point to synostosis of the spinous processes.

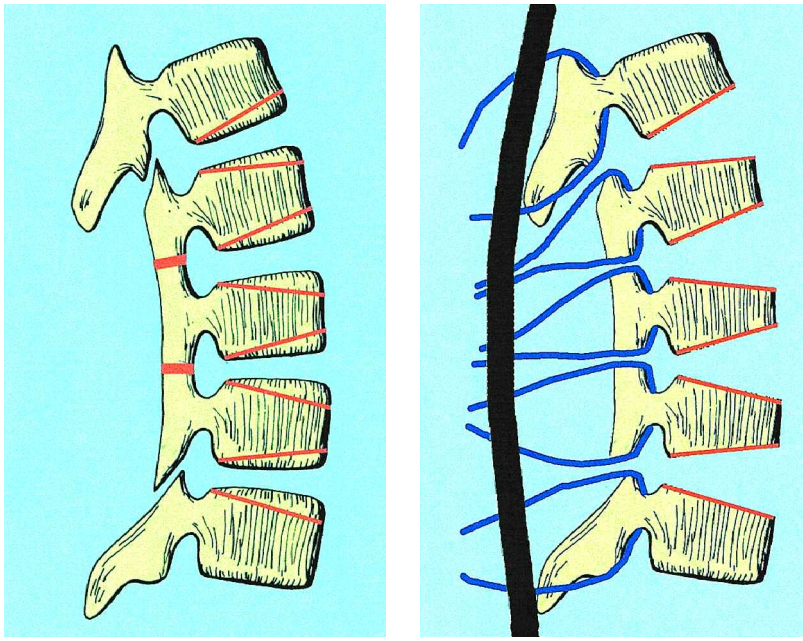
C. Standing posteroanterior radiograph taken 1 year postoperatively shows a slight increase in the lumbar curve and a pelvic imbalance of 2 cm. We therefore decided to perform a posterior spino-pelvic fixation.

D. The lateral standing radiograph on the same date shows correction of her lordosis to +5° and an increase in the spine-sternum distance of 9.6 cm.

E. Posteroanterior radiograph at 10 years postoperatively shows no improvement in pelvic obliquity (3 cm) and a trunk shift of 2.6 cm.

F. The thoracic kyphosis angle measured –7° (85° of correction). Note 12° loss of correction in 9 years.

Figure 2. Illustration of the surgical technique used in both of our cases



A. Anterior disc excision, planned anterior wedge osteotomies and corresponding posterior osteotomies of laminar synostosis are outlined by red lines. The disc spaces are not packed with bone chips because the wedges need to close anteriorly.

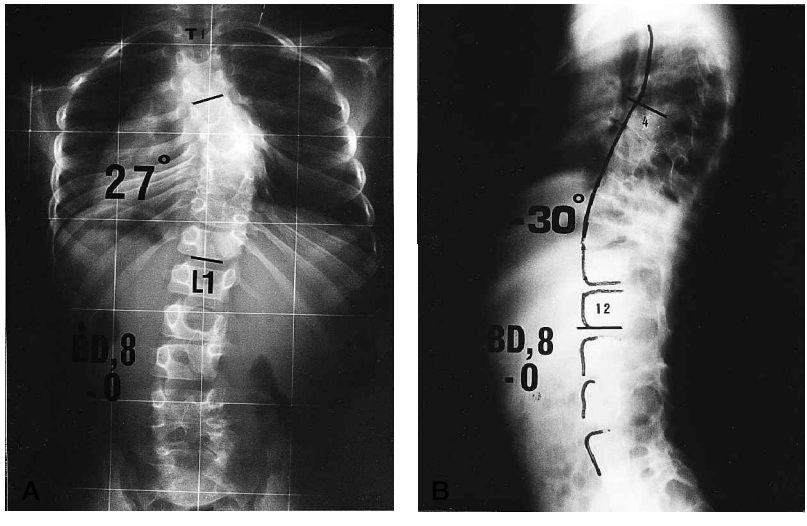
B. Passing of sublaminar wires and tightening over a kyphotically-contoured stiff rod converts lordosis into kyphosis.

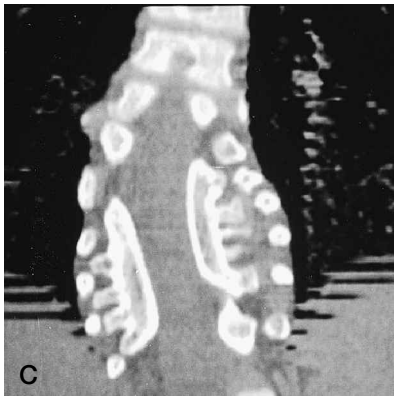
Wake-up test was normal and total duration of the operation 7 hours and 45 minutes. The blood loss was 0.6 L. She was weaned from an endotracheal tube on the first postoperative day and discharged on the 7th postoperative day. No brace was used postoperatively. She had a normal clinical appearance and was asymptomatic at the last follow-up after 36 months. Standing radiograms showed a solid anterior and posterior fusion, no scoliosis and the pathological –30° thoracic lordosis had been

