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For patients that died in hospital, staff perceptions of the last days of life of patients they cared for were evaluated using a Likert scale and free-text options. This allowed three key themes to be identified; coordinated care, culture of practice and complexity of care. These three themes can be used to identify areas of the dying process that went well and areas that could be improved. This tool could be used to improve the dying process for patients, family/whānau and staff.

Endoscopic retrograde cholangiopancreatography in the comorbid elderly: a retrospective comparative study in New Zealand

Kirsty Macfarlane, Reuben Wilson, Nicholas J Fischer, Henry Wei

Endoscopic retrograde cholangiopancreatography (ERCP) is a procedure used to treat disorders of the liver and bile duct (drainage of the liver). It is debated whether ERCP is safe for the elderly compared to younger patients. This study found ERCP to be relatively safe, although careful consideration is required when selecting patients with comorbidities to avoid subjecting those with short life expectancy to unnecessary procedures.

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Nastassia Randell, Suzanne Barker-Collo, Kathryn Murrell, Callum Wilson

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Documented incontinence after stroke. A secondary analysis of a cohort study. Reducing Ethnic and Geographic Inequities to Optimise New Zealand Stroke Care (REGIONS Care)

E Jean C Hay-Smith, Stephanie G Thompson, Mark Weatherall, Annamarei Ranta

A previous large New Zealand study of acute stroke care had collected data about bladder and bowel leakage. We examined these data to find out how common it was to have incontinence after stroke, whether it was more common for some ethnicities, and whether people with incontinence had worse outcomes. We found 14% of people had documented incontinence, which is much less common that previous New Zealand or international studies. People living with stroke and incontinence were more likely to die or be living in a residential care up to 12 months after stroke. People living with stroke and incontinence also had less good quality of life, due to the incontinence.

We still don't count: the under-counting and under-representation of Māori in health and disability sector data

Ricci B Harris, Sarah-Jane Paine, June Atkinson, Bridget Robson, Paula T King, Jennifer Randle, Anja Mizdrak, Melissa McLeod

This paper explores the undercounting of Māori in health data. It shows that Māori are under-represented in health care user data and in primary care data. When a Māori individual's self-identified ethnicity on the 2018 Census was compared with their ethnicity on primary care enrolment data, 21% of people who identified as Māori on the Census were not recorded as Māori on their health data. This has important implications for individuals' access to care and in the estimation of rates of health outcomes, service use and inequities. Improving ethnicity data quality should be an urgent priority for the health system.

Mental health inequities for Māori youth: a population-level study of mental health service data

Reremoana Theodorea, Nick Bowdenb, Jesse Kokauac, Troy Ruhed, Matt Hobbse, Sarah Hetrickf, Lukas Marekg, Jesse Wikih, Barry Milnei, Hiran Thabrewj, Joseph Bodenk

In this population-wide study we examined administrative mental health service data for Māori youth compared to non-Māori/non-Pasifika youth (10–24 years). Using specialist mental health service, hospital discharge and pharmaceutical dispensing data, we found that Māori were less likely to be identified for emotional conditions (anxiety and/or depression) than NMNP. They were more likely to be identified for substance problems and self-harm. Māori living in high deprivation areas compared to Māori in least deprived areas were significantly more likely to be identified for substance problems, but less likely for emotional conditions. These findings suggest that rangatahi Māori as compared to NMNP youth are less likely to be identified at earlier stages of distress until distress symptoms become more severe with explicit markers (e.g., self-harm injuries).

The past, present and future of liver cancer control for Māori

Sydney Clough, Tara Cleverley, Clarence Kerrison, Matire Henwood, Jonathan Koea, Jason K Gurney

Liver cancer is a substantial health problem for Māori. Māori are more likely to be diagnosed with liver cancer than non-Māori, and less likely to survive once diagnosed. This disparity is primarily driven by difference between Māori and non-Māori in exposure to the viruses hepatitis B and hepatitis C. In this manuscript, we call for a national primary care-based programme to detect and treat hepatitis B and C and to screen for liver cancer among high-risk patients, along with renewed effort to maximise hepatitis B vaccination rates, to reduce the burden of liver cancer for Māori in Aotearoa.

Whakairo: carving a values-led approach to understand and respond to the mental health and substance use of the New Zealand population

Helen Lockett, Cameron Lacey, Angela Jury, Talya Postelnik, Amanda Luckman, Richie Poulton

Good data in the hands of the people is imperative for supporting systems change, upholding Te Tiriti o Waitangi, and addressing inequities.

Epidemiological data provide reliable information on the nature, range, extent, frequency, geographical spread, and duration of health conditions. Good data also provide an understanding of the factors contributing to and protecting against these conditions, and information on the accompanying impact on peoples' lives.[1]

We (the authors) are proposing an innovative approach to defining what good epidemiological data is in the Aotearoa New Zealand context; how to collect it, and most importantly how it can be used to design and offer supports and services that respond to peoples' needs.

National prevalence surveys

In 2006, the landmark epidemiological study, *Te Rau Hinengaro: The New Zealand Mental Health Survey*, was published.[2] For the first time this gave Aotearoa New Zealand population level information on the prevalence of a range of mental health conditions and substance use disorders. The findings from this survey have been used extensively to inform planning a system-wide response to peoples' needs.[3–5]

The data informing *Te Rau Hinengaro* was collected in 2003 and 2004, so New Zealand's latest prevalence data is nearly 20 years old. The recommended period between national prevalence surveys is every 8 to 10 years.[6] Currently we have no accurate and comprehensive population prevalence data to inform policy, service, and workforce planning now and in the future. This is a massive systems-level data gap.

We are very concerned about this lack of robust up-to-date prevalence and impact data. As a group we come from different perspectives and backgrounds including lived experience, psychiatry, research, Māori, and non-Māori. We know that for some people and communities, things are getting tougher, not easier.[7] An understanding of the different needs of our diverse communities is imperative to effectively support people.

We must move forward with gathering this information. It is unethical not to invest in high quality data to inform such an important area of health service delivery. Robust data on mental health conditions and addiction in the population are crucial to upholding Te Tiriti o Waitangi, achieving equity, and is a Government responsibility. This data gap has been recognised and highlighted as a priority, notably in the *Data Investment Plan for Aotearoa New Zealand*, the plan commissioned by the Government Chief Data Steward to guide government investment in data, and in *Kia Manawanui: The Long-Term Pathway to Mental Wellbeing*.[8,9]

A comprehensive study, or series of studies will quantify and identify: (1) the distribution of mental health conditions and problematic substance use in the population; (2) the factors that both protect against and contribute to these conditions; (3) the impact these conditions have on people, whānau, and communities; and (4) where there is need and unmet need. This level and type of information can inform the distribution of resources and support now, and in the future.

Appraising existing data

In July 2022, researchers from Te Pou, a national workforce centre for mental health, addiction and disability, and the Department of Māori Indigenous Health Innovation (MIHI) at the University of Otago started engaging with stakeholders to advocate for a national epidemiological survey. Discussions are being held with a wide range of stakeholders including people involved in *Te Rau*

Hinengaro, Māori academics and clinicians, lived experience and whānau advisors, as well as policymakers, researchers, and clinical professional bodies. The intention is to pull together a collaborative group to advocate for, design, and identify funding for this work.

Te Pou have published a series of reports and resources exploring existing data and commonly used measures for the adult population.[10–12] Similarly, Theodore and colleagues in this issue highlight the inequities for Māori youth compared to non-Māori from an examination of routinely available service use data.[13] What these, and other recent surveys show is that something different is happening emotionally, particularly for rangatahi, and that there are growing inequities across different priority groups which we must pay attention to.[7,14,15]

To respond appropriately and effectively, a better understanding of the person, their whānau, community and their needs is required. This means collecting data that goes beyond the reliance on brief screening measures, like the Kessler-10 (K10), World Health Organization – Five Well-Being Index (WHO-5), or the Alcohol Use Disorders Identification Test (AUDIT). These measure psychological distress, symptoms of anxiety or depression, hazardous drinking, or general wellbeing, which tell us something is going on, but not any wider contextual information or the impact. Brief screening tools are intentionally designed to identify more people, and therefore overestimate prevalence.[16,17] In addition, symptoms such as psychological distress often have peaks and troughs in a population, without similar rises in underlying prevalence of mental health conditions and substance use disorders.[12,18]

Global and national events, such as the COVID-19 pandemic and subsequent lockdowns are known to have psychological impacts on the population, but it remains unknown whether there has been a greater shift in underlying prevalence of mental health conditions or substance use disorders, or whether the rises we are seeing in levels of psychological distress are a reactionary peak.

It is not sufficient to rely on service use data as there is large scale underreporting of mental health conditions and substance use disorders, as only around one-third of people experiencing these conditions will seek help.[2,6] This means service use data is inherently biased towards the people seeking and people accessing help, and will be under-representative of particular groups, such as young people, Māori, people from lower socio-economic groups, and people in rural areas. An overreliance on service use data is highly problematic for planning purposes as its use will tend to maintain the status quo.[1]

Developing a values-based evaluative framework

While people have told us that they generally support an in-depth epidemiological survey, there are multiple perspectives to consider. To effectively integrate these perspectives, we are proposing an evaluative framework to support the design of the research questions and methods, as well as the analysis, presentation, and dissemination of results. Research methods will be evaluated against criteria within each of the six domains proposed below. This will support critical review of the utility and appropriateness of the design, methods, and dissemination of findings.

From conversations with stakeholders, six domains for the framework have emerged:

1. Taking a Te Tiriti o Waitangi led approach—advancing Māori health.
2. Valuing and including people with lived experience.
3. Utilising scientific methods.
4. Practicality and utility—using and enhancing existing data sets.
5. Inclusive of diverse population groups.
6. Accessible and responsive, with timeliness of data feedback.

We are in the process of working with different stakeholder groups to operationalise these domains and start applying them to the design phase of this work.

Data for change

This epidemiological work is integral to bringing effective and equitable changes to the way we plan, purchase, and deliver mental health and addiction supports. Prevalence and impact data can inform the targeting of current and future investment to support better outcomes for people, whānau and communities, and provide information on the effectiveness of investments.

Prevalence data generate information to help identify where support should be targeted based on need. We know that experiences of distress are increasing, but what this means for mental

health and addiction services and other health and social services is unknown.

To speculate on something as important as peoples' emotional health and wellbeing is not acceptable. The process we are proposing is to carve out a plan to deliver accessible, usable, and reliable knowledge on mental health and substance use that can be used in the hands of people, communities, and the Government to invest well in our population's mental health and wellbeing now and in the future.

