LeDeR
Learning from lives and deaths – People with a learning disability and autistic people

Research Digest: Summer 2023

Welcome to the summer edition of our planned quarterly research digests for 2023. In this edition, we bring you a selection of 10 papers covering a wide range of topics relevant to the needs of people with a learning disability and autistic people (or service users), care providers and NHS commissioners to give an overview of the current research landscape, this time with a focus on constipation. Constipation is a sometimes overlooked, sometimes difficult to talk about subject that impacts the lives of many people with a learning disability. By highlighting this important area, we hope to increase awareness about the work that is currently being done to address constipation, as well as giving inspiration for the work that still needs to be done.

As always, for each paper we have provided a summary of 1) the population, 2) the setting and 3) the rating of the level of evidence provided, based on the 5 point rating summary provided here, where 1 is highest level of evidence and 5 is expert opinion. Please feel free to reach out with any questions or feedback on these digests.

In this edition

Focus on: Constipation

1. Euthanasia and assisted suicide in people with intellectual disabilities and/or autism spectrum disorders: investigation of 39 Dutch case reports (2012-2021).

2. Definitions, signs and symptoms of constipation in people with severe or profound intellectual disabilities: A systematic review.

3. Multiple morbidity across the lifespan in people with Down syndrome or intellectual disabilities: a population-based cohort study using electronic health records.

4. Constipation prevalence and perceptions: Comparison of children and adolescents with ASD and other developmental-behavioural disorders.

5. Association between early and current gastro-intestinal symptoms and co-morbidities in children and adolescents with Angelman syndrome.

6. Prevalence of gastro-intestinal symptoms among autistic individuals with and without co-occurring intellectual disability.

7. Medicine use in people with intellectual disabilities: a Finnish nationwide register study.

8. Reducing risks associated with medicines and lifestyle in a residential care population with intellectual disabilities: evaluation of a pharmacy review initiative in England.

9. Gastrointestinal and Sleep Issues in Toddlers with Autism versus other neurodevelopmental disorders.

10. Global prevalence of autism spectrum disorder and its gastrointestinal symptoms: A systematic review and meta-analysis.

1) Sample: 39 cases of autistic people and/or people with a learning disability.
2) Setting: People with a learning disability or autistic people who applied for euthanasia assisted suicide in the Netherlands (2012-2021)
3) Level of evidence: 4

Euthanasia and physician-assisted suicide in people with intellectual disabilities and/or autism spectrum disorders: investigation of 39 Dutch case reports (2012-2021).

Tuffrey-Wijne et al. (2023)
DOI: https://doi.org/10.1192/BJO.2023.69

In the Netherlands, approvals for euthanasia and physician-assisted suicide (EAS) are determined based on whether individuals are deemed as having 'unbearable suffering without improvement'. Typically, the majority (89%) of EAS requests in the general population are based on unbearable or untreatable physical symptoms, compared to 15% of individuals with a learning disability or autistic people. 39 cases where EAS was granted for autistic people and/or people with a learning disability were examined. Common themes of suffering were loneliness, social isolation, lack of flexible coping strategies and an oversensitivity to stimuli. Many of these factors could be deemed as social causes rather than medical causes. This paper demonstrated a need to examine whether appropriate social support is available for autistic people and those with a learning disability, and whether the presence of these conditions alone is an appropriate reason for EAS to be granted.

1) Sample: 24 studies of people with intellectual disability and constipation.
2) Setting: Peer reviewed papers on five electronic databases, published between 1998 and 2018
3) Level of evidence: 3

Definitions, signs and symptoms of constipation in people with severe or profound intellectual disabilities: A systematic review.

Wagenaar et al. (2022)
DOI: https://doi.org/10.1016/J.HELIYON.2022.E09479

Constipation is more common in people with a learning disability than in the general population. Constipation is a complex condition which can cause further health issues and even contribute to death. There is no agreed definition or diagnostic tool for constipation for people with a learning disability, despite this population being at a higher risk. In this systematic review, 24 studies were selected and reviewed to identify different signs, symptoms and definitions for constipation in people with a learning disability. Studies which used self-composed definitions, which were specifically adapted for this population, scored higher on the quality assessment than studies using standardized definitions. However, the results were inconsistent, and no well-evidenced definition could be agreed upon. This study demonstrated the need for agreed signs, symptoms and definitions for the diagnosis of constipation in this at-risk population.

