Author’s response to reviews

Title: Arm rotated medially with supination: Description of its surgical correction

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Author's response to reviews: see over
Reviewer’s report:
I would suggest some minor revisions:
1. the term "Mallet movements" is unclear- it certainly relates to the arm movements represented in the modified Mallet scale.

The term “Mallet movements” has been removed from the text on page 8.

2. the discussion includes repetitions on the described strategy. SHEAR and TT surgery have already been published by the author; I would like him to reference in the discussion some other surgical strategies, like Birch’s release surgery and Özkan’s contribution on prosupination: this would enhance the value of discussing other techniques with this novel and interesting approach.

We have expanded the discussion of alternate surgical strategies, and have cited Birch’s release surgery and Ozkan’s contribution on prosupination in the discussion section.

3. as the author did rather static bone corrective procedures, he should clearly state where he improved passive and/or active ROM.

Passive ROM was slightly improved and active ROM was not improved. The most important result of the surgery was improved positioning of the hand. A statement reflecting this was added to the results section.

4. I appreciate the author’s strategy, but so far long term results for TT surgery is lacking (the range here was 10 to 28 months). This does not diminish the value of the paper, but should be clearly outlined.

We have now made it more clear what results have been shown over a longer time frame and removed some statements based on unpublished and unreviewed data. (see also our response to the other reviewer’s comments 8, 17, and 27)

The presented ARMS association is a good clinical observation and the surgical strategy presented is logical, intended to correct a bad posture. The author could comment on the improvement of active motion and add some comments on the strategies for MRC and SD presented within the last 10 years, where he presents one further - and logic- option.

These comments on ROM and comparison with other strategies have been added to the results and discussion.
Reviewer’s report:
MANUSCRIPT REVIEW
“Arm Rotated Medially with Supination (the ARMS deformity): Description of a New Clinical Entity and Its Surgical Correction”
SUMMARY:
The article entitled “Arm Rotated Medially with Supination (the ARMS deformity): Description of a New Clinical Entity and Its Surgical Correction has been reviewed for publication in BMC Musculoskeletal Disorders. In addition, I reviewed a previous article entitled “Surgical Correction of the Medial Rotation Contracture in Obstetric Brachial Plexus Palsy published in the British Journal of Bone and Joint Surgery in 2007. After reviewing both these articles, my concerns regarding this article are expressed in the point by point basis. In its current form, the article is not acceptable for publication.
In summary, the article attempts to define a new clinical entity and its surgical correction. However, this is not really a new clinical entity and the patient numbers are quite small. The authors make a point that it is uncommon to have this combination of internal rotation of the shoulder and supination of the forearm. The authors may be right, but give very little evidence to support this position. Furthermore, the number of patients is quite small and the follow-up is even less. My particular concerns are expressed below:

ABSTRACT:
1. The abstract is written in structural form. The authors have previously described SHEAR deformity in previous publication. The authors now attempt to add an additional acronym for arm rotated medially with supination (a.k.a. the ARMS deformity). I think this acronym represents jargon and its better just to describe the deformity as a fixed supination deformity of the forearm. This would comply with previous publications regarding supination posture of the forearm.

We believe that the co-occurrence of the medial rotation at the shoulder and fixed supination deformity is an important phenomenon that deserves a singular description rather than the double-barreled moniker of “medial rotation contracture with fixed supination deformity.” We have chosen the term ARMS because it represents both the shoulder and forearm rotational issues that exist in one patient with a single term. Simply using the term fixed supination deformity of the forearm does not describe the interaction of the forearm deformity with the shoulder deformity (medial rotation contracture).

In deference to the reviewer’s opinion, we have removed the acronym ARMS from the title of this article. If the editors believe it is jargon, we can also remove this term (ARMS, arm rotated medially with supination) from the text and replace it with the above combination of terms (medial rotation contracture with supination deformity, or MRC with SD).
2. The methods section in the abstract should include more detail regarding the number of patients. This is listed in the results section but the demographics should be moved from the result section up to the methods section.

The description of patients was moved to the more appropriate methods section of the abstract.

3. The results section in the abstract indicates that glenohumeral anatomy was also improved, but not statistically significant. I not sure this belongs in the results section, but better left in the manuscript as it is non-significant.

We have removed this statement from the abstract. It is not an important point.

BACKGROUND:
4. On page 3, the authors indicate there are two common secondary deformities are the medial rotation contracture of the arm, and supination deformity of the forearm. The authors have previously stated that the supination deformity of the forearm is uncommon and here they stated it as common. This needs to be clarified.

We intended to state that each deformity alone is commonly diagnosed, but that the two deformities together are rarely seen. This has been clarified in the text with removal of the vague term “common” from this context.

5. The SHEAR deformity has recently been described by these authors and should be better delineated in the background. Most readers will be unfamiliar with the SHEAR deformity and a better definition describing this finding is necessary.

We have revised this section of the background to better introduce this deformity to the reader.

