

A Multi-dimensional Online Survey of Experts, Patients and the Public on Early Detected Scoliosis in Fetus Caused by Spinal Hemivertebra

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Abstract

In recent years, with the increasing number of women in advanced maternal age and standardized protocol of prenatal examination in China, more fetuses have been detected with scoliosis caused by hemivertebra (HV). The objective of this study was to obtain the responses of different populations on early detected scoliosis in fetuses caused by spinal hemivertebra through a multi-dimensional survey. A self-administered anonymous web and social media-based multi-dimensional online survey were conducted. Four short questionnaires concerning fetuses with scoliosis secondary to hemivertebra were sent to spine/orthopedic surgeons, obstetricians/ultrasonologists, scoliosis patients, and the non-medical general public separately. Survey items mainly included whether continue the pregnancy, the responses of different populations, whether they have knowledge of HV and what they wanted to know most about HV among the general public group, etc. All the questions were single choice questions, and additional responses could be added if needed. A total of 5586 respondents including 647 spine surgeons, 227 orthopaedic surgeons, 350 obstetricians, 54 ultrasonologists, 246 scoliosis patients and 4062 general public participated in the survey. 41.89% spine surgeons, 25.99% orthopedic surgeons, 6.57% obstetricians, 17.48% scoliosis patients and 19.23% general public chose to continue pregnancy and seek treatment after birth. 4.02% spine surgeons, 7.49% orthopedic surgeons, 3.71% obstetricians, 26.83% scoliosis patients and 14.28% non-medical general public suggested or chose to terminate the pregnancy. 54.10% spine surgeons, 66.52% orthopedic surgeons suggested explain the pros and cons to the parents and let them make their own choices.

Introduction

Congenital spinal deformity is the lateral curvature of the spine caused by vertebral malformations that result in an imbalance of longitudinal growth of the spine. And it is not an uncommon condition which was noted in about 1 in 1000 live births [1-5]. Hemivertebra (HV) is the most common type of congenital spinal deformity which is caused by failures of formation with the prevalence rate of 0.1-1% [6-7]. The rate of deterioration and the severity of the spinal deformity depended on the number, type, and site of HV [1-2,8].

In recent years, with the increasing number of elderly pregnant women and the popularization of three-dimensional ultrasound prenatal examination in China, more and more fetal HV has been identified antenatally between 12 and 28 weeks' gestation. And the fetus is usually accompanied by central nervous system abnormalities, urinary system abnormalities, and congenital heart diseases [9-12].

It is difficult to make a decision when faced with a fetus with congenital spinal deformity secondary to Hemivertebra in the medical ethics environment in China. As professionals, we surgeons or physicians have the desire to determine the outcomes of the intervention, whether keeping the baby and seeking treatment after birth or terminating the pregnancy. Also, the parents have the desire of whether to keep the baby or not. Under these not uncommon conditions in China with great numbers of populations, it is really difficult to choose.

The purpose of this study was to conduct a multiple dimensions survey to investigate the decision making among people of different professions when faced with a fetus with congenital spinal deformity secondary to Hemivertebra.

Results

Overall, 5586 people including 647 (11.58%) spine surgeons, 227 (4.06%) orthopaedic surgeons, 350 (6.27%) obstetricians, 54 (0.97%) ultrasonologists, 246 (4.40%) scoliosis patients and 4062 (72.72%) non-medical general public responded the survey.

Table 1 shows the numbers and percentages of different people with different choices or suggestions. 41.89% (271/647) spine surgeons, 25.99% (59/227) orthopedic surgeons, 6.57% (23/350) obstetricians, 17.48% (43/246) scoliosis patients, and 19.23% (781/4062) non-medical general public chose to keep the baby and seeking treatment after birth. 4.02% (26/647) spine surgeons, 7.49% (17/227) orthopedic surgeons, 3.71% (13/350) obstetricians, 26.83% (66/246) scoliosis patients and 14.28% (580/4062) non-medical general public suggested or chose to terminate the pregnancy. 54.10% (350/647) spine surgeons, 66.52% (151/227) orthopedic surgeons suggested explain the pros and cons to the parents and let them make their own

choices. Moreover, 81.48% (44/54) ultrasonologists, 66.49% (2701/4062) non-medical general public, and 54.88% (135/245) scoliosis patients chose to make their decisions after consulting the spine surgeons and obstetricians. 34% (119/350) obstetricians suggested the parents consult the spine surgeons. 55.71% (195/350) obstetricians and 18.52% (10/54) ultrasonologists suggested do further examinations.

