VIEWPOINT

Child with multiple problems: Clinical complexity and uncertainty

Michael McDowell1,2,3

1School of Medicine, University of Queensland, 2Child Development Network, Brisbane, Queensland, Australia and 3Neurodevelopmental and Behavioural Paediatric Society of Australasia [https://nbpsa.org]

Abstract: When practising neurodevelopmental–behavioural paediatrics, responsibility rests with the individual clinician to interpret research evidence in their daily work. This is not necessarily an easy task. Children do not usually present with single, easily definable problems. Complexity and uncertainty are present from the outset due to heterogeneity inherent in the diagnoses available, patterns of comorbid problems and the unique challenges of each child's social context. This article examines the resulting clinical challenge and offers potential responses. Rather than striving to eliminate uncertainty with strategies such as extensive assessment, the goal is to work effectively and efficiently within the limits of what can be readily known. Suggested strategies address complexity by considering the components of what is going on (biological, developmental, transactional) as hypotheses shared within each child's system of care. These hypotheses can then be tested and revised systematically as further information arises along the journey of treatment and longitudinal care.

In this edition of the Journal, much of the information is organised within individual diagnostic categories. Clinical practice, however, is not often as straightforward. This paper considers the clinical challenge of neurodevelopmental–behavioural paediatric (NBP) services when individual cases are complicated (e.g. simultaneous problems) and uncertain (e.g. areas of overlap, multiple potential causes for presenting problems). How is existing evidence used to best help children? To begin with, an example case is presented.

Kane, aged 6, presents with his mother Kelly, who is worried about Kane’s learning and his behaviour at home and at school. Parent and teacher rating scales completed in the referral paperwork support a diagnosis of combined-type attention deficit hyperactivity disorder (ADHD). To begin with, this seems like a straightforward clinical challenge with the potential for successful, evidence-based treatment options.

You find out more. Kane’s parents have separated. Domestic violence was likely, although there was no history of abuse against Kane himself. His parents are still in conflict. Kane sees his father Luke every week. Kelly seems demoralised, not in the best of physical or mental health. You wonder how much she drinks. You wonder about Kane’s pregnancy and early childhood. Kelly mentions that Kane spends too long on his iPad, and there is substantial conflict when she asks him to turn it off. She adds that he has trouble getting to sleep and sometimes soils his pants.

Kane’s learning profile is uneven. He has a reasonable understanding of maths but struggles with literacy. His father is described as ‘dyslexic’. Kane is resistant to writing. His social behaviour with peers is unpredictable, often interpreted by other children as bullying. He does not appear to appreciate nor care about their feelings when behaving this way. The variability of Kane’s behaviour on history sounds related to his emotional state. Despite appearing healthy and active, Kane’s skills in structured sports such as soccer are reportedly weak, a situation not helped by his seeming ambivalence to coaching.

Kane presents as a personable young man. He is interactive and cheeky, somewhat unencumbered by fear of the professional relationship. You suspect, however, that his teacher does not find him so likeable. Her referral information rates most behaviours on your questionnaire at the highest level of concern. Her narrative interprets intention behind Kane’s behavioural choices.

Kelly has waited some time to see you. Her expectations are high. You realise that Kane may be one of the many children you see for whom treatment will be neither straightforward nor quick.

Kane’s presentation exemplifies a set of challenges common to NBP clinical practice. How is it possible to best make sense of Kane’s situation diagnostically and use this diagnostic understanding to inform your treatment and management choices?

Uncertainty in Diagnosis

Medical training is traditionally built on a model of sequential practice: presentation → history → examination → differential diagnosis → investigations → diagnosis → treatment. This approach assumes the diagnosis to hold sufficient truth to explain the presenting problem(s) and to successfully inform treatment.1

However, like the blind men and the elephant,2 each diagnosis in NBP may provide only partial information. Specifically, these diagnoses may inform only part of the clinical picture. Some are biologically founded (certain or presumed), whilst others aggregate presenting clinical problems into integrated conceptual categories. As a result, each diagnostic type provides different...
patterns of clinically relevant information, each with differing degrees of certainty (Table 1).

