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ORIGINAL ARTICLE

Multilevel spinal reconstruction in pediatric patients under 4 years old with non-congenital pathology (10-year single-center cohort study)

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Abstract

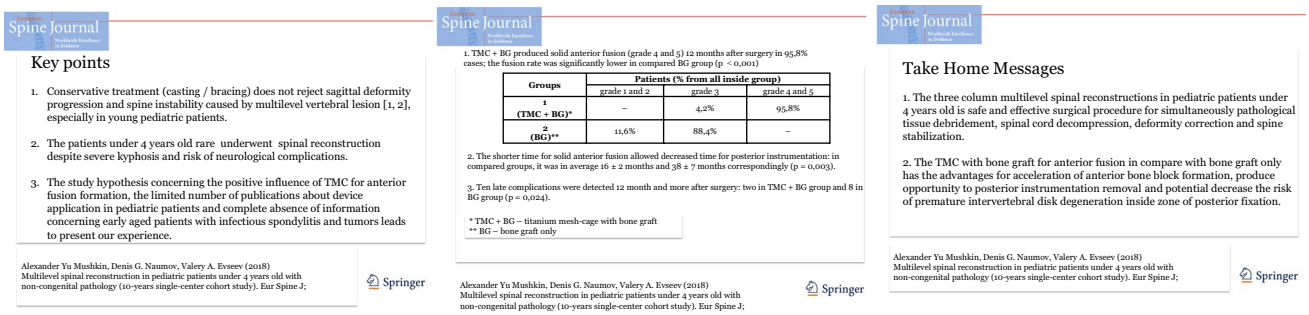
Purpose To evaluate the influence of anterior fusion option on the short- and long-time outcomes on multilevel spinal reconstructions in young children.

Methods Forty-five patients aged under 4 years old (2 years 2 months ± 11 months in average) underwent spinal reconstruction due to tuberculosis spondylitis (35), pyogenic spondylitis (9) and spinal tumors (1) complicated by angular kyphosis exceeded 20° (49.2° ± 14.3° in average). All lesions involved two or more spinal motion segments. Clinical and radiographic data were compared in two groups depended on the types of anterior fusion: titanium mesh cage with bone graft (TMC + BG) (19 patients) and cortical BG only (26). The average follow-up was 5 years 10 months ± 2 years 8 months (min = 3 years; max = 12 years).

Results The deformity correction was similar in groups. The operation time and blood loss were less in TMC + BG group ($p = 0.001$) as a times for anterior bone block formation ($p < 0.001$) and posterior instrumentation removal ($p = 0.003$). Ten late post-op complications registered include disease's recurrence (1), pseudoarthrosis (6), deformity progression (1) and graft resorption (1). The complication rate was less in TMC + BG than in BG group: two and eight cases consequently, $p = 0.024$.

Conclusions Multilevel spinal reconstruction in early aged patients is safe and effective procedure. The anterior fusion by TMC with bone autograft has advantages of reducing blood loss, operation time, time for anterior block formation and complications rate compared with bone autograft only.

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.



Keywords Pediatric spine · Spondylitis · Spinal surgery · Titanium mesh cage

Introduction

Intensive growth and active vascular support promote high risk of vertebral destructions and deformity progression in early aged patients with infectious (pyogenic and granulomatosis) spondylitis and spine tumors [1]. The neurological

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Extended author information available on the last page of the article

complications are rare in these patients due to relatively large size of spinal canal [2–5]. The main risk factors for severe kyphosis' progression include (1) the age below 10 years, (2) multilevel/transitional spine zone involvement and (3) local kyphosis exceeding 20° [6]. The drug therapy is a base for these diseases but casting (bracing) does not reject sagittal deformity progression and spine instability caused by multilevel vertebral lesion [7, 8].

