

ARTERIOMESENTERIC COMPRESSION OF THE DUODENUM AS A COMPLICATION OF THE SURGICAL CORRECTION OF TYPICAL IDIOPATHIC SCOLIOSIS: CLINICAL CASE

S.O. Ryabykh¹, E.G. Scryabin², V.P. Chevzhik³, E.Yu. Filatov¹

¹Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, Kurgan, Russia

²Tyumen State Medical University, Tyumen, Russia

³Regional Clinical Hospital No. 2, Tyumen, Russia

The presented rare clinical case of the development of arteriomesenteric compression of the duodenum is described with the aim of informing a wide audience of specialists (orthopedic trauma surgeons, vertebrologists, neurosurgeons, surgeons, neurologists and pediatricians) about a rare complication of surgical correction of typical adolescent idiopathic scoliosis — intestinal obstruction due to compression of the lower horizontal part of the duodenum by the superior mesenteric artery. Publications answering two posed questions were analyzed: is there a correlation between surgical correction of scoliosis and development of abdominal pathology in a patient in the immediate postoperative period, and should the instrumentation be completely or partially removed at the level of the thoracolumbar junction in the case of formation of duodenal arteriomesenteric compression in a patient operated on the spine? Abdominal visceral complications are extremely rare and represented by a very few publications even in modern electronic databases of medical information, therefore we consider it important to bring a case from our practice to the attention of readers. **Key Words:** superior mesenteric artery syndrome, arteriomesenteric compression, complication, deformity, scoliosis, duodenal obstruction.

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Surgical correction of idiopathic scoliosis using multiple anchor instrumentation has been actively used for the past three decades. Such aspects as the effectiveness of 3D correction of spinal deformities and reconstruction of the surface anatomy of the trunk, as well as shorter treatment duration, potential early verticalization, and lifestyle rehabilitation, are beyond dispute. Meanwhile, the rate of complications of surgeries to manage idiopathic scoliosis over the past two decades has been no higher than 7 %, while the typical structure of complications is in most cases represented by vertebrogenic ones (implant-dependent problems, surgical site infection, and neurological deficit). The rate of other complications is not higher than 1.7 % [1, 2]. Idiopathic scoliosis is known to affect the skeletopia of visceral organs, including the gastrointestinal tract organs [3, 4]. In most clinical case reports, the surgeries significantly improved patients'

anatomical and functional condition of visceral organs, including the gastrointestinal organs [5–7]. Meanwhile, after surgical correction some patients have severe visceral complications that are primarily caused by impairment of circulation and innervation in the stomach, intestine, and lungs [8–10].

Sporadic publications have focused on the pathogenetic situations leading to various dysfunctions and structural disorders of the digestive system in patients operated on for scoliosis [11, 12]. The fact that there are only few publications (mostly reporting a single clinical case) also demonstrates that visceral complications are very rare [13, 14].

The objective of this study is to inform the broad audience of specialists (orthopedic trauma surgeons, spinal surgeons, neurologists, and pediatricians) about the rare complication of surgical correction of typical [15] adolescent idiopathic scoliosis: intestinal obstruction caused by

compression of the inferior horizontal part of the duodenum by the superior mesenteric artery.

Clinical case. A female patient (aged 13 years and 6 months) was admitted to hospital for performing a scoliosis correction surgery. It is known from the patient's medical history that scoliosis was progressing since the girl was 5 years old; conservative treatment (exercise therapy, swimming, massage sessions, and corset wearing) yielded no positive results; contrariwise, the deformity was increasing. During the year before the surgery, the child started complaining of vertebrogenic pain. Such factors as progression of vertebrogenic deformity, vertebrogenic pain, ineffectiveness of conservative treatment, and willingness of the patient and her parents to treat the spine deformity were indications for surgical correction. Prior to surgical treatment, the patient was diagnosed with typical idiopathic scoliosis (grade IV according to Chaklin's classification,

Lenke 2BN [16], and moderate severity according to the James' classification [17]) based on patient's complaints, past medical history, as well as the clinical and radiographic diagnostic data.

Two-view radiographic examination of the spine in a standing position revealed a double scoliotic curve in the thoracic spine, with the principal right-sided curvature at T4–T12 (Cobb angle, 65°), the apex at T8, the proximal main thoracic curve (30°) and compensatory (nonstructural) lumbar curve (37°) (Fig. 1). The thoracic kyphosis angle was 12°. The functional radiographic examination of the spine (the images being recorded when the patient was bending to the left and right side to the maximum extent) showed that mobility of the lumbar curvature was > 40 %.