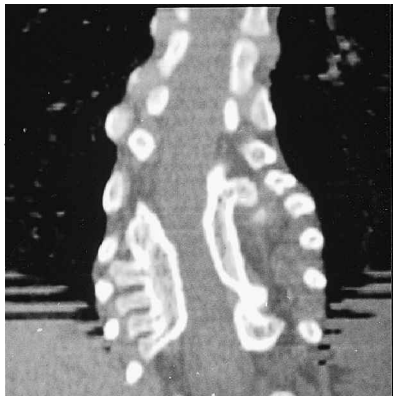
Figure 3. Case 2

A. Preoperative standing posteroanterior radiograph at age 8 shows right thoracic scoliosis of 27° between T4 and T12.

B. Preoperative standing lateral radiograph shows thoracic lordosis of –30° between T4 and T12.





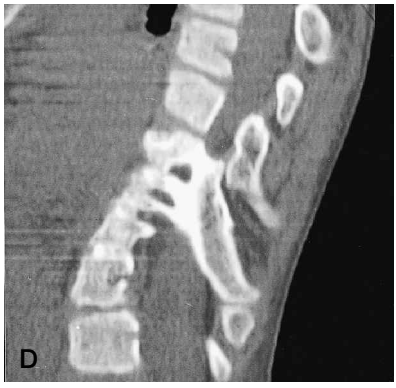


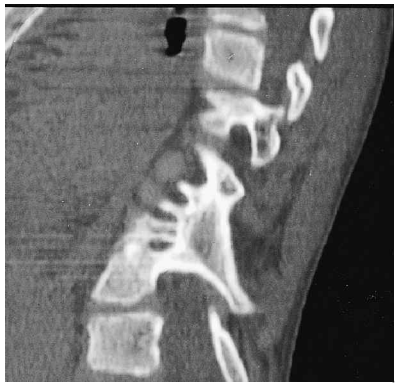
C. Coronal reformat CT-scans show very clearly fusion of the facet joints of T6-T10 on the left and of T7-T11 on the right side.

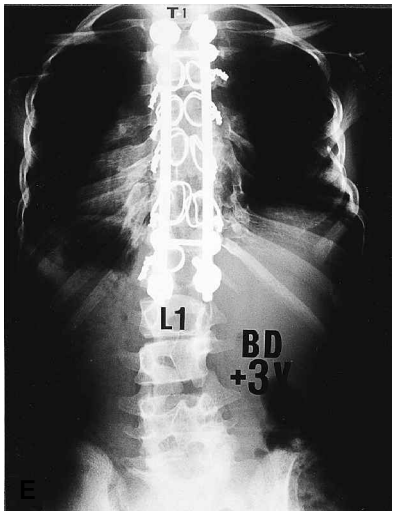
D. Sagittal reformat CT scans show fusion of the spinous processes between T5-T6 and T7-T11.

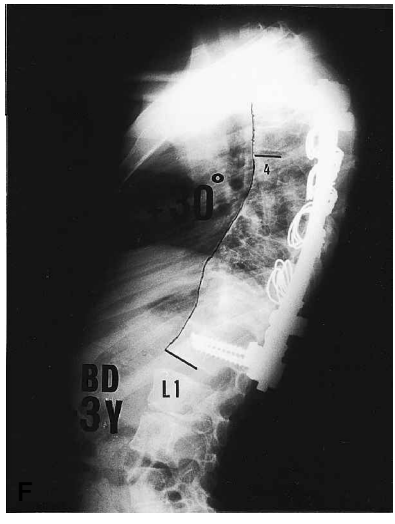
E. Standing posteroanterior radiograph at 3 years postoperatively shows a solid anterior and posterior fusion and no scoliosis.

F. Lateral standing radiograph showing a physiological thoracic kyphosis of 30°.









converted to physiological kyphosis of 30° (60° of correction) (Figure 3). Repeat pulmonary function test revealed about the same vital capacity of 1.1 L (four-fifths of normal) as preoperatively. She still has regular clinical and radiological follow-ups because of her multiple congenital anomalies above and below the fused segments, which have not progressed during the 3-year follow-up.

Discussion

Congenital lordosis, the least common of congenital spine deformities, is caused by a failure of posterior segmentation; cases with failure of posterior formation have not been reported. Most patients have some degree of scoliosis because the unsegmented bar is located posterolaterally, but the main deformity is lordosis (Winter et al. 1996, Lonstein 1999). The lordosis in the thoracic vertebrae can reduce the pulmonary reserve markedly and often causes dyspnea, which makes early treatment mandatory (Winter et al. 1975, Winter 1992). Even difficulty in swallowing of solid food, due to esophageal compression, can be a problem (Winter et al. 1978).

A thoracic kyphosis of less than 20° is considered evidence of a relative lordosis by the Scoliosis Research Society. Winter et al. (1975) first pointed out the harmful effect of thoracic lordosis on pulmonary function in their patients with idiopathic lordoscoliosis. He treated them all by posterior fusion with Harrington distraction rods, but found the amount of correction of the lordosis disappointing and stated that a better method of treatment was needed. Bradford et al. (1983) corrected more rigid idiopathic lordoscoliosis with anterior multilevel diskectomy, anterior convex rib resection, and posterior Harrington rods using Luque sublaminar wires and concave rib resection.