The key to successful outcome is to provide balanced spinal growth in such patients as much as possible. Management priority goes to procedure that can reconstruct three spinal columns. The decision-making process concerning the early aged patients could be difficult: On the one hand, vertebral column resection (VCR) offers the greatest correction, and on the other hand, this procedure increases blood loss, risk of neurological complication and leads to spine shortening [9]. Combination of anterior fusion by bone graft and posterior fixation provides deformity correction without multilevel vertebral reduction, but a limitation of the method is a long course of posterior multilevel instrumentation resulting in premature intervertebral disk degeneration inside fusion zone [10].

The titanium mesh cages (TMC) have a wide applications in the adult spine surgery: This device helps to decrease time of anterior bone block formation [11, 12]. The hypothesis concerning the positive influence of TMC for anterior fusion formation, the limited number of publications about this device application in pediatric patients and complete absence of information concerning early aged patients with infectious spondylitis and tumors lead to present our experience.

Objective

To explore influence of anterior fusion material (TMC with bone graft or bone graft only) over spine reconstruction outcomes in early aged patients.

Study design

This is a 10-year single-center retrospective cohort study.

Patients and methods

Forty-five of the 523 (8.8%) pediatric patients who underwent primary spinal surgery in our clinic between January 2005 and December 2014 were selected in accordance with inclusion criteria:

- patients' age under 3 years 11 months 30 days at the moment of primary surgery;
- the initial angular local kyphosis exceeding 20° (acc. Cobb's angle) caused by vertebral body(-es) destructive lesion;
- multilevel (two and more vertebral motion segments) anterior spine reconstruction;
- the morphological and/or bacteriological diagnosis' confirmation. The material was harvested from lesion site; the bacteriology study includes cultural isolates and molecular genetics tests (polymerase chain reaction, PCR);
- the pre-admission study including skin tests (TST), bacteriological blood tests, IGRA tests and a closed transcutaneous or thoracoscopic biopsy of affected areas followed by at least 2-month anti-bacterial chemotherapy before surgery (for infectious spondylitis) corresponding to the therapy regimen or drug resistance tests including national recommendations based on WHO guidelines, 2010 [13];
- follow-up post-op period exceeding 36 months;

Tuberculosis spondylitis (TB sp.) was verified in 35 cases; pyogenic spondylitis, in nine; and tumor, in one.

The average age was 2 years 2 months ± 11 months (min 8 months, max 3 years 11 months) at the time of surgery. The average follow-up period was 5 years 10 months ± 2 years 8 months (min 3 years; max 12 years).

The operation technique included pathological tissue debridement, anterior spinal cord decompression, deformity correction, anterior fusion and posterior instrumentation. Forty-two from 45 cases of "two stages one-narcosis surgery" were applied with anterior reconstruction followed by posterior instrumentation. Anterior and posterior stages were divided by 2 weeks in one case (Table 1, #13) due to initial severe pathology and high anesthesiology risk (the patient previously underwent neonatal sepsis with spinal involvement). The posterior instrumentation was not applied in two cases (Table 1, ##10, 18) due to complete deformity correction and spinal stability reached from anterior approach only.

An anterior approach for anterior spine is always used for cervical lesions; the lateral–posterior trans-costal approach without *processus transversus* resection was preferable for thoracic spine; lateral retroperitoneal, for lumbar. The anterior spinal cord decompression as a stage of anterior procedure applied in all cases as *with* (Table 1) as *without* neurological disorders. The additional advantage from anterior release was the possibility to elongate the anterior spinal column during anterior fusion procedure.

