Author's response to reviews

Title: Reversal of childhood idiopathic scoliosis in an adult, without surgery: a case report and literature review

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Author's response to reviews: see over
May 9, 2009  
To: Dr. Theo Grivas, Editor, Scoliosis Journal  
From: William J. Brooks, D.O.; Elizabeth Krupinski, Ph.D.; Martha C. Hawes, Ph.D.  
RE: response to reviewers

Dr. Negrini requested a more succinct abstract, with Cobb angles, and this has been done. A brief summary of our response to Dr. Rigo's feedback follows (box); point-by-point responses to specific concerns are provided on p. 3-4. Reviewer comments are in boldfaced type; author responses in italics.

Dr. Rigo's verbatim concluding remarks are as follows. X-rays show a different curve with a clear improvement from the first one to the second. This improvement is radiologically and clinically (as reported by the authors) significant but not in correspondence with what the authors report. I strongly recommend the authors to send again the X-rays to One-Independent-Scoliosis-Expert. Just and expert, showing with lines how the X-rays have been measured, is enough with no more needs. The methodology used by the authors demonstrates a clear interest to prevent bias but unfortunately this present reviewer cannot accept the reported results in table 2 and figure 4. Reporting accurate angular values this paper would be an article of importance in this field, but considering all the above mentioned points I must declare myself unable to decide on acceptance or rejection for a second time.

We again appreciate the time taken with our manuscript, and the concerns regarding accurate measurement of Cobb angle. We recognize that despite ongoing efforts by clinicians and scientists over many years to render Cobb angle measurement and interpretation more scientific and precise, it remains intrinsically an imprecise and indirect assay of only one of the three dimensions of scoliosis (e.g. 1-18, detailed below). A measurement error of 5-10 degrees or more is recognized as a limitation to interpreting whether improvement or deterioration has occurred in a particular patient (1-18). In response to this concern, we emphasize that, as described in the methods section, the readings provided in Table 2 represent means and standard deviations of replicated readings by each by three independent professionals (who are NOT authors of the study and whose identity is unknown to the first and third authors), each of whom was selected based on having had >15 years experience in reading musculoskeletal radiography in a university hospital clinical setting. They were recruited by a fourth independent professional (the second author) whose expertise is in the field of medical image perception and observer performance (biosketch below). The statistical analysis and interpretation also was done not by the readers but by the second author, who teaches statistical methods.

In the current case, the reviewer has said that his reading of the major curve in the 2005 radiograph, of primary concern in a study focused on respiratory function changes, is 30 degrees. Table 2 reports a value of 22-26 degrees (24 plus/minus 2 degrees) for the same curve. This 4- to 8-degree discrepancy between the reviewer's reading and that of the three experts who took part in this study is well within the margin for error seen in studies comparing reader consistency in Cobb angle measurement (1-18). Likewise for the secondary lumbar curve, which the reviewer reads as 'around 18 degrees' and blinded readings by the team of experts read as 8-12 degrees (10 plus/minus 2).

The radiographs from 1990-2001 in this study previously were read by two other experts who are independent of the current study and also have long experience in measuring and interpreting musculoskeletal radiography. These readings were reported in a preliminary study presented at the IRSSD meeting in Athens (inserted below, Table 2). Thus, these four radiographs (1990, 1994, 1998, 2001) have now been read, independently, by two groups of professionals with long-standing expertise in musculoskeletal radiography, with virtually identical values obtained. The final x-ray (2005) is provided as Figure 4, minus the confusing lines that were provided in response to the first set of reviews, so that readers can carry out their own readings and interpretations of how much improvement was obtained.

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**Elizabeth Krupinski, PhD**

Dr. Krupinski is a Research Professor at the University of Arizona in the Departments of Radiology and Psychology where she has been since 1992. She received her undergraduate degree from Cornell University and PhD from Temple University. She completed her early training in Radiology at the University of Pennsylvania. Her main interests are in medical image perception, assessment of observer performance, and human factors issues. She is the Associate Director of Evaluation & Assessment for the Arizona Telemedicine Program. She has published extensively in these areas as they pertain to both Radiology and Telemedicine, and has presented at conferences both nationally and internationally. She is President of the Medical Image Perception Society and serves on the Editorial Boards of a number of journals in both radiology and telemedicine. Dr. Krupinski is the North American Co-Editor of the Journal of Telemedicine & Telecare. She serves regularly on review panels for the NIH, DoD, FDA and TATRC. She is currently the Vice President of the ATA.

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**Table 1.** Changes in Cobb angle magnitude in thoracic and lumbar curvatures.