For further information on this collaborative work visit the Te Pou website: Understanding population mental health and substance use | Te Pou; or contact Helen Lockett at helen.lockett@tepou.co.nz or Cameron Lacey: cameron.lacey@otago.ac.nz

COMPETING INTERESTS
Nil.

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Dying in hospital—staff perceptions on providing quality care

Claire Whitehead, Kate Grundy, Rachel Wiseman, Suzanne Pitama

ABSTRACT

AIM: To understand what healthcare staff perceive contributes to the quality of patient and family/whānau experiences of dying and death on a hospital inpatient ward.

METHOD: A survey was created, piloted and sent to all staff members who had cared for a deceased patient within two working days of their death, at Christchurch Hospital (CH), New Zealand. The survey comprised questions evaluating whether the patients physical, emotional, social or family/whānau needs were met, using both a Likert scale and free-text options. The survey was sent over a three-month period in 2016/2017.

RESULTS: A total of 169 staff responded to the deaths of 51 patients. The majority (71.3%) of staff agreed that "end-of-life care was of a high standard", with the physical symptoms domain holding the highest score for both agreement (68%) and disagreement (13%) that "physical symptoms were well managed". Qualitative analysis of free-text responses revealed three themes: coordinated care (service delivery, complex case or communication needs, teamwork); culture of practice (dignity, trust, respect and relationships); and complexity of care (encompassing complex physical symptoms or patient or family/whānau interpersonal dynamics).

CONCLUSION: Evaluation of quality of death in hospitals can be enhanced by routine use of surveys of staff who cared for the deceased person. Such surveys could comprise part of a suite of tools to provide a holistic view of dying and death, complementing methods such as retrospective audits and family/whānau interviews.

Given the universality of death, examining the quality of clinical care around dying should be part of routine healthcare.[1] A poor quality experience can lead to a more complex grieving process for family/whānau and distress or burnout for staff. When the death is anticipated, family/whānau distress can be reduced by allowing the patient, the whānau and the healthcare team to understand and prepare for what is happening.[2]

There is no agreed consensus on what constitutes a "good death" and how to define quality with respect to the dying process. Quality measures must take account of the *process of* dying as well as the death as an *acute event*.[1] Developing international standards is intrinsically difficult as dying is a dynamic process, deeply personal to the individual.[3] Often, a proxy is required to speak for the deceased, such as a family member or health professional.[1,4–6] These opinions vary greatly depending on their relationship to the patient and their own personal values.[1,3,6,7] The retrospective nature of exploring a person's death also makes defining exact outcomes subject to retrospective bias.[7,8]

Most tools evaluating dying and death are based on a mixture of literature reviews and expert opinion, including both qualitative and quantitative research.[6] Scales are based on differing frameworks and definitions, developed in different cultural settings and most use quantitative measures such as Likert scales.[1,5] Reliability and validity has not been robustly tested for many of the measures.[1,6] Meier et al., in their 2016 literature review of "defining a good death", found that physical symptoms are well studied in comparison to other aspects of the dying process.[5] They suggest that future work should include a wider variety of ages and cultures.[5] Furthermore, it is acknowledged that any instrument designed to measure dying and death must encompass physical, psychological, cultural, and spiritual aspects.[7,9]

A previously published quality of dying audit tool was created as part of a comprehensive framework of clinical audit in Canterbury District Health Board (CDHB).[10] CDHB services a 0.5 million population and has a single tertiary centre, Christchurch Hospital (CH), which is the largest hospital in the South Island of New Zealand. CH has an 800 bed capacity, with over 1,000 inpatient deaths per year.

The quality of dying audit tool is used on a regular basis across the CDHB, and involves a detailed review of clinical documentation looking for factors known to contribute to quality patient and family/whānau experiences of dying and death.

It focuses specifically on holistic, cultural and spiritual domains. As an extension to the chart audit, staff directly involved in caring for the person were surveyed regarding their own perceptions and experiences of that death and the dying process that preceded it.

The purpose of this study was to explore whether a survey of this type was feasible and acceptable to staff, and to understand what they perceive contributes to the quality of patient and family/whānau experiences of dying and death in an acute hospital setting.

Method

In 2016, the Canterbury Regional Blood and Cancer Service at Christchurch Hospital (CH) developed a survey with the aim of retrospectively capturing staff members' views of recent deaths of patients under their care.

This survey was piloted with staff members who cared for 13 patients who died in the Oncology ward in mid-2016. A second round of pilot testing followed in December 2016 on a further nine patients, to test the survey distribution and operationalisation processes. Deaths of inpatients in the oncology, respiratory and haematology departments were identified from daily mortuary records. The survey was sent via a web link to all staff in the respective departments within two working days of the death, inviting those who had cared for the patient prior to death to respond. The survey was hosted on RedCap (RedCap Consortium, Vanderbilt) and filled out anonymously, other than identifying their role in the patients care. There were no patient identifiers contained in the email to preserve confidentiality, but upon completing the survey, staff were asked to enter the National Health Index (NHI) number and name of the patient, so that responses for the same patient could be collated.

Feedback received from this round of pilot testing refined the delivery method, such that individual staff involved in the care of the patient in the last 48 hours of life were identified from the clinical record and survey web links subsequently targeted to these individuals only. In addition, as a result of this round of pilot testing, the two questions around family/whānau support needs were merged (Items 8 and 9, Appendix 1). A separate free-text section was added which allowed staff to comment on the needs of the family/whānau. An option for staff to request further support was also added. Appendix 1 shows the final version of the survey, indicating where changes were made after review.

The survey was implemented across selected departments in CH (Oncology, Bone Marrow Transplant Unit, Respiratory, Emergency Department (ED) and General Surgery) between November and 2016 and January 2017. A single researcher (CW) was responsible for identifying deaths on a daily basis and coordinating emails to staff.

Quantitative analysis included descriptive statistics. Answers that were ranked, "strongly agree" and "agree" were added together, as were "strongly disagree" and "disagree".

Qualitative analysis was used to order data extracted from the free-text questions (10, 11 and 12). Inductive thematic analysis was determined as appropriate to accurately give voice to participants' commentary. Four members of the research team (CW, RW, KG and SP) agreed the design and development of two cycles of coding.[11] The first cycle of coding involved descriptive analysis that examined responses to each free-text question and identified similarities and differences between participants' perspectives and experiences on aspects of care. The second cycle of coding involved two further review processes, firstly pattern coding was utilised to collate and triangulate overall responses where the content being discussed was similar. Descriptive coding was then utilised to explore the depth of content areas being discussed by participants. The inclusion criteria for each code, drawn from the descriptive, and category, drawn from pattern coding, was discussed and defined by the research team during data analysis. The research team then further identified which categories articulated the same phenomenon and grouped these as themes. Two of the coders have extensive experience in palliative care as physicians (RW, KG), three of the coders have qualitative expertise (RW, KG and SP), and one of the coders was being mentored in the qualitative methodology and was a medical student (CW).

This study was out of Health & Disability Ethics Committee scope of review. Internal ethics approval was granted by the CDHB Oncology Department Low Risk Ethics Board.

Results

Quantitative analysis

A total of 169 staff responded to the deaths of 51 patients. The majority of survey respondents were nurses (50%), followed by doctors (39%). A small percentage of staff identified themselves as allied health professionals or other (8% and 3%, respectively).

Overall, 71.3% of the staff surveyed agreed or strongly agreed that "end-of-life care was of a high standard". The highest scoring domain was for physical symptoms, with 68% of respondents agreeing or strongly agreeing that physical symptoms were well managed. The emotional and support/social needs domains recorded lower scores, at 65% and 50% respectively (see Table 1). Due to the changes made after the second pilot testing, the totals seen in Table 1 for each item is not constant, and represents the total number of answers for that item. The most disagreed with item was that the "patient's physical symptoms were well managed" at 13%, followed by the "patient's family/whānau emotional needs were met" at 12.2%.

There were three free-text questions that staff could choose to answer. The overall response rate of answering at least one free-text question was 86.4%. Of those that only answered one free-text question, 80% answered the "what aspects of the end-of-life care went well" text option. The response rate of each free-text box ranged from 69.2% ("what aspects of the end-of-life care could be improved") to 81.7% ("what aspects of the end-of-life care went well"). Of note, the free-text box "comments on family/whānau emotional/support/social needs met" was added after the second round of testing, and there was a 74.5% response rate for this option.

The last item on the survey allowed staff to request extra feedback in more detail if they required. Thirteen staff requested extra feedback. An email was sent to these staff offering additional support, but no staff took up the offer.

Qualitative analysis

Three themes were identified through the coding processes; coordinated care, culture of practice and complexity of care.

1. Coordinated care

Coordinated care encompasses levels of service delivery seen by staff as pivotal to support both the patient and their family/whānau through their last days of life. Coordinated care was inclusive of complex case needs, communication of a diagnosis of dying, supporting family/whānau at the point of death and the importance of teamwork.

Complex case needs describes the balance between staff providing support, whilst allowing the patient and family/whānau to feel empowered to make decisions and be involved in care. This was challenging when patients presented late

Table 1: Likert scale results of agree/strongly agree.

Item	Total	Agree/ strongly agree (n) %	Neutral (n) %	Disagree/ strongly disagree (n) %
Patient physical symptoms well managed	169	(115) 68.0	(32) 18.9	(22) 13.0
Patient emotional needs met	168	(110) 65.5	(45) 26.8	(13) 7.7
Patient support/social needs met	169	(85) 50.3	(66) 39.1	(18) 10.7
Patient family/whānau emotional/social/ support needs met*	51	(32) 62.7	(14) 27.5	(5) 9.8
Patient family/whānau emotional needs met*	115	(69) 60	(32) 27.8	(14) 12.2
Patient family/whānau support/social needs met*	113	(69) 61.1	(36) 31.9	(8) 7.1
End-of-life care is of a high standard	167	(119) 71.3	(32) 19.2	(16) 9.6

*Totals appear smaller because these items merged after second round of testing.

or in a rapidly changing situation. Participants acknowledged issues such as lack of cohesion amongst clinical teams, declining services such as specialist palliative care and reduced resources on weekends. For example:

> *"It was extremely difficult to communicate to the family, as many didn't speak English, and only one member had a fluency in English. It was very hard to explain why we were doing things, access to the patient was often limited by family (having to translate) which made it hard to assess the physical needs of the patient. Some of the nursing team were reluctant to observe the cultural and religious requirements the family had surrounding death." – P1*

Communication of the diagnosis of dying was viewed as being pivotal to supporting the patient and their family/whānau. This included ensuring the trajectory of anticipated death was clear and appropriate supports were put in place to allow the patient and their family/whānau to make decisions. Participants were concerned that when an uncertain or unstable trajectory was identified, this was more likely to impact on the experience of dying. Conversely, participants also identified the need to ensure that for patients with a longer dying trajectory, the dying process was still discussed.