The posterior instrumentation in all cases was associated with posterior fusion (PF) by bone autograft. The extension of PF was the same as extension of anterior reconstruction

Table 1 Patients included into study

Patient no.	Age (years + months)	Sex	Disease	Level	Surgical approach	Posterior instrumentation (CDI)	Blood loss (ml/% of CBV)	Operation time (min)	Frankel scale pre-/post-op
Group 1									
1	1 + 3	m	TB sp.	Th2-4	LT	Th1-8	150/19.4	225	E
2	2 + 10	m	TB sp.	Th3-5	LT	Th2-6	120/10.1	150	E
3	2 + 11	f	TB sp.	Th5-7	LT	Th3-9	150/14.6	175	E
4	1 + 3	m	TB sp.	Th6-8	LT	Th5-9	150/19.2	150	E
5	2	f	TB sp.	Th6-8	LT	Th6-8	160/17.6	240	E
6	3 + 1	f	TB sp.	Th7-11	LT	Th5-L1	120/11.5	205	E
7	2 + 9	m	TB sp.	Th9-11	LT	Th6-L1	120/11.9	165	E
8	3 + 2	m	TB sp.	Th9-11	LT	Th8-11	200/19.1	240	E
9	2 + 10	f	TB sp.	Th10-12	LT	Th8-L1	180/17.6	230	E
10	3 + 1	m	TB sp.	Th11-L1	LT	no	200/19.1	180	E
11	3	f	TB sp.	Th11-L1	LT	Th10-L2	135/12.9	180	E
12	3 + 9	f	TB sp.	L 3-5	LR	L2-S1	115/12.8	250	E
13	2	m	Pyogenic spondylitis	Th4-7	LT	Th3-8	115/12.6	210	E
14	1 + 2	m	Pyogenic spondylitis (*S. aureus*[a])	Th4-9	LT	Th3-10	110/14.5	215	E
15	0 + 8	f	Pyogenic spondylitis	Th6-9	LT	Th5-10	110/18.1	215	E
16	0 + 8	f	Pyogenic spondylitis	Th8-10	LT	Th5-L1	80/13.1	200	E
17	1 + 6	f	Pyogenic spondylitis	L1-3	LR	Th12-L3	50/6.1	200	E
18	1 + 5	m	Pyogenic spondylitis (*S. aureus*)	L3-5	LR	no	60/7.4	90	E
19	3	f	Retroperitoneal ganglio-neuroblastoma	Th12-L2	LT	Th11-L3	250/23.9	240	D/E
Group 2									
20	1 + 8	f	TB sp.	C3-4	A	C2-5	150/17.3	235	E
21	0 + 11	m	TB sp.	C2-5	A	C2-7	70/10.1	135	E
22	3 + 6	m	TB sp.	Th2-7	LT	C3-Th9	270/24.5	315	C/E
23	1 + 3	m	TB sp.	Th3-4	LT	Th2-6	150/22.9	250	B/C
24	2 + 1	f	TB sp.	Th3-5	LT	Th1-8	300/35.7	290	E
25	1 + 6	f	TB sp.	Th3-5	LT	Th2-6	150/18.1	210	D/E
26	2 + 9	m	TB sp.	Th3-5	LT	Th1-6	200/27.2	220	D/E
27	0 + 11	f	TB sp.	Th3-8	LT	Th2-9	190/27.5	220	E
28	2 + 5	m	TB sp.	Th4-7	LT	Th1-9	250/20.6	300	E
29	1 + 6	f	TB sp.	Th5-7	LT	Th2-9	220/26.6	225	E
30	0 + 8	f	TB sp.	Th6-9	LT	Th5-10	200/35.5	240	E
31	3 + 10	f	TB sp.	Th7-10	LT	Th4-11	200/19.6	230	E
32	1 + 5	m	TB sp.	Th7-10	LT	Th3-L1	200/26.1	220	E
33	2 + 1	f	TB sp.	Th8-10	LT	Th5-L2	200/32.9	160	E
34	1 + 11	f	TB sp.	Th10-L1	LT	Th10 – L2	210/22.4	213	D/E
35	3 + 6	m	TB sp.	Th11-L1	LT	Th10-L2	190/15.8	195	E
36	2 + 2	m	TB sp.	Th12-L1	LT	Th8-L3	195/22.4	225	E
37	3 + 1	m	TB sp.	L1-3	LR	Th11-L3	200/20.3	240	E
38	1 + 10	f	TB sp.	L1-3	LR	Th12-L3	215/23.3	240	E
39	3 + 10	m	TB sp.	L3-5	LR	L3-5	120/10.7	90	E
40	3	f	TB sp.	L3-5	LR	L2-S1	230/22.1	135	E
41	2 + 3	f	TB sp.	L3-6	LR	L1-6	200/21.3	335	E
42	3 + 1	m	TB sp.	L5-S1	LR	L4-S1	400/37.8	275	E
43	0 + 11	m	Pyogenic spondylitis (*S. aureus*)	C3-4	A	C2-5	75/12.3	170	D/E
44	1 + 5	m	Pyogenic spondylitis	Th11-12	LR	Th11-L1	217/26.7	276	E
45	3 + 4	m	Pyogenic spondylitis	L1-2	LR	Th11-L3	195/18.1	215	E