Uncertainty in NBP diagnosis further arises due to the heterogeneity intrinsic to each individual diagnostic category. To begin with, each neurodevelopmental diagnosis may encompass a wide range of potential severity.

Second, many neurodevelopmental diagnoses are constructed from clinical phenotype information, rather than from biological data. The central diagnostic concept, for example, attention, autism, learning or anxiety, refers to an abstract idea. The psychiatric term for these concepts is ‘latent constructs’, where ‘latent’ refers to unobservable, and ‘constructs’ refers to ideas created to encapsulate the set of symptoms.5 Heterogeneity occurs when a latent construct diagnosis is made from sets of criteria, and different children within the same diagnostic category may have differing patterns of component problems.4

A third source of within-diagnosis heterogeneity is the diversity of aetiology. Biologically based research indicates multiple potential pathways of causation within single diagnostic categories.3 A final source of within-diagnosis heterogeneity is the imprecision that arises at diagnostic boundaries, when arbitrary cut-off points are used to define dichotomous diagnoses within spectrum conditions. As a result, children whose clinical presentation falls near these boundaries may move into, or out of, diagnostic categories over time.6

In addition to the variation due to diagnostic type and within-diagnosis heterogeneity, further complexity arises from individual patterns of comorbid problems. Comorbid problems are common in NBP practice.7 The relationship between problems may be complex, and the presence of comorbid problems appears to influence not only a child’s immediate challenges but also their long-term clinical journey (e.g. ADHD with reading and/or motor problems).8,9

This brief consideration of the complex, heterogeneous nature of neurodevelopmental diagnosis is presented to support this paper’s central assertion: For a child such as Kane, diagnostic uncertainty is intrinsic and fixed. It exists in addition to any consequence of insufficient diagnostic rigour.

This limitation does not have to be a difficult problem to accept. In more precise areas of academic endeavour, limits to what is knowable have been clearly demonstrated. In quantum mechanics, for example, Heisenberg identified an intrinsic limit to what can be measured.10 Another example is mathematics, where Gödel proved that any mathematical system contains truths that are not provable using mathematical techniques.11 In a similar manner, there is a pre-existing limit to what can be diagnostically known for many NBP problems.

In clinical practice, a common response to diagnostic uncertainty is to build more extensive, more complex assessment methodologies. Whilst these may provide a comprehensive snapshot of the child at that time and within that assessment context, there is a limit to clinical benefit that cannot be overcome with additional information. For this reason, it can be argued that further cross-sectional assessment, beyond a certain point, does not add significant value. It may even be detrimental, consuming time and resources when these may be better directed, for example, towards intervention.

In academic research, diagnostic uncertainty can be quantified as properties of assessment strategies such as validity and reliability. To achieve accuracy sufficient for research purposes, these measures allow the calculation of necessary power, usually the numbers of children needed to be tested before there is sufficient certainty to meet predetermined levels of likelihood. This approach, however, is not available to the clinician, who must make diagnostic and treatment decisions about individual children and do his or her best to manage the inevitable uncertainty.

### Table 1 What a diagnosis tells us. A transfer-of-care referral includes a diagnosis without elaboration. What can be known from the diagnosis alone?

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Aetiology</th>
<th>Expected current clinical problems</th>
<th>Clarity regarding what to do now</th>
<th>What the future may hold</th>
</tr>
</thead>
<tbody>
<tr>
<td>Causal (tested): For example, fragile X</td>
<td>Certain</td>
<td>Limited range of type and severity</td>
<td>Medium</td>
<td>Somewhat predictable depending on the child’s current picture</td>
</tr>
<tr>
<td>Causal (clinical): For example, FASD</td>
<td>Presumed</td>
<td>Broad range of possible consequences</td>
<td>Low</td>
<td>Presumed</td>
</tr>
<tr>
<td>Criterion-based clinical phenotype:</td>
<td>Uncertain</td>
<td>Likely to be one or more of a limited range</td>
<td>Medium</td>
<td>Uncertain</td>
</tr>
<tr>
<td>For example, SLD, ASD, ADHD</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Single functional problem:</td>
<td>Presumed</td>
<td></td>
<td>Medium</td>
<td>Mostly treatment determined</td>
</tr>
<tr>
<td>For example, sleep, encopresis</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

ADHD, attention deficit hyperactivity disorder; ASD, autism spectrum disorder; FASD, fetal alcohol spectrum disorder; SLD, specific learning disability.