1) Sample: 10,204 people with Down syndrome, 39,814 general population controls, 69,150 people with intellectual disabilities.
2) Setting: UK electronic health records (1990-2020)
3) Level of evidence: 2

Multiple morbidity across the lifespan in people with Down syndrome or intellectual disabilities: a population-based cohort study using electronic health records.

Baksh et al. (2023)
DOI: https://doi.org/10.1016/S2468-2667(23)00057-9

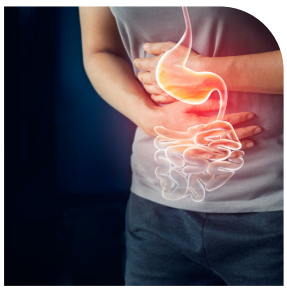
This study aimed to compare patterns of health conditions between people with Down syndrome, people with intellectual disabilities and people from the general population. People with Down syndrome are shown to be at an increased risk for dementia, hypothyroidism, epilepsy and hematological malignancy and a reduced risk for asthma, cancer, ischemic heart disease and hypertension than the general population. When compared to other people with intellectual disability, people with Down syndrome are at an increased risk of dementia, hypothyroidism and obstructive sleep apnea, and a reduced risk for asthma, cancer, certain dental issues, sleep disorders, diabetes, mood disorders and anxiety. These results suggested that people with Down syndrome have different health patterns than both the general population and others with intellectual disability. This is important for health screening, prevention and intervention.

1) Sample: 56 autistic children and 139 not autistic children (ages 2-17)
2) Setting: Children's hospital in the USA
3) Level of evidence: 3

Constipation prevalence and perceptions: Comparison of children and adolescents with ASD and other developmental-behavioural disorders.

Cuffman & Burkhart. (2021)
DOI: https://doi.org/10.1016/J.RASD.2020.101710

Constipation is a commonly reported health condition in autistic people and can be difficult to diagnose. This study investigated constipation in autistic children, compared to children with other developmental-behavioural conditions. Constipation surveys were given to children and caregivers, involving questions about the Rome IV criteria and personal perceptions. Toilet-trained autistic children were more likely to have experienced constipation and had a higher likelihood of being on constipation medication. There were differences between child and parent ratings of the Rome IV criteria for constipation. These results highlighted the importance of considering the child's perception of constipation to ensure a thorough assessment.

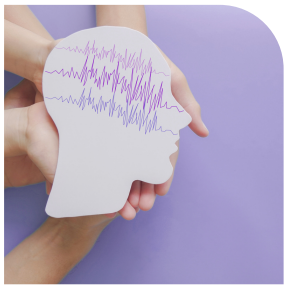


1) Sample: 173 people with Angelman syndrome, aged 3-17 years.
2) Setting: Global Angelman Syndrome Registry.
3) Level of evidence: 3

Association between early and current gastro-intestinal symptoms and co-morbidities in children and adolescents with Angelman syndrome.

Leader et al. (2022)
DOI: https://doi.org/10.1111/JIR.12975

Angelman syndrome is a developmental condition that can cause intellectual disability, deficits in language and motor skills, sleep issues and epilepsy. Angelman syndrome is also associated with gastrointestinal (GI) symptoms. This study compared childhood histories, toileting, behaviour, communication and sleep in children with Angelman syndrome who experienced either a high or low frequency of GI symptoms. Constipation occurred in 84% of the sample and was the most commonly reported GI symptom. GI symptoms were associated with several childhood factors, like vomiting and difficulty gaining weight. GI symptoms were also associated with sleep and toileting issues. GI symptoms were not associated with challenging behaviour or communication. This study adds to our understanding of Angelman syndrome and constipation. The results may inform professionals about co-occurring symptoms of GI in children with Angelman syndrome and highlight possible predictors of constipation for future research.



1) Sample: 308 autistic children, 110 with co-occurring intellectual disability
2) Setting: Specialist clinic for autistic children in the USA (2010-2019)
3) Level of evidence: 3

Prevalence of gastrointestinal symptoms among autistic individuals, with and without co-occurring intellectual disability.

Holingue et al. (2023)
DOI: https://doi.org/10.1002/AUR.2972

The measurement of gastrointestinal (GI) symptoms can be difficult in autistic children. This may be more challenging in the presence of co-occurring intellectual disability. This study explored whether parents of autistic children with an intellectual disability were less certain in their reporting of GI symptoms than parents of children without an intellectual disability. After rating a list of GI signs and symptoms, parents of autistic children with an intellectual disability were less certain about reporting 'internal' symptoms like nausea or bloating. There was no difference in reporting 'external' symptoms, like diarrhoea, between groups. The authors reported that among parents who were certain about symptoms, there were no major group differences in the number of symptoms. The results suggest that co-occurring intellectual disability, in addition to being autistic, does not increase the likelihood of GI symptoms but that parental reporting alone for GI symptoms is not sufficient for reliable diagnosis.

