Correlation between Age and Surgical Approach for Thoracic and Lumbar Hemivertebra

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Abstract

Objective Hemivertebra (HV) is a congenital defect of the formation of the spinal vertebra, which can result in scoliosis or kyphosis along with the related symptomatology of spine deformity. More often than not, it is linked to other abnormalities and requires attention. Its management is surgical and it is of great importance for the physician to choose the right approach at the right time, due to its deteriorative prognosis.

Methods Due to the interest of the subject, the authors investigated the world literature between 1990 and 2018 and found 45 articles, reporting thoracic, thoracolumbar, and lumbar HV in children and its postsurgical outcome, aiming to show whether the approaches are equal in terms of the final outcome.

Results The chosen surgical method depends much on the level of the pathology. Despite this fact, after analyzing the included data, we found that the surgical techniques are unequal with regard to the purpose of achieving improvement. Age, caudal and cranial curves, segmental kyphosis, and scoliosis are factors playing a major role in this.

Conclusion If not treated, HV leads to deterioration and dysfunction. The most optimal result, however, is achieved only when the surgical approach is applied according to age and rest of the accompanying factors, which should be considered in future management planning.

Keywords
- hemivertebra
- quality of life
- spine surgery

Introduction

Embryologic period is essential for the correct growth of tissues and organs. During this period, many errors and abnormalities can occur which will accompany the human during its life. Such a congenital condition is hemivertebra (HV), where only half of the vertebral body is developed. Other expressions for hemivertebra are "congenital scoliosis," "unilateral aplasia of the vertebral body," and "complete unilateral failure of formation of the vertebral body."[1]

Its first mention in medicine was not long ago by Winter et al[2] and their classification system on congenital scoliosis (CS). Current statistics evaluate the incidence of the hemivertebra to be around 0.5 to 1/1000 births.[3,4]

Usually, around the sixth week of gestation, independent centers of chondrification are developing the vertebrae. [5] A few weeks later, these centers merge to provide ossification centers for the development of the notochord remnant. Lack of these centers lead to the development of lateral hemivertebra, while the posterior hemivertebra is caused by a lack of ossification.[6,7] Currently, according to the disk space between the bony structures, there are four known types of hemivertebra.[8]

The HV acts as a triangular-shaped construction within the spine, causing contralateral divergence of the spine.[9] More often than not, HV is associated with other deformities, disorders, and imbalances.

Many reports describe this abnormality and conclude that surgery is a vital procedure for the therapy of the HV.

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Available approaches for HV include anterior and posterior techniques with or without fusion.[8]

Hemivertebra is a condition that piqued our interest to a high degree, because of its symptomatology and capability of treatment with the proper technique. The most commonly used are anterior and posterior approaches; however, no other study reports whether there is a connection between the age, outcome, and technique, which represents the aim of this study.

Methods

For the study, all data published by MEDLINE (Cochrane, Wiley) and EMBASE on the subject of HV in the thoracic, lumbar, and sacral spine between 1990 and 2018 were collected to examine the role of the approach in the final result. The used MeSH terms for the search are as follows: "spine surgery," "hemivertebra," "congenital disorders," "scoliosis."

The inclusion criteria were the following: articles in English and German; type of articles–case report and clinical study; articles with sufficient information on the segment and approach, demographic data, as well as the postoperative outcome; and articles reporting the main curve postoperatively. Case reports are included because of the rare incidence of the condition and the fact that the outcome, either way, represents the average score.

On the other hand, from the review are excluded all articles: written in other than English and German language; letters to editor and comments; reviews; articles with insufficient information on the subject; studies on animals; articles reporting cervical HV; articles reporting fetal or embryological HV; articles reporting infants (younger than 1 year) or older than 18 years; articles reporting more than one approach; articles with no demographic data (age and sex) or postoperative outcome (►**Fig. 1**).

After filtering the data, we divided the included reports into two main groups: group A–age under 9 years and group B–age equal or over 9 years. This age is considered as middle point or median value (18/2 = 9) and was chosen without any other additional settings. The included articles are further analyzed with *t*-test, linear regression analysis, and correlation matrix on statistical program, based on the used approach–posterior versus anterior + posterior.

Improvement in this analysis is based on the outcome. No complications and reoperations are evaluated as 100% improvement. Any complications and reinterventions are evaluated as 75% improvement, stable conditions as 50%, and any deterioration or worsening as 0%.