### Uncertainty in Clinical Formulation

Formulation is a clinical strategy developed to help overcome diagnostic uncertainty.12 It refers to the clinical process of combining relevant diagnostic considerations into an individualised, structured set. One example has been the multi-axial structure of DSM-IV.13

Formulation approaches have been proposed for NBP.14 Whilst formulation provides a powerful contribution to addressing clinical uncertainty, it does not fully address the complexity of clinical interactions and component contributions to presenting clinical challenges.

Using Kane’s case as an example, the individual presenting problems are not adequately explained by individual diagnoses. They are likely to arise from multiple, interacting processes. Consider Kane’s behaviour at school. In peer relationships, he exhibits strong social desire, but recurring descriptors of his social
behaviour include ‘immature’ and ‘aggressive’. How is his social behaviour best understood? It seems more than just the impulsivity of ADHD. He does not meet criteria for an autistic diagnosis. It does not help that he is teased for his learning and soiling. The volatility of his behaviour appears driven by his emotional state. An intuitive path for understanding Kane’s behaviour is developmental or, more specifically, as a developmental delay. He seems like a frustrated 4-year-old, who can sometimes settle and act with greater maturity. Putting all this together, there are likely to be several contributing factors: poor impulse control, frustration with poor emotional regulation and delayed development in social understanding and skills. Diagnostic formulation may better identify contributing factors but does not fully explain how these result in the presenting problems.

Kane’s behaviour in class and at home is equally complex. In his grade 1 class, his behaviours of concern include avoidance, inattention and what his teacher interprets as disrespect. It is likely that these behaviours arise from weak attention, impulse and mood control, combined with frustration from his learning struggle and a lack of social maturity. These factors contribute also to Kane’s behaviour at home, combined with potential contribution of parental acrimony and behavioural strategies that are not aligned. It is not possible at this stage to know the extent to which he may also be fearful (in response to current threat or subsequent to past experiences), angry or insecure.

Kane’s learning struggle centres around literacy. His reading decoding skills fall below the range accommodated by the curriculum. He has not mastered full letter recognition, and his penmanship is poor. He may well meet the criteria for a specific learning disability in literacy, along with the consequences of executive function and possible fine motor weaknesses. Even at the age of 6, Kane appears to experience frustration and a fear of failure that contribute to his current avoidance, along with the accumulated academic consequences of limited learning experience.

The challenge with diagnostic formulation is that Kane’s presenting symptoms of concern (behaviour, learning, sleep, soiling) are unlikely to reflect just the linear consequences of individual diagnostic processes. Each symptom set could be considered a final common pathway for multiple simultaneous causal processes. Diagnostic assessment at a single point of time, even communicated as formulation, is unlikely to fully inform how multiple potential components contribute to a child’s presenting problems.

Uncertainty in Treatment

In Kane’s case, there is sufficient confidence in an ADHD diagnosis to recommend stimulant medication, prescribed cautiously because of his young age and sleep problems. It is not possible to predict how much difference this will make. Based on his current reading level, it is reasonable to recommend literacy intervention. Identified differences in parenting style may be addressed with referral for behavioural training. Functional problems with sleep, soiling and even media usage can be addressed with specific medical and behavioural protocols. These are all treatment options associated with individual diagnoses, supported with evidence-based practice (EBP) research.15 Is this enough?

After some time, Kane and Kelly return for review. His situation has improved, but the pattern of problems persists. His behaviour remains unpredictable and unstable. His learning has not substantially improved. How do you address Kane’s need for mood stability, his confidence as a learner, his social maturity? How can you achieve the consistency required across households to manage his sleep, media usage and enuresis? This situation is worsened by the fact that his stimulant medication wears off during the afternoon. How can Kane’s learning needs be met when it appears that he is unlikely to ‘catch’ up, even with some additional assistance at school and a tutor once a week at home?