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Medicine use in people with intellectual disabilities: A Finnish nationwide register study.

Nurminen et al. (2022)
DOI: https://doi.org/10.1111/JIR.12988

1) Sample: 37,196 people with intellectual disabilities and 37,196 sex and age matched controls
2) Setting: Finnish healthcare registers (2017-2019)
3) Level of evidence: 2

Compared to the general population, people with an intellectual disability tend to have poorer health, require more support from healthcare services and need more medication. This study aimed to define the population of people with an intellectual disability in Finland and to compare their medicine use with age and sex matched controls from the general population. People with an intellectual disability used more medication and spent more money on medication than the comparison group. The difference in medication use was largest in children and adolescents, and smallest in older adults. In people with intellectual disability, antipsychotic use was four times higher and medication for epilepsy, constipation, supplements and anxiolytics were four to seven times higher. The findings highlighted the potential risk of over and underusing medication and other drug-related issues. These differences in prescription rates need to be understood to ensure that medication use in this population is effective, safe, cost-effective and rational.

Reducing risks associated with medicines and lifestyle in a residential care population with intellectual disabilities: evaluation of a pharmacy review initiative in England.

Thayer et al. (2021)
DOI: https://doi.org/10.1136/BMJOPEN-2020-046630

1) Sample: 160 care home residents with intellectual disabilities (ages 24-79)
2) Setting: Residential care homes in the Wirral, England.
3) Level of evidence: 3

This study evaluated pharmacist intervention for care home residents with an intellectual disability. The aim was to understand medication and health outcomes of pharmacist reviews and to understand whether these recommendations aligned with LeDeR priorities. 160 care home residents were prescribed 1207 medicines. 74% of these residents were prescribed over 5 medicines. 507 pharmacist recommendations were made, and of the reviews discussed with general practitioners or psychiatrists, 86% were accepted. The most common recommendations were lifestyle-risk related, changing medication or stopping medication. These findings highlight the extent of multiple medication prescriptions in care home residents with intellectual disabilities. The high recommendation acceptance rate demonstrated the success of the pharmacist intervention programme. Widespread use of this programme could have positive outcomes for people with an intellectual disability and reduce the pressure on other health services.

Gastrointestinal and sleep issues in toddlers with autism versus other neurodevelopmental disorders.

Zlatnik et al. (2023)
DOI: https://doi.org/10.1177/00099228231156774

1.) Sample: 500 autistic children and 146 children with other neurodevelopmental conditions
2) Setting: Outpatient clinic in the US.
3) Level of evidence: 3

Autistic children are likely to have co-occurring gastrointestinal (GI) symptoms and sleep issues. However, there is little evidence comparing the rates of GI symptoms in autistic children to children with other neurodevelopmental conditions. This paper aimed to compare the prevalence and type of GI symptoms and sleep difficulties between a group of autistic toddlers and a group of toddlers with other neurodevelopmental conditions. Of the entire sample, 46% had GI symptoms and 22.6% had sleep difficulties. Toddlers with other neurodevelopmental conditions were more likely to have GI symptoms and sleep difficulties than autistic toddlers. The most commonly reported GI symptoms were food aversions, picky eaters, reflux and constipation. These results demonstrated the need to assess all children with developmental conditions for GI symptoms and sleep difficulties, not just autistic children.

Global prevalence of autism spectrum disorder and its gastrointestinal symptoms: a systematic review and meta-analysis.

Wang et al. (2022)
DOI: https://doi.org/10.3389/FPSYT.2022.963102

1) Sample: 126 primary research studies
2) Setting: Studies on Pubmed, Web of Science, and Embase, in March 2022
3) Level of evidence: 1

The global prevalence of autism has increased over the last 40 years. In autistic patients, gastrointestinal (GI) symptoms are common. The aim of this study was to provide an updated estimate of the global prevalence of autism and the prevalence of GI symptoms in autistic patients. A systematic review and meta-analysis of papers from multiple databases was conducted. The worldwide prevalence of autism was 98 in every 10,000 people, autism was more common in males than in females and autism was more prevalent in developing countries than in developed countries. The pooled prevalence of GI symptoms in autistic patients was 48.67% and the main symptom was constipation (26.17%). Rates of both autism prevalence and GI symptoms are increasing in both developed and developing countries.