> *"The death was quick and sudden even though expected, and although family were not emotionally prepared they had been supported and informed that he was dying." – P2*

Poor communication fostered mistrust, which included patients not understanding their prognosis and condition severity, an inability to navigate language barriers, cultural differences and inability to comprehend information. The opposite was also the case; good communication clearly helped family/whānau navigate the dying process more easily.

> *"A family meeting was held where the patient and their family were made well aware that the patient was in his final days. There was a clear plan of which family member to call if the patient deteriorated, which was done." – P3*

Participants regarded teamwork as an important aspect of a good death. As multiple staff were involved in the care of a patient, it was important that teams had good cohesion and were able to work together, even when language and communication issues were present.

2. Culture of practice

The culture of practice encapsulates the concepts of dignity, respect and trust in the relationship between staff and a patient, i.e., values that shape the culture of practice. This culture of practice was inclusive of: after death and bereavement care, spiritual and cultural needs/practices, engagement with the family/whānau and building a relationship with patients and family/whānau which enabled trust, dignity and respect.

Participants identified that when cultural needs were respected, it often required support and resource, and not all were easy to access due to resource constraints:

> *"It was unfortunate that there are limited community resources catering for Muslim needs as X's situation was very unique…" – P5*

Dignity and respect of the patient and their family/whānau was a critical element. Participants described various roles that demonstrated how they enacted dignity and respect. Examples included the opportunity to assist a patient to get married while in hospital, through to care of the patient's body after death.

> *"X's partner was able to sleep in an adjoining bed which we positioned so that they could cuddle together." – P6*

Participants discussed the challenges in identifying spiritual needs of a patient, and therefore the difficulty in knowing whether these needs were met. Participants shared spiritual wellbeing signposts, such as words like "blessed" within their clinical encounters or a feeling that the family were prepared for death. A frequent area of concern was lack of privacy for the patient and family/whānau. The lack of appropriate facilities within their work places meant family/whānau were left to grieve in hallways or public spaces.

> *"Facilities are inadequate to support families in the ward environment. It*

is always sad to see families needing to grieve in public places." – P8

Participants identified that family/whānau engagement and involvement was a marker of a positive culture of practice. Participants also noted a less ideal culture of practice when patients were unable to communicate, had no family/whānau to guide them, or took a long time to build trust with staff.

"Karakia, himene performed at the request of patient who was fully comprehensive & focussed… last wishes of X were the responsibility of the daughters with the manaaki, tautoko, awhi of all those present and administered by X's brother… X's immediate whānau were able to stay overnight with X if they wished…" – P10

3. Complexity of care

This theme consolidates content which describes both the complexity of physical symptoms and complexity of patient or family/whānau interpersonal dynamics. This theme specifically encapsulates the influence of distress and denial, family/whānau conflict and symptom control in end-of-life care.

"His partner appeared to have been displeased by previous care in the hospital prior to this admission which I felt made things difficult for them." – P11

Participants explained that distress and denial could occur in patients, family/whānau and staff. Participants described how dealing with their own distress could be improved if there was more clinical support provided in difficult situations, and if resource was allocated so that they could better care for family/whānau in a busy department.

Participants also described that their job became more complex when family/whānau dynamics resulted in conflict within the clinical space in the negotiation of end-of-life care. Participants noted that the situation could escalate if the conflict was not resolved around the time of death. Conflict events created environments which were difficult for participants to navigate without appropriate system support.

"[It's] challenging family to talk things through with as they struggled with her illness and deterioration." – P12

Participants saw symptom control as an important success factor for a patient's care.

"Symptom control was not dealt within a time that was appropriate to the symptoms that arose and that were reported to medical team." – P13

Discussion

This is the first study in New Zealand to explore staff views of what contributes to quality of dying. This study aimed to assess the feasibility and acceptability of such a survey, and to obtain their perspectives of what contributes to the quality of patient and family/whānau experiences of dying and death in an acute hospital setting. A mixed method approach has enabled a richness of detail to the dataset, allowing key themes to be identified.

Obtaining staff feedback via survey was feasible and effective. Not only were staff willing to give their feedback, but the information provided was considerable. The survey allowed staff from diverse areas of the hospital an anonymous outlet for expressing concerns, and an option to request more support.

Themes emerging from this study are similar to those previously described in the literature, suggesting the survey is drawing out appropriate information from staff. A 2014 literature review focusing on nurse's experiences of providing end-of-life care in acute hospital settings found 16 studies, using a mixture of quantitative and qualitative methods.[12] Themes resonated strongly through all studies, the issues around culture of the organisation being paramount, as well as difficulties around diagnosing dying, communication/collaboration and lack of time or privacy. A further study by Reid et al. of multidisciplinary focus groups found that these themes encompassed the wider healthcare team.[13]

Assessing quality of dying and death is not straightforward and there is no consensus in the literature as to the best tools to use. A strength of this study is that it was developed in New Zealand for a New Zealand demographic. Previous instruments have been developed in the United States of America, Canada, Taiwan and Japan.[6] Most quality of dying and death instruments measure symptom control on a Likert scale, with little qualitative data reported. One of the most robust and widely used, the Quality of Dying and Death (QODD) instrument, is based on quantitative data

only.[1,6] A detailed evaluation of the quality of a death can be made by auditing the clinical record in retrospect, looking for evidence that aspects of care known to enhance quality at the end-of-life were provided.[10] Opinions from family/whānau members and/or healthcare professionals (as the person's proxy) are also useful. Each of these has a role to play in providing a comprehensive view of the performance of the organisation as a whole.

As this survey was voluntary, uptake was not universal. The number of survey requests sent was not recorded, so a response rate cannot be provided. Changes were made to the survey after a second pilot, with addition of a comments section to address family/whānau concerns. This limited data available for qualitative analysis. In addition, this survey did not include surgical sub-specialities, aside from General Surgery, or the Intensive Care Unit (ICU). This survey did not include paediatric wards, but some paediatric patients were captured through ED. Whilst retrospective, since responses were collected within two working days of the death, memories were still fresh.

This study has two main implications. Firstly, it provides an insight to a New Zealand specific population. This will enable improved care of both patients and family/whānau during the dying process and after death. Secondly, this survey allowed staff an anonymous avenue for reflection, without repercussion. This is important as it allows staff to process and reflect on their own and their teams practice around dying and death.

Conclusion

A survey of staff could be used routinely in acute hospitals as part of a suite of tools to provide a holistic view of dying and death across the organisation. It would complement other methods such as retrospective audits and family/whānau interviews/questionnaires. Outcomes may inform policy development and quality initiatives, informing service leads about areas requiring attention and highlighting individual staff members who might benefit from additional training and support.

COMPETING INTERESTS
Nil.

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Appendix 1

Item descriptor	Response
Patient name (last, first)*	Free text
Patient NHI	Free text
Your role in the health care team*	Doctor, nurse, allied health, other
The patient's physical symptoms were well managed*	Strongly disagree, disagree, neither agree nor disagree, agree, strongly agree
The patient's emotional needs were met (i.e., aspects surrounding holistic care that honours dignity, spiritual, cultural needs).*	Strongly disagree, disagree, neither agree nor disagree, agree, strongly agree
The patient's support/social needs were met (e.g., fulfilling last wishes, financial support, creating an appropriate environment for care, preferred place of death).*	Strongly disagree, disagree, neither agree nor disagree, agree, strongly agree
The patient's family/whānau/significant others emotional needs were met^	Strongly disagree, disagree, neither agree nor disagree, agree, strongly agree
The patient's family/whānau/significant others support/social needs were met^	Strongly disagree, disagree, neither agree nor disagree, agree, strongly agree
Comments on family/whānau emotional/support/social needs met^	Free text
Overall, the end-of-life care was of a high standard.*	Strongly disagree, disagree, neither agree nor disagree, agree, strongly agree
What aspects of the end-of-life care went well?	Free text
What aspects of the end-of-life care could be improved?	Free text
Would you like to be contacted by one of the audit team to discuss your feedback in more detail?^	Yes, no

* Indicates mandatory field
^ Indicates items added or changed after review.

Endoscopic retrograde cholangiopancreatography in the comorbid elderly: a retrospective comparative study in New Zealand

Kirsty Macfarlane, Reuben Wilson, Nicholas J Fischer, Henry Wei

ABSTRACT

AIM: To ascertain if endoscopic retrograde cholangiopancreatography (ERCP) in the elderly is associated with an increased risk of complications.

METHODS: Retrospective study of 509 consecutive ERCPs on 338 patients in one year (2019–2020). Patients were categorised as >75 years old (elderly test group) or ≤75 (controls). The primary outcome was ERCP complications. Secondary outcomes were the length of hospital stay after complications, intensive care admissions, and all-cause mortality at 30 and 90 days.

RESULTS: Forty-four complications occurred in a group of 42 (8%) patients; 11 (2%) were severe, including four deaths. The most common complication was pancreatitis n=33 (6%). There was no difference in complication rates between the elderly and younger controls. Length of stay after complications was similar (median five versus four days; p=0.354). All-cause mortality was higher in the elderly at 30-days (8.5% versus 2%; p=0.002) and 90-days (19.7% versus 6.9%; p=0.001), mostly attributed to malignancy. Logistic analysis showed that neither age >75 years nor Charlson Comorbidity Index (CCI) ≥5 was associated with post-ERCP pancreatitis, but a CCI≥5 strongly increased the odds of death at 90-days (AOR=74.44; 95% confidence interval (CI): 9.78- 566.38, p<0.001).

CONCLUSION: ERCP is relatively safe in elderly patients, but comorbidities should be considered to avoid subjecting vulnerable individuals with a short life expectancy to unnecessary procedures.