As with diagnosis, these considerations are presented to underlie a central conclusion about the reality of clinical treatment. EBP, however accurate, is unlikely to provide sufficient information to inform treatment over time for a case such as Kane’s. As with diagnosis, this uncertainty of treatment is not simply due to a lack of evidence and knowledge. This uncertainty is also intrinsic to the clinical complexity of Kane’s situation.

Managing Diagnostic Uncertainty

The essential contribution of a medical specialist is to incorporate an understanding of biology into the clinical consideration. There are potentially three areas of aetiological consideration that underpin diagnostic categorisation and formulation.

The first is biological. Even without diagnostic certainty, what is likely to be the expression of an intrinsic biological weakness? In Kane’s case, it is clear he has problems with attention and impulse control, along with the executive function skills that underlie learning. He will probably meet criteria for a specific learning disability in literacy, presumably of genetic origin. History, along with clinical observations, identifies poor motor control and motor learning, impacting most noticeably on pencil control. Kane demonstrates limited emotional regulation, along with responses to regular daily challenges that are disproportionately large. All these are likely to reflect, to some degree, a biological contribution to causation.

The second area of consideration is developmental. This refers to the component of Kane’s clinical struggle attributable to skills that have not yet been learned but are potentially able to be. The mechanism behind this ‘developmental delay’ may be biological; however, the functional problem is the current consequence of slower skill acquisition over time. Examples in Kane’s case may include his social maturity, learning skills, self-control of thought, mood and behaviour, his proprioceptive self-awareness (soiling) and even his self-soothing skills, presenting clinically as problems with screen use and sleep.

A final area of consideration is ecological and transactional. This refers to the proportion of current problems that arise from and are readily modified by the interactions within his world. It is not possible to know the extent to which Kane’s behaviour at school results from the teasing or teaching behaviours such as unpredictability or shouting. Equally, it is not possible to know the extent to which inconsistencies of individual parenting behaviour, and between parents, contributes to the current picture. It is likely, however, that these transactional factors contribute to some degree.
Managing Treatment Uncertainty

This three-level model of analysing Kane’s situation lends itself to treatment strategies that explore what is possible. By using medication, the underlying biological component is symptomatically modified, reducing the impairment and impact of the ADHD symptoms. This treatment helps define the extent that may be biologically driven. By providing reading remediation, there can be exploration of the potential for neuroplastic improvements in Kane’s delayed literacy development. By encouraging strategic changes in teaching and parenting behaviours, information may emerge that clarifies the extent to which Kane’s problems are intrinsically flexible and situation-responsive.

This approach is necessarily experimental. It begins with a hypothesis, works towards a predetermined goal, tests this with treatment strategy and then modifies both hypothesis and strategy depending on outcome. This adaptation of scientific method has been referred to in developmental practice as ‘response to intervention’.16,17

Sharing a hypothesis is not the same as explaining a diagnosis. A hypothesis enables all those involved to understand the set of initial presumptions, with the understanding that time will further clarify. These presumptions include what is likely to be medically based, with the implication that it is unreasonable to expect Kane to do what is just too difficult, or unsustainable, for him. In a similar way, if part of the clinical picture is attributable to a developmental delay, it means Kane cannot do what is expected at an age-appropriate level, that he will need support to learn, and if there is an underlying biological weakness, his progress may continue to be slow, and he may not close the developmental gap. An explanation of what is likely to be situation-reactive may help to align management behaviours towards strategies more likely to achieve the desired goal.