MRI of the thoracic and lumbar spine revealed a 2.2×0.3 cm syringomyelic cyst at the T7–T8 level; the patient's neurological status was not impaired. Lung CT scanning revealed no focal or infiltrative changes.

Taking into account the type and severity of the deformity, the patient underwent correction and posterior instrumented fusion at the T3–L2 level, and posterior fusion within the instrumented fixation zone using the standard procedure without any technical difficulties.

During the first postoperative week, the patient's condition was satisfactory and corresponded to the severity of the surgery. On day 8, the girl started having abdominal pain, nausea, and vomiting. Having examined the patient's digestive system, the pediatrician made a diagnosis of nonspecific reactive hepatitis and gastroduodenitis. The recommended treatment consisted of infusion therapy (normal saline), antimicrobial therapy (Cefosin), analgesics (Tramadol), spasmolytic agents (drotaverine hydrochloride), antiemetics (Metoclopramide), antianemic drugs (Ferrum Lec), normalization of the water-electrolyte balance (Rehydron), an antiflatulent agent (Espumisan), diet, and water intake schedule. After two days of this therapy, the patient's condition was normalized. After the control pelvic ultrasound examination, the girl

was discharged from the inpatient orthopedic department to receive outpatient care.

The postoperative wound in the projection of the spine was allowed to heal by primary intention. The postoperative angle of the major curvature decreased to 17°, so the degree of deformity correction was 74 % (Fig. 2).

The well-being of patient's gastrointestinal organs was delusive: abdominal pain and dyspeptic symptoms were resumed after two days of being at home. This made the patient's parents call an emergency care team, which transferred the girl to a specialized inpatient surgery center.

The patient's condition at admission was regarded as serious because of dehydration manifesting itself as extreme weakness. The patient was taking up the forced (side-lying or crawling) position. Her skin and mucosa were pale. Normal breathing was heard over all the lung fields (no rales; respiratory rate, 20 breaths per minute). Heart tones were well-defined and rhythmic; heart rate was 93 bpm; BP was 110/80 mm Hg. The abdomen was distended, palpation was causing pain in all the quadrants. At examination, the patient was experiencing profound vomiting (dark green vomit). According to the patient and her mother, the patient was having black stool. Immediately after a nasogastric tube had been inserted, 3,000 mL of

dark stagnant gastric contents without blood was removed through it.

Because of the severity of her condition, the patient was admitted to the intensive care unit. Complete blood count test showed leukocytosis $(25 \times 10^9/L)$, with a shift in differential WBC count toward 9% of band neutrophils and 79 % segmented neutrophils. The child had severe metabolic impairment requiring intense syndromic treatment and examination.

Abdominal ultrasound revealed signs of enteroplegia; the intestine was enlarged and congested with the gastric contents. Esophagogastroduodenoscopy revealed profound dark contents and food debris in the gastric lumen. The view was limited; no signs of blood were detected. Thickened gastric folds and active gastric motility before the stomach exit were observed. The gastric mucosa was diffusely reddened; the vascular pattern seemed unchanged. The pylorus was round-shaped (the opening size being 10 mm). The duodenal bulb: round-shaped lumen; reddened mucosa; profound dark stagnant contents were detected in the gastric lumen. Barium radiographic examination revealed that the stomach was significantly enlarged, with the greater curvature of the stomach located in the pelvis, without any signs of evacuation of stomach contents (Fig. 3).



Fig. 1
Radiographs of the thoracic and lumbar spine recorded telemetrically and two-view functional radiographic examination (see comments in the text)

Conservative treatment was ineffective; abdominal pain and distention persisted. Stagnant gastric contents (500 mL) were evacuated via the gastric tube daily. The patient was diagnosed with chronic duodenal obstruction.

Treatment failure and the manifestations of severe intestinal obstruction persisting for the two days that the patient was remaining in the inpatient surgical department were indications for surgical intervention. Laparoscopy revealed enlarged stomach and duodenum; the jejunum was collapsed. Feeding normal saline into the stomach was accompanied by filling and significant dilatation of the duodenum; the normal saline did not move further into the jejunum; and pancreatic head enlargement was observed. Conversion to laparotomy was performed. Revision of the abdominal cavity revealed that compression of the inferior (third) horizontal part of the duodenum by the superior mesenteric artery was the reason for the obstruction (Fig. 4). Taking into account the clinical situation, retrocolic duodenojejunostomy (to form a side-to-side Roux-en-Y anastomosis using two-layer suture) was performed.