Other factors such as main curve, segmental kyphosis/scoliosis, and compensatory cranial/caudal curve are measured from the latest follow-ups.

The probability of bias of the current study is very small, if present, since the results and assumptions are based only on statistics. The latter are result of analysis of already published data, all of which lack reports on bias.

Results

In the study are included 45 articles with a total number of 971 patients (►**Tables 1** and **2**). Mean age of all the participants is 7.16 years with sufficient male dominance.

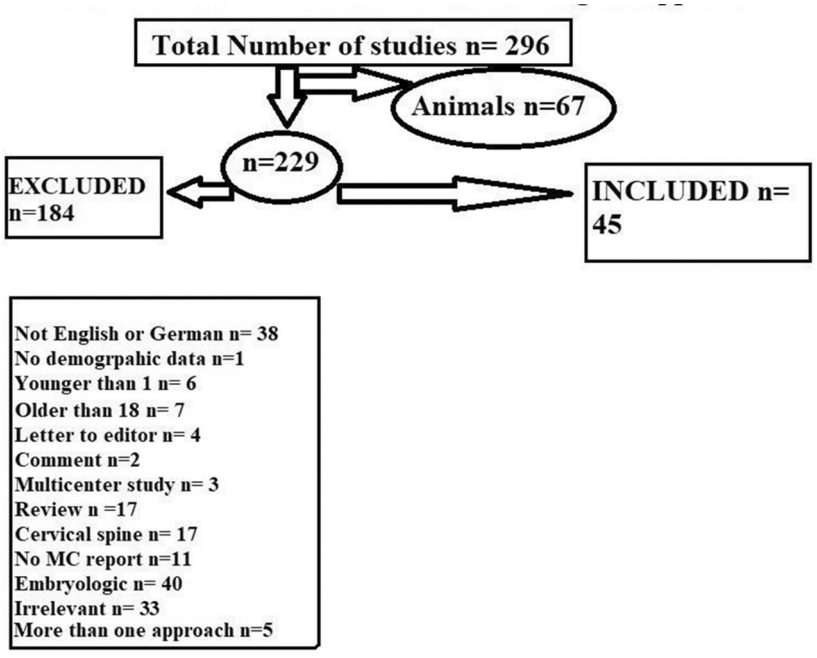

Fig. 1 Flow chart—Hemivertebra and surgical approach..

Table 1 List of studies reporting posterior approach

Study	Age	Number of patients	Improvement rate
Li et al[10]	9.4	24	100
Hardequist et al[11]	4.2	10	100
Ruf et al[12]	3.5	33	91.67
Ruf et al[13]	3.5	28	83.93
Shono et al[14]	5.7	12	97.92
Ruf et al[15]	3.2	25	94
Ruf et al[16]	5.8	20	92.5
Nakamura et al[17]	10	5	100
Zhang et al[18]	9.9	56	97.77
Li et al[19]	17	12	100
Peng et al[20]	3.3	10	100
Erturer et al[21]	9.2	9	100
Lyu et al[22]	13.2	17	94.18
Basu et al[23]	11.2	22	97.73
Guo et al[24]	5.4	39	96.79
Feng et al[25]	5.8	19	90.79
Chang et al[26]	11.3	45	91.11
Yang et al[27]	11.4	9	83.33
Guo et al[28]	9.8	116	96.77
Chang et al[29]	2	1	75
Chang et al[30]	6.6	20	96.25
Zhuang et al[31]	10	14	98.21
Chu et al[32]	10.8	17	100
Zhu et al[33]	7	60	97.08
Crostelli et al[34]	5.5	15	96.67
Kose et al[35]	5	12	91.66
Crostelli et al[36]	7.5	30	99.17
Jeszenszky et al[37]	2	1	100
Huang et al[38]	11.9	15	86.67
Chen et al[39]	9.5	18	98.61
Wang et al[40]	6.5	37	100
Basu et al[41]	9.2	20	96.25

T-test was performed on the four groups: posterior approach in younger than 9; posterior approach in older than 9; posterior + anterior in younger than 9; and posterior + anterior in older than 9. The results of the *t*-tests reveal that the techniques are unequal between the groups (►**Tables 3** and **4**).

