

Technical Report

Bilateral vascularized rib grafts to promote spinopelvic fixation in patients with sacral agenesis and spinopelvic dissociation: a new surgical technique

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Abstract

BACKGROUND CONTEXT: Sacral agenesis is a rare congenital disorder that may have spinopelvic instability due to sacroiliac joint malformation. Surgical indication in patients with sacral agenesis is to improve their sitting balance and protect the visceral organs. Achieving solid arthrodesis across this congenital malformation is challenging and prone to non-union.

PURPOSE: The purpose of this study was to describe a novel surgical technique with vascularized ribs for management of sacral agenesis and complex spinopelvic dissociation.

STUDY DESIGN: Retrospective study.

PATIENT SAMPLE: Six patients with sacral agenesis were reviewed and followed for a mean of 8.5 years after spinopelvic fusion augmented with vascularized rib graft spanning the lumbo-pelvic junction.

OUTCOME MEASURES: The primary outcome measure was the presence or absence of a stable spinopelvic junction and fusion across the spine-vascular rib grafts-pelvis interface. The secondary outcome measures were maintenance of pelvic obliquity, lumbosacral kyphosis, and overall sagittal balance.

METHODS: The surgical procedure consisted of two-stage surgeries performed 6–12 weeks apart. The first stage consisted of spinal instrumentation and correction of the deformity via a posterior approach and impaction of one of the vascularized ribs from the spine to the iliac crest. The second stage consisted of an anterior thoraco-lumbar approach for spinal fusion and the second vascularized rib spanning the spine to the iliac crest.

RESULTS: All six patients eventually achieved a solid spinal and spinopelvic fusion. All vascularized ribs increased in diameter over time. A high complication rate consisted mainly of spinal infections and prominent hardware requiring revision surgeries (a total of seven procedures in four patients). Two patients had decreased mobility secondary to spinopelvic surgery at last follow-up.

CONCLUSIONS: Spinopelvic fusion can be successfully achieved with this novel surgical technique using vascularized rib grafts. This technique allows for biological long-term maintenance of the sagittal deformity correction. Fusion across the lumbosacral junction in patients with sacral agenesis may place them at risk of losing the ability to mobilize independently. Recent lower profile implants have prevented implant-related complications. © 2015 Published by Elsevier Inc.

Keywords:

Congenital scoliosis; Lumbosacral kyphosis; Pedicle screws; Sacral agenesis; Spinopelvic fusion; Vascularized rib graft

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Introduction

Sacral agenesis or caudal regression syndrome is a rare congenital disorder with unclear etiology. It has been associated with maternal diabetes [1–13], genetic factors, and vascular hypoperfusion [2,5,7,8]. An autosomal dominant form of the disorder also exists, known as Currarino syndrome. It consists of anorectal malformations, presacral mass, and urogenital malformations [4,8,14,15]. Patients with sacral agenesis can have spinopelvic instability and even frank spinopelvic dissociation due to sacroiliac joint malformation, not to mention the multitude of genitourinary and gastrointestinal malformations [2,5,6,9,10,14,16–18]. In complete lumbosacral agenesis, the lower thoracic vertebrae are linked with the pelvis only via soft tissue, creating an unstable spinopelvic junction and thus an unstable lumbosacral junction, which can lead to severe kyphosis in a sitting position or on forward bending [9,10]. These mechanical problems are a result of a spectrum of developmental failure involving the lumbar, sacral, and coccygeal vertebrae [1,2,5,6,8,9,12,14,16,18,19] typically caused by a neural tube defect before 28 days of gestation [2,5,7,12,14,18,20]. Typically, the agenesis also involves the spinal cord at the same levels [1–3,6,7,14], resulting in neurologic deficits that range from mild impairment of bladder control to total loss of motor and sensory function of the lower extremities [1–3,6,7,9,12,14,17,21–24]. Scoliosis, myelomeningocele, lower limb weakness or paralysis, hip and knee contractures with popliteal webbing, lower limb and foot deformities, and hip dislocations are frequently seen in patients with sacral agenesis [1–3,6,9,12–14,16,17], along with the inability to ambulate. Treatment of these patients is challenging and involves a multidisciplinary team consisting of a urologist, a pediatric surgeon, a pediatrician, a physiotherapist, a pediatric orthopedic surgeon, and a spine surgeon [7,12].

