



ARTICLE

# The prevalence of scoliosis within Belgian myelomeningocele population and the correlation with ambulatory status and neurological comorbidities: a chart audit

Arne Heyns<sup>1</sup> · Stefano Negrini<sup>2,3</sup> · Katrien Jansen<sup>4</sup> · Pierre Moens<sup>5</sup> · Sebastiaan Schelfaut<sup>5</sup> · Koen Peers<sup>1</sup> ·  
Carlotte Kiekens<sup>1,6</sup>

Received: 11 November 2020 / Revised: 22 December 2020 / Accepted: 23 December 2020 / Published online: 25 January 2021  
© The Author(s), under exclusive licence to International Spinal Cord Society 2021

## Abstract

**Study design** Retrospective chart audit.

**Objectives** Firstly determining the prevalence of scoliosis in myelomeningocele (MMC) patients of the University Hospitals Leuven. Secondly analyzing whether there are differences concerning distribution of radiological level, ambulatory status, hydrocephalus, tethered cord, and syringomyelia in MMC patients with/without scoliosis.

**Setting** University Hospitals Leuven, spina bifida convention.

**Methods** The following data were collected: age, gender, radiograph type, age at the time of the radiograph, position during radiograph, presence of fusion, age at the time of fusion, diagnosis of hydrocephalus, tethered cord, or syringomyelia, radiological level of MMC, ambulatory status, main Cobb angle, main curve convexity, and main curve location. Correlation between prevalence of scoliosis and ambulatory status, neurological comorbidities, and radiological level were investigated.

**Results** There were 116 patients remaining, after excluding patients without MMC or useful images. The scoliosis prevalence in MMC patients was 78.4% (95% CI, 71.0–85.8) for Cobb angle  $\geq 10^\circ$ ; 60.3% (95% CI, 51.4–69.2) for  $\geq 20^\circ$ , 52.6% (95% CI, 43.5–61.7) for  $\geq 30^\circ$ , and 36.6% (95% CI, 27.7–45.5) for an angle  $\geq 40^\circ$ . Wheelchair users had 4 to 8 times more chance of having scoliosis than patients able to walk on all surfaces without aid. Thoracolumbar and lumbar radiological levels had a slightly higher prevalence of scoliosis than sacral levels.

**Conclusions** The high prevalence of scoliosis warrants a thorough screening and follow-up for MMC. There was no statistically significant difference between hydrocephalus, tethered cord, or syringomyelia regarding scoliosis. Future studies should focus on the interactions of the neurological comorbidities associated with MMC and scoliosis.

## Introduction

The estimated prevalence of spina bifida is 34.0–48.4 per 100,000 live births worldwide [1] and myelomeningocele (MMC) is considered the most clinically significant subtype of spina bifida [2]. Many medical comorbidities such as paraplegia, hydrocephalus, neurogenic bladder,... are associated with the birth defect [3]. Scoliosis is the second most frequent medical complication in spina bifida patients preceded only by urinary tract infections [4]. The spinal deformation itself can cause other complications, chronic restrictive lung disease with respiratory failure leading to death [5], psychological problems, and decreased participation because of appearance [6]. The fact that scoliosis can have a serious impact on functions, activities, and participation indicates the importance of screening for scoliosis in spina bifida patients to ensure adequate management and follow-up.

✉ Arne Heyns  
arne.heyns@uzleuven.be

<sup>1</sup> Physical and Rehabilitation Medicine, University Hospitals Leuven, Leuven, Belgium  
<sup>2</sup> Department of Biomedical, Surgical and Dental Sciences, University La Statale, Milan, Italy  
<sup>3</sup> IRCCS Fondazione Don Gnocchi, Milan, Italy  
<sup>4</sup> Department of Development and Regeneration, University Hospitals Leuven, Leuven, Belgium  
<sup>5</sup> Orthopaedic Surgery, University Hospitals Leuven, Leuven, Belgium  
<sup>6</sup> Spinal Unit, Montecatone Rehabilitation Institute, Imola, Italy

