Exploring the validity of screening for learning disability – using the Wilson-Jungner screening programme criteria

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Abstract

Recommendations have been made about how to identify people in primary care services who are likely to meet the criteria for learning disability, as one step in reducing health inequalities. In this paper we use the Wilson-Jungner appraisal criteria for screening programmes, to explore whether the introduction of such screening for learning disability can be considered to be feasible and valid. We consider this in the context of the currently recommended screening checklist and alternative evidence-based screening tools. We conclude that, while learning disability is a social construct and the Wilson-Jungner criteria were originally designed to be applied to screening for specific diseases, introducing the routine use of accurate and evidence-based screening tools, into primary care is largely consistent with the Wilson-Jungner criteria.

Keywords: learning disability; screening; Wilson-Jungner criteria; nurses; primary care

Background

Initiatives which are designed to address the health inequalities which continue to be experienced by people with a learning disability require the person's learning disability to be recognised in the first place. Research has consistently shown that people with a learning disability experience delayed or missed diagnosis, especially if their learning disability is mild, and that the majority do not have their learning disability recorded at all, meaning that they remain 'hidden' (Emerson and Glover 2012). As knowledge about learning disability is low in many staff groups, including health (Emerson et al 2012) and social care staff (McKenzie et al 2022), indicators of learning disability are likely to be missed. This means that many people with a learning disability will not be offered appropriate and targeted healthcare. Their health is likely to suffer as a result. As an example, in order to benefit from additional support offered by primary care services, such as annual health checks, flu vaccinations and other reasonable adjustments in health care provision, the person has to be registered with the practice as having a learning disability.

There have been attempts to address some of these issues by introducing screening methods to identify people who are likely to meet the criteria for learning disability into primary care. Such methods are not designed to replace full diagnostic assessment, but if they are evidence based and accurate, they can offer a way of identifying those people who have an increased likelihood of meeting the criteria for learning disability. As such, they can help health staff make decisions about who would benefit from further assessment, placement on the learning disability register, and appropriate adjustments in their support and healthcare.

In this paper, we will use the Wilson-Jungner (Wilson and Jungner 1968) appraisal criteria for screening programmes, to explore whether the introduction of routine screening for learning disability would be considered to be feasible and valid. We will consider this in

the context of the ‘Learning Disability Register Inclusion Tool’ that has been signposted to primary care staff by NHS England and NHS Improvement, (2019) and alternative the evidence-based Learning Disability Screening Questionnaire (LDSQ) which has additionally been recommended by the Royal College of General Practitioners (RCGP, n/d).

The validity of screening for learning disability – the Wilson-Jungner criteria

The Wilson-Jungner criteria (Wilson and Jungner 1968) were developed over 40 years ago. They are still considered the gold standard for appraising the validity of a screening programme, although there have been some suggested adaptations to take account of current advances and debates in healthcare provision (Andermann et al 2008). The criteria were originally developed in relation to screening programmes which were designed to identify specific diseases or genetic conditions. When used as a framework in relation to learning disability they highlight some important differences between specific diseases and a condition that is fundamentally a social construct. A summary of the criteria and some of these associated issues, if considering a routine screening programme for learning disability, are outlined below.

The Wilson-Jungner criteria

1. **The condition being screened for should be an important health problem**

While having a learning disability does not in itself constitute a health problem, it is associated with health inequalities. Many people with a learning disability experience serious and/or chronic health conditions, with common causes of death in the years prior to the Covid-19 pandemic including epilepsy and bacterial pneumonia (NHS England and NHS Improvement 2021). Covid-19 also had a greater impact on people with a learning disability, with a death rate that was over 3 times greater than that for the general population (Public

Health England 2020). The seriousness with which these health issues are taken is reflected in the introduction, over the years, of a number of initiatives. These have been developed to try and improve the health of people with a learning disability and have included the introduction in primary care of learning disability registers and associated reasonable adjustments in health care, such as annual health checks (e.g., NHS England 2018). Research suggests that such approaches can be successful. Health checks, carried out within primary care, have been found to increase health promotion and clinical interventions, such as vaccinations, at least in the short-term (Byrne et al 2016). They have also led to the identification of previously unrecognised health conditions (Robertson et al 2014).

Despite such initiatives, people with a learning disability continue to experience a number of health inequalities, the most significant of which relates to life expectancy. Recent figures suggest that dying from an avoidable medical cause was three times more likely for people with a learning disability than for the general population (The LeDeR Team 2021). This suggests that screening for learning disability would offer a route into addressing significant health inequalities, thereby addressing an important health issue.