The postoperative period was uncomplicated; abdominal pain and dyspeptic symptoms were eradicated. The passage of intestinal contents was restored, and patient's stool was normalized. The post-laparatomy wound healed by primary intention. When staying at the surgical department, the patient had no subjective complaints about the operated spine.

Discussion

The main question has arisen when analyzing the clinical situation: whether the surgical correction of scoliosis and development of the abdominal pathology in the patient in early postoperative period were interrelated.

Thus, the reported rare complication of surgical correction of scoliosis is most often equated with the term «superior mesenteric artery syndrome» (also known as the Wilkie's syndrome, SMA, or cast syndrome) in the literature. An analysis of the literature data has revealed two main aspects: (1)

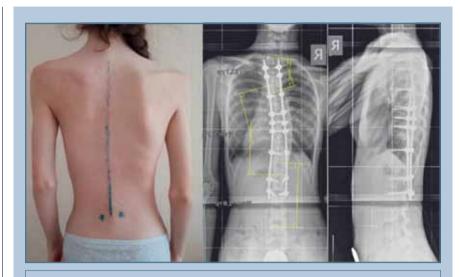


Fig. 2

Appearance of the posterior trunk surface and radiographs of patient's spine in the early postoperative period

in most cases, surgical correction of spinal deformity (including surgeries involving distraction as the key component of correction) does not trigger the acute operative pathology in adolescents and (2) asthenic constitution is the main background factor that has caused the superior mesenteric artery syndrome, followed by intestinal obstruction. Thus, prior to the surgery, the patient's weight was 43 kg, while height was 165 cm. The calculated body mass index was 15.8, which is a pronounced body mass deficit.

Based on their clinical morphological studies, M.V. Repin and E.S. Patlusova [18] stated that arteriomesenteric duodenal compression is a disorder primarily caused by patients' constitution. These patients usually lack adipose tissue in the mesenterium and the retroperitoneal space, which acts as some kind of buffer protecting the inferior horizontal part of the duodenum from pressure exerted on it by the superior mesenteric artery and preventing it from being pressed against the aorta and spine at the L1 level [19-21]. In abdominal surgery, it is considered that a factor triggering this pathology is the angle between the superior mesenteric artery and the abdominal aorta being more acute ($<28^{\circ}$) than the normal one $-38-60^{\circ}$ (Fig. 5).

In addition to the acute aortomesenteric angle, patients with asthenic constitution have shorter distance between the aorta and the superior mesenteric artery at the level of the duodenum (0.5–0.7 cm vs the normal 1.5–2.0 cm). Barium duodenography showed that the compressed part of the duodenum at the L1 level can be as long as 1.5–3.0 cm [19].

It is considered that one of the possible reasons for the abnormal vascular anatomy in this region is surgical correction of multiplanar vertebral deformity accompanied by distraction and detorsion, thus causing vertical distraction of the mesenteric artery and a decrease (being quite natural) in the aortomesenteric angle and the distance between these vessels [14, 23]. Researchers believe that connective tissue dysplasia plays a significant role in the pathogenesis of the superior mesenteric artery syndrome followed by duodenal compression in adolescents [18]. Despite the discussions held around the pathogenesis of idiopathic scoliosis, some researchers suppose that connective tissue dysplasia is one of the factors responsible for its progression [24].

According to the literature data [13, 25], the rate of superior mesenteric artery syndrome in patients who had



Fig. 3
Abdominal survey radiograph (barium sulfate used as a contrast agent).
The greater curvature of the stomach is located in the lesser pelvis

undergone spine surgeries is 0.8–4.7 %. Having reviewed the available publications focused on this topic over the past 45 years (up to 2017), Louie et al. [26] reported on only 19 published cases of this abdominal pathology that had developed after surgical correction of scoliosis. Even if taking into account the fact that the authors have retrieved and analyzed not all the publications focused on this problem, it still must be admitted that it has not been adequately covered in modern medical literature.

