The meta-analysis by Osborn and colleagues\(^1\) of neurodevelopmental studies in children with isolated sagittal craniosynostosis offers a comprehensive review of findings and highlights the limitations of existing research. The authors identified 32 peer-reviewed studies comparing the neurodevelopment of children with sagittal synostosis with that of control participants without synostosis or with test norms. Their review\(^1\) included studies conducted in the US, Europe, Asia, and Australia and represented 1422 individual participants. As the first meta-analysis of this body of research, the study\(^1\) makes an essential contribution to the literature and may guide future studies.

The most striking result from the analysis\(^1\) is the heterogeneity in findings, showing that neurodevelopmental outcomes for children with sagittal synostosis range from worse to slightly better than those of control participants or test norms. They observed this variability regardless of study timing (eg, before vs after surgery) and neurodevelopmental domains assessed. Disparate findings may be expected given the small samples in published studies, variable outcome measures used, and the reliance on test norms instead of control groups for comparison (ie, only 9 of the 32 studies included a control group). As the authors point out,\(^1\) the comparison to test norms is problematic because of well-documented Flynn effects resulting in inflated scores. This effect creates variability across studies that relied on test norms depending on how current the normative data were for their comparison. In addition, Osborn et al\(^1\) correctly note that comparison to test norms assumes that patient samples are demographically similar to published norms and that any differences can be attributed to their medical condition. This assumption is often flawed, and unfortunately, the studies reviewed often did not include the data needed to examine the influence of demographic characteristics.

Another gap illustrated by this review\(^1\) relates to the reporting on surgical and other clinical care that children receive. Factors such as age at surgery, type of surgical procedure (eg, cranial vault reconstruction, endoscopic assisted), and duration of anesthesia exposure have been hypothesized to affect neurodevelopmental outcomes.\(^2-4\) Similarly, children with sagittal synostosis may receive developmental interventions in the community or school expected to improve outcomes. Unfortunately, reporting on these factors was insufficient in published studies for inclusion in this meta-analysis.\(^1\) Collecting these data is complicated and time-consuming and is ideally done prospectively rather than relying on abstraction from medical records, which are often inconsistent in reporting. Further study of clinical care parameters may identify modifiable factors to improve care and clarify health disparities and potential remedies. For example, recent studies\(^5-7\) suggest that race, ethnicity, and socioeconomic status are associated with the duration and timing of surgery for children with craniosynostosis. Specifically, White, non-Latino children undergo surgery at an earlier age than their peers of other racial and ethnic groups. Timely access to care for craniosynostosis is critical, resulting in shorter operations and allowing for surgical options that must be completed at a younger age (eg, endoscopic-assisted surgeries).\(^7\)

Ideally, the meta-analysis by Osborn et al\(^1\) would motivate efforts to establish an agreed-upon set of demographic and clinical parameters for future research and large-scale studies to better understand the associations of craniosynostosis with neurodevelopmental outcome. In practical terms, these data would provide parents of children with sagittal synostosis the information needed to make decisions regarding their child's care.
Heterogeneity in Neurodevelopmental Outcomes Associated With Isolated Sagittal Craniosynostosis

REFERENCES