

Ethics in paediatric neurology

Bernard Dan^{1*}, Peter L. Rosenbaum², Gabriel M. Ronen³, Jennifer Johannesen⁴, Eric Racine⁵

¹ Professor of Neuroscience, Université libre de Bruxelles (ULB), Brussels; Director of Rehabilitation, Rehabilitation Hospital Inkendaal, Vlezenbeek, Belgium; e-mail: bernard.dan@ulb.ac.be

² Professor of Paediatrics, McMaster University, Hamilton, Ontario, Canada; e-mail: rosenbau@mcmaster.ca

³ Professor of Paediatrics, McMaster University, Hamilton, Ontario, Canada; e-mail: roneng@mcmaster.ca

⁴ Author and Patient Advocate, Toronto, Ontario, Canada; e-mail: jen.johannesen@gmail.com

⁵ Director, Neuroethics Research Unit, Institut de Recherches Cliniques de Montréal (IRCM); Associate Research Professor, Department of Medicine, Université de Montréal; Adjunct Professor, Department of Neurology and Neurosurgery, McGill University; Affiliate Member, Department of Medicine, Division of Experimental Medicine, and Biomedical Ethics Unit, McGill University, Montreal, Quebec, Canada; e-mail: eric.racine@ircm.qc.ca

*Corresponding author

ABSTRACT

The field of paediatric neurology has evolved considerably in parallel with the enhanced longevity of children with neurodevelopmental conditions, advances in technology, and cultural changes, notably about the meaning of living with a disability. New concepts have been promoted on health and quality of life as well as beyond the context of healthcare in the legal realm leading to novel approaches to the child in society, to disease and disability, professional-family relationship, and the role of the paediatric neurologist. This has brought into focus several issues including dealing with uncertainty of diagnosis and prognosis, harmonising the goals of clinical care between different professions and stakeholders, implementing shared decision-making, supporting evidence-informed practice, research, and partnerships with families and young people with neurological impairments. The principles of bioethics, namely autonomy, beneficence, non-maleficence and justice, resonate well with paediatric neurology. Together with a relational and contextual approach to clinical and ethical practice, they help in identifying and addressing ethical issues on daily (clinical) life. They are also pillars for revisiting the interface between good clinical practice and empathic ethical practice.

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Medical ethics has long historical roots that have grown strong through countless individual and societal challenges. From centuries past, medical ethics was often captured in three Latin words freely adapted from the Hippocratic Oath: *primum non nocere* – ‘Do no harm’. In fact, the Oath rather states the following: ‘I will, according to my ability and judgement, prescribe a regimen for the health of the sick; but I will utterly reject harm and mischief’ [1]. Still, ‘*primum non nocere*’ aptly epitomises the non-maleficence principle that currently guides ethical reasoning and clinical practice across clinical professions alongside three other principles, namely respect for autonomy, beneficence, and justice [2]. These ethical principles have developed a particular resonance in paediatric neurology because of new challenges that have emerged over the past few decades, such as enhanced longevity in many childhood-onset neurological conditions and technological advances with regard to both diagnosis and management [3-5]. In parallel, marked developments have occurred in the cultural perception of the child in society, as well as the concepts of disease and disability, thereby creating a need to revisit the equilibria struck between those principles.

HEALTH

The concept of health was long defined as a state of complete physical, mental and social well-being [6]. It has shifted towards a more dynamic, personal perspective with an emphasis on function. Wilder Penfield, American-Canadian pioneering neurosurgeon, for example, explored at length the goals of treatments with his patients before surgery [7]. Health is now rather seen as a person’s ability to function in a manner that is acceptable to themselves, in ways that include their expectations and values, even in the presence of impairments [8]. The very notion of self-evaluated functioning has come to complement the more objective characterisation of neurological impairment [9,10]. Beyond asking what is wrong, clinicians now consider what is right, what works, what this child’s and family’s strength are, and what goals and interventions they would value.