Furthermore, in the group A for anterior + posterior approach, there is a negative correlation between main curve and improvement rate ($R^2 = 0{,}288$) (►**Table 5**). More detailed regression analysis reveals the correlation between age and compensatory caudal curve, compensatory cranial curve, segmental kyphosis and segmental scoliosis for the four groups (►**Table 6**). Based on the approach and the

Table 2 List of studies reporting posterior and anterior approach

Study	Age	Number of patients	Improvement rate
Bollini et al[42]	3.3	21	96.43
Garrido et al[43]	2.8	31	97.58
Klemme et al[44]	1,7	6	100
Hadequist et al[45]	3.1	18	98.61
Ginsburg et al[46]	10.5	96	100
Cheung et al[47]	3,4	7	91.67
Bradford et al[48]	4.14	34	92.86
Xu et al[49]	12,1	34	100
Holte et al[50]	12	37	89.19
Lazar et al[51]	1.5	11	93.18
Winter et al[52]	12	1	100
Chang et al[53]	4	1	75
Wang et al[40]	5.4	18	100

Table 3 Paired t-test same approach different age

Paired samples T-Test			
	t	df	*p*-Value
Posterior	969.53	1	< 0.001[a]
A + P	− 46.00	1	0.014[a]

Abbreviation: A+P: anterior + posterior.
[a]Significant value.

Table 4 Paired *t*-test same age, different approach

Paired samples T-Test				
		t	df	*p*-Value
A + P under 9 y	Posterior under 9 y of age	3259.0	1	< 0.001[a]
Posterior over 9 y	A+P over 9 y of age	124.8	1	0.005[a]

Abbreviation, A + P: anterior + posterior approach.
[a]Significant value.

Table 5 Correlation between age, improvement rate, and main curve for group A

		Age	Improvement
Improvement	Pearson's r	− 0.170	—
	p-value	0.662	—
Main curve	Pearson's r	− 0.129	− 0.537
	p-value	0.741	0.136

age group, the correlations between the factors vary, but surely the age plays a very important role. The improvement, as lack of complications and reintervention, seems to be also connected to the latter (►**Table 7**). According to the analysis, every approach should be considered based

Table 6 Correlations between age, segmental kyphosis/scoliosis, and compensatory cranial/caudal curve

Approach	Age	Segmental kyphosis R^2	Segmental scoliosis R^2	Compensatory cranial curve R^2	Compensatory caudal curve R^2
Posterior only	Age < 9	0.2434	− 0.0538	− 0.2434	− 0.1667
	Age > 9	−0.0346	− 0.7562	0.3407	0.0882
Posterior + anterior	Age > 9	− 1	–	− 0.957	− 0.2844
	Age < 9	0.2601	–	–	1

Table 7 Correlations between improvement, segmental kyphosis/scoliosis, and compensatory cranial/caudal curve

Approach	Improvement	Segmental kyphosis R^2	Segmental scoliosis R^2	Compensatory cranial curve R^2	Compensatory caudal curve R^2
Posterior	< 9	0.2001	0.4834	0.1045	0.0116
	> 9	− 0.4219	0.3367	0.451	0.3128
Anterior+ posterior	< 9	1	–	0.9158	− 0.0035
	> 9	− 0.9955	–	–	–

on the age of the patients to achieve better results. A statement is based on the fact that in the anterior + posterior approach, there is a strong link between age/improvement and segmental kyphosis (follow-up results), while in the posterior only approach, there is a solid link between age/improvement and segmental scoliosis (follow-up results).

Although the approaches seem to be identical, they appear to be unequal with regard to improvement, and every technique according to the concrete age group has different correlations and values of the most optimal outcome. They are not the same and the surgery of HV should be personalized in the future.