This group of authors performed a systematic review determining the prevalence of scoliosis in different spina bifida subpopulations [7]. We found four articles concerning MMC (479 patients, 283 females, and 196 males) with an overall weighted prevalence of scoliosis (20° Cobb angle cutoff) of 52.5%. Most studies examined in the systematic review had (methodological) flaws. The most important flaw was a lack of reporting the cutoff value Cobb angle that was used to define scoliosis or limiting the study to one specific Cobb angle (e.g., 10°). The International Scientific Society on Scoliosis Orthopedic and Rehabilitation Treatment (SOSORT) defines scoliosis with a Cobb angle of 10° [8]. This 10° angle might be important for epidemiological research but clinicians might be more interested in higher Cobb angles, for example the threshold for surgery often lies around 40°–50° Cobb angle [9]. Another flaw was that the mean age of included patients was not mentioned or was quite low. Muller et al. [10] showed that the most progression of scoliosis is expected before the age of 15 years, so groups with a mean age <15 years might underestimate scoliosis prevalence and/or severity.

This study excludes patients aged <15 years old to minimize the number of patients which might develop scoliosis in a later phase. Secondly we will report multiple cutoff values of Cobb angles (10°, 20°, 30°, and 40°) to provide data that is both epidemiologically and clinically relevant and to make comparison with other research more feasible. The main aim of the current study is to determine the prevalence of scoliosis in patients with MMC, who are part of a convention of spina bifida patients in multidisciplinary follow-up, in the University Hospitals Leuven. After acquiring the number of spina bifida patients in other Belgian centers, we will calculate whether the group of the University Hospitals Leuven form a good representation of the Belgian spina bifida population. The secondary aim is to determine whether there are differences concerning the distribution of gender, the radiological level of MMC, the ambulatory status, hydrocephalus, tethered cord, and syringomyelia in the MMC patients with or without scoliosis.

## Methods

### Participants

All patients' records from the patients included in the spina bifida convention of the University Hospitals Leuven in Belgium were examined. The spina bifida convention is an agreement between the hospital and the national health service. Only three hospitals in Belgium have this convention: University Hospitals Leuven, University Hospitals Ghent and University Hospitals Saint Luc. The convention

finances a specialized multidisciplinary team, available for children with spina bifida and for spinal cord injuries incurred within the first 2 years of life. Adults who are transitioning from pediatric care or who were not yet in multidisciplinary follow-up can also access the convention. Patients visit the spina bifida clinics at least once a year. The following inclusion criteria were used, inclusion in the spina bifida convention of the University Hospitals Leuven, a diagnosis of MMC, age  $\geq 15$  years and availability of useful radiographs (full spine or a combination depicting the entire spine taken on the same day). Patients needed to be  $\geq 15$  years at the time of the radiograph unless they had a scoliosis fusion, the latter were included irrespective of the age at the time of the radiograph. Patients whose Cobb angle could not be determined (not mentioned in the radiology report and not measurable on radiographs) were excluded from the study unless they underwent a scoliosis fusion in the past.

A group "MMC full spine" consisting only of the selected MMC patients with a full spine radiograph was created. The "MMC full spine" group was used to examine a difference in the distribution of radiograph position in MMC patients with/without scoliosis.

### Data collection

The following data were collected: age, gender, radiograph type, age at the time of the radiograph, position during radiograph, presence of fusion, age at the time of fusion, diagnosis of hydrocephalus/tethered cord/syringomyelia, radiological level of MMC, ambulatory status, main Cobb angle, main curve convexity, and main curve location. Useful radiographs comprised full spine radiographs, a combination of abdominal and thorax radiographs, a radiograph depicting the course of a ventriculoperitoneal drain (abdominal, thorax, and cervical) or a combination of lumbar and dorsal radiographs. The full spine was preferred if present. The most recent one was used in case of multiple radiographs. If patients underwent a scoliosis fusion in the past, the preoperative radiograph was taken, if available, instead of the most recent one. The radiograph position was defined as standing, sitting, or supine. The patients' records were examined to see whether patients were diagnosed with hydrocephalus, tethered cord, or syringomyelia. The diagnosis was deemed positive if it was mentioned in the medical history or in the radiology reports of spinal or cerebral imaging. The radiological level, derived from the radiology report, was divided in the following categories: "cervical", "cervicothoracic", "thoracic", "thoracolumbar", "lumbar", "lumbosacral", and "sacral". The ambulatory status was based on the description of the gait pattern in the patients' records. The gait pattern was divided into six groups of the Functional Mobility Scale [11]: "wheelchair