QUALITY OF LIFE

Accordingly, the issue of quality of life has become central [11] in clinical evaluation and management planning. Some studies have thus demonstrated similarities in self-reporting

of various aspects of quality of life between selected groups of patients with significant neurological impairment and unimpaired peers [12-14]. This has led to a recognition of the so-called 'disability paradox' that many individuals with serious and persistent disabilities report experiencing a good quality of life whereas to most external observers it would appear as undesirable [15]. These studies do not suggest that high levels of quality of life should be expected in the presence of impairment but rather that proxy stakeholders, including paediatric neurologists, cannot confidently anticipate an individual patient's self-perceived life quality by the level of impairment alone.

CHANGING SCOPE

Societal changes that have influenced the ethical approach in paediatric neurology also concern the relationship and reciprocal expectations of professionals and families, and thus the role of paediatric neurologists. The need to plan and implement increasingly complex investigations and therapies, and to counsel individuals and families about them in this new relational setting, have resulted in increased responsibility on the part of paediatric neurologists. The scope of effective follow-up has also extended from foetal life, as a result of advances in antenatal diagnostic and even therapeutic possibilities, through adolescence and transition to adult care, thanks to improved life expectancy and better clinical services.

EVIDENCE-BASED PRACTICE

Evidence-based medicine has also become increasingly important in paediatric neurological practice. This approach implies that practitioners maintain continuing education and use their clinical expertise in retrieving, interpreting and applying the results of scientific studies to individuals in everyday practice. In many domains of paediatric neurology, the evidence is currently patchy [16]. This situation implies a responsibility for clinicians towards the development of clinical research in our field to accumulate and strengthen evidence to understand, prevent and treat disease, as well as preserve life-long health. Extrapolation from studies conducted in adults often has limited relevance and may be harmful, as in stroke management. Occasionally, there may be a tension between the two roles, of clinical practitioner and researcher. Healthcare duties of the clinician are directed towards the individual patient, while the scientist's obligation is to contribute to the growth of knowledge. In practice, whenever a conflict is identified between the two roles, the patient's inalienable right to the best available clinical management must always take precedence. Clinical research in children raises a number of more specific ethical issues [17]. Children show specific vulnerability and require protection. They also have the right to the best standard of healthcare, to information and participation in decision-making. The issue of consent following appropriate information remains fundamental to the conduct of research. Over the past decades, young people and parents have increasingly been involved in all aspects of research. Ethical and legal codes describe proxy-consent on behalf of children lacking the capacity to provide appropriately informed consent, i.e. the ability to understand what is involved and the consequences. Consent is obtained from a parent, or legally authorized representative, on behalf of the child. Yet, the child's assent, i.e. active affirmative agreement should also be sought from the child [18]

just line with clinical practice [19]. This occurs in the context of relationship between the physician, the parents and the child in which child medicine is carried out, which commonly involves moderate paternalism on the part of the physician [4].

In addition, ethical issues have been identified with respect to the risk for shortcomings of the right to health following studies in which children participated, particularly in resource poor settings, e.g. post-trial access to HIV pre-exposure prophylaxis [20]. In 2013, this requirement was integrated into the World Medical Association's Declaration of Helsinki that suggests ethical principles for medical research involving humans: *"In advance of a clinical trial, sponsors, researchers and host country governments should make provisions for post-trial access for all participants who still need an intervention identified as beneficial in the trial. This information must also be disclosed to participants during the informed consent process."* [21]. More generally, the issue of the social value of health research has been addressed increasingly from a global health perspective, not eluding resource poor settings [22]. Studies have highlighted the importance of sharing the benefits of research with participants, but also some remaining debate about the researchers' responsibility to ensure sharing of benefits from the research with the participants' communities and host nations [23].