6. On page 4 the authors write that it is important to know that apparent loss of supination in patients with MRC is actually a manifestation of a misaligned humeral head related to the glenoid fossa, which rotates the wrist and hand into a resting position that is medial of neutral. This sentence is awkward and needs to be better written. The authors should state that the internal rotation contracture of forearm masks the inability to supinate.

We have revised the entire background section to focus and shorten it, and have also changed the wording in the sentences describing apparent supination ability in patients with MRC alone.

7. On page 4 the last paragraph uses the term “disability”. This should be changed to impairment.
The wording has been changed as suggested.

8. On the bottom of page 4 and the top of page 5, the authors indicate that triangle tilt operation has been shown to be anatomically and functionally superior to traditional techniques. This sentence has very little evidence and is based on a single article in 2007 that was reviewed. The article makes no comparison to other techniques and therefore this statement is too strong.

This section has been reworked to include only published evidence for efficacy and remove the opinion of the technique's outcome.

We do have evidence for the anatomic improvement and continued functional gains, but these two results are currently under peer review and have not yet been published.

9. On page 5 the authors attempt to define the imbalance across the forearm resulting in a fixed supination posture. The standard anatomy needs to be re-reviewed. The pronator teres and pronator quadratus are not solely innervated by C7. Certainly the pronator teres is the C7 muscle, but the pronator quadratus is innervated by the anterior interosseous nerve, which has substantial lower C8-T1 innervation. Furthermore, the authors needs to be better explain why the patients listed in Table 1 had primarily upper trunk involvement, especially patients 1-5.

We acknowledge that the pronator quadratus does receive innervation from the lower roots (C8-T1). The patients we have seen, however, show no other signs of lower root injury and must thus conclude that it is possible for a primarily upper root injury to cause supination deformity. We believe it is possible for this to occur when the injury to C7 is transient and the biceps (C5-C6) recover power before the pronator teres (C6-C7), leaving it unable to overcome the supination force of the biceps. Although the pronator quadratus may also be contributing some pronation force in this scenario where the lower roots are unaffected, it is apparently not great enough to prevent the imbalance from developing into a fixed supination deformity. A study by Gordon et al of EMG signal during supination and pronation [21] suggests that “during resisted pronation [in our case: early biceps recovery of supination power], the pronator teres muscle is the primary agonist.”

We have moved this section to the discussion.

METHODS:
10. The methods includes a series of patients from 2005-2006. In a previous publication the authors had patients between February and August of 2005 that underwent triangular tilt operation. Were any of these patients included in the first publication? If so, this needs to be mentioned.
There are only 3 patients common to these two studies. The published study gave results only before forearm surgery was performed, so the pre-surgical data are common between the two studies, but the post-surgical data are not.

11. On page 6, the authors indicate that the sequence is recognized clinically by the parallel positioning of the volar surface of the forearm to the anterior surface of the arm. I'm assuming that they are talking about the supination posture of the forearm as it relates to the arm. This needs to be clarified.

This section has been rewritten to improve clarity.

12. The authors indicate on page 6, that to the untrained eye the hand is a neutral position and symmetric with the uninjured side. This is condescending and needs to be toned down.

We have changed the phrasing to remove any judgment of the observer’s training or ability, and focus on the appearance of the child.

13. On eight of the fourteen patients had sufficient follow-up data for the study. In addition, when you peruse Table I there is even less follow-up available. Patients 3, 5, 7, and 8 were all missing radiographic data that would be important to substantiate the authors’ conclusions.

The radiographic data were not the key to this study. We were unable to obtain post-operative follow-up for all patients. If the reviewers and editors agree, we are happy to remove this post-operative data from Table I (and sections in the results regarding post-operative radiology marked in green), and to also remove the final figure (7 in the original submission, or 8 in the revision).

The important data to note are the post-operative functional results. We include the pre-operative radiology, because it is critical to pre-surgical planning.

14. A variety of patients had previous surgeries including mod quad, nerve grafting, posterior glenohumeral capsulorrhaphy and biceps tendon lengthening. The authors relate the improvement in glenohumeral alignment in their small number of patients to the triangular tilt. However, the posterior glenohumeral capsulorrhaphy and modified quad may have changed glenohumeral alignment. This needs to be at least discussed in more convincing terms.

All of these operations were performed before the pre-operative scores were taken. This has been clarified in the methods section. We agree that some benefits to glenohumeral alignment can be obtained from PGHC or Mod Quad procedures. The improvements, however, already occurred before the data presented in this paper.
15. On page 7 the authors discuss measurements of glenohumeral joint and SHEAR deformity. The authors spent a great deal of time on how to measure subluxation and how to measure glenoid version. However, no time was spent on how to measure SHEAR deformity. When one reads the 2007 article, there is a similar critique. The authors at least need to have a diagram demonstrating the measurement technique for SHEAR deformity.

The methods sections describing the measurements have been revisited, and balanced. We also prepared a figure showing measurement of the scapular elevation that describes the extent of the SHEAR deformity (added as Figure 3).