Endoscopic retrograde cholangiopancreatography (ERCP) is the first-line therapy for choledocholithiasis and associated pancreaticobiliary pathology, with over half a million procedures annually in the United States alone.[1] Pancreaticobiliary pathology is more common in the elderly, with an estimated 10–20% of elderly patients presenting with cholecystitis having coexisting choledocholithiasis, compared to 5% in the general population. In addition, the incidence of ductal stones in elderly patients undergoing emergency cholecystectomy approaches 50%.[2,3] Due to an ageing worldwide population, ERCP is increasingly utilised in the elderly as a therapeutic tool.[3–6] Despite this, ERCP in the elderly is purported to be associated with increased procedural difficulty and periprocedural adverse events. The increased risk might be expected due to a higher prevalence of frailty and comorbidities in this population, and the safety of ERCP in the elderly is often debated, with age frequently cited as a contraindication.[7]

Despite the perceived risks of ERCP in the elderly, there are limited comparative studies on outcomes. Literature on the safety and complication rates of older patients undergoing ERCP is conflicting. A systematic review of 18 studies by Harmeet et al. in 2017 concluded that ERCP was safe in the elderly, with higher rates of post-ERCP pancreatitis reported in younger patients.[2,7] Another study of octogenarians undergoing ERCP with a mean age of 84 years cited a mortality rate of 3.1%, which is considerably lower than the standard quoted mortality rate of 5%.[3,8] In contrast, high rates of post-ERCP mortality were reported by Kalaitzakis et al. in the very elderly >80 years, mainly due to underlying diseases, notably cancer.[8] Sedation for ERCP is also associated with increased risks in the elderly and may contribute to periprocedural morbidity and mortality.[9]

To our knowledge, there is no comparative outcomes data for elderly patients undergoing ERCP in New Zealand. Increased understanding of the complication and outcomes of ERCP could help clinicians select the best therapeutic approach and potentially avoid exposing vulnerable patients to risky interventions.

Methods

The records of 509 consecutive ERCP procedures performed between September 2019 and September 2020 were retrospectively reviewed. Procedural details were captured from endoscopy reports via ProVation® MD software. In addition, a comprehensive assessment of clinical records, including radiology and laboratory data, was completed. Specific data collected included demographics, procedural indication, American Society for Gastrointestinal Endoscopy (ASGE) complexity score,[10,11] procedural techniques and whether non-steroidal anti-inflammatories (NSAIDs) were prescribed. Comorbidities were classified using the Charlson Comorbidity Index (CCI) below. The cause of death was obtained via clinical records. Patients were divided into elderly >75 years and controls ≤75 years old for analysis and comparison. Ethics approval was obtained from the Health and Disability Ethics Committee.

All procedures were completed by four consultant gastroenterologists and a senior endoscopy fellow at Middlemore Hospital. Middlemore Hospital is a tertiary referral centre for Counties Manukau District Health Board in Auckland, New Zealand and services approximately 567,000 people, representing 11% of the total New Zealand population in 2018.[12] Both emergency and elective ERCP were included. The majority utilised conscious sedation with a combination of midazolam and fentanyl as directed by the endoscopist. A small number of procedures were conducted with propofol or general anaesthesia. Rectal NSAIDs with diclofenac were prescribed at the start of each procedure for post-ERCP pancreatitis (PEP) prophylaxis at the endoscopist's discretion. Cannulation was typically carried out using a guidewire-assisted technique with a sphincterotome; other interventions such as needle-knife or trans-pancreatic precut sphincterotomy, dilatation and stenting were carried out depending on the requirements of each procedure and the endoscopist expertise.

Charlson Comorbidity Index

CCI is an internationally recognised method of categorising comorbidities based on the International Classification of Diseases. Comorbidities are weighted based on the adjusted risk of mortality or resource use at one year, and the sum of all the weights results in a single comorbidity score for each patient.[13,14] Nineteen conditions are used in the scoring system and given a weight based on the estimated mortality hazard ratio from a Cox proportional hazards model. A score of zero indicates no comorbidities, whilst a score of five or more may be considered highly comorbid. Studies have shown that using the CCI as a composite score is a better predictor of prognosis than individual comorbidities alone.[14]

Primary and secondary outcomes

The primary outcome was complications from ERCP based on clinical records, laboratory findings and radiology results. Pancreatitis was classified using the revised Atlanta classification for pancreatitis, which requires that two or more of the following criteria be met for the diagnosis of acute pancreatitis: (a) abdominal pain suggestive of pancreatitis; (b) serum amylase or lipase level greater than three times the upper normal value; or (c) characteristic imaging findings.[15] In addition, the Cotton criteria were used for grading bleeding, perforation and sepsis.[10,16] We also examined variables that predicted PEP in a multivariate analysis model.

Secondary outcomes were the length of hospital stay after complications from ERCP, intensive care admissions, and all-cause mortality at 30 and 90 days following ERCP.

Statistics

Our sample size was greater than the estimated sample size of 400 procedures required to detect a 10% difference between study groups for combined adverse events from ERCP (α 0.05; statistical power 80%; 1:3 enrolment ratio).

Fisher's exact test was used for categorical variables, and the Mann–Whitney U test for non-categorical variables was performed to compare study groups. Statistical significance was set at the p value <0.05 (two-sided) level. Potential variables for ERCP complications were entered into a multivariate logistic regression analysis with odds ratio adjusted for potential confounders. Mortality results were presented using Kaplan–Meier survival curves. Data analysis was completed using IBM Statistics 26 program (SPSS Inc, Chicago, III).

Results

A total of 509 ERCP procedures performed on 338 patients were included. The mean age was 61 years (range 16–93). The elderly >75 years cohort comprised 23% (117/509) of the total procedures. Compared to the younger cohort, the elderly were more likely to be male (53.8% vs 39.8%; p=0.007)

and of NZ European ethnicity (51.3% vs 35.2%; *p*=0.002). The elderly cohort also had a significantly higher CCI (mean 6.25 vs 3.17). There was no significant difference in ERCP indications and ASGE complexity score between the elderly and young groups. The most common indication for ERCP was choledocholithiasis (43%), ascending cholangitis (16.3%), pancreatic cancer (9.4%), gallstone pancreatitis (6.9%), biliary cancer (5.5%) and jaundice—cause not specified (5.5%). The elderly group were less likely to receive NSAIDs at ERCP (9.4% vs 19.3%; *p*=0.002). Other procedural characteristics were similar between groups (Table 1).

Complications from ERCP

Forty-four complications occurred in 42 (8%) patients, 11 (2%) of which were severe, including four deaths. Pancreatitis occurred in six (5.1%) patients of the elderly versus 27 (6.9%) patients in the younger cohort, with no statistical difference (*p*=0.669). In addition, there was no difference in the total number of complications, intensive care admissions and length of hospital stay after complications (median four days in elderly vs five days in the younger cohort; *p*=0.354) (see Table 2). Complication rates did not differ between senior endoscopists or in cases involving a fellow.

Predictors of PEP

Multivariate logistical regression showed that neither age >75 years nor CCI ≥5 was associated with an increased risk of PEP. After adjusting for other variables, an increased risk of PEP was observed in females (adjusted odds ratio AOR=2.93; 95% confidence interval (CI):1.12–7.67; *p*=0.028), pre-cut sphincterotomy (AOR=6.82; 95% CI:1.82–25.6; *p*=0.004) and pancreatic sphincterotomy (AOR=23.65; 95% CI: 3.74–149.7; *p*=0.001). Interestingly, ASGE complexity score was not associated with an increased risk of PEP; however, NSAID use was associated with PEP (AOR=4.28; 95% CI: 1.89–9.7; *p*=0.001) (see Table 3).

30- and 90-day all-cause mortality

At 30-days post ERCP, there were 22 deaths from any cause, representing 4.32% of the study population. Fifty deaths (9.8%) were observed at 90 days post ERCP. All-cause mortality was significantly higher in the elderly group at 30 days (8.5% vs 2%; *p*=0.002) and 90-days (19.7% vs 6.9%; *p*<0.001). Causes of death at 90 days are summarised in Figure 1.

After adjusting for sex and ethnicity, multivariate logistical regression showed that a CCI ≥5 significantly increased all-cause mortality at 90 days (AOR=74.44; 95% CI: 9.78–566.38; *p*<0.001), followed by malignant biliary disease (AOR=2.89; 95% CI: 1.44–5.79; *p*=0.003) and age >75 years (AOR=2.28; 95% CI: 1.14–4.55; *p*=0.02). Complications from ERCP did not significantly increase the odds of death at 90-days in our model (see Table 4). Considering patients in both groups, CCI ≥5 increased the odds of death at 90 days (see Figure 2).

Discussion

Our finding is the first of its kind in New Zealand and adds to the growing literature on ERCP in the elderly. There was no difference in ERCP complication rates between the elderly and younger patients. This is consistent with an earlier systematic review that demonstrated incidence rates of ERCP complications for elderly patients similar to most reported literature.[17] While several studies have established the safety and efficacy of ERCP in the elderly, they had limitations with lacking a comparator group or including outcomes for elderly patients as young as 60 years old.[18–20]

Counter-intuitively, NSAID use was associated with increased pancreatitis in multivariate analysis. At the time of this study, NSAID use was at the endoscopist's discretion and typically only for patients considered at high risk of PEP. The overall NSAID use was significantly lower in the elderly cohort, and this should increase the risk of pancreatitis in the elderly, but our findings show similar rates of pancreatitis versus the younger cohort. Advancing age has been shown to have a protective role against the development of PEP in prior studies and, perhaps, offset the effects of lower NSAID use amongst the elderly.[2]

While ERCP-related mortality is well-described, there is a lack of comparative data on all-cause mortality in the elderly and few studies that incorporate a standardised comorbidity index. We reported a 9.8% all-cause mortality at 90 days post ERCP, comparable to published data from international registries.[8,21,22] Mortality post ERCP was driven primarily by comorbidities, and a CCI of ≥5 was the strongest predictor of death. 90-day mortality was rarely observed in those with a CCI of <5, irrespective of age. We did not find an increased risk of ERCP complications in those with CCI ≥5, in contrast to a study of 614 patients where a CCI of ≥2 was recorded in 21% of the cohort and was associated with increased odds of adverse events from ERCP.[23] This is despite our cohort being more comorbid, with a CCI ≥5 recorded in

Table 1: Patient and procedural characteristics.

