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We have to talk about HRQOL

(This is a commentary on the article by Ghotra *et al.* (1) in this issue)

"Happiness does not depend on outward things, but on the way we see them." Leo Tolstoy, *Childhood, Boyhood and Youth*.

Can we ever know the mind of another? A few years ago, the Study of Participation of Children with Cerebral Palsy Living in Europe (SPARCLE) researchers published a seminal paper (2) on the "quality of life" of young people with cerebral palsy (the reason for the quotation marks will become clear). The unarguable message of this work was that - so long as they were free from pain - these young peoples' self-assessments of their emotional well-being matched those of the general population, despite sometimes very severe objective impairments. In another object lesson in this paradox, the introduction over the last couple of decades of non-invasive ventilation for boys with Duchenne muscular dystrophy - which had been a highly controversial proposal seen as "prolonging wretched lives" - was supported by a wealth of evidence in the boys' own voices that they valued their existence and situation (3). Life expectancies have almost doubled since as a result of the widespread acceptance of this intervention.

These are important lessons in the value of hearing the patient's voice. Objective (externally observable) assessments of a person's health can differ widely from subjective valuations of the same health state, and these different viewpoints can lead to very different priorities both in intervention development (what about my situation do I most want changed?) and measurement (what aspects of my situation should a meaningful health status measure reflect?). The Patient Reported Outcome revolution in health care research is long overdue.

In this issue of Archives Ghotra *et al.* report on "health-related quality of life" (HRQOL) in children after paediatric arterial ischaemic stroke and show that it is lower than in healthy controls (1). The unthinking reader might think this is hardly surprising and turn the page, but in doing so would miss important underlying questions.

If we are to make much progress in our thinking here we need to agree our concepts. There is an uneasy truce between two approaches to thinking about health status that whilst not incompatible do not always sit well together. The World Health Organisation (WHO) International Classification of Functioning (ICF) (4) emphasises Participation ("involvement in life situations") as a key priority of any health intervention. A major insight of the ICF theoretical framework is that Participation arises from important interactions between (i) processes located within the child (referred to under headings of Activity and Function) and (ii) properties of her setting or environment (in the broadest sense: not just dropped kerbs, but the attitudes of her classmates, the adequacy of the family benefits system, etc, etc). Even after the enormous effort that went into the development of the ICF there remain residual differences in views of what exactly is meant by Participation, but most would agree that it is (or at least includes) an objective

(externally verifiable) assessment of involvement in society. This in turn implies shared, societal norms for the evaluation of that health state: a boy with autism may have low levels of participation and indeed prefer things that way, but from an ICF perspective his Participation is still low (5). The SPARCLE researchers (2) wanted to highlight how different Participation could be from a child's subjective (personally reported) emotional well-being.

As Ghotra *et al.* rightly start their paper by stating, "a clear and thorough understanding of HRQOL...is especially important to guide health-improving interventions" but I've been using quotation marks around the "quality of life" phrase as it remains an even slipperier concept than Participation (6).

There are at least two important, and related, areas in which instruments which all purport to measure "quality of life", differ. The first – and most fundamental – is the extent to which they focus on aspects of an individual's situation that are in principle observable by another. Many measures, including the PedsQL used by Ghotra *et al.* include items such as whether an individual has "problems running". This, in ICF terms, is a question about Activity, verifiable by any observer. The second important area in which instruments differ is whether and how the salience of that problem to the individual is captured. As originally conceived, HRQOL's distinctive selling point is precisely that it gives the self-reported salience perspective ("I do have problems running, but I'm OK about that"): this is what was so valuable in the SPARCLE (2) and Duchenne (3) papers. Researchers are increasingly using the term "subjective well-being" as a synonym for this particular meaning of HRQOL. But in paediatric research (and other settings where hearing the patient's own voice is might be anticipated to be a challenge) there is a temptation to seek proxy reports. What are we to make of a proxy-reported HRQOL, however well-intentioned and well-informed the reporter might be? The purists would say that proxy-reported HRQOL is a contradiction in terms. Pragmatists might say that it's a valuable perspective, though perhaps it should have a different name to clarify its non-personal perspective. Proxy-reported "HRQOL" is generally, parent- or carer reported Activity and Participation.

Some of these issues come through in Ghotra *et al.*'s findings. Where both parent and young person perspectives are jointly available, perspectives differ. Parents report lower overall scores and emphasise emotional issues; the young people report their neurological impairments, although we have little sense of what this *means* to them. It would have been interesting to have had more qualitative exploration of these differing perspectives, but one interpretation could be that parents are making the same error as those physicians caring for boys with Duchenne thirty years ago (albeit to a much lesser degree). Previous work by the same group (7) reported that neonatal-onset stroke was associated with poorer "HRQOL" than stroke occurring later in childhood. But it is worth recalling these neonatal-onset stroke survivors would have been eligible for Colver *et al.*'s cohort (2) (i.e. neonatal stroke is an important cause of cerebral palsy). Why do Ghotra *et al.* have a gloomier view of HRQOL after neonatal stroke than Colver *et al.*? The resolution of this contradiction (and others referred to in Ghotra *et al.*'s Discussion) is likely in part to reflect the fact that PedsQL and KIDSCREEN (the tool used by Colver *et al.*) are really measuring different constructs (KIDSCREEN is much more weighted toward "subjective well-being") (6). It's tempting to hypothesise that age at onset, and particularly the extent to which acquisition of a stroke in later life disrupts a previously-established self-identity, lies at the heart of some of the paradoxes that research of this nature identifies.

Again, Participation arises from the interactions between processes located within the child and properties of her context. This is important because it determines where efforts to improve Participation are best directed: change the child, or change her world? One slightly tongue-in-cheek conclusion that could be drawn from SPARCLE's work is that the most effective intervention one could propose for children with cerebral palsy is to move them all to northern Denmark!(8). Distinctions emphasised in the ICF between attributes of the child ("problems running") and attributes of her setting ("being teased at school") tend to be blurred in HRQOL measures such as the PedsQL . It is perhaps unsurprising that Ghotra *et al.* find socioeconomic status (SES) is an important determinant of HRQOL independent of intrinsic neurological impairment. Drilling down into precisely how deprivation is affecting participation for these children might identify important new candidate interventions.

Finally as a neurologist and neuro-rehabilitationist I cannot let one assumption pass unremarked. The authors justify their twelve month assessment time point on the basis that "peak recovery" after stroke will have occurred within a year. Such statements can become self-fulfilling prophecies. We have shown important gains in gross motor function years after acquired brain injury including stroke (9), and there is strong evidence that we are simply not providing the rehabilitation doses required to effect meaningful change after acquired brain injury. Nor is it helpful to think of recovery after brain injury in children as progress to a stable plateau that is then maintained indefinitely: as with cerebral palsy, the effects of a brain injury in early life evolve over time as ongoing development interacts with a disrupted brain architecture (10).

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