user”, “walking with walking frame”, “walking with two crutches”, “walking with two sticks or one crutch”, “walking without aid on level surfaces”, and “walking without aid on all surfaces”. The Cobb angle was defined as the angle between the cranial endplate from the upper vertebra and the caudal endplate of the lower vertebra from of the scoliotic curve. Only the main (biggest) curve was determined for double or triple curves. One author (AH) measured the Cobb angle if it was not mentioned in the radiology report. Only the preoperative Cobb angle was determined in patients with a fusion. The patient who underwent a fusion in the past and whose preoperative Cobb angle could not be obtained was considered a patient with a Cobb angle  $\geq 30^\circ$ . This was based on the premise that in our hospital scoliosis surgery is very rarely indicated with a Cobb angle  $< 30^\circ$ . The cutoff was limited to  $\geq 30^\circ$  instead of  $\geq 40^\circ$  because sometimes fusion is performed between  $30^\circ$  and  $40^\circ$  degrees depending on the evolution of the curve and the age of the patient. Those patients without a preoperative Cobb angle will be excluded concerning the calculations with cutoff value Cobb angle  $40^\circ$ . The patients were not required to sign an informed consent procedure by the ethical committee due to the retrospective nature of the study. The study was approved by the ethical committee of the University Hospitals Leuven.

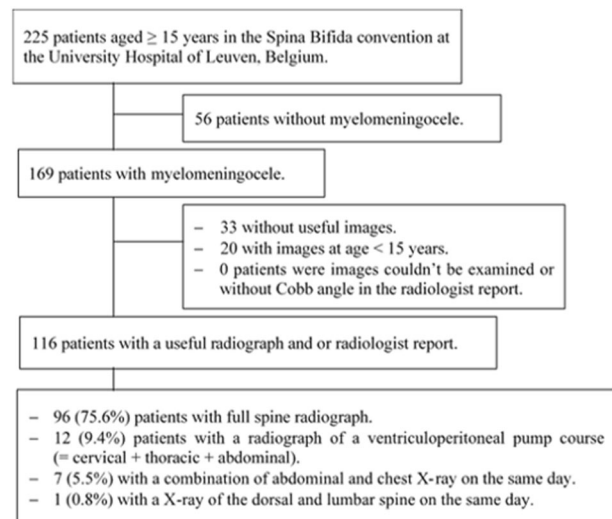
## Data analysis

Statistical analyses were performed using SPSS (IBM Corp. Released 2017. IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp). The following statistical tests were used with a 95% confidence interval and with  $P < 0.05$ : Kruskal–Wallis test, Mann–Whitney  $U$ -test and Fisher-exact test. The multiple Fisher’s-exact test was used with an adjusted  $P$  value (Bonferroni correction) and with a 95% confidence interval. The margin of error for proportions was calculated using the following formula:

$$E = \pm z \sqrt{\frac{\hat{p}(1 - \hat{p})}{n}} \quad (1)$$

With  $E$  = margin of error (%),  $z$  = z-score,  $\hat{p}$  = sample proportion, and  $n$  = sample size with a z-score of 1.96 representing a 95% confidence interval.

The prevalence of scoliosis was calculated for different cutoff values of the Cobb angle ( $10^\circ$ ,  $20^\circ$ ,  $30^\circ$ , and  $40^\circ$ ). Fisher-Exact test was performed to examine a potential difference in distribution concerning gender, hydrocephalus, tethered cord, syringomyelia, radiological level, and ambulatory status between MMC patients without or with scoliosis. These analyses were done for three different cutoff values of the Cobb angle ( $10^\circ$ ,  $20^\circ$ ,  $30^\circ$ , and  $40^\circ$ ). Only patients with a Cobb angle  $\geq 10^\circ$  were taken into account to calculate the median Cobb angle.