This is likely to work most successfully when all relevant parties share the hypothesis, the strategy, the intended goal and the processes for revision. Who is involved? What do they need to know? How is the information best shared, the concerns of the involved parties best heard and a shared understanding developed? In Kane’s case, for example, there is conflict between parents. The medical specialist is in a unique position to reduce this impasse. If Kane had diabetes or cancer, medical information would hold power to align parental differences. Whilst more

<table>
<thead>
<tr>
<th>Table 2</th>
<th>Suggested strategies to manage treatment complexity</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Strategy</strong></td>
<td><strong>Examples</strong></td>
</tr>
<tr>
<td>Explanation as advocacy and treatment</td>
<td>As a medical specialist, recommend that relevant people (e.g. teacher, principal) are informed that you are seeing the child and effectively communicate what you believe to be the child’s medically based and developmentally acquired problems.</td>
</tr>
<tr>
<td>Alignment within the child’s system</td>
<td>Each child lives in a system (home, school, family, community). Who are the key influences on that child’s development and well-being? The system will be most effective in supporting the child if there is an alignment of causal belief (why this is happening), purpose (what we are trying to achieve), strategy (who is doing what), communication (sharing new knowledge along the way) and review (how do we know if this is working).</td>
</tr>
<tr>
<td>Low-hanging fruit</td>
<td>What can be achieved easily and is most likely to get the greatest outcome? In Kane’s case, this might be:</td>
</tr>
<tr>
<td></td>
<td>• Parental agreement about ADHD and cooperation with treatment</td>
</tr>
<tr>
<td></td>
<td>• Focus on encopresis, so Kane can be confident of continence</td>
</tr>
<tr>
<td></td>
<td>• Management of sleep</td>
</tr>
<tr>
<td></td>
<td>• Curriculum adjustments at school (take the pressure off)</td>
</tr>
<tr>
<td>Step by step</td>
<td>It is tempting to make multiple diagnoses and initiate corresponding treatments all at once. Parents then commute between therapies, counselling, tutoring and whatever else you recommend. Each component may act as if they were the only relevant intervention.</td>
</tr>
<tr>
<td></td>
<td>Instead, consider the total workload. What comes first? How are interventions best sequenced in terms of what is most important, and what is most likely to work? How can workload be reduced and balanced (e.g. permission to avoid unnecessary curriculum)?</td>
</tr>
<tr>
<td>Scientific method: Hypothesis, goal, strategy, review</td>
<td>Engage those involved with your hypothesis. An example may be that the child will make more mature behavioural choices if he is calm.</td>
</tr>
<tr>
<td></td>
<td>Test the hypothesis (e.g. what happens if you keep a child such as Kane calm first and then manage the behaviour). Review the results in light of the intended goals and adjust as needed.</td>
</tr>
<tr>
<td>Encourage innovation</td>
<td>If the doctor makes authoritative statements about what needs to be done, others may step aside and not contribute.</td>
</tr>
<tr>
<td></td>
<td>Instead, hypothesis and goals can be shared. It is then possible to give them permission to experiment, encourage them to generate ideas and evaluate them. They may find strategies that you would never have considered.</td>
</tr>
<tr>
<td>Try several things, monitor, reinforce what works best</td>
<td>It is initially reasonable to try medication, adjustment of schoolwork, social skills programmes, tutoring and so on. Of these, the medication and curriculum adjustment help, but the social skills and tutoring seem to lack value for effort. It is then possible to let go of what does not seem to work and build what does.</td>
</tr>
</tbody>
</table>
complex in NBP practice, a similar approach to explanation may be successful if Kane’s problems were communicated in a set of explanations (medical, developmental, transactional) that more directly inform what is, and is not, likely to work.

In summary, the management of treatment uncertainty may require experimentation within a ‘response to intervention’ model, built on scientific strategies of hypothesis testing. It can be built on a systems model, where the stakeholder group, guided by the hypothesis and associated goals, explores solutions and learns from these experiments. Finally, if there is no clear evidence-based path, decisions about treatment may instead be helpfully guided by principles rather than by algorithms. Some principles for management suggested from the author’s clinical observations are summarised in Table 2.

Uncertainty in Management Over Time

Presuming a significant biological contribution to Kane’s problems, the possibility exists that his underlying difficulties will persist, in some form, over time. This raises the challenge of how best to manage the clinical journey and achieve the best long-term outcomes most effectively. The risk is that each visit will be dominated by the need to address current problems. This can be a tiring and clinically unsatisfying method of practice. It can be difficult to look into the future with optimism. How might the paediatrician overcome this impasse?

