Author's response to reviews

Title: Prevalence of hip dislocation in regions with and without a surveillance programme: a cross sectional study in Sweden and Norway

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Author's response to reviews: see over
BioMed Central Editorial
Abigail Quiniquini
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Dear Editor,

Thank you for the positive evaluation and giving us the opportunity to resubmit a revised version of our manuscript (MS: 1625273843576268) on “Prevalence of hip dislocation among children with cerebral palsy in regions with and without a surveillance programme: a cross sectional study in Sweden and Norway” by Areej I Elkamil, Guro L Andersen, Gunnar Hägglund, Torarin Lamvik, Jon Skranes and Torstein Vik.

We appreciate that both reviewers evaluated our paper as important in the field, and we also appreciate the remarks and advices given by the editor and reviewers. Below you will find our point-by-point response to these remarks.

Firstly, we sincerely apologise for an error we discovered during the revision of the manuscript. The error was related to a child who moved to the regular care area at the age of five years, and who had undergone adductor tenotomy for hip displacement at the age of two. This patient had a migration percentage of 78 at the age of four, and after arrival in Norway he developed a complete dislocation and was considered to be inoperable. It was our intention, as described in the original paper, to exclude this child, however, by mistake
another patient had been excluded. The latter patient had been operated on twice; first he underwent tenotomy later followed by pelvic osteotomy. Thus, the total number of children with dislocation in the regular care area should have been 18 (15.1%) instead of 19 (16%) as originally described. Nonetheless, the difference in prevalence of hip dislocation between the regular and the surveillance area is still highly statistically significant (p < 0.001). Beside this change, there were some accompanying minor changes in the numbers and p-values in Tables 2 and 3 as well as in the mean age at surgery in the regular care area, increasing from 7.5 to 7.6 years (page 2, paragraph 3 and page 10, paragraph 3). However, the main results and the conclusion are unchanged.

Our point-by-point comments:

**Referee 1:**

**Minor Essential Revisions**

1. P7, Para 2 beginning ‘In the Norwegian counties....’ and ending ‘...data were further analysed’. This would be better placed in Para 1 of the Results section

   ➢ **Comment:** The point is well taken and we placed the sentence in the Results section as well (page 9, paragraph 2).

2. P11, Para 2, Line 19: '(refs)' seems to indicate that some reference citations are missing from the text

   ➢ **Comment:** We apologise for that. We have added the appropriate references in the revised manuscript (page 13, paragraph 1).

3. P13, Para 1, Line 8: ‘...GMFCS III-IV... ’. Should this be ‘...GMFCS III-V...’?

   ➢ **Comment:** We apologise for the misprint, we corrected the roman number four to five.

**Discretionary Revision**

4. Adductor surgery. Although the main focus of the report is not on surgery, it would be helpful for the surgical reader to know what the authors mean by ‘adductor surgery’.
Was this a myotenotomy of adductor longus and gracilis only? Was psoas included? Or was it a more variable procedure? A couple of sentences to clarify this would be helpful.

- **Comment:** In the Swedish population soft tissue operations always included myotenotomy of adductor longus, gracilis and iliopsoas. In the Norwegian counties gracilis and psoas were more variably included. We have added this information (page 8, paragraph 2 in Methods section). For more information regarding surgery, see also comment 2 to reviewer 2.

**Referee 2:**

**Minor points:**

1. **Abstract:** Not sure this methodological design can define both groups of patients in a clear way
   - **Comment:** We have re-written the methods description in the abstract and we hope that it is clearer in the revised abstract (page 2, paragraph 2).

2. **Results:** it is not clear for me how many orthopaedic surgeries underwent each hip joint with total dislocation observed during the last follow-up.
   - **Comment:** These are the details of surgeries:
     - i. 7 children did not undergo hip surgery, 3 of them died.
     - ii. 9 children were operated on once:
       1. 5 children had adductor tenotomy (including psoas in 3)
       2. 1 child had varus derotation femur and pelvic osteotomy
       3. 3 children had femoral head resection
     - iii. 2 children were operated on twice:
       1. Adductor tenotomy (later) femoral head resection.
       2. Adductor & psoas tenotomy (later) varus derotation femur osteotomy & pelvic osteotomy.
   - **Comment:** We have added a summary of this information in the revised paper (page 10, paragraph 1).
3. Discussion: page 13 line 9 - there is: --- children with GMFCS III – IV…. Should be changed on “ children with GMFCS III – V

Comment: We apologise again for this misprint.

Editor's comments:

1. Add "among children with cerebral palsy" to the title

Comment: Thank you for the advice. We totally agree and have changed the title accordingly.

2. Do you have data on the number of x-rays per patient (probably only available for Sweden, but ideally it would be good to see it for both Sweden and Norway). There may be a down-side to repeated x-rays, especially in infants/young children, so it would be good provide whatever data you have on how frequent (particularly in first year) X-rays (or other imaging) is performed in these patients.