For spine surgeons and orthopedic surgeons, 37.76% suggested the parents keep the baby and seek treatment after birth, 4.92% suggested the parents terminate the pregnancy, and 57.32% Explain the pros and cons to the parents and let them make decisions themselves.

For obstetricians and ultrasonologists, up to 63.61% (257/404) of them never met a fetus with HV during their antenatal examinations. 36.88% (149/404) of them didn't know that HV could result in spinal deformity (Table 2).

When faced with the question "what do you most want to know about congenital spinal deformity?", the scoliosis patients and the general public had almost the same percentages of choices. 47.75% of them wanted to know the causes and prevention methods. 4.92 % of them wanted to know the development of postnatal spinal deformity without treatment. 14.86% wanted to know the treatment and its effect, risks, expenses, and so on after the child is born. 32.47% wanted to know the effect on children's future after effective treatment. (Table 3)

47.15% (116/246) scoliosis patients already have child, different choices and what they wanted to know most about congenital spinal deformity between those have a child and those don't have are listed in Table 4. More of them chose to keep the baby than those without child (23.28% vs 13.08%).

For the general public, 23.83% (968/4062) have some knowledge of spinal deformity, 18.88% (865/4062) know a spinal deformity patient, and 69.92% (2840/4062) already have a child. Different choices and what they wanted to know most about congenital spinal deformity between those who have a child and those who don't have are listed in Table 5. For the male/female general public, their choice, and what they want to know most are listed in Table 6. For different education background, their choice and what they want to know most and whether have some knowledge of spinal deformity are listed in Table 7.

Discussion

Hemivertebra is the most common cause of the congenital spinal deformity and many patients also manifest deformities in other systems. Beauregard-Lacroix [13]. conducted a retrospective study from 2004 to 2015 in 300 congenital spinal deformity patients and found up to 67.1% of them had associated anomalies of different systems. This is one of the important reasons why it is difficult to make decisions whether keeping the fetus with HV in China.

The etiology of congenital spinal deformity is considered to be multifactorial, containing both genetic and environmental factors. The genetic factors remain much unknown and some studies found it a multi genetic disease. Studies also demonstrated that environmental aspects played an important role in the development of congenital spinal deformity involving vitamin deficiency, hypoxia, alcohol use, exposure to hyperthermia, boric acid, and valproic acid [5,16-19].

About the natural history of congenital spinal deformity, 25% of the curves do not progress, 25% progress slowly and 50% progress rapidly [14]. And fully segmented HV has the highest potential for progression [15]. The rate of deterioration and the severity of the spinal deformity depends on the number, type, and site of HV.

Involving the treatment, many studies with long term follow-up concluded that early HV resection was a safe and effective way to correct the spinal deformity and restore the spinal balance [2,20-25]. But there are also long-time follow-up studies found that spinal deformity secondary to HV could be treated conservatively and some patients even had gradual improvement without treatment [26-27].

This is the only multi-dimensional and well-attended online survey on this topic. In our survey, among the medical groups, most of the ultrasonologists (81.48%) suggested the parents see a spine surgeon and obstetrician for advice. And most of the

obstetricians (89.71%) suggested the pregnant women do further examinations to exclude accompanied abnormalities and consult the spine surgeons/orthopedic surgeons for suggestions. Among the non-medical related groups, most of them also chose to consult the spine surgeons and obstetricians. So finally the spine surgeons and orthopedic surgeons need to make the final suggestion, and most of them recommended keeping the baby or explain the pros and cons to the parents. As professional doctors treating spinal deformity, most spine surgeons/orthopedic surgeons understand the causes and treatment of this condition, basically support to keep the baby and will communicate with the patient to explain the situation in detail.

Moreover, 62.86% of the surveyed obstetricians never met a fetus with HV at work, 40.29% of obstetricians don't even know that HV could result in spinal deformity. They paid more attention to observe whether the fetus had other abnormalities. That is also why 89.71% of them suggested the parents do further examinations and see a spine surgeon/orthopedic surgeon. Therefore, it is also necessary for obstetricians to understand the impact of spinal abnormalities on the child's future as an important part of fetal development. Also, the multi-professional cooperation consultation will help to make the best decision

Also as an important part of fetal development, although 68.52% of the ultrasound doctors never met a fetus with HV at work, 85.19% of them know the harm of the hemivertebra. And if the examination found that the fetus has a hemivertebra, the pregnant women will be recommended to the department of obstetrics and spine/orthopedics for further consultation.

For scoliosis patients, there is still a large amount of concern about spinal deformity caused by hemivertebra, other study indicated that the scoliosis patient had an increased incidence and risk of depression [28]. so maybe this is why up to 26.83% of the scoliosis patients chose to terminate the pregnancy. They also wanted to understand the causes, prevention methods, treatment of spinal deformity, and the children's future.