TECHNOLOGICAL IMPERATIVE

Another increasing trend that raises serious ethical concerns has resulted from dramatic technological advances in possibilities for diagnosis and clinical management in paediatric neurology. These advances have reshaped clinical practice as well as research, and have led to new societal expectations and demands. The technological imperative may thus imply, quite simplistically, that *if something can be done, it must be done*. The technological imperative will gravely threaten the child's interest if paediatric neurologists feel compelled to use new and emerging technologies without adequate reflection on whether the use they are making contributes to the goals of medical care [24]. Though new diagnostic and therapeutic possibilities may appear to reduce the responsibility of the clinician (e.g. when prescribing modern neuroimaging or genetic testing creates the illusion that clinical examination can be cursory), clinical skills remain paramount to relevance and integration of information. On the contrary, the technological imperative enhances the clinician's ethical responsibility, especially when investigations are undertaken that may be 'interesting' for the clinician but less important to the family [25]. The question *'whose needs are being served?'* must remain a guide in clinical practice.

SOCIETAL ROLES

Increased ethical responsibility of paediatric neurologists concerns not only individual patients as they have acquired increasingly recognised societal roles. Among these, advocacy, support of lay patients' organisations and education of the public have developed in parallel with societal changes and progress in communication technologies. The paediatric neurologist's guidance is also sought in court and in the process of law-making. In addition, it may be necessary to take a proactive role in anticipating the social impact of new technologies and play a whistle-blowing role, e.g. with respect to neglect or abuse of vulnerable populations.

MEDICAL INDICATIONS	PATIENT PREFERENCES
<p>Beneficence and Nonmaleficence</p> <ul style="list-style-type: none"> • What is the patient’s medical problem? History? Diagnosis? Prognosis? • Is the problem acute? Chronic? Critical? Emergent? Reversible? • What are the goals of treatment? • What are the probabilities of success? • What are the plans in case of therapeutic failure? • In sum, how can this patient be benefited by medical and nursing care, and how can harm be avoided? 	<p>Respect for Patient Autonomy</p> <ul style="list-style-type: none"> • Is the patient mentally capable and legally competent? Is there evidence of capacity? • If competent, what is the patient stating about preferences for treatment? • Has the patient been informed of benefits and risks, understood this information, and given consent? • If incapacitated, who is the appropriate surrogate? Is the surrogate using appropriate standards for decision making? • Has the patient expressed prior preferences (eg, advance directives)? • Is the patient unwilling or unable to cooperate with medical treatment? If so, why? • In sum, is the patient’s right to choose being respected to the extent possible in ethics and law?
QUALITY OF LIFE	CONTEXTUAL FEATURES
<p>Beneficence, Nonmaleficence, and Respect for Patient Autonomy</p> <ul style="list-style-type: none"> • What are the prospects, with or without treatment, for a return to normal life? • What physical, mental, and social deficits is the patient likely to experience if treatment succeeds? • Are there biases that might prejudice the provider’s evaluation of the patient’s quality of life? • Is the patient’s present or future condition such that his or her continued life might be judged as undesirable? • Is there any plan and rationale to forgo treatment? • Are there plans for comfort and palliative care? 	<p>Loyalty and Fairness</p> <ul style="list-style-type: none"> • Are there family issues that might influence treatment decisions? • Are there provider (physician, nurse) issues that might influence treatment decisions? • Are there financial and economic factors? • Are there religious or cultural factors? • Are there limits on confidentiality? • Are there problems of allocation of resources? • How does the law affect treatment decisions? • Is clinical research or teaching involved? • Is there any conflict of interest on the part of the providers or the institution?

Figure 1: The four topics approach to clinical ethical case analysis, after Jonsen et al. [29].

To some extent, the physician’s increased responsibilities have been delineated by legal rights, such as national laws on rights of patients or the *United Nation Convention on the Rights of Persons with Disabilities* [26]. It must be remembered, though, that legal texts may at best clarify the margins to practice within but are rarely at the core of ethical practice. Indeed, ethics is often concerned with more subtle questions related to the balancing of legal requirement with other interests and competing ethical principles.

PRINCIPLE OF BIOETHICS

The general analytical framework that can be used in clinical practice to disentangle the ethical complexity of the clinical situations includes principles that are widely applied in health care, such as respect for autonomy, beneficence, non-maleficence, and justice [2]. These principles do not impose attitudes or provide ready-made solutions. They cannot *a priori* identify ‘good’ and ‘bad’ in a specific clinical situation. Rather, they can help organize reflections on moral issues that often arise in clinical practice and support a constructive dialogue about those issues [5].