**Fig. 1** Flowchart showing the patient selection procedure. There were 116 patients who fitted all the inclusion criteria. From those 116 patients, 96 had a full spine radiograph and 20 of them had a combination of useful radiographs.

## Results

There were 255 spina bifida patients aged  $\geq 15$  years, of which 169 were diagnosed as MMC. The algorithm of patient selection is shown in Fig. 1. There were 96 patients with a full spine radiograph (= “MMC full spine” group) and 20 with useful radiographs. Table 1 presents the gathered data of the 116 included patients (“MMC group”). There were four patients with fusion which had an unknown preoperative Cobb angle. These four patients were considered as having a  $\geq 30^\circ$  Cobb angle as explained in the “Methods” section.

The overall prevalence of scoliosis in patients with MMC was 78.4% (95% CI, 71.0–85.8) respecting the most used definition of scoliosis with a Cobb angle  $\geq 10^\circ$  [8]. The prevalence was 60.3% (95% CI, 51.4–69.2) for an angle  $\geq 20^\circ$ ; 52.6% (95% CI, 43.5–61.7) for  $\geq 30^\circ$ ; and 36.6% (95% CI, 27.7–45.5) for an angle  $\geq 40^\circ$ . Eight of the 20 patients (40%) with useful radiographs were diagnosed with scoliosis (Cobb angle  $\geq 10^\circ$ ); ranging from  $11^\circ$  to  $20^\circ$ . The convention of the spina bifida patients in the University Hospitals Leuven ( $n = 338$ ) represent 48.2% of the spina bifida patients in multidisciplinary follow-up. There are 223 (31.8%) patients in University Hospitals Ghent and 140 (20.0%) in University Hospitals Saint Luc. The used sample represents 48.2% of the Belgian spina bifida population in multidisciplinary follow-up with a margin of error of  $\pm 3.7\%$ .

The median Cobb angle was  $36.0^\circ$  [20.0–58.0] for the “MMC group” and  $39.0^\circ$  [23.0–60.0] for “MMC full spine”. The median preoperative Cobb angle of the patients who underwent a scoliosis fusion was  $54.0^\circ$  [40.0–81.0].

**Table 1** The collected data of the final 116 MMC patients who fitted the inclusion criteria, forming the “MMC group”.

Age patients (years)	Median age at time of radiograph	22.0 (17.8–29.0)
	Median age at time of the fusion	13.0 (12.0–15.0)
Gender	Males	45.7% ( <i>n</i> = 53)
	Females	54.3% ( <i>n</i> = 63)
Radiograph type	Full spine	82.8% ( <i>n</i> = 96)
	Ventriculoperitoneal drain	10.3% ( <i>n</i> = 12)
	Abdominal + thoracic	6.0% ( <i>n</i> = 7)
	Dorsal + lumbar	0.9% ( <i>n</i> = 1)
Radiograph position	Standing	49.1% ( <i>n</i> = 57)
	Sitting	45.7% ( <i>n</i> = 53)
	Supine	5.2% ( <i>n</i> = 6)
Ambulatory status	Walking on all surfaces without aid	23.3% ( <i>n</i> = 27)
	Walking on level surfaces without aid	4.3% ( <i>n</i> = 5)
	Walking with a walking stick	0.0% ( <i>n</i> = 0)
	Walking with crutches	6.0% ( <i>n</i> = 7)
	Walking with a walking frame	3.4% ( <i>n</i> = 4)
	Wheelchair user	62.9% ( <i>n</i> = 73)
Fusion	Patients with fusion	37.1% ( <i>n</i> = 43)
	Patients without fusion	62.9% ( <i>n</i> = 73)
	Age of fusion ≤10 years	9.3% ( <i>n</i> = 4)
	Age of fusion >10 and ≤15 years	65.1% ( <i>n</i> = 28)
	Age of fusion >15 years	11.6% ( <i>n</i> = 5)
	Age of fusion unknown	14.0% ( <i>n</i> = 6)

The group “wheelchair user” had a 4–8 times, depending on the cutoff, more chance of having scoliosis compared to those “walking on all surfaces without aid” (see Table 2). Odds ratios were 7.5 [2.6–21.6], 4.2 [1.7–10.7], 6.7 [2.4–18.8], and 8.5 [1.9–39.0] for the cutoff Cobb angles of 10°, 20°, 30°, and 40° respectively.