CBV circulated blood volume, *LT* lateral transthoracic approach, *LR* lateral retroperitoneal approach, *TB sp*. tuberculosis spondylitis, *A* anterior approach

[a]Result of bacteriological test at the time of surgery

to decrease the asymmetric growth of anterior and posterior spinal column.

The active vacuum drainage continued 1 or 2 days; it is removed if the content of drainage became < 20 ml per day.

Two study groups formed according to material for anterior fusion:

group 1 ($n = 19$)—TMC and bone graft ("TMC + BG");
group 2 ($n = 26$)—bone graft (BG) only.

The result of operation was registered. All patients were observed up routinely with X-ray and computed tomography (CT) at 6 and 12 months post-op; followed radiological examination performed once a year and at the time of posterior instrumentation removal. Seven patients with neurological disorders had obligatory MRI before and 1 year after surgery.

The patient's data are in Table 1.

Figures 1 and 2 illustrate the results of surgeries in patients operated at ages 8 month (cases #16) and 2 years 1 month (case #34).

The study design is in Fig. 3.

Next, clinical and radiological parameters were evaluated:

1. operation time and blood loss
2. kyphosis correction (degrees acc. to pre- and post-op Cobb angle);
3. dynamic of anterior fusion formation (according to five-grade scale, Table 2) [14]*;
4. time of posterior instrumentation removal (months after initial surgery);
5. complications rate.

Statistical analysis

Statistical analysis was performed using Statistical Package for the Social Sciences (SPSS), version 22.0 (SPSS Inc., Chicago, IL, USA). All evaluation parameters had normal distribution. Independent t test was used for contrasting means in two study groups: time of anterior fusion formation; complications rate; loss of deformity correction; operation time; and blood loss. A p value < 0.05 was considered statistically significant.

Results

The next two parameters were significantly less in TMC + BG group in comparison with BG group:

The operation time was 3 h 2 min ± 44 min versus 3 h 58 min ± 49 min, respectively ($p = 0.001$);
The absolute blood loss volume was 129 ± 53 ml versus 204 ± 72 ml and in percent of CBV—16.4 ± 6.2% versus 26.1 ± 9.2%, respectively ($p = 0.001$).

It was not statistically significant difference ($p = 0.539$) in deformity correction between groups, but the loss of correction was significantly higher in BG group at the end of follow-up period (Table 3).

TMC + BG produced solid anterior fusion (grades 4 and 5) in 95.8% cases 12 months after surgery; the fusion rate was significantly lower in BG group (Table 4).

