effects may be particularly notable in certain populations, such as those with neurological disorders. Using sensation as an example, level 1 in HUI indicates "able to see, hear and speak normally for age," whilst level 2 "requires equipment to see or hear or speak." It is plausible that an aphasic or dysarthric subject could be rated as level 1 because he/she does not use an assistive device. Thus, the HUI may not be appropriate for populations with severe disabilities since floor effects are unlikely given the scale's design.

In summary, Glaser and colleagues reach overly optimistic conclusions about the utility of the HUI in paediatric brain tumour patients. The fundamental psychometric issues outlined above need to be addressed in more detail before the instrument can be recommended for use in clinical studies.

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# Response from D. Feeny, *et al.*

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We thank Dlugos and colleagues for raising important issues concerning the report by Glaser and associates [1] about the usefulness of the Health Utilities Index Mark 2 (HUI2) and Mark 3 (HUI3) systems in the context of survivors of tumours of the central nervous system in childhood. We will deal with each of the points raised in their letter.

It is agreed that the assessment of test–retest reliability of the modified question used to determine HUI2 and HUI3 levels of emotion is not evidence of test–retest reliability for the HUI. Indeed Glaser and associates [1] 1999 indicate (p. 260) that the modification is 'inadvisable'. Evidence on test–retest reliability for HUI is found elsewhere [2–4]. Evidence on inter-rater reliability for HUI in this context is available also [2,5–8] whilst evidence on the construct validity of HUI in the context of brain tumours or other conditions involving cognitive impairment has been published [2,5,8–20].

Dlugos and colleagues do not state what level of agreement among assessors they expected to find. Perhaps they expected a high level of agreement. Yet abundant evidence for paediatric and adult measures of health-related quality of life (HRQL) indicates that substantial and systematic differences in opinions among assessors are not at all uncommon. Herjanic and Reich [21] administered simultaneously a structured diagnostic interview consisting of 168 questions to 307 children and their mothers. High agreement (kappa $\geqslant 0.50$) was observed for 16 (9.5%) of the questions; middle agreement (kappa 0.30–0.49) was observed for 30 (17.9%); low agreement (kappa $\leqslant 0.29$) was observed for 122 (72.6%). Many of the latter dealt with factors that are not readily observable, such as emotional health. We feel that our lack of surprise in not observing high agreement is well founded; evidence on a lack of agreement among assessors in paediatric settings using other instruments is substantial [22–25].

Dlugos and colleagues argue that aphasic or dysarthric subjects might be categorised as level 1 sensation in HUI2. Indeed it is possible, but it is very unlikely if the data were collected using sensation-related questions from the standard 15-item HUI2/3 questionnaire as in Glaser and associates [1]. It is unlikely because the level for HUI2 sensation is not determined directly from one question of the 15-item questionnaire, but rather from previously determined HUI3 levels of vision, hearing and speech. To be classified as level 1 sensation, a subject must have already been classified as level 1 vision and level 1 hearing and level 1 speech, based on combinations of responses to 6 questions. It seems highly unlikely that these types of subjects would be classified as level 1 speech in HUI3.

Dlugos and colleagues express concern about a suspected ceiling effect in that 21% of patients had no

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problems. Dlugos and colleagues do not state their expectation of prevalence for problems in this population of brain tumour survivors. Using HUI2, Saigal and associates [10] found that 34% of adolescent survivors of extremely low birthweight (a known group with a high burden of morbidity) were without problems, compared with 58% in a control group matched for age, sex and socio-economic status. Glaser and associates [1] results do not appear to be anomalous.

Finally, we agree that the results reported by Glaser and associates [1], and interpreted in isolation, do not support a definitive conclusion that HUI2/3 measures are reliable and valid in the context of assessing survivors of brain tumours in childhood. As Streiner and Norman (p. 157) [26] note, the validation of any instrument involves the ongoing accumulation of evidence. However, the findings of Glaser and associates [1] are consistent with results from a number of other studies involving survivors of brain tumours in childhood which lends support to their conclusions.

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