Characteristic	Overall (n=509) % (total)	≤75 years (n=392) % (n)	>75 years (n=117) % (n)	*p* value
Age (median, years)	64	58	81	
Male	43% (219)	39.8% (156)	53.8% (63)	0.007[b]
Female	57% (290)	60.2% (236)	46.2% (54)	0.007[b]
Ethnicity				
NZ European	39% (198)	35.2% (138)	51.3% (60)	0.002[b]
Pacific	24% (122)	25.5% (100)	18.8% (22)	0.141[b]
Māori	11% (56)	12.5% (49)	6% (7)	0.063[b]
Asian	9% (47)	9.7% (38)	7.7% (9)	0.589[b]
Indian	8% (39)	7.9% (31)	6.8% (8)	0.844[b]
Othersa	9% (47)	9.2% (36)	9.4% (11)	1[b]
Charlson Comorbidity Index				
CCI ≤4	63.3% (332)	71.4% (280)	35.9% (42)	<0.001b
CCI ≥5	36.6% (187)	28.5% (112)	64% (75)	<0.001b
Indication for ERCP				
Choledocholithiasis	43% (219)	43.5% (170)	42.2% (49)	0.832[b]
Ascending cholangitis	16.3% (83)	15.6% (61)	19% (22)	0.396[b]
Pancreatic cancer	9.4% (48)	9% (35)	11.2% (13)	0.474[b]
Gallstone pancreatitis	6.9% (35)	6.9% (27)	6.9% (8)	1[b]
Biliary cancer	5.5% (28)	5.1% (20)	6.9% (8)	0.490[b]
Jaundice—cause not specified	5.5% (28)	5.6% (22)	5.2% (6)	1[b]
Chronic pancreatitis	2.9% (15)	3.6% (14)	0.9% (1)	0.210[b]
Benign biliary strictures	2.8% (14)	2.8% (11)	2.6% (3)	1[b]
Bile leak	2.4% (12)	2.8% (11)	0.9% (1)	0.312[b]
Ampullary cancer	2.4% (12)	2.3% (9)	2.6% (3)	1[b]
Other indications	1% (5)	0.5% (2)	0	1[b]
Sphincter of Oddi dysfunction	0.8% (4)	0.5% (2)	1.7 % (2)	0.228[b]

Table 1 (continued): Patient and procedural characteristics.

Characteristic	Overall (n=509) % (total)	≤75 years (n=392) % (n)	>75 years (n=117) % (n)	*p* value
Pancreatic duct assessment	0.8% (4)	1% (4)	0	0.578[b]
ASGE complexity scale				
1	15.5% (79)	15.8% (62)	14.5% (17)	0.884[b]
2	54.2% (276)	55.1% (216)	51.3% (60)	0.526[b]
3	29% (148)	27.8% (109)	33.3% (39)	0.249[b]
4	1.2% (6)	1.3% (5)	0.9% (1)	1[b]
Procedural characteristics				
Fellow involvement	36.1% (184)	36.7% (144)	34.2% (40)	0.662[b]
NSAIDs for PEP prophylaxis	19.3% (98)	22.2% (87)	9.4% (11)	0.002[b]
Previous pancreatitis	19% (97)	19.9% (78)	16.2% (19)	0.377[b]
Biliary sphincterotomy	47.9% (244)	48.5% (190)	46.2% (54)	0.660[b]
Precut sphincterotomy	3.3% (17)	3.6% (14)	2.6% (3)	0.595[b]
Endoscopic balloon sphincteroplasty	12.2% (62)	11.5% (45)	14.5% (17)	0.376[b]

[a]Latin American, Middle Eastern, African and other ethnicities not specified.
[b]Fisher's exact test, two-sided.

Table 2: Comparison of ERCP complications between younger and older groups.

Complication	≤75 years n (% total)	>75 years n (% total)	*p* value
Pancreatitis	27 (6.9%)	6 (5.1%)	0.669[b]
Bleeding	5 (1.4%)	0	0.594[b]
Perforation	2 (0.5%)	1 (0.9%)	0.544[b]
Sepsis	1 (0.3%)	2 (1.7%)	0.134[b]
Death	2 (0.5%)	2 (1.7%)	0.228[b]
Total complications	35 (8.9%)	9 (7.7%)	0.851[b]
Severe complications	8 (0.02%)	3 (0.01%)	0.721[b]
Length of hospital stay after complication (median, days)	5	4	0.354[c]
Intensive care admission	0.5% (2)	0	1[b]

[a]Grading of complications defined by the revised Atlanta classification for pancreatitis and Cotton criteria for bleeding, perforation and sepsis.
[b]Fisher's exact test, two-sided.
[c]Mann–Whitney U test.

Table 3: Multivariate regression analysis of risk factors for post-ERCP pancreatitis.

Risk factors	Adjusted odds ratio	95% Confidence interval	*p* value
Age >75 years	1.498	0.522–0.4299	0.453
Female	2.930	1.120–7.667	0.028
CCI ≥5	0.581	0.216–1.567	0.283
NSAIDs for PEP prophylaxis	4.277	1.886–9.702	0.001
Endoscopic balloon sphincteroplasty	0.952	0.248–3.658	0.943
Precut sphincterotomy	6.822	1.818–25.601	0.004
Pancreatic sphincterotomy	23.649	3.737–149.748	0.001
Biliary sphincterotomy	1.139	0.499–2.599	0.757
Previous pancreatitis	1.300	0.445–3.797	0.632
ASGE complexity score 4	0.536	0.140–2.048	0.362

Figure 1: All causes of death within 90 days post-ERCP.

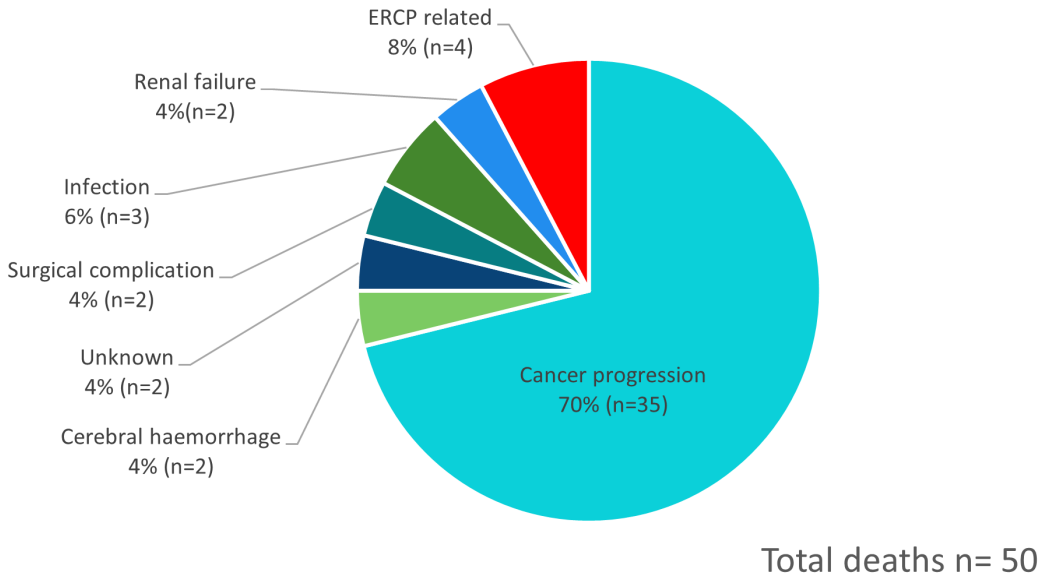
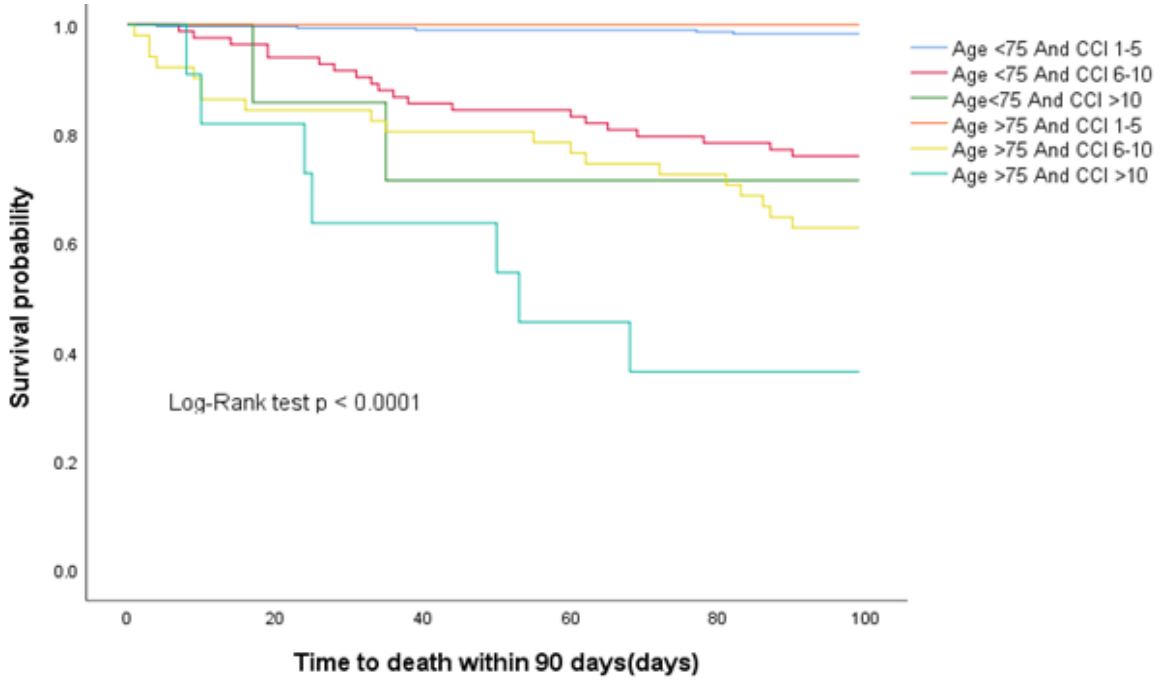


Table 4: Multivariate regression analysis of risk factors for 90-day all-cause mortality.

Risk factors	Adjusted odds ratio	95% Confidence interval	*p* value
Any complications from ERCP[a]	2.201[b]	0.695–6.978	0.180
Age >75 years	2.278[b]	1.142–4.551	0.020
Hepatic or pancreaticobiliary malignancy	2.890[b]	1.442–5.792	0.003
CCI ≥5	74.441[b]	9.784–566.381	<0.001

[a] Complications of ERCP, including PEP, bleeding, perforation, and cholangitis.
[b] Adjusted for sex and ethnicity.

Figure 2: Kaplan–Meier curve of all-cause mortality 90 days post-ERCP according to age and CCI category.