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<tr>
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<tbody>
<tr>
<td>T4-T12</td>
<td>47±1</td>
<td>38±2</td>
<td>34±2</td>
<td>28±1</td>
</tr>
<tr>
<td>L1-L4</td>
<td>26±1</td>
<td>19±1</td>
<td>17±1</td>
<td>13±1</td>
</tr>
</tbody>
</table>

**The methodology used in this case report when measuring the Cobb angles of the scoliotic curves is at least questionable. These readings were done according to standard of care by three radiologists each with >15 years' experience in reading and interpreting musculoskeletal radiography in a university-based hospital. As the reviewer recognizes, the rigorous methodology was used to avoid bias, a matter of importance in any study but especially critical in a case report where potential biases of participants in the study need to be strictly and formally separated from interpretation and analysis of assays like Cobb angle measurement. Dr. Krupinski was recruited to handle the process because of her expertise in this area. Goldberg et al 1988 was used as our standard because they specifically explore variation between and among readers, including 'scoliosis experts' vs technician (more experimental variability was found among 'experts' than non-experts). The fact that Cobb angle is intrinsically subject to experimental variation and interpreter error has been detailed in numerous studies over many years (1-18, as representative samples)). Intraobserver variation for a single X-ray may be less than two degrees (e.g. Fernando et al. 2001, Goldberg et al. 1988, Peterson and Nachemson 1995), but interobserver variation ranges from two degrees to >10 degrees among different surveys (e.g. Gross et al. 1983, Kittleson and Lim 1970, Loder et al. 1995, Stokes and Aaronson 2006). The measurement error of secondary minor curves can be even higher (Goldberg et al. 1988). Treatments such as surgery can introduce additional variation by changing the shape of the curve and its apex. Since long-term benefits of conservative care have not been reported, it is impossible to predict how such changes might be manifested in Cobb angle measurements of the aging spine in correlation with increased sagittal curve and improved function. Emerging studies documenting improvement in magnitude of curvature in adults will allow monitoring over time. In the meantime, it is not unexpected that measurement variability will occur.

**No matter how high is the certification of those measuring the X-rays, the question is how experts are they in the specific issue of scoliosis. I think that an expert in the issue do not need to re-view the Moe's text book in order to measure the Cobb angle with accuracy and reliability. Providing the Cobb angle chapter is not an indication of a need to educate anyone, only a part of a formal procedure designed to ensure that all of the anonymous readers receive the same instructions, without a need to break anonymity.

**The authors state that the main thoracic curve has been measured from T4 to T12 and the lumbar minor curve from L1 to L2. The lines drawn on the X-rays (figure 4) show a different thing. No, the authors do not state that the minor curve has been measured from L1 to L2. The lumbar minor curve is stated as being from L1 to L4. See Table 2.

**First X-ray: The upper end vertebra taken by the authors seems to be T4 (just counting from caudal and assuming that there is no anatomical variation) but the line on the upper end plate is overmeasuring the inclination of the vertebra to my point of view. I cannot see one vertebra in this area with such an inclination. As described previously, care was taken to remove identifying marks from x-rays between readings. Those provided in response to the reviewer's previous concerns were residual from the last reader. This appears to have provided only additional confusion and concern, and have been removed.

**The lower end vertebra should be, according to the authors, T12, but the line is marked on the lower end vertebra of L1. In any case I personally would take T5 as the upper end vertebra and most probably T11 as lower end vertebra of the main thoracic curve. The Cobb angle is around 47 degrees in agreement with what the table 2 indicates but the lines on the X-ray are confusing. In the lumbar curve, both lines are overmeasuring in the same X-ray, with a real Cobb angle near to 30 degrees. The same picture (figure 4) shows the last X-ray taken in 2005. In this X-ray, counting from cranial, it seems that the upper end vertebra is well selected on T4, however, when counting
from caudal it can be observed 5 lumbar vertebrae and from T-12 the toracic curve defined by the authors include 8 but not 9 vertebrae. Thus, it seems that in this second case the selected vertebra was T5. In the lower part, the authors again mark a line on the lower end plate of L1 and this line should be drawn on the lower end plate of T12, according to their own explanations. Even more, there is a second couple of lines on L2, creating still more confusion. The limits of the main thoracic curve seems to be also T5 and T12 or T11. Cranially it could be taken even T6 as the most tilted vertebra to the concavity. The angular values in the second X-ray are over 30 degrees and around 18 degrees for the main thoracic and the minor lumbar curve respectively. The line drawn by the authors on the upper end plate of their selected upper end vertebra is undermeasuring clearly. The confusing and obfuscating lines have been removed and the radiographs are provided as scanned directly from films, by the Chief Medical Photographer, Arizona Health Sciences Center, University of Arizona, Tucson AZ.