The shorter time for solid anterior fusion allowed decreased time for posterior instrumentation: In compared groups, it was in average 16 ± 2 months and 38 ± 7 months

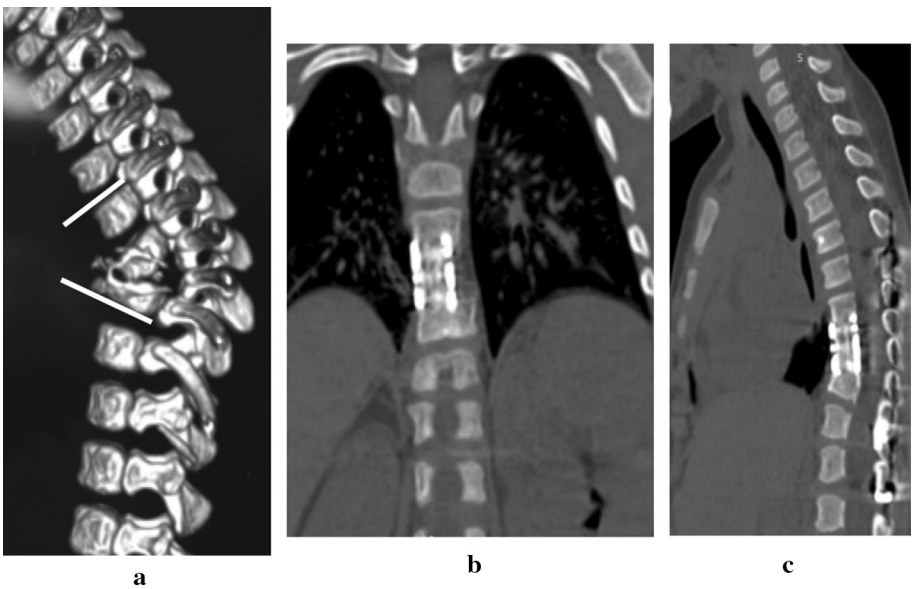

Fig. 1 Patient no. 16 (acc. Table 1). Pre-op 3D–CT (**a**) and post-CT (**b**, **c**): 1 year, 4 months after surgery