36.6%. Although CCI may have a role in risk stratification, we agree with Galeazzi et al. that a multidomain, comprehensive geriatric assessment should be considered to understand the patient beyond medical history or fitness level.[24]

A prior systematic review has demonstrated cardiopulmonary complications as the most common ERCP complication among nonagenarians.[25] Our study did not attribute deaths to cardiopulmonary complications; however, we collected data from hospital records and death certificates which may overlook short-term cardiopulmonary events. We should note that most procedures in our study were completed with conscious sedation utilising a combination of midazolam and fentanyl, which may differ from international literature where propofol or general anaesthesia may be employed more frequently. In one randomised study, propofol sedation was associated with a shorter recovery time and less hypoxic events among high-risk octogenarians undergoing ERCP.[26]

The main limitation of our study was the single-centre retrospective design. Future prospective studies would help explore the full spectrum of risks in the elderly, including cardiopulmonary adverse events. Due to a lack of a non-interventional control group, the clinical course of elderly patients not undergoing ERCP was not explored. While this may lead to selection bias, a randomised study of this nature is unlikely to happen as it would not be ethical to withhold critical ERCP routinely.

In conclusion, ERCP is relatively safe in elderly patients and should not be withheld based on age alone. The higher prevalence of comorbidities in the elderly should be considered to avoid subjecting vulnerable individuals with a short life expectancy to procedures where potential benefits outweigh the risk of harm.

COMPETING INTERESTS
Nil

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Outcomes in mild hyperphenylalaninemia: a comparison with PKU and healthy controls across cognition, behaviour, and quality of life

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ABSTRACT

AIMS: Considering the cognitive, behavioural and quality of life (QoL) consequences of high phenylalanine levels in early treated phenylketonuria (PKU), this study examined whether monitoring and active management of individuals with the mild form of the condition hyperphenylalaninemia (HPA) would be advisable.

METHOD: Six individuals (aged 6 to 15) with untreated HPA were compared with six age and gender matches with PKU, and six healthy controls on the Wechsler Intelligence Scale for Children, 5th edition; Wechsler Individual Achievement Test, 2nd edition; Trail-Making test; Contingency Naming Test; and Oral Fluency test. Self- and parent-report rating scales administered included the Conners Comprehensive Behavior Rating Scales; Behavior Rating Inventory of Executive Function, 2nd edition; the Pediatric Quality of Life Inventory, and the Phenylketonuria Quality of Life (PKU group only) questionnaires.

RESULTS: Early treated PKU participants demonstrated normal intelligence, pointing to the efficacy of dietary management. Quality of life and behavioural difficulties were observed including more severe externalising problems. HPA participants showed normal ability, including executive ability. Power was limited by the small sample.

CONCLUSION: This was the first study of the New Zealand population with HPA. While there was insufficient evidence to warrant treatment, there was also insufficient evidence to safely exclude the presence of cognitive impairment.

Hyperphenylalaninemia (HPA) and phenylketonuria (PKU) are variants of the same genetic condition whereby activity of liver enzyme, phenylalanine hydroxylase (PAH), is reduced.[1] This results in an inability to metabolise the amino acid phenylalanine (Phe).[1] Phe accumulates in the blood and tissues and becomes neurotoxic. In PKU, PAH activity is severely reduced or absent, whereas in HPA some activity is retained.[1–2]

When untreated, PKU results in intellectual disability, microcephaly, epilepsy, and psychiatric and behaviour problems.[1–2] Since the introduction of newborn screening and subsequent early treatment, this severe phenotype is rarely seen. However, even with treatment individuals with PKU have IQ scores 5–15 points lower than their unaffected siblings and parents (though within normal range) and show executive deficits, behavioural and psychological problems, and poorer quality of life (QoL).[3–6]

In contrast, individuals with HPA are thought to have normal neuropsychological function and typically receive no active follow-up.[7–9] However, early research supporting this approach focused on global intelligence, rather than the more subtle executive deficits seen in early treated PKU.[3] In a 2005 study comparing 35 children with HPA, 37 children with PKU, and 29 healthy controls, HPA participants produced significantly worse executive scores than controls.[10] Thus, further studies are still needed to safely exclude cognitive impairment and especially executive impairment in HPA, and to establish what Phe levels require dietary treatment. Considering cognition, executive function, and QoL, this study sought to add to the evidence by comparing children with HPA to age and gender matches with PKU and healthy controls.

Method

Participants

Three groups of children aged 6 to 16 years were recruited from across New Zealand—a group with HPA, a group with PKU, and a group of healthy controls. Groups were matched for age (+/-2 years), gender and ethnicity. In New Zealand, since the 1990s, infants with day two Phe levels

exceeding 400umol/L at confirmatory testing (Guthrie card blood spot) have been classified as having PKU. Treatment consists of strict dietary protein restriction, amino acid supplementation, and regular monitoring of blood-Phe levels. Individuals with levels between 150 and 400umol/L at confirmatory testing are classified as having HPA and receive no long-term follow-up. Females are advised of the need for possible dietary treatment during pregnancy, where levels over 300umol/L can cause foetal damage. Of all cases of HPA identified between 2007 and 2014 in New Zealand, 19.2% were classified HPA (1:87,726 births), and 80.8% PKU (1:20,887 births).

Group demographic and metabolic characteristics appear in Table 1. All participants were aged between 6 and 15 and all were female. While we sought to recruit males and females aged 6 to 16 there were no males with HPA and no 16-year-olds eligible to complete the study. Within each group, five participants self-identified as New Zealand European/Pākehā, and one as Māori. Average blood Phe at birth was 281.50umol/L (SD=83.38umol/L) for the six HPA participants, and 1,248.00umol/L (SD=541.83umol/L) for the six PKU participants. As expected, levels were significantly higher amongst PKU participants. No significant demographic differences were found between groups.

Measures

Initial and historical blood Phe levels for HPA and PKU participants were obtained from clinical records. Phe was also measured on the day of testing. All participants completed a test battery assessing intelligence, achievement, executive functioning, and processing speed. Pen and paper questionnaires were completed by participants and a parent/guardian to produce a behavioural profile. Health-related QoL life was also examined, with PKU participants additionally completing a questionnaire about PKU-related QoL.

Cognitive tests

Wechsler Intelligence Scale for Children, 5th Edition, Australia and New Zealand (WISC-V)

The WISC-V assesses cognitive ability in children aged 6 to 16.11 Internal consistency for the subtests ranges from 0.74 to 0.95, and for the Primary Indices, from 0.86 to 0.96. Subtest scale scores and index scores, including the Full Scale Intelligence Quotient (FSIQ), were used in the analysis.

Wechsler Individual Achievement Test, 2nd Edition, Australian Abbreviated (WIAT-II-A)[12]

An abbreviated version of the WIAT-II ability measure, the WIAT-II-A Australian contains three subtests: Word Reading, Numerical Operations, and Spelling. It has demonstrated adequate reliability and validity. Subtest standard scores and the overall composite score were used in the analysis.

Trail Making Test for children (TMT)

The TMT is a simple, two-part "connect-the-dots" task.[13] Part A provides an indication of basic speed of processing, while Part B places demand on sequencing and ability to shift set. The children's version published by Reitan shows good ecological validity and adequate reliability.[13] Z-scores were calculated using age-related normative data for analysis.[14–15]

Oral fluency

The oral fluency test is a short test of verbal, executive and speed of functioning. Both semantic (animals) and phonemic (FAS) fluency are assessed.[16] Test-retest correlations are high (>0.70) for both tasks, with the phonemic fluency task additionally showing good internal consistency.[17] Z-scores were calculated using age-related normative data for analysis.[14,17–18]

Contingency Naming Test (CNT)

The CNT measures processing speed, attention shift, and response inhibition in children.[19] Participants complete two baseline tasks, then two switching tasks evaluating rapid memory retrieval, inhibition and set shifting. The CNT is sensitive to brain maturation and damage, including that resulting from PKU. Z-scores for time taken and efficiency were included in the analysis.

Questionnaires

Conners Comprehensive Behavior Rating Scales (CBRS)

These assess a range of behavioural, emotional, social, and academic issues in school aged youth.[20] The CBRS demonstrates good test-retest reliability, internal consistency and discriminative validity between different diagnoses.

Behavior Rating Inventory of Executive Function, 2nd Edition (BRIEF2)

These scales assess executive behaviours in children and adolescents.[21] The scales have shown good test-retest reliability.

Pediatric Quality of Life (PedsQL) Inventory

This measures functioning and health-related QoL in healthy children and those with health conditions across four domains: physical, emotional, social, and school.[22] The PedsQL has demonstrated fair reliability and construct validity.

Phenylketonuria-Quality of Life (PKU-QoL) Questionnaires

These assess the physical, emotional, and social impacts of PKU and its treatment on individuals with PKU and their families. Validity and reliability estimates are fair to good.[23]

Procedure

This study was approved by the Health and Disability Ethics Committee (HDEC; 17/CEN/272). Ten individuals with HPA were identified and contacted by the National Metabolic Service. One male was excluded due to severe autism, two had moved overseas, and another could not be contacted. The remaining six agreed to participate and were sent consent forms and questionnaires to complete prior to testing. Once an individual with HPA had completed the study, an age and gender matched individual with PKU was identified. For PKU participants, the questionnaire pack additionally included the PKU-QoL forms. Where possible, healthy controls were nominated by the family of participants, or otherwise recruited via word of mouth. Assessment of controls was the same, with the exception of not requiring a Phe level.

Testing took place at the hospital, university, outpatient metabolic clinic, or the child's home or school. All tests were completed in a single session using a standardised procedure. Participants completed the WISC-V, followed by the executive tasks (oral fluency, TMT, CNT), and then the WIAT-II-A. Breaks were offered as and when required. The total duration of testing was between 85 and 160 minutes. Participants received a $40 supermarket voucher as a thank you for their participation. Parent report data were missing for two participants (one control, one PKU) whose parents did not respond to a third contact attempt.

All tests were scored in accordance with standardised procedures. Anonymised data was exported into SPSS 26.0 for analysis. Independent sample t-tests and Chi-squared tests were used to determine group differences in terms of demographic factors. Multivariate analyses of variance (MANOVA) were conducted to identify differences between the groups across outcome measures. Pearson's bivariate correlations were generated to examine the relationship between performance and Phe levels for the HPA and PKU groups only.

Table 1: Demographic and Phe characteristics of the groups.