2. The natural history of the condition should be well understood

There is a growing understanding of the genetic and environmental factors that can lead to learning disability. Likewise, the nature of many of the chromosomal conditions, such as Down Syndrome and Fragile X Syndrome, and metabolic disorders, such as Phenylketonuria that are associated with learning disability is increasingly well documented. Learning disability does, however, have multiple causes including chromosomal abnormalities, infection and environmental, and in many cases the cause is unknown (Shree and Shukla

2016). This means that there is not one natural history of the condition that can be understood.

While screening already exists for specific conditions that are associated with learning disability, such as Down Syndrome, the purpose of screening for learning disability as a social construct is not to prevent it, but to help identify those who are likely to have it, in order that the many associated health problems and other difficulties can be effectively addressed. In this respect, this appraisal criteria may not be considered to be met in relation to learning disability as a broad construct.

3. There should be a detectable early stage

Developmental delays, for example in motor skills or early language may be apparent by the time a child is two, particularly if the learning disability is more severe (American Psychiatric Association [APA] 2021). As learning disability is defined by significant impairment in intellectual and adaptive functioning, with childhood onset (APA 2013) the condition is diagnosed on the basis of standardised assessments of these components, as well as developmental history. Very early assessment is less accurate because of the rapid development of children and so diagnosis, particularly of mild learning disability, may not occur until school age, when the child's difficulties become apparent when faced with increased academic demands (Voigt and Accardo 2016). The fact that many people experience missed or delayed diagnosis (Emerson and Glover 2012) reflects that learning disability can be challenging to diagnose. It is, however, possible to detect at a relatively early age.

A suitable test should be devised for the early stage

One way for primary care staff to identify people who are likely to meet the criteria for learning disability (NHS England and NHS Improvement 2019) is from diagnostic codes combined with a 21 item 'Learning Disability Register Inclusion Tool.' This method does, however, have limitations. It is not underpinned by a published evidence base, no information is provided about accuracy, reliability or validity, and users are not provided with a cut-off score that would help guide them in their decision about whether the person is likely to meet the criteria for learning disability or not. It is also unclear to what extent it is sensitive to the age of the person who is being screened. This makes the outcome difficult to interpret, making the tool of potentially limited benefit. This would suggest that it is not an entirely suitable tool.

Alternative recommended screening methods do, however, exist. The LDSQ is recommended as part of the RCGP (n/d) 'Health Checks for People with Learning Disabilities Toolkit'. The LDSQ has been found to accurately identify adults who are likely to meet the criteria for learning disability (e.g., McKenzie et al 2015) and can be used with people aged 16 years and above. There is also a version which can identify children who are likely to meet the criteria of learning disability at a relatively young age. The Child and Adolescent Intellectual Disability Screening Questionnaire (CAIDS-Q) can accurately distinguish between children with and without learning disability from ages 6-18 (e.g., McKenzie et al 2019).

Both the CAIDS-Q and LDSQ have an established evidence base, good psychometric properties and are accurate at differentiating between individuals with and without a learning disability in a number of different service settings. They have been successfully used to identify individuals who were not previously known to have a learning disability (see McKenzie et al 2021 for an overview). As such, they could be considered to be suitable

screening tools. They will be used as example screening tools in relation to the subsequent Wilson-Jungner criteria.

4. **The test should be acceptable**

Being diagnosed with a learning disability can result in the person being stigmatised and feeling shame, which in turn can impact negatively on mental health (Clapton et al 2018). It is, therefore, important that any screening test is acceptable to those involved in its use. Research suggests that the use of both the CAIDS-Q and LDSQ as screening questionnaires are acceptable to professionals, parents, and service users. Fewer than 20% considered that screening would cause the person to feel stigmatised (McKenzie et al 2021) and many benefits to using screening questionnaires were identified (see below).

5. **Intervals for repeating the test should be determined**

Research suggests that the CAIDS-Q and LDSQ have good psychometric properties and the outcome on the screening tests have high correlations with clinical diagnoses of learning disability (e.g., McKenzie et al 2015, 2019). This suggests that the outcome is generally accurate and would indicate that there is not an ongoing need for repeated screening. The CAIDS-Q does, however, have different cut-off scores for children under 8 years and children aged 8 years and above, to reflect the changes in development and skill acquisition that occur throughout childhood. It may, therefore, be helpful for screening to be repeated if the child was initially screened at a younger (e.g., age 6). Screening again at age 8 could help confirm if the outcome remained the same as it was at the earlier age.