The bioethical principle of autonomy describes the patient’s right to make their own choices. This must be supported by informed consent, which implies special attention to the notion of competence and eventually surrogacy. The principle of beneficence grants much importance to the child’s best interest and welfare and includes health prevention. Though literally meaning ‘do good’, in practice it refers to ‘doing more good than harm’. It is not identical to the prin-

ciple of non-maleficence, referred to above as ‘do no harm’, though the latter also emphasises the patient’s interest, and in particular quality of life. It may be thought that the non-maleficence principle can be met by ‘doing nothing’. However, in clinical practice rather than doing nothing or straightforward doing ‘no harm’, this principle applies when striving to not to take action that would result in more harm than good. The fourth biomedical principle is justice, a concept that emphasises fairness and equity among individuals, implying, for example, fair distribution of care by the health professional as well as by the health system. This raises complex discussions, particularly when it involves management of scarce resource [27].

Currently, many clinicians consciously value these ethical principles, but most do not systematically use them directly when making clinical decisions [28]. An alternative, more practical and contextual approach to medical ethics has suggested a ‘four topics chart’ to deal with clinical situations [29] (Figure 1). These topics include medical indications, patient preferences, quality of life and contextual features. Under medical indications, questions relating to the principles of beneficence and maleficence are examined, such as the goals of treatment, how this patient can benefit from care and how harm can be avoided. Similarly, patient preferences relate to autonomy, to the quality of the information that is provided, and to cooperation. The topic of quality of life concerns beneficence, non-maleficence, and autonomy. It questions, for example, on what grounds one can judge desirable or undesirable quality of life for a patient who cannot make or express such a judgement. Finally, contextual fac-

tors relate to the principle of justice and cover topics such as conflicts of interest that could thwart the fair consideration of the interests of a patient.

Whether an approach based on principles or topics is preferred, ethical reasoning and communication are grounded in features identified in individual situations. Clinical encounters are inherently particular, both humanly and contextually, yet they provide us with useful clues to address other, more or less related situations. These individual situations and their 'resolution' cannot act as precedent cases as they do in a legal discussion, where a principle (or its interpretation) put forth in a previous legal case becomes binding or at least a persuasive reference when deciding on subsequent cases involving similar issues. The limitations of the jurisprudential approach originate in the irreducible singularity of clinical cases, which have to reflect the clinical notion that similar facts do not reliably yield predictably similar outcomes. Indeed, any individual clinical situation comprises multiple realities corresponding to views and projections of each of the implicated individuals. These should not be expected to coincide with previous cases. In fact, they often come into conflict with other similar cases. Respect, trust, honesty, empathy and commitment are instrumental to articulating the different ways of reasoning but also to allow the emergence of an ethical decision resulting from meaningful dialogue. The exercise of dialogue and open-minded ethical problem solving is, however, difficult. One of the pitfalls is the danger of alienation through cultural relativism, based on the assumption that an individual's beliefs and attitudes are determined by the specifics of his or her culture. Cultural aspects require to be decoded not only where different geographic origins seem to call for this but based on the recognition that every individual relates to his or her own culture in different ways. In practice, the presence of cultural factors (together with other social factors) should not overshadow the importance of more general, non-specific issues such as communication, lack of adaptiveness of the system, or uncertainty in diagnosis, outcome, management planning, etc. Indeed, the latter factors that are often at the very core of the presentation of clinical problems. Communication on ethical issues can often be usefully supported by sharing and weaving together a narrative, a story in which 'the patient is the hero'. At times, this allows for envisioning alternative stories and processes by which possibilities of a situation can be considered. Such narratives are ultimately based on observed signs, facts and empirical data, and they must also deal with incomplete knowledge, conjecture and situational judgement [30]. In many ethical discussions, the focus appears to be on decisions between options of action, as though this were the crux of ethical thinking. However, decision-making in paediatric neurology is mostly a gradu-

al, stepwise process that must be inscribed in time with the setting of priorities as they plausibly manifest themselves. This may imply postponing difficult questions later along the unfolding of the situation and leaving the narrative open, so to speak.