The natural history of these caudal regressions varies depending on the level of involvement. Van Buskirk and Ritterbusch [11] illustrated that a good understanding of these patients' overall function must be considered before embarking on surgical correction as surgery may hinder the patients' independence rather than improving their quality of life. A deformity does not necessarily need a correction. For the spine surgeon involved in the treatment of these patients, an important consideration is the potential of these patients to become ambulators. The main problem, if any, to be addressed by the spine surgeon in the cases of sacral agenesis is the spinopelvic instability or obliquity. A spinopelvic fusion and stabilization in such situations may help the sitting balance of these patients and protect the visceral organs from compression by reducing the kyphotic deformity [16,17]. In the potentially ambulatory patient, it is even more important to stabilize their spinopelvic junction to improve their function [7,11]. According to Renshaw [3], patients with sacral agenesis of Renshaw classification types III and IV more commonly develop signs of progressive spinopelvic kyphosis or instability, and may require lumbo-pelvic fusion, which is recommended to be carried out after the age of 4 years [3].

However, it is difficult to achieve a solid lumbo-pelvic fusion in these cases because of the lack of bone stock in the area prone to instability [19]. A limited number of techniques have been described in the literature to address spinopelvic fusion in patients with sacral agenesis [7,17,19,22,25]. Those techniques are associated either with non-unions [7,17,22] or with morbidity from bone graft donor sites distant from the surgical site [17,19].

In this article, a new technique is described for consistently achieving arthrodesis across the spinopelvic section with the creation of a biological strut that bypasses the dysplastic lumbosacral-pelvic junction. The technique involves achieving initial stabilization with the use of spinopelvic instrumentation and creating a biological strut by using bilateral vascularized rib grafts spanning the spine to the pelvis to provide long-term stability. The purpose of this study was to describe the safety and efficacy of a novel surgical technique with vascularized ribs for management of sacral agenesis and complex spinopelvic dissociation.

Materials and methods

This is a technical report of a retrospective review of cases, over a period of 19 years, of six patients who underwent spinopelvic instrumentation and fusion using bilateral vascular rib grafts (surgical technique developed by VA), with the exception of one patient who had a unilateral rib graft. During the same period, we managed four patients with sacral agenesis non-operatively. There were three boys and three girls in the operative series. The mean duration of follow-up was 8.0 years (range: 3.1–14.3 years) and the mean age at surgery was 5.6 years (range: 2.9–6.7 years). The review was done using the patients' hospital charts, operating room reports, clinical notes, and plain radiographs. All patients had a diagnosis of sacral agenesis and were treated at our institution by either one of the two senior authors. The types of sacral agenesis were classified according to the Renshaw classification [3] (Fig. 1). Perioperative parameters such as blood loss, duration of surgery, type of fixation, and complications were recorded. Radiologic assessment was performed by evaluation of the preoperative, postoperative, and final follow-up plain radiographs. Standard scoliosis imaging, which included postero-anterior and lateral radiographs of the entire spine, was performed preoperatively and at each postoperative visit. The radiologic parameters measured were pelvic obliquity, lumbosacral kyphosis, overall sagittal balance, and fusion across the spine-rib-pelvis interface. The clinical and radiologic follow-ups of one patient (Case 5) are illustrated in Figs. 2 and 3, respectively.

Surgical technique

The surgical procedure consisted of two-stage surgeries performed 6–12 weeks apart. In the first stage, three specific objectives had to be met: (1) correction of the spinal deformity with solid spinopelvic instrumentation, (2)

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