There was a statistical difference between patients with “sacral” radiological levels compared to those with “thoracolumbar” or “lumbar” radiological levels. Odds ratios vary between 0.04 [0–0.4] and 0.13 [0.03–0.47] (see Table 3). There was no difference concerning the radiological level at the 10° or 40° cutoff (see Table 3).

Ninety-eight patients of the “MMC group” were diagnosed with hydrocephalus (84.5%), 59 with tethered cord (50.9%) and 13 with syringomyelia (11.2%). There was no statistically significant difference in the distribution of hydrocephalus, tethered cord, or syringomyelia and scoliosis with Cobb angle cutoffs at 10° ( $P = 1$ ,  $P = 0.747$ , and  $P = 0.731$  respectively), 20° ( $P = 0.551$ ,  $P = 0.596$ ,  $P = 0.561$  respectively), 30° ( $P = 0.811$ ,  $P = 0.269$ , and  $P = 0.493$  respectively), or 40° ( $P = 0.783$ ,  $P = 0.228$ , and  $P = 0.528$  respectively). The distribution of gender didn’t differ concerning scoliosis with Cobb angle cutoffs at 10° ( $P = 0.118$ ), 20° ( $P = 0.341$ ), 30° ( $P = 0.576$ ), or 40° ( $P = 0.536$ ).

There was no difference concerning the radiograph position for the mean Cobb angle in the “MMC full spine” group,  $P = 0.458$ .

## Discussion

The 116 included MMC patients had a prevalence of scoliosis 78.4% (95% CI, 71.0–85.8) for a Cobb angle  $\geq 10^\circ$ ; 60.3% (95% CI, 51.4–69.2) for an angle  $\geq 20^\circ$ ; 52.6% (95% CI, 43.5–61.7) for an angle  $\geq 30^\circ$ ; and 36.6% (95% CI, 27.7–45.5) for an angle  $\geq 40^\circ$ . The prevalence of 60.3% (95% CI, 51.4–69.2) concerning the 20° cutoff is comparable with the results found in the literature (52%) [7], with  $P = 0.146$ . The examined convention represents about half (48.2%  $\pm$  3.7%) of the Belgian spina bifida population in multidisciplinary follow-up.

The research of Müller et al. [10] and Trivedi et al. [12] formed the basis to set the age limit at  $\geq 15$  years as both of them showed that most progression of scoliosis happens before the age 15 years. Patients who underwent a scoliosis fusion in the past and whose preoperative radiograph could not be obtained were regarded as patients with a Cobb angle  $\geq 30^\circ$  as explained in the methodology. The fact that the median preoperative Cobb angle in this study was 54.0° [40.0–81.0] reinforces this statement.

**Table 2** Distribution of the ambulatory status of the patients from the “MMC group” for the different Cobb angles (10°, 20°, 30°, and 40°).

Ambulatory status	Cobb angle <10°	Cobb angle ≥10°	P value	Cobb angle <20°	Cobb angle ≥20°	P value	Cobb angle <30°	Cobb angle ≥30°	P value	Cobb angle <40°	Cobb angle ≥40°	P value
Walking on all surfaces without aid	13	14	1	17	10	1	21	6	0.578	25	2	0.105
Walking on level surfaces without aid	2	3		3	3		3	2		3	2	
Walking on all surfaces without aid	13	14	0.426	17	10	1	21	6	1	25	2	0.511
Walking with crutches	2	5		4	3		5	2		6	1	
Walking on all surfaces without aid	13	14	0.12	17	10	0.284	21	6	0.063	25	2	0.070
Walking with a walking frame	0	4		1	3		1	3		2	2	
Walking on all surfaces without aid	13	14	<0.001 <sup>*a</sup>	17	10	0.003 <sup>*b</sup>	21	6	<0.001 <sup>*c</sup>	25	2	0.001 <sup>*d</sup>
Wheelchair user	8	65		21	52		25	48		41	28	
Walking on level surfaces without aid	2	3	1	3	3	1	3	2	1	3	2	0.523
Walking with crutches	2	5		4	3		5	2		6	1	
Walking on level surfaces without aid	2	3	0.444	3	3	0.524	3	2	0.524	3	2	1
Walking with a walking frame	0	4		1	3		1	3		2	2	
Walking on level surfaces without aid	2	3	0.12	3	3	0.166	3	2	0.344	3	2	1
Wheelchair user	8	65		21	52		25	48		41	28	
Walking with crutches	2	5	0.49	4	3	0.545	5	2	0.242	6	1	0.491
Walking with a walking frame	0	4		1	3		1	3		2	2	
Walking with crutches	2	5	0.21	4	3	0.196	5	2	0.096	6	1	0.241
Wheelchair user	8	65		21	52		25	48		41	28	
Walking with a walking frame	0	4	1	1	3	1	1	3	1	2	2	1
Wheelchair user	8	65		21	52		25	48		41	28	