GOOD PRACTICE

Finally, it must be underlined that medical ethics has long been a crucial part of good clinical practice. However, the last decades have brought many changes within and outside of clinical practice that have generated unforeseen ethico-clinical situations as well as new ways of engaging with those situations. In the process of tackling ethically problematic situations, one should, therefore, avoid the temptation of 'understanding too quickly' since a comprehensive and generous understanding of clinical situations is an important starting point. Another consideration is that regardless of the moral reasoning approaches employed, ethical thinking often offers non-definitive resolutions of ethical problems, and thus humility is warranted. The interested reader will find examples of the application of this approach in paediatric neurodisability in our recent edited volume featuring discussions of numerous scenarios faced by international clinicians in their daily practice [5].

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Author Contributions

All authors discussed together the themes included in the manuscript and all wrote chapters in a book that served as a primary base for the manuscript (ref. 5). BD drafted the manuscript, which was critically revised by all the authors.

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REFERENCES

1. Pavur C. The Hippocratic Oath in Latin with English translation. URL = http://www.academia.edu/2638728/The_Hippocratic_Oath_in_Latin_with_English_translation, retrieved 20 July 2016
2. Beauchamp TL, Childress JF. Principles of Biomedical Ethics. 5th edition. Oxford University Press, Oxford, 2001
3. Shevell MI: A brief synopsis of the “why, what, and how” of teaching ethics to pediatric neurology trainees. *Semin Pediatr Neurol* 2011, 18 (2):79-80. DOI: 10.1016/j.sp.en.2011.05.009 PMID: 22036484.
4. Ronen GM, Dan B: Ethical considerations in pediatric neurology. *Handb Clin Neurol* 2013, 111 (1):107-14. DOI: 10.1016/B978-0-444-52891-9.00011-7 PMID: 23622156.
5. Rosenbaum PL, Ronen GM, Racine E, Johannesen J, Dan B (eds.). *Ethics in Child Health: Principles and Cases in Neurodisability*. Wiley, London, 2016.
6. Frank P. The Preamble of the Constitution of the World Health Organization. *Bull World Health Organ* 2002; 80: 982.
7. Sedney CL, Bernstein M: Wilder Penfield - bio-ethicist. *Can J Neurol Sci* 2014, 41 (2):177-81. PMID: 24534027.
8. Huber M, van Vliet M, Giezenberg M, Winkens B, Heerkens Y, Dagnelie PC et al. Towards a ‘patient-centred’ operationalisation of the new dynamic concept of health: a mixed methods study. *BMJ Open* 2016, 6 (1):e010091. DOI: 10.1136/bmjopen-2015-010091 PMID: 26758267.
9. World Health Organization. International classification of functioning, disability and health. World Health Organization, Geneva, 2001
10. World Health Organization. International classification of functioning, disability and health: children & youth version. World Health Organization, Geneva, 2007
11. Ronen GM, Rosenbaum PL. *Life Quality Outcomes in Children and Young People with Neurological and Developmental Conditions: Concepts, Evidence and Practice*. Wiley, London, 2013.
12. Colver A, Rapp M, Eisemann N, Ehlinger V, Thyen U, Dickinson HO et al. Self-reported quality of life of adolescents with cerebral palsy: a cross-sectional and longitudinal analysis. *Lancet* 2015, 385 (9969):705-16. DOI: 10.1016/S0140-6736(14)61229-0 PMID: 25301503.