Characteristics		HPA (n=6)	PKU (n=6)	Control (n=6)	Significance of difference
Age (years)	Mean (SD)	10.80 (4.31)	10.53 (3.82)	10.96 (3.99)	F(2,15)=0.017; p=0.983
	Range	6.05–15.87	6.46–15.38	6.05–15.24	
Gender n (%)	Female	6 (100.00)	6 (100.00)	6 (100.00)	χ^2=0.000; p=1.000
	Male	0 (0.00)	0 (0.00)	0 (0.00)	
Ethnicity (%)	NZ European	5 (83.33)	5 (83.33)	5 (83.33)	χ^2=0.000; p=1.000
	Māori	1 (16.67)	1 (16.67)	1 (16.67)	
Current Phe	Mean (SD)	167.50 (69.91)	450.00 (136.75)	NA	F(1,10)=20.30; p=0.001
	Range	65–265	306–621	NA	
Phe at birth	Mean (SD)	281.50 (83.38)	1,248.00 (541.83)	NA	F(1,10)=18.64; p=0.002
	Range	153–382	670–2,000	NA	

Note: Phe levels measured in umol/L.
Abbreviations: HPA = mild non-PKU hyperphenylalaninemia, PKU = phenylketonuria, SD = standard deviation, Phe= phenylalanine, NA= Not assessed.

Results

Cognitive functioning

Table 2 presents group performance on the WISC-V and WIAT-II-A. Mean scores for all three groups fell within the average range. Two MANOVAs were conducted with group (HPA, PKU, control) as the grouping variable and performance across subtests and indices of the WISC-V; and then the WIAT-II-A, as dependent variables. No significant effect of group on WISC-V [F(30,4)=2.495; p=0.327] or WIAT-II-A [F(8,24)=0.606; p=0.764] performance was identified.

Group mean Z-scores across executive measures generally fell between -1 and 1 (oral fluency being the exception; see Table 3). All three groups performed similarly on the CNT simple naming (Trial 1), however, as demand on shifting capacity increased (Trials 3–4), more frequent errors and slower speed of processing were observed in HPA and PKU groups relative to controls. The HPA group produced time and efficiency Z-scores more than 1 SD below the normative mean for Trial 4 and for the total score (Trials 1–4). A MANOVA with time and efficiency scores as dependent variables did not identify any significant effect of group. Similarly, for the TMT, HPA and PKU participants were slower to complete both Parts A and B than were controls. Group mean scores for phonemic fluency (FAS) were generally equivalent (+/-2 responses). On semantic fluency, however, the HPA group produced fewer correct responses than the control and PKU groups. A second MANOVA with scores on the phonemic and semantic fluency tasks, and Parts A and B of the TMT as dependent variables similarly did not identify any significant effect of group.

Behavioural functioning

Conners CBRS

Group mean scores on the CBRS are presented in Table 4. Rates of behavioural and emotional difficulties were highest amongst the PKU group, with mean scores falling in the clinically elevated range on four of the parent-reported scales. The control group produced the lowest scores (with the exception of the Math scale), with the HPA group scoring somewhere in the middle. Given the large number of scores produced by the CBRS, content and symptom scales for the self- and parent-report forms were analysed using separate MANOVA. These analyses did not identify any significant effect of group.

A frequency table was produced to examine the presence of clinically significant scores within the sample (see Table 5). Parents of PKU children endorsed "Very Elevated" difficulties on 15.6% of the total subscale scores, exceeding that expected based on normative data (i.e., expected in 2% of normal population). The frequency of "Very Elevated" scores amongst HPA participants was much lower, but still double that expected (4.2%), while control participants rates (2.5%) were proportionate to the normal population.

BRIEF-2

Table 6 shows mean group scale and index scores for the BRIEF-2. Reports of executive difficulties were most frequent amongst PKU participants and least frequent amongst controls. A MANOVA with index scores as dependent variables revealed a significant effect of group [F(2,12)=32.24; p=0.030], with self-reported Shift and Emotion Regulation Index scores contributing significantly to the difference. Post hoc analyses indicate that PKU participants scored significantly higher than controls (p=0.044) on the shift scale; and significantly higher than both control (p=0.003) and HPA groups (p=0.008) on the Emotion Regulation Index.

Quality of life

PedsQL

Table 7 presents group mean QoL scores on self- and parent-reported PedsQL. The PKU group generally reported the lowest quality of life, with differences in school functioning being most pronounced. A MANOVA examining the impact of group on the PedsQL fell short of statistical significance [F(16,10)=1.97; p=0.140].

PKU-QoL

PKU participants self-reported severe tiredness, stomach aches, concentration difficulties and moodiness, as well as moderately severe headaches, slowed thinking and irritability. Parents endorsed similar difficulties, but less severely than participants themselves. Participants and parents emphasised the emotional burden of treatment. Participants reported strongly disliking the taste of supplements, low food enjoyment, extreme food temptation, difficulties with adherence, and guilt for non-adherence. Parents endorsed being greatly impacted by their child's anxiety related to blood testing and elevated Phe levels, difficulties ensuring adherence, and guilt related to non-adherence. Adherence to supplements was identified as more challenging than adherence to the low protein diet.

Table 2: Performance on WISC-V and WIAT-II-A for each group.

Measure	Index	HPA (n=6)	PKU (n=6)	Control (n=6)
WISC-V	Verbal Comprehension Index	98.00 (14.48)	102.00 (10.28)	105.17 (9.11)
	Visual Spatial Index	103.83 (15.64)	98.83 (14.78)	100.17 (15.01)
	Fluid Reasoning Index	104.67 (14.98)	106.83 (19.76)	101.50 (8.64)
	Working Memory Index	94.00 (11.54)	103.67 (14.76)	105.50 (14.87)
	Processing Speed Index	102.83 (17.28)	104.00 (14.13)	98.33 (9.29)
	Full Scale Intelligence Quotient	100.83 (15.11)	104.33 (14.33)	103.00 (8.25)
WIAT-II-A	Word Reading	97.50 (17.39)	100.33 (12.93)	95.67 (14.12)
	Numerical Operations	95.67 (11.24)	87.67 (21.53)	89.50 (17.93)
	Spelling	101.67 (20.78)	97.83 (12.45)	95.33 (10.82)
	Composite Score	98.33 (19.37)	94.83 (15.25)	91.67 (12.99)

Notes: Mean (SD). No group differences reached significance (p>0.05).
Abbreviations: HPA = mild non-PKU hyperphenylalaninemia; PKU = phenylketonuria;
WISC-V = Wechsler Intelligence Scale for Children, 5th edition;
WIAT-II-A = Wechsler Individual Achievement Test, 2nd Edition,
Australian Abbreviated; IQ = intelligence quotient.

Table 3: Mean performance on the executive measures (as Z-scores) for each group.

Measure		HPA (n=6)	PKU (n=6)	Control (n=6)
Contingency Naming Test	Trial 1 Time	-0.06 (0.54)	-0.19 (0.66)	0.01 (0.72)
	Trial 2 Time	-0.29 (1.04)	0.11 (0.65)	-0.20 (0.94)
	Trial 3 Time	-0.85 1.56)	-0.54 (1.94)	0.30 (0.91)
	Trial 4 Time	-1.27 (1.90)*	-0.97 (1.91)	-0.34 (1.39)
	Total Time	-1.31 (1.88)*	-0.79 (2.16)	-0.46 (1.29)
	Trial 1 Efficiency	-0.07 (0.36)	-0.22 (0.75)	0.06 (0.63)
	Trial 2 Efficiency	-0.37 (1.33)	-0.29 (1.23)	-0.10 (0.91)
	Trial 3 Efficiency	-0.33 (1.80)	-0.03 (1.75)	0.51 (1.36)
	Trial 4 Efficiency	-1.39 (1.77)*	-0.68 (1.67)	-0.03 (0.96)
	Total Efficiency	-1.22 (1.96)*	-0.52 (1.93)	0.15 (1.14)
Oral Fluency	Phonemic (FAS)	1.81 (3.96)	0.83 (4.04)	3.17 (5.71)
	Semantic (animals)	-0.26 (1.12)	0.72 (1.94)	0.64 (1.49)
Trail Making Test	Part A Time	0.43 (0.32)	0.77 (0.71)	0.97 (0.42)
	Part B Time	0.27 (0.69)	0.13 (0.81)	0.00 (1.37)

Note: Mean (SD). *p<0.05
Abbreviations: HPA = mild non-PKU hyperphenylalaninemia; PKU = phenylketonuria.

Table 4: Group mean performances on scales of the self- and parent-reported CBRS.

	Conners CBRS-SR			Conners CBRS-PR		
	HPA (n=3)	**PKU (n=4)**	**Control (n=4)**	**HPA (n=5)**	**PKU (n=4)**	**Control (n=5)**
Content scale						
Emotional distress	51.33 (8.08)	53.00 (11.05)	45.00 (8.52)	50.20 (7.26)	61.60 (15.87)	49.00 (7.78)
Upsetting thoughts	-	-	-	46.00 (0.71)	57.00 (22.00)	45.80 (1.30)
Worrying	-	-	-	48.40 (8.20)	53.75 (12.34)	52.20 (13.70)
Social problems	-	-	-	52.00 (10.91)	59.80 (20.50)	46.20 (9.39)
Defiant/aggressive	44.67 (2.89)	59.00 (19.87)	45.00 (6.73)	49.40 (6.15)	56.20 (19.29)	47.00 (3.74)
Academic difficulties	53.33 (2.52)	54.00 (9.56)	53.50 (9.88)	49.40 (11.87)	54.00 (16.43)	48.80 (11.67)
Language	-	-	-	47.40 (6.19)	47.80 (6.94)	44.60 (6.99)
Mathematics	-	-	-	55.60 (19.84)	54.60 (10.69)	57.80 (21.46)
Hyperactivity/impulsivity	46.00 (1.73)	50.75 (9.03)	50.75 (10.50)	44.80 (3.63)	54.60 (20.78)	49.00 (7.68)
Separation fears	47.67 (6.03)	55.00 (10.71)	45.00 (6.06)	55.40 (11.15)	57.75 (10.44)	44.00 (3.74)
Perfectionistic/compulsive	-	-	-	44.60 (3.85)	51.75 (7.93)	44.00 (3.32)
Violence potential	44.67 (1.15)	55.75 (14.57)	46.00 (5.35)	49.80 (5.36)	49.00 (3.65)	47.20 (1.10)
Physical symptoms	52.33 (6.66)	58.75 (15.52)	44.50 (9.95)	53.40 (7.16)	61.75 (14.38)	47.00 (6.44)
Symptom scale						
ADHD PI	50.00 (5.29)	54.75 (10.31)	49.50 (15.55)	44.00 (4.06)	57.40 (20.17)	45.20 (4.09)
ADHD PHI	46.00 (1.73)	50.75 (9.03)	50.75 (10.50)	44.80 (3.63)	56.60 (19.46)	49.00 (7.68)
CD	43.33 (0.58)	59.50 (17.14)	46.25 (5.85)	47.80 (6.91)	56.20 (19.14)	45.00 (2.45)
ODD	47.33 (10.12)	50.75 (11.79)	48.00 (10.61)	54.40 (17.76)	58.60 (19.35)	50.20 (7.66)
Symptom scale						
MDE	52.67 (5.51)	52.75 (13.79)	45.75 (7.80)	48.40 (6.80)	58.80 (12.52)	47.80 (7.05)
Manic	47.33 (6.66)	50.75 (15.65)	44.75 (10.56)	44.20 (4.15)	53.00 (9.63)	45.80 (10.33)
GAD	54.00 (7.81)	52.25 (8.54)	44.50 (8.06)	51.80 (13.03)	61.00 (10.56)	52.40 (8.44)
Separation anxiety	48.67 (4.16)	54.75 (14.55)	46.00 (7.87)	54.80 (9.83)	60.50 (13.58)	43.60 (3.91)
Social anxiety	50.67 (9.87)	52.00 (10.49)	48.50 (15.76)	47.40 (6.66)	54.00 (9.56)	48.00 (9.87)
OCD	48.00 (4.58)	50.50 (11.39)	45.00 (4.69)	45.40 (0.89)	53.50 (11.62)	45.00 (0.00)
ASD	-	-	-	47.20 (5.76)	53.00 (4.83)	47.80 (9.01)