None of the patients walked with a walking stick (excluded in table). There is a significant difference between “walking on all surfaces without aid” and “wheelchair user” for the three different Cobb angles. Significant *P* values, using multiple Fisher’s exact tests (2 × 2) with a new  $\alpha$ -level (<0.01), are marked with an asterisk. Addendum: Four patients which preoperative Cobb angle could not be obtained were left out of the calculations concerning the 40° cutoff value as explained in the “Methods” section.

Odds ratios:

<sup>a</sup>7.5 [2.6–21.6].

<sup>b</sup>4.2 [1.7–10.7].

<sup>c</sup>6.7 [2.4–18.8].

<sup>d</sup>8.5 [1.9–39.0].

**Table 3** Distribution of the radiological level among the different groups with different Cobb angle cutoffs (10°, 20°, 30°, and 40°).

Radiological level	Cobb angle <10°	Cobb angle ≥10°	<i>P</i> value	Cobb angle <20°	Cobb angle ≥20°	<i>P</i> value	Cobb angle <30°	Cobb angle ≥30°	<i>P</i> value	Cobb angle <40°	Cobb angle ≥40°	<i>P</i> value
Thoracic	0	3	/	1	2	0.231	2	1	0.108	3	0	0.167
Thoracolumbar	0	10		0	10		1	9		2	4	
Thoracic	0	3	1	1	2	0.504	2	1	0.231	3	0	0.214
Lumbar	2	20		4	18		6	16		8	10	
Thoracic	0	3	1	1	2	1	2	1	1	3	0	0.547
Lumbosacral	14	43		23	34		28	29		39	18	
Thoracic	0	3	0.529	1	2	0.201	2	1	1	3	0	1
Sacral	9	15		18	6		18	6		20	4	
Thoracolumbar	0	10	1	0	10	0.283	1	9	0.387	2	4	1
Lumbar	2	20		4	18		6	16		8	10	
Thoracolumbar	0	10	0.106	0	10	0.012	1	9	0.035	2	4	0.171
Lumbosacral	14	43		23	34		28	29		39	18	
Thoracolumbar	0	10	0.034	0	10	<0.001 <sup>*a</sup>	1	9	0.001 <sup>*b</sup>	2	4	0.029
Sacral	9	15		18	6		18	6		20	4	
Lumbar	2	20	0.211	4	18	0.071	6	16	0.127	8	10	0.094
Lumbosacral	14	43		23	34		28	29		39	18	
Lumbar	2	20	0.038	4	18	<0.001 <sup>*c</sup>	6	16	0.003 <sup>*d</sup>	8	10	0.019
Sacral	9	15		18	6		18	6		20	4	
Lumbosacral	14	43	0.285	23	34	0.007	28	29	0.049	39	18	0.273
Sacral	9	15		18	6		18	6		20	4	

There was a significant difference between “thoracolumbar” and “lumbar” levels with the “sacral” level in the groups with a 20° and 30° cutoff. Significant *P* values, using multiple Fisher’s-exact tests (2 × 2) with a new alpha level (<0.005), are marked with an asterisk. Addendum: Four patients which preoperative Cobb angle could not be obtained were left out of the calculations concerning the 40° cutoff value as explained in the “Methods” section.

Odds ratios:

<sup>a</sup>Unable to compute (cell contains zero).