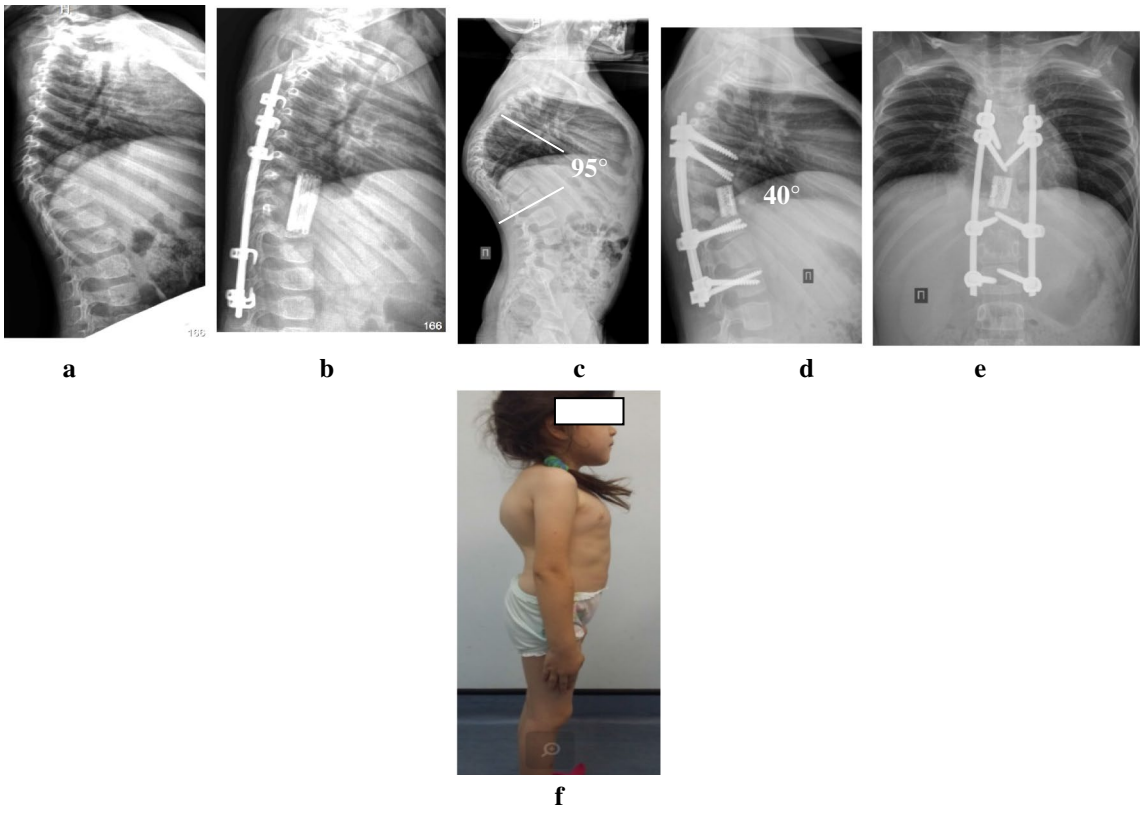

Fig. 2 Patient no. 34 (acc. Table 1). Lateral X-ray before (**a**) and after (**b**) surgery at the age of 2 years, 1 month; X-ray (**c**) and photograph (**f**) 4 year, 6 months after surgery (complete bone graft resorption; the posterior instrumentation was removed 1 year before not in our clinic; the reason is unclear); **d**, **e**—6 month after revision surgery

correspondingly ($p = 0.003$). The indication for posterior instrumentation removal was solid anterior and posterior fusion confirmed by CT.

There were not complications in early post-op period before 30 days after surgery. Ten late complications were detected 12 months and more after surgery: two in TMC + BG group and eight in BG group ($p = 0.024$). The average time for complications appearance was 3 years 3 months (min 12 months; max 9 years) after surgery, and only three of them were explained by primary pathology recurrence (Table 5).

Revision surgery was performed for all patients with complications and included TMC + BG anterior fusion and posterior fixation. There were not any complications at final follow-up.

Discussion

Surgical management of spinal destructions caused by infections and tumors complicated by severe kyphosis in early aged patients is one of the most challenging, difficult and insufficiently studied areas in pediatric spine surgery. On the one hand, it is connected with a small number of diseases where spinal reconstruction is essential; on the other hand, there are limited numbers of clinics specialized on the given pathology. This makes difficult correct comparison of presented results and experience of other researchers.

According to three medical databases (PubMed, ClinicalKey and Medline), only ten articles described treatment principles for spine destruction in early aged pediatric patients and majority of them concern infectious (TB or pyogenic) spondylitis [15–17]. Rajasekaran [6] formulated the main indications for spinal TB surgery in pediatric patients: progressive neuro-deficit, persisting pain due to instability and severe deformity. The author demonstrates good clinical outcomes of three-column spine reconstruction. The most numerous series by He et al. [18] included 54 TB spondylitis' children with average age 9 years 2 months. The authors have limited follow-up parameters by dynamic of neurological disorders and postoperative kyphosis correction and focused on the high level of postoperative complications (31.5%; 17 from 54 patients) most often related to apical deformity recurrence and PJK (proximal adjacent kyphosis)—six and eight cases from 17, respectively. Unfortunately, the follow-up period does not exceed 4 years.