6. **Adequate health service provision should be made for the extra clinical workload resulting from screening**

Screening with the CAIDS-Q and LDSQ generally takes less than 5 minutes, meaning that the screening process itself is likely to have minimal impact on the workload of clinical staff. Both questionnaires have 7 core items, which is 14 fewer than the 21 item 'Learning Disability Register Inclusion Tool.' The former tools also have easy to use evidence-based cut-off scores. The recent development of online versions of the screening tool, which automatically calculate the score and provide immediate feedback about whether the person is likely to meet the criteria for learning disability or not, means that the workload required for conducting screening is reduced further (see https://learningdisabilitymatters.co.uk/tools/)

There is limited research into the extent to which identifying more people with a learning disability would impact on the workload of clinical staff. It could, however, be argued that the use of accurate, evidence-based screening tools is likely to reduce workload by increasing awareness of learning disability, allowing referrals for full diagnostic assessment to be prioritised (McKenzie et al 2019a) and preventing associated health conditions from becoming more chronic and expensive to treat. For example, evidence indicates that annual health checks offered to those on GP learning disability registers result in the detection and targeted treatment of previously undiagnosed conditions, many of which are serious or life-threatening (Robertson et al 2014).

7. **Treatment at an early stage should be of more benefit than at a later stage**

As people with a learning disability have a developmental condition that is associated with significant difficulties with their intellectual abilities and day to day skills, there are a number of areas where they could potentially benefit from early support and interventions. This might

be in relation to day-to-day skills (e.g., Sheppard and Unsworth 2010), academic skills, such as reading (Kemal, Wilkerson, and Ruppar 2018) or health interventions (Byrne et al 2016, Robertson et al 2014). Research suggests that early intervention can both help reduce the decline in the intellectual development of children with learning disabilities and help improve their social and cognitive skills in the longer term (Guralnick 2017).

Early identification as a result of screening has also been found to have a number of direct and indirect benefits (McKenzie et al 2021), including identifying and promoting increased understanding of the support needs of people with a learning disability and their families/carers, and facilitating access to resources, such as additional support at school.

8. The risks, both physical and psychological, should be less than the benefits

There are no medical risks associated with the use of the CAIDS-Q and LDSQ screening questionnaires, as the process is physically non-invasive. While there is the possibility that those who are identified as likely to meet the criteria for learning disability may feel stigmatised, as outlined previously, research suggests that this was identified as a potential psychological risk by less than 1 in 5 parents, service users, and professionals. The LDSQ has also been identified as a means by which the issue of prisoners being ashamed to highlight their difficulties can be addressed (McKenzie et al 2021).

9. The costs should be balanced against the benefits

Overall, research suggests that use of the CAIDS-Q and LDSQ has few disadvantages and many advantages (McKenzie et al 2021). The most obvious advantage and key purpose of the screening tools is that they accurately identify people who are likely to meet the criteria

for learning disability. This then opens the possibility for further assessment, support, and intervention. In the case of primary care services, this can trigger a range of reasonable adjustments to health care, including the offer of annual health checks.

Implications for practice

Recent recommendations by NHS England and NHS Improvement (2019) and RCGP (n/d) were aimed at helping primary care staff to identify those people who were likely to meet the criteria for learning disability in their practice. In turn, the purpose of this screening exercise was to address the significant health inequalities faced by people with a learning disability. This paper explored the validity of the premise of introducing routine screening for learning disability, using the Wilson-Jungner validity criteria (Wilson and Jungner 1968). This exercise indicated that, while learning disability is a social construct, rather than a specific disease or health problem, routine screening for learning disability met many of the validity criteria outlined by Wilson and Jungner (1968).

Learning disability is associated with many complex, significant and life-threatening health problems; it can be diagnosed at a relatively young age; suitable, acceptable screening tools exist which can accurately screen for learning disability from age 6 upwards; repeated use is unlikely to be needed except for in the case of the youngest children; their use is likely to have minimal impact on workloads; the risks and costs would appear to be outweighed by the benefits and there are significant benefits of early identification.

While the initiative to identify people with a learning disability has been located in primary care, learning disability nurses are also well-placed to administer, or signpost colleagues to the screening questionnaires, given their expertise in learning disability, links with colleagues in primary care, and involvement in annual health checks for people with a

learning disability (see Robertson et al 2014). While the CAIDS-Q and LDSQ don't require the person using them to have a particular specialist professional background or training, the learning disability nurse will have the experience to put the results of the screening questionnaires in context for the person being screened and liaise with colleagues if further action is required, such as further specialist assessment.

Conclusion

The paper suggests that introducing the routine use of accurate and evidence-based screening tools, such as the CAIDS-Q and LDSQ, into primary care is largely valid, when considered against the Wilson-Jungner criteria. This may offer an appropriate way of helping to identify those who are likely to meet the criteria for learning disability and help reduce the health inequalities experienced by this group of people.

Implications for practice
• The Wilson-Jungner appraisal criteria for screening programmes helped identify whether the introduction of screening for learning disability could be considered to be feasible and valid. • The use of short, accurate, screening questionnaires can provide an evidence-based method to identify people in primary care who are likely to meet the criteria for learning disability. • This may offer an important first step in addressing the health inequalities experienced by people with a learning disability.

- Learning disability nurses are well placed to use such screening tools and to support their primary care colleagues in doing so.

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