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## Editorial

## Post-COVID syndrome and adults with intellectual disability: another vulnerable population forgotten?

Rohit Shankar, Bhathika Perera, Ashok Roy, Ken Courtenay, Richard Laugharne and Manoj Sivan



An area of interest presently is the lingering symptoms after COVID-19, i.e. post-COVID-19 syndrome (PCS). Specifics of diagnosis and management of PCS are emerging. However, vulnerable populations such as those with intellectual disabilities, who were disproportionately affected by the pandemic, risk being 'left behind' from these considerations.

**Keywords**

Post-COVID-19 syndrome; long COVID; developmental disabilities; discrimination.

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**Intellectual disability**

Intellectual disability (also known as learning disability in UK health services) is characterised by significant impairments of intellectual and adaptive functioning with onset in childhood. People with intellectual disabilities (PwID) have higher rates of physical, mental and behavioural disorders compared with the general population. Communication deficits and impaired cognition can impede people from identifying their difficulties or distress and communicating them to get the right support and advice. PwID can react to changes in their physical, mental and emotional states as well as to any changes in their social or environmental circumstances. This can be presented as challenging behaviours towards themselves, towards property or others. PwID are also disadvantaged by premature mortality. Increased prevalence of physical and mental health conditions, low health literacy, diagnostic overshadowing combined with challenges accessing healthcare are some of the factors that have led to premature mortality among PwID.

**Intellectual disability and COVID-19**

PwID represent one of the most vulnerable populations to any pandemics for many different reasons. These include multimorbidity, low levels of health literacy, difficulties in understanding and communication, reliance on other people for care, low compliance with complex hygiene rules, the strong need for routine/sameness and

low adaptive skills. This became more evident during the COVID-19 pandemic. As the pandemic unfolded, an increased risk of hospital admissions and mortality from COVID-19 among PwID became clearly evident.<sup>1,2</sup> A threefold higher risk was recognised for PwID than the general population in succumbing to COVID.<sup>3</sup> Studies looking at mortality in this cohort have identified several associated factors such as Down syndrome, epilepsy, mental illness, dysphagia and severity of ID.<sup>2,3</sup> These alarming statistics have resulted in PwID being prioritised for the COVID-19 vaccination programme.

Apart from the direct effect of COVID-19 illness, the disproportionate indirect impact of the pandemic on the mental health of PwID and their carers has also become evident. Travel restrictions and change in human contact has led to increased levels of distress and behaviour change in these individuals. The impact of the pandemic on the mental health well-being of PwID is yet to be studied in large samples. Small-scale studies have shown the possibility of increased mental health difficulties as well as increased prescribing of psychotropic medications in PwID.<sup>4</sup> One also has to acknowledge the impact on the mental health of PwID when their families, carers and staff get COVID-19.

**Post-COVID-19 syndrome (PCS)**

Although most patients recover completely after COVID-19 infection, some continue to have persistent symptoms or develop new symptoms which is known as PCS or long COVID. The National Institute for Health and Care Excellence defines it as signs and symptoms that develop during or after an infection consistent with COVID-19, continuing for more than 12 weeks (3 months), and not explained by an alternative diagnosis. Commonly reported symptoms are breathlessness, fatigue, neuropathic and musculoskeletal pain, cognitive difficulties and psychological distress. There are more than 200 symptoms reported involving more than ten organ systems making it a challenging multisystem condition to manage. It is estimated that there are more than 1 million adults in England alone with PCS and over 40 million worldwide. The underlying pathophysiology is not yet clearly established but an underlying exaggerated immune response, endothelial damage causing hypercoagulation, direct and indirect brain and central nervous system damage, dysautonomia and psychological response are all contributory and linked mechanisms driving these symptoms.

Studies have shown around 10% of people who was positive for COVID-19 developed PCS. The self-reporting COVID-19 Yorkshire Rehabilitation Scale (C19-YRS) has been validated in the general population.<sup>5</sup> It captures a range of subjective and objective symptoms.