<sup>b</sup>0.04 [0–0.40].

<sup>c</sup>0.07 [0.02–0.31].

<sup>d</sup>0.13 [0.03–0.47].

This study showed a clear correlation between scoliosis and the ambulatory status and a limited correlation with the radiological level. Wheelchair users had 4–8 times more chance of having scoliosis compared to those capable of walking on all surfaces without aid. This supports earlier studies [12, 13] who described a correlation between functional status and scoliosis. Patients with a “sacral” level had a lower prevalence of scoliosis than those with “lumbar” or “thoracolumbar” level. However the strength of this correlation was very weak with OR ranging from 0.04 to 0.13. Previous research [12] stated that the radiological level can predict scoliosis but that this prediction might be limited.

There was no statistically significant difference between the presence of hydrocephalus, tethered cord or syringomyelia with respect to scoliosis even though these interactions are described in other articles. This could be conflicting because others state a correlation between for

example tethered cord and scoliosis [14]. Our results are more in line with Dias who also stated that there is only limited evidence supporting a relationship between scoliosis and syringomyelia as well as tethered cord in a MMC population [15]. This indicates that the neurological status and/or ambulatory status might have a greater impact on the development of scoliosis than associated comorbidities in the central nervous system such as hydrocephalus. One factor that might interfere with statistical analysis in in this study is the high number of patients with hydrocephalus (84.5%) and the low number of patients with syringomyelia (11.2%).

There were some limitations, mainly because of the retrospective nature of the study. Firstly, the neurological level was absent in most patients files and could therefore not be incorporated in our analysis. Secondly, fifty-three patients were excluded for the lack of images ( $n = 33$ ) or because they were aged <15 years old at the time of the most recent



radiograph ( $n = 20$ ). One would expect that this group without images would have a low chance of having scoliosis, which would decrease the prevalence, since it is common practice in the convention to take a full spine radiograph if there are clinical signs of scoliosis. However, eight of the 20 patients (40%) with useful images but without a full spine radiograph (i.e., clinical signs were absent or missed) had a Cobb angle  $\geq 10^\circ$  (ranging from  $11^\circ$  to  $20^\circ$ ). So it is likely that there are cases of undiagnosed scoliosis among the 33 excluded patients. Thirdly, there was no uniform radiograph position/protocol and different radiograph positions (standing, sitting, and supine) could have altered our results. Yazici et al. [16] measured IS in both supine and standing position with the Cobb method and found a difference of 29.8% between them, with the standing radiograph having a higher Cobb angle. There were no studies found which measured the difference between a sitting and standing position. The impact of the different positions could be minimized in this study since there were only three supine radiographs with two of them having a Cobb angle of  $\geq 30^\circ$  and 40 of the 52 sitting radiographs (76.9%) had a Cobb angle  $\geq 30^\circ$ .

Despite its limitations this study forms a good representation of the Belgian spina bifida population concerning patients with MMC. The results show that scoliosis screening is important in these patients and especially in those patients who are wheelchair users. It is important to notice that even with our clinical screening we missed some patients with scoliosis, diagnosed on other radiographs in this study, but those curves did not exceed  $20^\circ$ . We provided different Cobb angle cutoff to allow optimal comparison with other studies for both epidemiological and clinical research. The role of tethered cord, syringomyelia and hydrocephalus seems limited in our study but warrants future investigations since there are conflicting results in the literature.

## Conclusion

The high prevalence of scoliosis 78.4% (95% CI, 71.0–85.8) for a Cobb angle  $\geq 10^\circ$  and with 36.2% (95% CI, 27.5–44.9) having a Cobb angle  $\geq 40^\circ$ , warrants a thorough screening and follow-up for patients with MMC. The 60.3% (95% CI, 51.4–69.2) of patients with a Cobb angle  $\geq 20^\circ$  is comparable with the 52% found in the literature. There was no statistically significant difference between MMC patients with or without hydrocephalus, tethered cord or syringomyelia regarding scoliosis even though these interactions are described in other articles. Future studies should focus on the interactions of the neurological comorbidities associated with MMC and scoliosis.