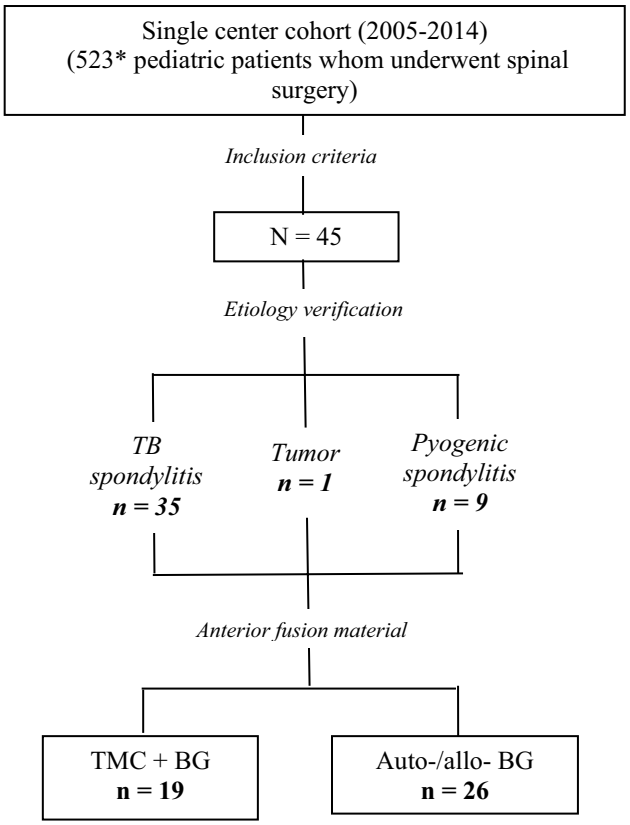

Fig. 3 Scheme of the study. (Asterisks) The clinic historically specializes in the surgical treatment of destructive bone lesions in pediatric patients

Schulitz et al. [19] analyzed growth changes of anterior bone fusion in pediatric patients with tuberculosis; they concluded negative effect of bone grafts including restricted anterior growth and limited the capacity for spine remodeling more than 30%. After that, the bone graft alone intensified loss of deformity correction. Other articles have a small number of patients and do not have opportunities for comparing results [20, 21].

It is difficult to compare our data with other types of surgery in early aged pediatric patients: As a rule, they concern the congenital abnormalities, but it is important to indicate the high rate of blood loss and long operation time for VCR [22]. The additional advantages from the procedure, which we used in our patient, in comparison with VCR technique from posterior approach only, are the possibilities…

(a) to elongate the anterior spinal column before anterior fusion;
(b) to minimize the shortening of posterior column during procedure and the risk of significant spinal shortening in follow-up;
(c) to exclude the necessity to scarify the neural root.

It is understandable that the best way to prevent severe kyphosis in early aged patients is early diagnosis and targeted anti-bacterial treatment for primary infection. As to concern our series, the kyphosis was the first manifestation of spinal lesion in all cases. It could be of interest that in our country the pediatric patients with TB spondylitis and complications of infectious spondylitis (with spinal deformities

Table 2 Five-grade scale for anterior fusion (bone block) estimation [acc. 14]

Grade	Graft position		Graft–vertebrae contact zone			
	No dislocation	Dislocation	Distance between graft and vertebrae (mm)		Incorporation of the graft into the vertebrae without any distance	
			≥ 3 mm	< 3 mm	Unstructured (unclear bone trabeculars)	Clear bone structure
1		+				
2	+		+			
3	+			+		
4	+				+	
5	+					+

Table 3 Changes in kyphosis deformity

Group	Pre-op (degree)			Post-op (degree)			Correction		Loss of correction*
	Min	Max	M ± m	Min	Max	M ± m	Abs.	%	abs.
1	23.0	80.0	50.1 ± 16.5	10.0	44.0	21.7 ± 6.7	28.4 ± 9.8	43.3	3.7 ± 1.4
2	34.0	62.0	48.0 ± 12.1	14.0	37.0	22.4 ± 6.9	25.6 ± 5.2	46.6	15.4 ± 3.2

*$p = 0.039$

Table 4 Anterior fusion rate estimated 12 months after surgery

Groups	Patients (% from all inside group)		
	Grades 1 and 2	Grade 3 (%)	Grades 4 and 5
TMC + BG	–	4.2	95.8%
BG	11.6%	88.4	–

$p < 0.001$ for all grades

and/or neurological disorders) were traditionally concentrated for surgical stage into our clinic only—that is why we had the definite experience in this field.