An analysis of medical data on the problem of development of severe intestinal obstruction in adolescents operated on for scoliosis yielded summarized data on the essential aspects of formation and course of this severe condition. It turned out that the disease onset was observed within the 1st week after spine surgery in approximately half of all cases. About 35 % of pathology cases develop during the 2nd week after the fusion; 15 % of cases, during the 3rd week [26, 27].

In our case report, the disease onset was observed on day 8 after the intervention; the pathology manifested itself



Fig. 4

An intraoperative image: the duodenum enlarged to 15 cm in diameter was delivered into the wound

as abdominal pain, nausea, and vomiting. Many authors have mentioned these very symptoms at disease onset [15, 27, 28]. Next, usually within several days, other manifestations of severe intestinal obstruction appear: early satiety, epigastric heaviness, abdominal distension, absence of bowel movement or passage of flatus, and constipation [14, 23]. Late symptoms of the pathology include eating disorders leading to anorexia and even more significant body weight loss [20, 26].

In addition to the assessment of patient's complaints, past medical history, as well as clinical and paraclinical symptoms, physicians used such methods as abdominal ultrasonography [15, 28], contrast radiography of the stomach [15, 23, 27, 28], CT scanning [14, 28], and endoscopy [26, 28] to objectively diagnose superior mesenteric artery syndrome. Differential diagnosis is performed to distinguish between superior mesenteric artery syndrome and duodenitis or intussusception [26].

All the aforelisted objective diagnostic techniques except for CT scanning were used for diagnosing our patient and during the examination.

It is recommended that duplex ultrasound of abdominal vessels for studying

their anatomy and blood flow parameters is performed during the preoperative preparation of children with scoliosis in order to prevent arteriomesenteric duodenal compression [12].

Conservative management should be started with inserting a nasogastric tube to evacuate the stagnant stomach contents [26] and infusion therapy to normalize the water-electrolyte balance and treat protein-energy insufficiency [26, 28]. Antibiotics are used to eliminate the inflammation symptoms identified using the paraclinical examination methods [15, 27]. Symptomatic pharmaceutical treatment is indicated to correct the existing dyspeptic symptoms and the general ill-being caused by them [23]. Feeding (usually enteral) is performed under strict medical supervision [26].

Ineffective conservative therapy is an indication for abdominal intervention [29, 30]. The aim of the surgery is to perform duodenojejunostomy using various procedures (Robinson's, Albrecht–Stavely's with Boppe's modification, etc.) [20]; gastrojejunostomy is performed less frequently [28].

In the reported clinical situation, the conservative therapy was ineffective. A decision was made to perform a surgery, which was conducted on day 3 after the

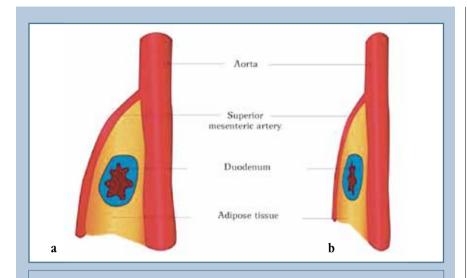


Fig. 5
Schematic view of anatomical relations of the abdominal aorta, the superior mesenteric artery, and the duodenum in the sagittal plane: normal variant (a) and arteriomesenteric duodenal compression (b)

onset of abdominal pain, nausea, and repeated vomiting. As already mentioned, the surgical intervention involved performing Roux-en-Y retrocolic duodenojejunostomy. Infusion, antimicrobial, and symptomatic treatments were continued during the postoperative period; patient's well-being, appetite, and stool were normalized. On day 18 after the abdominal surgery, the girl was discharged from the surgical department to receive outpatient care; her condition was satisfactory. An analysis of the short-term results (within 6 months) revealed no recurrence of gastrointestinal manifestations. The condition of the operated spine was satisfactory and corresponded to the severity of intervention and the period that had passed after it.

