To the Editor:

The recent article by Callanan et al.[1] was intended to explore adverse psychological effects, reproductive uncertainty, and risk perception in 52 "carrier by non-carrier" CF parental pairs. The paper included a description of a five point scale used to grade the perceived probability that a child of these parental pairs could be a CF carrier or affected by CF. The scale is described as ranging from ". . .extremely high (1) to extremely low (5)." They displayed an average value for the results of that scale in the various groups of the study in Figure 1. Although the methodology for calculating the average was not specified in the methods section, it was presumably an arithmetic mean. Averages can only be determined for cardinal numbers, not for numerals that are ordinal labels for categories. An "average" value has no meaning because the differences between successive categories are not necessarily equal. Because such an average has no mathematical meaning, the performance of a *t* test also generates a meaningless result. Thus no conclusions can be drawn from the analysis of these data in this paper.

A second issue is that of the small sample size in this study. It would have been useful for the authors to have performed and described a power analysis prior to the execution of the study or at least describe the precision (confidence intervals) of the negative results after they were determined. The paper is essentially a description of a negative study. There are two common explanations for this result: either the groups do not differ or the study was too small to be likely to find a clinically significant difference. The latter interpretation would mean that this paper may have nothing to tell us about whether or how to implement CF screening.

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Reference

1. Callanan NP, Cheuvront BJ, Sorenson JR. CF carrier testing in a high risk population: Anxiety, risk perceptions, and reproductive plans of carrier by "non-carrier" couples. *Genetics in Medicine* 1999;1:323–327.

In Response:

Dr. Biesecker is correct that there are more technically appropriate statistical tests than the *t* test for the data we presented in Figure 1 of our article.[1] We have rerun the data using the Wilcoxon signed ranks test, a test designed for ordinal data.[2] The *p* value for the "children carrier" comparison is 0.451 and the *p* value for the "children affected" comparison is 0.581. The results are consistent with those obtained using the *t* test. Regarding Biesecker's comments on statistical power, our Ns were very small. Obviously with our sample size we could not detect small or minor changes, and we appreciate Biesecker's concern about cautiously interpreting the data. This said, the data suggest to us that concerns about very severe negative consequences of CF carrier testing in carrier by test negative couples may be overstated.

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References

1. Callanan NP, Cheuvront BJ, Sorenson JR. CF carrier testing in a high risk population: Anxiety, risk perceptions, and reproductive plans of carrier by 'non-carrier' couples. *Genetics in Medicine* 1999;1:323–327.
2. Hollander, M and Wolfe, DA, (1972) Nonparametric Statistical Methods, New York, John Wiley and Sons.