13. Mezgebe M, Akhtar-Danesh GG, Streiner DL, Fayed N, Rosenbaum PL, Ronen GM: Quality of life in children with epilepsy: How does it compare with the quality of life in typical children and children with cerebral palsy? *Epilepsy Behav* 2015, 52 (Pt A):239-43. DOI: 10.1016/j.yebeh.2015.09.009 PMID: 26469801.
14. Pangalila R: Quality of life in Duchenne muscular dystrophy: the disability paradox. *Dev Med Child Neurol* 2016, 58 (5):435-6. DOI: 10.1111/dmcn.12959 PMID: 26534893.
15. Albrecht GL, Devlieger PJ: The disability paradox: high quality of life against all odds. *Soc Sci Med* 1999, 48 (8):977-88. PMID: 10390038.
16. Noritz G: How Can We Practice Ethical Medicine When the Evidence Is Always Changing? *J Child Neurol* 2015, 30 (11):1549-50. DOI: 10.1177/0883073814563141 PMID: 25660134.
17. Modi N, Vohra J, Preston J, Elliott C, Van't Hoff W, Coad J et al. Guidance on clinical research involving infants, children and young people: an update for researchers and research ethics committees. *Arch Dis Child* 2014, 99 (10):887-91. DOI: 10.1136/archdischild-2014-306444 PMID: 24914095.
18. Lepola P, Needham A, Mendum J, Sallabank P, Neubauer D, de Wildt S: Informed consent for paediatric clinical trials in Europe. *Arch Dis Child* 2016, 101 (11):1017-1025. DOI: 10.1136/archdischild-2015-310001 PMID: 27226526.
19. Informed consent, parental permission, and assent in pediatric practice. Committee on Bioethics, American Academy of Pediatrics. *Pediatrics* 1995 (2):314-7. PMID: 7838658.
20. Singh J: How Bioethics is Complementing Human Rights in Realizing Health Access for Clinical Trial Participants: The Case of Formative PrEP Access in South Africa. *Health Hum Rights* 2015, 17 (1):E58-62. PMID: 26204584.
21. World Medical Association, Declaration of Helsinki: Ethical Principles for Medical Research Involving Human Subjects. 64th WMA General Assembly, Fortaleza, Brazil, October 2013.
22. Bekker LG, Slack C, Lee S, Shah S, Kapogiannis B: Ethical issues in adolescent HIV research in resource-limited countries. *J Acquir Immune Defic Syndr* 2014, 65 Suppl 1 (1):S24-8. DOI: 10.1097/QAI.0000000000000006 PMID: 24321980.
23. Lairumbi GM, Michael P, Fitzpatrick R, English MC: Ethics in practice: the state of the debate on promoting the social value of global health research in resource poor settings particularly Africa. *BMC Med Ethics* 2011, 12 (1):22. DOI: 10.1186/1472-6939-12-22 PMID: 22085702.
24. Kluge EH: Ethical and legal challenges for health telematics in a global world: telehealth and the technological imperative. *Int J Med Inform* 2011, 80 (2):e1-5. DOI: 10.1016/j.ijmedinf.2010.10.002 PMID: 21067967.
25. Rasmussen LA, Bell E, Racine E: A Qualitative Study of Physician Perspectives on Prognostication in Neonatal Hypoxic Ischemic Encephalopathy. *J Child Neurol* 2016, 31 (11):1312-9. DOI: 10.1177/0883073816656400 PMID: 27377309.
26. United Nations General Assembly. Convention on the Rights of Persons with Disabilities. Resolution adopted by the General Assembly, 24 January 2007, A/RES/61/106, available at: <http://www.refworld.org/docid/45f973632.html>, retrieved on 20 July 2016.
27. Eyal N: Pediatric heart surgery in Ghana: three ethical questions. *J Clin Ethics* 2014, 25 (4):317-23. PMID: 25517570.
28. Page K: The four principles: can they be measured and do they predict ethical decision making? *BMC Med Ethics* 2012, 13 (1):10. DOI: 10.1186/1472-6939-13-10 PMID: 22606995.
29. Jonsen AR, Siegler M, Winslade WJ. *Clinical Ethics: A Practical Approach to Ethical Decisions in Clinical Medicine*. 8th edition. McGraw-Hill Education, New York, 2015.
30. Dan B: Medical rhetoric and rhetoric medicine. *Dev Med Child Neurol* 2014, 56 (10):916.