### Concerns about recognising PCS in PwID

Given that acute COVID-19 is worse in PwID, it can be argued that the rate of PCS in PwID is likely to be worse or at the least similar to that in the general population. However, there is no published evidence on the prevalence rate of PCS in PwID. Furthermore, recognising PCS in PwID can be challenging as the ability to articulate symptoms or distress can be very limited because of communication difficulties and lower intellectual functioning in this cohort. As neuropsychiatric symptoms including depression, anxiety, delirium and psychosis are already prevalent in PwID they can be difficult to separate out as sequelae of PCS. It also creates clinical challenges and diagnostic dilemmas in identifying whether the symptoms are a sequelae of COVID-19 or not. Key symptoms of PCS such as fatigue and pain are difficult to describe for people with communication problems leading to diagnostic overshadowing.

### Why capturing this and researching this area is important

Given the disabling symptoms or functional difficulties in PCS, the importance of estimating the scope of the problem in PwID is the need of the hour. It can be argued recognition of PCS in PwID may help to better understand the behavioural and neuropsychiatric manifestations of PCS in this population and improve management including reducing usage of psychotropic medications.<sup>4</sup> Comorbid neurodevelopmental conditions such as autism may make it more difficult to accurately tease out the contribution of PCS to the clinical presentation in these individuals. A clear understanding of the impact of PCS in PwID will also help for future planning for social care given the high rate of support needed in this population. Complications seen in PCS such as pulmonary fibrosis, myocarditis, pericarditis, cardiac arrhythmias, renal and hepatic dysfunction, deep vein thrombosis/pulmonary embolism may contribute to increased mortality and morbidity. Appropriate and timely management can be put in place with better understanding of the condition. PCS is a long-term condition and is also likely to make these individuals more vulnerable to other infections and complications further accentuating morbidity and mortality. Finally, given the significant healthcare utilisation and economic impact of PCS in the general population, the financial implications need to be estimated in PwID, which will inform service planning and commissioning.

### Recommendations for clinical practice and research

The World Health Organization has suggested the triple R approach to PCS – Recognition, Research and Rehabilitation. The Appendix outlines a logical approach to build evidence on the recognition and impact of PCS in PwID.

Recognising PCS needs intellectual disability clinicians and carers to look for subtle changes in behaviour and functional need. We need to develop PCS-specific screening tools for this cohort of people. Adapting the C19-YRS scale and validation in this cohort might help recognise the problem and get a real estimate of the burden. To do this caregivers need to acquire the skills to detect subtle changes in behaviour and function and identify changes in need for psychotropic medication where PCS could be suspected.

An earlier stage in building the evidence base may be through case reports and case series. Although a lower standard of evidence, these descriptions can give early clues on the consequences of COVID for PwID. Early case series of COVID deaths in PwID gave clues to the consequences of COVID-19 infection before larger studies were described.<sup>2,3</sup>

Change in carer burden can also indicate PCS. Without such recognition of the condition the residual manifestations would remain undetected and can lead to potential worsening of the presentation itself. Awareness of PCS could be achieved by bespoke training of front-line carers and family carers. Such training needs to be extended to front-line health professionals who are likely to meet PwID such as primary care staff, acute hospitals and specialist intellectual disability staff.

Research should focus on understanding whether the presentation of PCS in PwID is different from that being reported in the general population and whether the impact from symptom type, severity and functional is different. Research should also focus on long-term functional and neurobehavioural changes in prospective longitudinal studies. PwID are often excluded from research even for conditions to which they are especially prone, for example epilepsy. This exclusion from research should not prevail in PCS given the scale of the problem and potential implications for the patient, carer and the wider healthcare and social system.

Changes in prescribing before and after COVID infection may suggest the consequences of PCS. Case-control studies comparing patients who have had COVID and those who have not may help detect changes in prescribing that may be a proxy for PCS. Cohort studies examining changes prospectively would be ideal, but the changing nature of the virus makes this methodology very challenging. Ideally clinical rating scales such as the validated Royal College of Psychiatrists Health of the Nations quality of life outcomes tool for PwID (HONOS-LD) or similar, before and after infection could be used for assessing the effects of COVID-19 infection and repeated at 3–6 months intervals after infection.