As the largest group (4062/5586, 72.72%) participating in the questionnaire, there are up to 76.17% of the general public who have little or don't have any knowledge of spinal deformity. That is why 14.28% chose to terminate the pregnancy, and thus more of them wanted to know the causes, prevention methods, treatment of spinal deformity, and the children's future. Furthermore, the general public with or without a child had almost the same choices about the fetus and what they wanted to know most about congenital spinal deformity. With the advancement of educational background, the percentage of those knowing spinal deformity is increasing, and more chose to keep the baby, more wanted to know the treatment and future of their children (Tables 1, 2, and 3).

Methods And Materials

A self-administered anonymous web and social media-based multiple dimensional online surveys were conducted at a one-time point lasting for 3 days. Ethics approval and IRB Number was not applicable. Four short questionnaires concerning fetuses with congenital spinal deformity secondary to Hemivertebra were sent to spine/orthopedic surgeons, obstetricians/ultrasonologists, scoliosis patients, and the non-medical general public separately. All the questions were single choice questions, and additional responses could be added if needed. Survey items included gender, whether have a baby or not, professions, years of experience in his or her specialty, their suggestions or choices about the fetus with HV, etc.

The survey was designed and conducted using the online platform <https://www.wjx.cn> (WenJuanXing, CSX, CHN), and the four short questionnaires were sent through WeChat, the most commonly used communicating social media in China. Hundreds of specific WeChat groups with spine/orthopedic surgeons, obstetricians/ultrasonologists, scoliosis patients, or non-medical general public received different questionnaires. Part of the non-medical general public received his/her questionnaire via WeChat moments. We didn't offer any incentive for the participants joining the survey.

Data were analyzed using the software SPSS 19.0. T-test was used for comparison of means among groups, and the chi-square test or Fisher's exact test was used for counting data ($P < 0.05$ was statistically significant).

Declarations

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Author contributions statement Yong Hai conceived the survey, Jingwei Liu.and Yiqi Zhang conducted the survey, Jingwei Liu,Yiqi Zhang and Aixing Pan analysed the results. All authors reviewed the manuscript.

Conflicts of interest/Competing interests We don't have a financial or personal relationship with a third party. We have full control of all primary data and we agree to allow the journal to review their data if requested. On behalf of all authors, the corresponding author declares that there is no conflict of interest.

Statement All experimental protocols were approved by Office of Ethics, Beijing Chaoyang Hospital, Capital Medical University 100020, Beijing, China. All methods were carried out in accordance with International Ethical Guidelines for Biomedical Research Involving Human. All participants are over 18 and informed consent was obtained

Conflict of interest statement

We don't have a financial or personal relationship with a third party. On behalf of all authors, the corresponding author states that there is no conflict of interest.

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Tables

Table1: Numbers and percentages of different people with different choices or suggestions.							
	Keep the baby	Terminate the pregnancy	Explain the pros and cons to the parents and let them make decisions themselves	Consult the spine surgeons and obstetricians	Consult the spine surgeons/orthopedic surgeons	Have further examinations	NO. of Total
NO. of Orthopedic surgeons	59 (25.99%)	17 (7.49%)	151 (66.52%)				227
NO. of Spinesurgeons	271 (41.89%)	26 (4.02%)	350 (54.10%)				647
Ultrasonologists				44 (81.48%)		10 (18.52%)	54
NO. of Obstetricians	23 (6.57%)	13 (3.71%)			119 (34%)	195 (55.71%)	350
NO. of Scoliosis patients	44 (17.89%)	66 (26.83%)		136 (55.28%)			246
NO. of General public	781 (19.23%)	580 (14.28%)		2701 (66.49%)			4062
NO. of Total	397 (21.09%)	702 (12.57%)	501 (8.97%)	3000 (53.71%)		205 (3.67%)	5586

Table 2: Numbers and percentages of ultrasonologists and obstetricians whether knowing or meet HV			
	NO. of Ultrasonologists	NO. of Obstetricians	NO. of Total
Whether meet fetus with HV during antenatal examinations			
yes	17 (31.48%)	130 (37.14%)	147 (36.39%)
no	37 (68.52%)	220 (62.86%)	257 (63.61%)
Whether know that HV could result in scoliosis			
yes	46 (85.19%)	209 (59.71%)	255 (63.12%)
no	8 (14.81%)	141 (40.29%)	149 (36.88%)
NO. of Total	54	350	404