## Data availability

Data were retrieved from medical files of patients included in the Spina Bifida Convention, University Hospitals Leuven. The data are not publicly available due viewpoint of personal information protection but are available from the corresponding author on reasonable request.

**Acknowledgements** We would like to thank Myleen Christian, coordinating nurse of the spina bifida convention Leuven, Bart Thomas, coordinator the spina bifida convention Ghent, and Dr. Ann Renders, responsible for the spina bifida convention Saint Luc Bruxelles for providing data concerning the conventions.

**Author contributions** AH was responsible for the data collection, analyzing data, interpreting results, and writing the paper. CK and SN were responsible for analyzing the data, interpreting the data, and revising the paper. KJ, PM, SS, and KP provided feedback on interpretation of the data and revised the paper.

## Compliance with ethical standards

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

**Ethical approval** The study protocol was approved by the ethical committee of the University Hospitals Leuven.

**Informed consent** Informed consent form was waived by the ethical committee.

**Publisher's note** Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

## References

1. Atta CA, Fiest KM, Frolkis AD, Jette N, Pringsheim T, St Germaine-Smith C, et al. Global birth prevalence of spina bifida by folic acid fortification status: a systematic review and meta-analysis. *Am J Public Health*. 2016;106:e24–34.
2. Copp AJ, Adzick NS, Chitty LS, Fletcher JM, Holmbeck GN, Shaw GM. Spina bifida. *Nat Rev Dis Prim*. 2015;1:15007.
3. Werhagen L, Gabrielsson H, Westgren N, Borg K. Medical complication in adults with spina bifida. *Clin Neurol Neurosurg*. 2013;115:1226–9.
4. Kumar R, Singh SN. Spinal dysraphism: trends in northern India. *Pediatr Neurosurg*. 2003;38:133–45.
5. Dicianno BE, Sherman A, Roehmer C, Zigler CK. Co-morbidities associated with early mortality in adults with spina bifida. *Am J Phys Med Rehabil*. 2018;97:861–5.
6. Allam AM, Schwabe AL. Neuromuscular scoliosis. *PM R*. 2013;5:957–63.
7. Heyns A, Negrini S, Jansen K, Moens P, Schelfaut S, Peers K, et al. The prevalence of scoliosis in spina bifida subpopulations: a systematic review. *Am J Phys Med Rehabil*. 2018;97:848–54.
8. Negrini S, Donzelli S, Aulisa AG, Czaprowski D, Schreiber S, de Mauroy JC, et al. 2016 SOSORT guidelines: orthopaedic and rehabilitation treatment of idiopathic scoliosis during growth. *Scoliosis Spinal Disord*. 2018;13:3.

9. Weinstein SL, Ponseti IV. Curve progression in idiopathic scoliosis. *J Bone Jt Surg Am.* 1983;65:447–55.
10. Muller EB, Nordwall A, Oden A. Progression of scoliosis in children with myelomeningocele. *Spine.* 1994;19:147–50.
11. Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The functional mobility scale (FMS). *J Pediatr Orthop.* 2004;24:514–20.
12. Trivedi J, Thomson JD, Slakey JB, Banta JV, Jones PW. Clinical and radiographic predictors of scoliosis in patients with myelomeningocele. *J Bone Jt Surg Am.* 2002;84:1389–94.
13. Thomas JG, Hwang SW, Blumberg TJ, Whitehead WE, Curry DJ, Luerssen TG, et al. Correlation between shunt series and scoliosis radiographs in children with myelomeningoceles. *J Neurosurg Spine.* 2012;17:410–4.
14. McLone DG, Herman JM, Gabrieli AP, Dias L. Tethered cord as a cause of scoliosis in children with a myelomeningocele. *Pediatr Neurosurg.* 1990;16:8–13.
15. Dias MS. Neurosurgical causes of scoliosis in patients with myelomeningocele: an evidence-based literature review. *J Neurosurg.* 2005;103(Suppl 1):24–35.
16. Yazici M, Acaroglu ER, Alanay A, Deviren V, Cila A, Surat A. Measurement of vertebral rotation in standing versus supine position in adolescent idiopathic scoliosis. *J Pediatr Orthop.* 2001;21:252–6.