Awareness communication, training and rehabilitation provided in a timely manner may reduce the long-term functional deterioration. This has been well proven for other aspects of chronic condition management and should be extended to PCS management in general, particularly for those PwID. An integrated rehabilitation approach that has input from a range of specialists with expertise in managing the complications seen in this multisystem condition will prove useful in reducing long-term functional decline and helping family and carers manage the condition better. We need to ensure these vulnerable individuals are not left behind in our response to this devastating pandemic.

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B.P. has received speaker fees from Flynn pharma outside the submitted work. R.S. has received institutional and research support from LivaNova, GW pharma, UCB, Eisai, Veritron pharma, Averelle and Destin outside the submitted work. M.S. is a significant contributor to the development of the C19-YRS Scale discussed in the paper. No other author has declared any conflict of interest.

## Appendix

Potential plan to enable research about post-COVID-19 syndrome (PCS) in people with intellectual disabilities (PwID) and challenges to realise them		
Research item	Challenges	Advantages
1. Identify and publish a case series of PwID who present with change in physical and neuropsychiatric symptoms post-COVID infection and recovery.	<ol style="list-style-type: none"> <li>1. PwID are a heterogeneous group varying in cognitive and communicative abilities. This could lead to reporting bias from different perspectives such as observer bias, contextual influences and prejudice.</li> <li>2. There could be a skewing of sample (e.g. those with a noticeable behavioural change might be identified more than those who highlight tiredness).</li> <li>3. The case series could be distorted by diagnostic overshadowing.</li> <li>4. Selection bias.</li> </ol>	<ol style="list-style-type: none"> <li>1. It is a feasible study design to initiate a grouping of symptoms and signs.</li> <li>2. Easy to conduct.</li> <li>3. Requires less time and financial resources.</li> <li>4. Will use the UK clinical network and stakeholders supporting PwID.</li> </ol>
1. Implement the COVID-19 Yorkshire Rehabilitation Scale (C19-YRS) Checklist, which has been validated in the general population, in PwID.	<ol style="list-style-type: none"> <li>1. The issue of heterogeneity of the population and their cognitive and communicative abilities would make a direct implementation challenging.</li> <li>2. The C19-YRS has many items that require responses about one's subjective state.</li> </ol>	<ol style="list-style-type: none"> <li>1. With co-production, convert the validated Checklist to a suitable tool to be used verbatim with PwID and their carers.</li> <li>2. Use the adapted tool with the original in a cohort of people from the general population who have been diagnosed to have PCS to help validated the different items (<math>n = 18</math>) of the C19-YRS.</li> </ol>
1. Implement a modified/adapted C19-YRS checklist along with free-text communications for other suspect symptomology in PwID known to have had COVID.	<ol style="list-style-type: none"> <li>1. While the hope is that at least some factors from the C19-YRS would resonate in the presentations there is no guarantee of this.</li> <li>2. Concerns of significant information bias.</li> <li>3. The free text of other associated symptoms might not provide suitable themes.</li> </ol>	<ol style="list-style-type: none"> <li>1. The feedback on the items of the C19-YRS from the different groups could be reviewed using a pre-designed statistical analysis plan.</li> <li>2. The free text would be converted to themes and could be assessed with co-production partners on relevance.</li> </ol>
2. Compare C19-YRS items and free-text themes in those who had no noted change in function/behaviour post-COVID with those who developed changes, especially those retained after 6 weeks.		
1. Implement a new developed tool, a mix of validated factors from C19-YRS, thematic analysis and co-production into select clinical settings as a quality improvement project.	<ol style="list-style-type: none"> <li>1. Identifying a suitable clinical setting and services.</li> <li>2. There might be minimal items that emerge.</li> </ol>	<ol style="list-style-type: none"> <li>1. If there is too much divergence from the original CY19-YRS there might still be an opportunity to create a validated screening tool.</li> </ol>

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