It has only recently been appreciated that thoracic lordosis is the most lethal of congenital spine deformities (Winter and Leonard 1990). The natural history of congenital lordosis, especially in the thoracic spine, is not good since progressive deterioration in pulmonary function is inevitable and early death can result from respiratory insufficiency and cor pulmonale (Winter et al. 1978). If detected before severe deformity and loss of pulmonary function have occurred, then the treatment of choice is anterior fusion although it does not correct the existing deformity (Winter et al. 1996, Lonstein 1999). The first report in the literature by Winter et al. (1978) concerned the surgical treatment of 5 patients with congenital thoracic lordosis. 3 of them died because of respiratory insufficiency postoperatively and 2 had a single procedure, anterior or posterior spine fusion. Because the patients generally present late with more marked lordosis, as in our patients, and correction is mandatory, both anterior and posterior procedures are needed (Winter et al. 1996, Lonstein 1999). Winter and Leonard (1990) reported the first successful 2-staged surgery. They corrected the congenital thoracic lordoscoliosis, but did not perform a rib resection or rib osteotomy. This 13-year-old girl's thoracic lordosis was corrected from –25° to + 25° of thoracic kyphosis, and her scoliosis from 71° to 41°. Unlike scoliosis or kyphosis, there is no proper way to determine preoperatively how much thoracic lordosis can be corrected (Winter 1984). We found the combination of techniques described by Bradford et al. (1983) and Winter and Leonard (1990) very effective in converting a pathologic lordosis into kyphosis.

We know of no reports on correction of congenital thoracic lordosis at one sitting. We achieved 60° and 80° of correction. Like others (Bradford et al. 1983, Winter et al. 1996, Lonstein 1999) we believe that bilateral multiple rib osteotomy is necessary for correction of the lordosis. The correction will otherwise be hampered by the rib cage. The passing of sublaminar wires in the thoracic spine has inherent neurologic risks in the best of circumstances, but doing it in thoracic lordosis is especially worrisome since the spinal cord is much closer to the laminar surface than to the vertebral body (Winter 1992). No neurologic complications occurred in our patients.

In the first case, we extended the fusion area and performed a posterior spino-pelvic fixation for the lumbar curve, which was slightly increased at one year. We did this to prevent possible deterioration of the lumbar deformity, correct the existing trunk-shift of 2 cm and also increase the stability of the previous thoracic instrumentation construct, which consisted of sublaminar wires lacking axial stability. However, during the follow-up, hardly any improvement occurred in the overall spino-pelvic balance and trunk-shift after this second procedure. We now believe that it was probably unnecessary because she was mature enough and had a relatively balanced congenital lumbar deformity at that time. Thus, observation and close follow-up would have been another alternative, which we preferred for the second case.

Since the primary purpose of correction and fusion of congenital thoracic lordosis is preser-

vation and improvement of pulmonary function, it is preferable to do both anterior and posterior procedures under one anesthetic to increase lung volume immediately. Doing the thoracotomy without increasing the thoracic volume simultaneously can cause many problems (Winter and Leonard 1990, Winter 1992, Bagshaw and Jardine 1995). Fewer complications occur during a single longer anesthetic than after two anesthetics and two recovery periods (Winter 1992). Because of the preexisting respiratory compromise, thoracotomy and bilateral multiple rib resections, postoperative respiratory care is critical and a prolonged period of ventilatory support may be necessary. Accordingly, the first of our patients with the most serious thoracic lordosis of –92° and a preoperative vital capacity one third of normal needed 17 days of ventilatory support postoperatively.

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

Bagshaw O N, Jardine A. Cardiopulmonary complications during anaesthesia and surgery for severe thoracic lordoscoliosis. Anaesthesia 1995; 50: 890-2.

Bradford D S, Blatt J M, Rasp F L. Surgical management of severe thoracic lordosis. A new technique to restore normal kyphosis. Spine 1983; 8: 420-8.

Lonstein J E. Congenital spine deformities. Clin Orthop 1999; 30: 387-405.

Winter R B. Thoracic lordoscoliosis in neurofibromatosis: Treatment by a Harrington rod with sublaminar wiring. J Bone Joint Surg (Am) 1984; 66: 1102-6.

Winter R B. Surgical correction of rigid thoracic lordoscoliosis. J Spinal Disord 1992; 5: 108-11.

Winter R B, Lovell W W, Moe J H. Excessive thoracic lordosis and loss of pulmonary function in patients with idiopathic scoliosis. J Bone Joint Surg (Am) 1975; 57: 972-7.

Winter R B, Moe J H, Bradford D S. Congenital thoracic lordosis. J Bone Joint Surg (Am) 1978; 60: 806-10.

Winter R B, Leonard A S. Surgical correction of congenital thoracic lordosis. J Pediatr Orthop 1990; 10: 805-8.

Winter R B, Lonstein J E, Boachie-Adjei O. Congenital spinal deformity. Instr Course Lect 1996; 45: 117-27.