Consider the role of the medical specialist in the context of this long-term risk. Is the achievement of optimal long-term outcome a central part of the paediatric purpose, as is the case if Kane had a traditional chronic medical condition such as diabetes? Arguably, the greatest potential contribution of a medical specialist will be to positively alter Kane’s trajectory over time so that he has an outcome when graduating from school into adult life that could be considered his personal best.

In the management of medical chronic conditions, it is clear that a problem-reactive model of care is insufficient to alter trajectory towards the best long-term outcome. Diabetic services do not see children only when something goes wrong. If this method of practice is to change, it is the doctor’s responsibility to outline terms of reference for how this should work.

For a complex presentation such as Kane’s, one consideration is to contract paediatric services in two clinically defined phases. The first, treatment phase, is to address the immediate issues. The goal is for Kane to be in the best possible shape with regards to his development, behaviour and mental health. This may require a higher frequency of visits. It acknowledges that some problems may be overcome (e.g. sleep, encopresis), but others with a medical basis are likely to persist, with a constancy of impact on his developmental progress.

The second clinical phase is to consider and intentionally address long-term risk, with intended long-term outcomes that extend beyond problem care to include the child’s resilience and optimisation. Contracted as an explicit and mutually understood purpose above and beyond maintenance (e.g. management of medication), this has the potential to intentionally build a developmental trajectory towards the best possible long-term outcome. Strategically, this includes anticipating and preparing for potentially difficult times, such as new teachers, and other transitions. It includes finding sustainable balance over time. It seeks to build resilience, self-knowledge, self-efficacy and areas of personal strength for Kane. It considers his emerging sense of self and well-being, particularly during adolescence.

When proposing how to clinically contract this optimisation phase, the paediatrician may consider both the frequency of regular reviews and the pathways whereby potential or emerging problems can be dealt with early, before they become crises. On a continuing basis, this is likely to work most effectively when all those in Kane’s world remain actively involved, for example, his father.

Conclusions

The ‘medical model’ of clinical practice is powerful in the context of predictable, low uncertainty pathways for causation, intervention and related outcome. In the complexity of modern health care, this model is being questioned. This paper argues that our default model of care in NBP would also benefit from reconsideration.

The first proposition of this paper is that substantial levels of complexity and uncertainty lie at the very centre of NBP clinical practice in diagnosis, treatment and management. There is a limit that cannot be overcome, regardless of more extensive assessment. When consulting with a child such as Kane over time, a level of frustration and confusion is inevitable due to this uncertainty. Rather than this reflecting personal failing as a clinician, it is an inherent property of the clinical work.

The second proposition of this paper is that how we undertake clinical practice would benefit from mindful acceptance of this uncertainty and appropriate adaptation. As the current best interpretation of the data, diagnoses do not have to be statements of lifelong truth but can be understood and utilised as hypotheses to be tested. Treatment, whilst utilising EBP where possible, embraces each child’s individual circumstances, along with their family, educational and social systems. In the short term, intervention strategies should be undertaken intentionally as experimental, n-of-1 trials to explore how best to achieve predetermined goals. These strategies have the flexibility to learn and are modified with response-to-intervention review. They are undertaken within a systems context, with everybody working as much as possible in synchrony. In the longer term, management works intentionally towards the best possible outcomes, rather than reacting episodically to recurrent or emergent problems.

To think and practice in this way, the clinician must become comfortable working with uncertainty; have the humility to manage mistakes; and maintain the necessary flexibility to experiment, learn and adapt as information emerges. This is not a loss of medical authority. The doctor has a key guiding role, particularly as the central consideration for any child is an exploration of what is likely to be the symptomatic expression of biologically based differences, along with the resulting implications for how best to understand and manage each child’s individual set of needs.

Acknowledgement

The author acknowledges the helpful review comments from Dr Jane Lesslie, Dr John Wray and Dr Catherine Skellern in the preparation of this manuscript.
References


Coloured white birds by Saharah Scalas (age 11) from Operation Art 2017