Comment: This is an interesting and important comment. In Sweden the surveillance programme has a full overview, as correctly assumed by the editor, due to the prospective design of the follow-up program. In this area the mean number of plain antero-posterior x-rays of the pelvis was 9 per child. However in Norway we are only able to account for the number of x-rays per child taken in two counties (covering 28 children). The missing details in Norway are due to the retrospective design and the successive introduction of digitalized imaging. In the two counties with complete data, we counted an average of five plain pelvic x-rays per child. However, this figure is complicated by the fact that in three of these 28 children a pelvic CT-scan was taken for surgery planning (twice in one child, and once in two children) while in Sweden, no child had a pelvic CT-scan.

Comment: We have added a comment on this in the results section (page 10, paragraph 5) and in the discussion (page 15, paragraph 2).

3. Page 5 - do you have a reference for the statement the "The medical services and the level of care for children with CP in Scandinavia are quite similar."
Comment: We have added a reference (ref No 18 in the revised paper) to a large Nordic project on children’s health and well-being in the Nordic countries with emphasis on children with disabilities.

4. (also, probably would be more accurate to say "...for children with CP in the different Scandinavian countries are quite similar.")

Comment: We agree, it is more accurate and we have rewritten this in the revised manuscript (page 5, paragraph 2).

5. Page 6 - what do you mean by "regular" radiological screening (annual? or more often?)

Comment: The hip surveillance programme in Sweden has a protocol for follow-up in different age groups. They take an annual pelvic x-ray for children with CP in GMFCS III-V to the age of 8 years then individual evaluation of the frequency of radiological examination. This information has been included in the revised paper (page 6/ paragraph 2)

6. Page 7 - would be useful to note in the paragraph that begins "In the Norwegian counties there were 494... that the (primary?) reason clinical data is missing for 27% of the eligible children is because parents didn't provide consent for review of medical records.

Comment: The primary reason for the missing data in 27% of the CP population is actually work overload for local doctors who were not able to complete the CP-registration forms. Only one family has refused to participate in the CP-register. In this particular study, where an active consent was not required, patients/parents were given the opportunity to decline ("passive consent"). However, only one family refused to have their child’s medical records reviewed. We added a sentence to explain this in the Methods (page 7, paragraph 3) and in the discussion (page 12, paragraph 2).
7. Page 7 - was the x-ray reviewer blinded to the country? Any info on reliability of reviewer ratings (inter- or intra-person variability, within this study)?

- **Comment:** Unfortunately, the x-ray reviewer was not blinded to the area, and we agree that this may be a limitation of our study.
- However, all radiographs in Norway were assessed by radiologists or orthopaedic surgeons and no additional hips were deemed dislocated by the x-ray reviewer of the present study.
- Moreover, we have some children with borderline high MP where we could not obtain more recent x-rays (see table). These patients were not classified as having hip dislocation. Thus, in the Norwegian population the proportion of children with hip dislocation is more likely under- rather than over-estimated.
- Regarding the Swedish population, x-rays were evaluated prospectively, and before the present study was planned. Thus, a low number of hip dislocations due to a biased observer are also not likely to explain our results.
- It can, on the other hand not be excluded that the x-ray observer overlooked some hip dislocations in Sweden. However, even if we assume a six-fold increase in hip dislocations (six dislocations instead of one) the prevalence in Sweden (4.6%; CI: 2.1 – 9.7) would still have been significantly lower than the prevalence in Norway (15.1%; CI: 9.8 – 22.6).
- We have added a few sentences on this in the revised version (discussion/limitations: page 12, paragraph 1).

<table>
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<th>ID</th>
<th>x-ray year</th>
<th>x-ray date</th>
<th>MP right</th>
<th>MP left</th>
<th>Reason for missing x-ray</th>
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<td>08.01.09</td>
<td>38</td>
<td>74</td>
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<tr>
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<td>13.04.05</td>
<td>76</td>
<td>58</td>
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<td>28.05.08</td>
<td>53</td>
<td>78</td>
<td>Operated 2009 RH</td>
</tr>
<tr>
<td>15</td>
<td>2009</td>
<td>13.08.09</td>
<td>78</td>
<td>80</td>
<td>Inoperable, they will not take new.</td>
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<tr>
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<td>29.09.04</td>
<td>24</td>
<td>74</td>
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</tbody>
</table>
8. How recent were the most recent x-rays (those used in the study), and were there any differences in "recency" of x-rays for Norway vs Sweden?

- **Comment:** In Norway, 87 children (73%) had an x-ray that was less than two years old when the study was done. In Sweden, 114 (84%) of the 136 children had an x-ray from the last two years. Again, if this difference had affected our results, it would most likely have been through an underestimation of hip dislocations in Norway.

- **Comment:** For the Swedish cohort, pelvic x-rays were taken in the last year only in children still at risk of dislocation. For children with low risk, radiological follow up stops when the radiographs show no progress, no displacement and fused growth plates.

- **Comment:** We have added a comment on this issue in the discussion under limitations (page 11, paragraph 2).

9. Minor edits: Abstract, methods section: combine first 2 sentences into one sentence (with comma after "region") Abstract, last sentence: follow-up (instead of follow up)

- **Comment:** Thank you for the comment, we corrected that.

We hope you will find that we have amended the manuscript according to yours and the reviewers’ suggestions, and that you will find the revised version acceptable for publication.

Lund 11/11/2011

Gunnar Hägglund