Discussion

Scientifically proved, congenital abnormalities of the spine are formed in the embryonic period, either as formation or segmentation failure.[54] These abnormalities lead to the formation of congenital scoliosis and the most underlying factor is HV.[55] It has variable frequency from 1.33/10.000 (0.133/1000) to 10/10.000 (1/1000).[1,6,56]

Generally speaking, HV headed to local instability, decreased longitudinal development, as well as enhanced convolution of the spine.[2,57] The progression and its level could not be predicted.[40] However, for sure, there will be asymmetric growth, followed by secondary curvature and endorsed balance, requiring adequate therapy to prevent more severe deformity.[13] The main controversy here is whether to choose posterior or posterior and anterior approach. Theoretically, posterior approach is a method that can fully resolve the problem, but it is more insisting and might cause some neurological problems. Due to this, some physicians chose to proceed with the posterior–anterior approach, even though it requires more operation time, hospital stay and, currently stated complications.[16,58]

HV of the thoracic spine is more common than the HV of the cervical spine. Its symptomatology is mainly pain and myelopathy.[59] The surgical choices are in situ fusion, removal of HV, and epiphysiodesis.[45] The main target of the therapy is to accomplish spine with right direction and fewer difficulties. Due to the sensitivity of the thoracic spine, it is a recommended procedure with less complication risks.[45] But also taken under consideration is the possibility of good outcome, when spinal growth is present.[60,61] Regarding the technique, it could be a method with posterior only approach, posterior and anterior, or as Suzuki et al[62] reports, correction with 4-staged surgical therapy. Finally, total elimination of the HV with cervical pedicle. Based on the fragment of the spine, the outcomes are promising. Regarding the thoracic and thoracolumbar spinal HV. Zhang et al[18] reports improvement after only posterior approach with even lower rate of complication than expected. Better visualization, sense on the compression, and less invasiveness are reached with this approach. Qureshi et al[63] also reports 69% improvement of the kyphosis, 73% improvement of the sagittal, and 76% of the coronal shift after posterior approach for thoracic, lumbar, and thoracolumbar HV. In this relation, Mladenov et al[64] observed 59% improvement after posterior approach and 55% after combined method. It is believed that the posterior approach only for thoracic HV provides stabilization but not correction.[60] To achieve better correction, usually it takes the combined approach.

Lumbar spine is the second most common location to form HV. Its symptomatology seems to be very interesting. It consists of gait distortions, lordosis and bended hips with bowel, bladder abnormality, and back pain.[54] Lumbar stenosis and nerve extension might also be observed. If not treated, it is a condition that leads to functional loss.[54] Due to this, the operation has the aim of relaxing the nerves and providing stability.[54] There are three basic types: fusion, removal, and epiphysiodesis.[40,65] Based on the approaches, there are two choices: posterior and anterior. In some cases, the surgery is performed with anterior approach at the beginning, while in others, initially posterior.[40] Posterior approach is better for children.[66] Anterior approach, on the other hand, is believed to

be suitable for thoracolumbar HV.[65] In many cases, it can be provided in one or two stages. The first stage consists of removal, while the second comprises fusion.[65] One stage surgery is recommended because of the shorter time of operation, while their combination is recommended for bigger resection.[40,65,67] The operation of the lumbar HV leads to improvement of the symptomatology.[54] This is shown in the report of Bollini, who observed 59.9% enhancement for the total curve after surgical management of the lumbar HV after combined posterior-anterior approach.[42] Bradford et al[8] reports 68.1% improvement after HV removal with the same approach. A similar result on combined approach has Lazar et al[51] with 70.2% enrichment. Regarding only the posterior approach, Shono et al[14] reports 63.3% improvement and Nakamura et al[17] 54.3% after one-stage surgery for lumbar HV. After investigation of the published data, it seems that the posterior approach is as safe as the combined for the thoracic and lumbar spine[58,68] Finally, for the lumbar HV, Zhu et al[33] reports 87.3% improvement after posterior approach with fusion, which is encouraging results but this technique is very challenging and could lead to complications.

The outcome of the study presents a new insight in spine surgery for HV. After many years of reporting clinical outcomes, it has become clear that the techniques are unequal with respect to the improvement of the patients with thoracic and lumbar HV. According to the results of the study, the approaches are unequal for the both age groups, suggesting that age is a determining factor for the final results, and should be always considered before proceeding with the chosen technique. Since the pathology is rare, more detailed reports are needed.

In addition to this, if not treated, HV in children will worsen the condition and complicate the status itself and the surgery. It is a deteriorative pathology, leading to functional loss that requires treatment. So, in those cases and especially in dorsal HV, surgical intervention at an early stage is the highly recommended.[12,18,15,55,58,63] As to the approach, it is clear that the patient determines the method. So, choosing the right technique aids in improvement.

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Conflict of Interest
None declared.

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