The second question (whether the metal instrumentation should be removed, either completely or partially, at the level of the thoracolumbar junction if a patient who had undergone spine surgery develops arteriomesenteric duodenal compression) has been addressed in none of the medical publications focused on the problem being discussed in this study. The removal of instrumentation would reduce the

extent of spine distraction, thus relieving the tension of mesenteric vessels and increase the angle between the superior mesenteric artery and the abdominal aorta (i.e., eliminate the factors that are currently considered to be crucial in the pathogenesis of Wilkie's syndrome). In the literature, this question has been superficially addressed by Louie et al. [26]. They reported that if arteriomesenteric duodenal compression develops in patients operated on for scoliosis, there is usually no need to remove metal instrumentation, except for the cases when lumbar hyperlordosis was formed. This clinical situation requires closer attention and comprehensive analysis, since arteriomesenteric duodenal compression in adolescents is a potentially lethal complication [26, 27]. In support of this, we would like to mention the publication by Kennedy et al. [31]; they presented a case report of a 14-year-old boy with asthenic constitution who had undergone surgical correction of scoliosis using Harrington distraction followed by spine immobilization by using a plaster corset. After the surgery, the angle of the major spine curvature decreased from 73 to 54°. Forty days after the spine surgery,

the patient suddenly started presenting with abdominal pain and vomiting. After the emergency clinical and instrumental examinations, a decision was made to perform laparotomy. Revision revealed intestinal contents in the patient's stomach, distended stomach, and a large perforation defect in the fundus. The diameter of the proximal segment of the duodenum was increased up to 16 cm; the duodenum was compressed within the aortomesenteric angle area. The surgical interventions involved gastrectomy and esophagojejunostomy. Thirty minutes after the surgery was completed, the child had cardiac arrest; resuscitation attempts failed. The time between the first complaints and death pronouncement was 6 hrs. The authors of the publication pointed out the late development of clinical symptoms of severe intestinal obstruction (only on day 40) and the fact that the plaster corset did not allow patient's relatives to timely detect symptoms of the disease affecting the anterior abdominal wall (e.g., distended abdomen). The authors are absolutely right in their opinion that an important cause factor in this dramatic situation was that the child was mentally ill, so the rapport with him was poor.

Having analyzed medical publications devoted to the problem of arteriomesenteric duodenal compression in adolescents operated on for scoliosis, the following conclusion can be drawn: body weight index below 18 should be considered the essential risk factor for the development of this pathology [13, 15, 19, 20, 23, 27–30].

Furthermore, Braun et al. [21] reported that the baseline angle of the major curvature was larger on average by 12° in adolescents who developed this complication; the curvature type in the lumbar spine corresponded to lumbar modifiers B and C according to the classification proposed by Lenke et al. [16] Smaller (by 11 %) degree of scoliosis correction was attained in these patients. These spine characteristics can be regarded as the potential reasons for the onset of arteriomesenteric duodenal compression syndrome in patients operated on for scoliosis.

Conclusions

Spine surgeons are well familiar with common errors and complications in surgical correction of scoliotic deformities of the spine in children and adolescents. The most frequent types of these complications are the vertebrogenic ones: screw malposition, surgical site infection, disruption of the integrity of metal instrumentation, and long-term deformity progression. Other complications, including visceral

complications affecting the abdominal organs are much less frequent; even the modern medical databases contain only sporadic reports on them.

The reported clinical case of arteriomesenteric duodenal compression as a complication of surgical correction of typical idiopathic scoliosis is an additional method for informing the concerned specialists of the risk of extremely severe and potentially life-threatening visceral complications.

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Address correspondence to:

Ryabykh Sergey Olegovich Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, 6 Marii Ulyanovoy str., Kurgan, 640014, Russia, rso @mail.ru Received 04.10.2019 Review completed 31.10.2019 Passed for printing 06.11.2019

Sergey Olegovich Ryabykh, DMSc, Head of the Clinic of Spine Pathology and Rare Diseases, Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, AOSpine RF Education Officer Ortho, 6 Marii Ulyanovoy str., Kurgan, 640014, Russia, ORCID: 0000-0002-8293-0521, rso_@mail.ru;

Evgeny Gennadievich Skryabin, DMSc, Professor of the Department of Traumatology and Orthopedics, Tyumen State Medical University, 54 Odesskaya str., Tyumen, 625023, Russia, ORCID: 0000-0002-4128-6127, skryabineg@mail.ru;

Valery Petrovich Chevzbik, Head of the Children's Surgery Department No. 2 of the State Unitary Enterprise "OKB No. 2", 75 Melnikaite str., Tyumen, 625039, Russia, ORCID: 0000-0001-7426-9019, val615@yandex.ru;

Egor Yuryevich Filatov, MD, PhD, orthopedic trauma surgeon, junior researcher, Clinic of Spine Pathology and Rare Diseases, Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, 6 M. Ulyanovoy str., Kurgan, 640014, Russia, ORCID: 0000-0002-3390-807X, filatov@ro.ru.