Conclusion

The multilevel spinal reconstructions are rare in early aged patients. The indications for such surgery include vertebral body(-ies) lesions in spondylitis and tumors which are complicated by kyphosis. This deformity progresses in the natural history and could be complicated as by neurological signs as by severe cosmetic defect. This operation is not only effective for deformity correction in early aged patients. It is safe procedure despite simultaneous pathological tissue debridement, anterior spinal cord decompression, anterior fusion and posterior instrumentation with fusion.

The anterior fusion by TMC with bone graft has the advantages in comparison with bone graft only: The acceleration of anterior bone fusion formation produces opportunity to posterior instrumentation removal, and it could potentially decrease risk of premature intervertebral disk degeneration inside zone of posterior fixation.

Limitations

The authors understand that the study has a few limits:

The diagnosis was not confirmed by biological methods in some cases of spondylitis (especially pyogenic, Table 1). It could be explained by prescribed preoperative anti-bacterial drugs: It is effective for bacterial elimination but could not prevent orthopedic complications;
the retrospective study design has made impossible estimation the direct surgery effect on body alignment and total sagittal profile;
the follow-up period did not exceed 12 years (maximal) in our cohort. Most patient have not finished the growth

Table 5 Late postoperative complications

No.[a]	Gender	Age	Diagnosis	Initial surgery	Type and time of complications diagnosis (yrs + months)	Revision surgery	Follow-up after revision surgery
9	f	2 + 10	TB sp. Th10-12	AF Th10-12 TMC + BG, PI Th8-L1 (06.2013)	*Pseudoarthrosis* 1 + 0	PI Th8-L1	1 + 11
12	f	3 + 9	TB sp. L3-5	AF L3-5 TMC + BG, PI L2-S1 (12.2014)	*Pseudoarthrosis* 1 + 10	AF L3-5 TMC + BG, PI L2-S1	1
30	f	0 + 8	TB sp. Th6-9	AF Th6-9 BG, PI Th5-10 (02.2014)	*Deformity progression* 1 + 5	AF Th6-9 TMC + BG, PI Th5-10	2 + 5
33	f	2 + 1	TB sp. Th8-10	AF Th7-12 BG, PI Th5-L2, 04.2012	*Bone graft resorption* 5 + 6	AF Th7-12 TMC + BG, PI Th5-L2	1 + 1
37	m	3 + 1	TB sp. L1-3	AF L1-3 BG, PI Th11-L3 (04.2011)	*Pseudoarthrosis* 1 + 0	AF L1-4 TMC + BG, PI Th11-L3	5 + 8
39	m	3 + 10	TB sp. L3-5	AF L3-5 BG, PI L2-5 (07.2011)	*Disease recurrence* 1 + 4	AF L3-5 TMC + BG, PI L2-5	5 + 1
40	f	3	TB spondylitis L3-5	AF L3-5 BG, PI L2-S1 (12.2014)	*Pseudoarthrosis* 1 + 1	AF L3-5 TMC + BG, PI L2-S1	1 + 11
41	f	2 + 3	TB sp. L3, L5-6	AF L3-6 BG, PI L1-6 (02.2009)	*Disease recurrence* 3 + 3	Fistulotomy, AF L3-6 TMC + BG	5 + 7
42	m	3 + 1	TB sp. L5-S1	AF L4-S1 BG (11.2006)	*Pseudoarthrosis* 9 + 0	AF L4-S1 TMC + BG, PI L3-S1	2 + 1
44	m	1 + 5	Pyogenic spondylitis Th11-12	AF Th11-12 BG, PI Th11-L1 (11.2009)	*Pseudoarthrosis* 5 + 5	AF Th11-12 TMC + BG, PI Th11-L1	2 + 8

No. patient's number according to Table 1, *AF* anterior fusion, *TMC + BG* TMC with bone graft, *PI* posterior instrumentation, *BG* bone graft

and delayed problem could be realized later—the most disturbing will the pubertal period.

That is why the current data could be presented as a stage of continued study.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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