16. The authors indicate on page 8 and 9 that they use a variety of scales and surgical technique designed by the senior authors. I don’t think that the senior authors should be listed as Nath’s modified Mallet scale, but rather just modified Mallet scale. In addition, the adding of a variety of subcategories may affect the reliability of this measurement. Have the authors done any reliability measurements on these additional subscales?

We have removed phrasing related to “Nath’s modified Mallet scale.” We used the established modified Mallet scale to evaluate patients and then looked at several other functional parameters related to the forearm position and supination ability. The modified Mallet scale scores shoulder function, and it was key to include some assessment of forearm function for these particular patients. It was unintentional to imply that a new measurement or scale had been devised.

17. Similarly, on page 9 the authors indicate that the lead author has developed a novel and effective osseous procedure, named the triangle tilt. I agree this is a novel procedure with single publication has been noted. I think the authors should simply state that a novel osseous procedure called the triangular tilt has been developed, which has shown an earlier promising results.

We have changed the wording here to remove any opinion and let the published results speak for themselves.

18. On page 10, the authors discuss the second stage surgical correction was a derotational osteotomy of the radius and intramedullary pinning. There are no details concerning where the osteotomy was performed or radiographs. If the authors main point is to describe the combination of internal rotation contracture of the shoulder and fixed supination deformity, than this would be valuable information to the readership.

We agree, and have expanded the description of the forearm procedure.

19. The authors show a variety of pictures of patients before and after surgery. The authors did include one patient humeral osteotomy based upon her age. The authors comment on page 6 that the osteotomy was done because of her
advanced age. If the authors should define why they have an age cut-off? This would be useful information.

The surgical approach is not taken in older patients because of age-dependent ossification. A short sentence was added.

20. On page 12 the authors have a footnote that needs to be referenced. It is in the second paragraph, and needs to be taking out of ( ).

Done. Thank you for catching this.

21. On page 12 the authors indicate the angles of supination deformity significantly increased, as did the Mallet scales for supination also increased. This is confusing and I may have a misunderstanding. My understanding is once the arm was placed into external rotation, the forearm was corrected into pronation. If one accesses the Mallet scales for supination, I don’t see how these can improvement following a rotational osteotomy into pronation. Please explain.

You are correct, the arm was placed into external rotation and then the forearm was corrected to neutral (away from supination).

We mislabeled the columns in the original manuscript and have corrected this. The Mallet scores were for External Rotation and not supination.

22. Radiological criteria – Table 1 show the results of CT and MRI evaluation. Table 1 also shows considerable missing data. There so little data within Table 1, that I’m not sure any conclusion can be made. In fact, patient 1 showed a percentage increased in SHEAR following surgery. Patients 2, 3, 4 and 6 decreased and the remaining patients have no values at all. I think the authors need to temper their radiologic results.

Please see our response to query 13.

23. Both the discussion and introduction are too long. Both sections need to remove some extraneous material and get to the point. The introduction section should simply introduce the problem and the discussion section should compare the previous published results.

Both introduction and discussion sections have been reworked to shorten and focus the paper. Comparison with published results and techniques has been moved to the discussion.

24. On page 14, the authors indicate that they described for the first time the simultaneous occurrence of the medial rotation contracture and supination deformity of a group of patients with a history of brachial plexus birth palsy. I don’t think that this is the case. The authors would better indicate that they make
note of bringing attention to their simultaneous occurrence. Certainly, we have all seen this combination of deformities in our clinical population.

We have only seen one description of this combination of deformities in the literature [27]. Otherwise, the literature focuses on shoulder alone or forearm alone with little consideration for how the two deformities combine, and create an altogether different functional deformity. We do not doubt that other practitioners have observed this combination in their own patients and treated them appropriately. We have, thus, reworded this section to better acknowledge earlier contributions.

25. Again, I think the acronym ARMS needs to be removed. It does not provide any additional information, but rather is just a jargon.

Please see our response to query 1.

26. On page 15, the authors indicate that there are variety studies that correct supination posture of forearm. If one of the points of the article is that rotational osteotomy allows for correction of this deformity, then this needs to be expanded. The authors need to provide more information as to the following:
   a. Radial osteotomy was performed
   b. Techniques of fixation
   c. Amount of correction obtained.
   d. Information regarding how they determined how much correction was necessary.

We have added this information to the methods section.

27. On page 15, the authors indicate that they have successfully used a triangular tilt procedure to correct the shoulder in hundreds of patients over the last four years. The previous report in 2007 only includes forty-four patients and the others have not undergone peer review. Therefore, this statement needs reference only those patients that have undergone adequate peer review or follow-up. If the authors truly have hundreds of patients that have undergone the triangular tilt, then those numbers should be reported in a separate manuscript.

This statement has been revised to reflect only published results without reference to un-peer-reviewed work.

CONCLUSION:
28. The conclusion section is fairly well written. I agree that the shoulder should be corrected prior to the forearm. This section is the best written section of the entire manuscript and should be used as a platform to rewrite the introduction and the discussion sections in a shorten fashion.

Please see response to query 23. Thank you for your input.