Table 3: Numbers and percentages of what the scoliosis patients and the general public wanted to know most about congenital scoliosis			
	NO. of Scoliosis patients	NO. of Public	NO. of total
Causes and prevention methods	120 (48.78%)	1937 (47.69%)	2057 (47.75%)
What is the development of postnatal scoliosis without treatment?	11 (4.47%)	201 (4.95%)	212 (4.92%)
The treatment and its effect, risks, expenses and so on after the child is born	41 (16.67%)	599 (14.75%)	640 (14.86%)
The effect on children's future after effective treatment	74 (30.08%)	1325 (32.62%)	1399 (32.47%)
NO. of total	246	4062	4308

Table 4: Numbers and percentages of the scoliosis patients with or without a child of their choices and what they wanted to know most about congenital scoliosis		
	NO. of scoliosis patients having a child	NO. of scoliosis patients having no child
Choices about the fetus		
Keep the baby	27 (23.28%)	17 (13.08%)
Terminate the pregnancy	31 (26.72%)	35 (26.92%)
Cousult the spinesurgeons and obstetricians	58 (50%)	78 (60%)
What they wanted to know most about congenital scoliosis		
Causes and prevention methods	53 (45.69%)	67 (51.54%)
What is the development of postnatal scoliosis without treatment?	7 (6.03%)	4 (3.08%)
The treatment and its effect, risks, expenses and so on after the child is born	9 (7.76%)	32 (24.62%)
The effect on children's future after effective treatment	47 (40.52%)	27 (20.77%)
NO. of total	116	130

Table 5: Numbers and percentages of the general public with or without a child of their choices and what they wanted to know most about congenital scoliosis		
	NO. of the general public having a child	NO. of the general public having no child
Choices about the fetus		
Keep the baby	550 (19.37%)	231 (18.90%)
Terminate the pregnancy	416 (14.65%)	164 (13.42%)
Consult the spine surgeons and obstetricians	1874 (65.99%)	827 (67.68%)
What they wanted to know most about congenital scoliosis		
Causes and prevention methods	1356 (47.75%)	581 (47.55%)
What is the development of postnatal scoliosis without treatment?	141 (4.96%)	60 (4.91%)
The treatment and its effect, risks, expenses and so on after the child is born	379 (13.35%)	220 (18.00%)
The effect on children's future after effective treatment	964 (33.94%)	361 (29.54%)
NO. of total	2840	1222

Table 6: Numbers and percentages of the male and female general public of their choices and what they wanted to know most about congenital scoliosis		
	NO. of male general public	NO. of female general public
Choices about the fetus		
Keep the baby	264 (22.62%)	517 (17.86%)
Terminate the pregnancy	194 (16.62%)	386 (13.33%)
Cousult the spinesurgeons and obstetricians	709 (60.75%)	1992 (68.81%)
What they wanted to know most about congenital scoliosis		
Causes and prevention methods	548 (46.96%)	1389 (47.98%)
What is the development of postnatal scoliosis without treatment?	70 (6.00%)	131 (4.53%)
The treatment and its effect, risks, expenses and so on after the child is born	148 (12.68%)	451 (15.58%)
The effect on children's future after effective treatment	401 (34.36%)	924 (31.92%)
NO. of total	1167	2895

Table 7: Numbers and percentages of the general public with different education background of their choices, what they wanted to know most about congenital scoliosis and whether have some knowledge of scoliosis				
	High school or less	College	Bachelor	Master degree or above
Choices about the fetus				
Keep the baby	50 (11.04%)	143 (17.21%)	354 (18.19%)	234 (28.13%)
Terminate the pregnancy	68 (15.01%)	115 (13.84%)	288 (14.80%)	109 (13.10%)
Cousult the spinesurgeons and obstetricians	335 (73.95%)	573 (68.95%)	1304 (67.01%)	489 (58.77%)
What they wanted to know most about congenital scoliosis				
Causes and prevention methods	267 (58.94%)	437 (52.59%)	896 (46.04%)	337 (40.50%)
The development of postnatal scoliosis without treatment	20 (4.42%)	43 (5.17%)	101 (5.19%)	37 (4.45%)
The treatment and its effect, risks, expenses and so on after the child is born	38 (8.39%)	84 (10.11%)	293 (15.06%)	184 (22.12%)
The effect on children's future after effective treatment	128 (28.26%)	267 (32.13%)	656 (33.71%)	274 (32.93%)
Whether have some knowledge of scoliosis				
Have	85 (18.76%)	190 (22.86%)	452 (23.23%)	241 (28.97%)
Have little or don't have	368 (81.24%)	812 (77.14%)	1492 (76.77%)	591 (71.03)
NO. of total	453	831	1946	832