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## *Severity Grading Important in Radial Dysplasia Research*

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Murphy et al. should be congratulated for their comprehensive review of the literature and evaluation of results achieved in the treatment of radial dysplasia. However, as pointed out by the authors, most studies on this topic are hobbled by quality issues. The designated evidence level of IV for this review and meta-analysis corroborates that fact.

Improving future research quality is difficult when rare diseases such as congenital malformations are studied. Furthermore, the biological nature of any congenital anomaly in general, and radial dysplasia specifically, means that there are always marked differences between patients. The lack of patient subgrouping is a pervasive weakness of many studies, and Murphy et al. acknowledge this by writing, “Most of the primary studies did not report individual patient data, or stratify their results by severity of the radial dysplasia.”

Murphy et al. do not define or clarify the term “severity.” Severity grading is very important when evaluating results, as it allows the grouping of comparable patients. I am concerned that many readers are not familiar with the severity grading of radial dysplasia, and to improve future research, it would be important to always use severity grading to divide the patients into subgroups. This will facilitate acquiring valid and generalizable data that can be readily interpreted, universally understood, and easily compared to other results. Such a grading system has been published (1).

Additionally, the interpretation of one of the cited studies (reference 27) needs clarification. Murphy et al. say that “the hand-forearm angle in that study was recorded after ‘gentle stretching’ of the hand…The hand-forearm angle in that series is therefore not directly comparable with those in other series.” In fact, in the study cited in reference 27, the gentle stretching was used only in the preoperative phase. Preoperative stretching was performed to characterize the wrist tightness and determine the ease of wrist deformity correction. The stretching was *not* used when recording the long-term results. This clarification is

noteworthy because all the long-term results were measured taking normal PA and lateral radiographs of the resting hand, which, given a stable position of the radial-sided graft, may have allowed for even more sensitive and reproducible measurements of radial deviation or forearm angle than in other studies where there was no stable structure on the radial side of the wrist. I agree with the authors that, in general, “radiographic measurements are prone to errors,” but they are widely available and currently one of our best and most cost-effective tools for skeletal assessment.

## References

1. S.K. Vilkki: Severity grading in radial dysplasia The Journal of Hand Surgery (European Volume) 2014, Vol. 39E(9) 977–983

Conflict of Interest: None Declared

## Article Author Response

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***Article Author(s) to Letter Writer(s)***

**A Core Outcome Set Facilitates Both Severity Grading and Outcome Comparison**

We thank Professor Vilkki for his letter in response to our paper. We agree that defining the severity of disease is important for the fair comparison of outcomes between series, especially in conditions as heterogenous as radial dysplasia. Like any review, ours was limited by the quality of the primary literature. This meant that we were unable to stratify patients’ outcomes by severity, as these data were not uniformly reported in the original literature; nor did we manage to obtain them by correspondence with the authors of those papers.

Professor Vilkki also supplies a welcome clarification about how he measured the postoperative hand-forearm angle in his series; this brings his method of postoperative (but not preoperative) measurement into line with the other papers we included. The fact that this was not clear from either the original paper or our correspondence with him during our review highlights the need for a standardised approach to outcome measurement.

Happily, both these points are addressed by the forthcoming International Consortium for Health

Outcomes Measurement (ICHOM) standard set on congenital upper limb anomalies (1). This provides a core set of outcomes established using robust consensus methodology by an international expert panel, who were informed by both the literature and patient perspectives. It also provides standardised timepoints and instruments to measure these outcomes. By collecting data pre- and postoperatively, it will allow both stratification by disease severity and fair comparison of the effects of treatment. We recommend its use in future studies on radial dysplasia and on congenital upper limb anomalies more widely.

## References

1. ICHOM standard set on congenital upper limb anomalies. http://www.ichom.org/medical-conditions/congenital-upper-limb-anomalies/