Note: Mean (SD).
Abbreviations: CBRS = Comprehensive Behavior Rating Scale; PKU = phenylketonuria; HPA = mild non-PKU hyperphenylalaninemia; SD = standard deviation; ADHD PI = attention-deficit/hyperactivity disorder –inattentive type; ADHD PHI = attention-deficit/hyperactivity disorder – hyperactive-impulsive type; CD = conduct disorder; ODD = oppositional defiant disorder; MDE = major depressive episode; GAD = generalised anxiety disorder; OCD = obsessive compulsive disorder; ASD = autism spectrum disorder.

Table 5: Frequency and proportion of participants and observations in the clinically significant range on the CBRS self-report by group.

Participants n (%)	CBRS-SR			CBRS-PR		
	HPA (n=3)	**PKU (n=4)**	**Control (n=4)**	**HPA (n=5)**	**PKU (n=5)**	**Control (n=5)**
High Average	1 (33.33)	3 (75.00)	2 (50.00)	3 (60.00)	4 (80.00)	4 (80.00)
Elevated	0 (0.00)	2 (50.00)	1 (25.00)	0 (0.00)	3 (60.00)	0 (0.00)
Very Elevated	0 (0.00)	1 (25.00)	1 (25.00)	2 (40.00)	3 (60.00)	2 (40.00)
Observations n (%)	**(n=51)**	**(n=68)**	**(n=68)**	**(n=120)**	**(n=109)**	**(n=120)**
High Average	4 (7.84)	10 (14.71)	3 (4.41)	5 (4.17)	15 (13.76)	10 (8.33)
Elevated	0 (0.00)	4 (5.88)	2 (2.94)	1 (0.83)	5 (4.59)	0 (0.00)
Very Elevated	0 (0.00)	7 (10.29)	2 (2.94)	5 (4.17)	17 (15.60)	3 (2.50)

Notes: "Average" *T*=<60; "High Average" *T*=60–64; "Elevated" *T*=65–69; "Very Elevated" *T*>70.
Abbreviations: CBRS = Conners' Comprehensive Behavior Rating Scale; PR = parent report; SR = self-report; HPA = mild non-PKU hyperphenylalaninemia; PKU = phenylketonuria.

Table 6: Mean scores and effects of group on the BRIEF-2.

BRIEF-2 self-report	HPA (n=3)	PKU (n=3)	Control (n=3)	F	p	Partial η^2
Inhibit	44.33 (5.03)	54.00 (17.78)	50.67 (16.86)	0.92	0.47	0.32
Self-monitor	47.33 (3.21)	62.67 (10.26)	45.33 (8.39)	5.37	0.07	0.73
Shift	53.00 (7.55)	59.33 (14.57)[a]	52.33 (12.86)[a]	7.16	0.05	0.78
Emotional control	49.00 (10.82)	60.67 (12.90)	46.33 (9.29)	3.32	0.14	0.62
Task completion	52.67 (8.02)	64.00 (13.75)	53.33 (12.01)	3.04	0.16	0.60
Working memory	50.67 (6.81)	58.00 (14.73)	55.00 (13.11)	1.08	0.42	0.35
Plan/organise	45.00 (2.65)	55.00 (14.18)	53.67 (15.14)	1.16	0.40	0.37
Behaviour Regulation Index	45.00 (3.46)	58.00 (16.09)	41.67 (3.06)	1.85	0.27	0.48
Emotion Regulation Index	51.00 (4.36)[a]	62.00 (13.89)[a]	49.33 (12.86)[a]	34.45	0.00	0.95
Cognitive Regulation Index	48.67 (4.93)	59.00 (14.73)	54.33 (14.29)	1.78	0.28	0.47
Global Executive Composite	48.00 (5.20)	60.00 (14.80)	49.67 (11.93)	3.03	0.18	0.60
BRIEF-2 parent report	**(n=6)**	**(n=5)**	**(n=5)**	**F**	**p**	**Partial η^2**
Inhibit	50.00 (12.70)	51.40 (15.24)	43.40 (6.02)	0.89	0.89	0.06
Self-monitor	53.50 (8.31)	48.20 (9.52)	41.20 (4.38)	0.34	0.34	0.42
Shift	50.17 (11.05)	53.60 (10.14)	51.40 (15.90)	0.47	0.47	0.32
Emotional control	54.00 (11.45)	59.60 (12.46)	48.80 (6.98)	0.65	0.65	0.19
Initiate	47.30 (6.19)	53.20 (16.47)	40.20 (2.17)	0.73	0.73	0.15
Working memory	48.80 (9.41)	54.00 (17.41)	48.80 (16.10)	0.30	0.30	0.46
Plan/organise	47.30 (7.66)	53.20 (16.24)	46.20 (11.80)	0.57	0.57	0.25
Task monitor	41.83 (5.95)	51.60 15.42)	41.00 (4.80)	0.45	0.45	0.33
Organisation of materials	44.83 (5.85)	52.00 (14.75)	41.20 (3.70)	0.95	0.94	0.03
Behaviour Regulation Index	51.67 (10.60)	52.40 (12.20)	42.00 (6.20)	0.53	0.53	0.27
Emotion Regulation Index	52.17 (9.50)	55.20 (12.44)	50.60 (9.56)	0.72	0.72	0.15
Cognitive Regulation Index	45.50 (6.28)	54.00 (18.99)	42.80 (8.11)	0.62	0.62	0.21
Global Executive Composite	48.50 (8.76)	53.80 (14.79)	44.40 (7.37)	0.73	0.73	0.15

Notes: Mean (SD). [a] Groups with significant difference.
Abbreviations: BRIEF-2 = Behavior Rating Inventory of Executive Functioning, 2nd edition; HPA = mild non-PKU hyperphenylalaninemia; PKU = phenylketonuria.

Table 7: Group mean scores on self- and parent-reported quality of life.

	HPA	PKU	Control
PedsQL self	**(n=6)**	**(n=5)**	**(n=6)**
Physical	94.38 (6.79)	92.12 (13.02)	94.17 (14.29)
Emotional	68.33 (30.07)	62.25 (16.36)	84.58 (17.26)
Social	82.78 (18.31)	79.67 (14.40)	93.61 (13.35)
School	86.67 (9.01)	54.67 (36.20)	88.06 (22.98)
Psychosocial	79.26 (15.72)	65.53 (17.57)	88.75 (16.85)
PedsQL parent	**(n=6)**	**(n=4)**	**(n=5)**
Physical	88.13 (18.24)	89.06 (10.67)	98.38 (2.32)
Emotional	79.79 (20.16)	67.34 (13.05)	75.50 (14.83)
Social	78.06 (14.58)	85.00 (23.80)	100.00 (0.00)
School	89.44 (19.63)	72.08 (18.12)	93.33 (14.91)
Psychosocial	82.43 (13.92)	74.81 (16.78)	89.61 (7.08)

Note: Mean (SD). Higher scores indicate greater quality of life.
Abbreviations: PedsQL = Pediatric Quality of Life; HPA = mild non-PKU hyperphenylalaninemia; PKU = phenylketonuria; SD = standard deviation; Phe = phenylalanine.

Correlations with Phe levels

Due to the small sample size, the following findings need to be viewed with caution. Bivariate correlations were generated between all outcomes and Phe levels (birth and current). There was only one significant correlation with the cognitive measures; birth Phe was negatively correlated with WIAT-II-A numerical operations performance (r=-0.58; p=0.05).

In regards QoL, self-reported school functioning was significantly correlated to birth and current Phe (r=-0.64, p=0.04; and r=-0.62, p=0.04, respectively) as was parent-reported school functioning and current Phe (r=-0.70; p=0.02). Correlations with the PKU-QoL were also conducted but should be interpreted with additional caution given the measures were administered only to PKU participants. Birth Phe was significantly positively correlated with self-reported tiredness, lack of concentration, and overall health status. Higher current Phe was significantly positively correlated with parent reported aggression, social impact, and difficulties with adherence related to the strict diet and taste of formula.

In regards behavioural difficulties, birth Phe was significantly positively correlated with self-reported conduct disorder and parent reported perfectionistic and compulsive behaviour. Current Phe was significantly positively correlated with parent reported upsetting thoughts, OCD, ADHD –predominantly inattentive, major depressive episode, and manic episode. There was only one significant relationship with the BRIEF-2, whereby participants with higher current Phe self-reported greater self-monitor difficulties (r=0.86; p=0.03).

Discussion

All three groups performed in the normal range across the ability and achievement measures indicating intelligence was not significantly compromised in HPA or PKU groups compared to population norms or matched controls. This is consistent with the early literature on HPA, but in contrast with more recent findings.[9–10,24] Previous research on PKU has generally found IQ within the average range, but marginally lower than the general population.[6,25–26] In this study, the PKU group obtained a mean IQ score one point higher than the mean for controls, and four points higher than the HPA group. We hypothesised that working memory and processing speed difficulties

would be present in both HPA and PKU groups, given previous studies of PKU and the vulnerability of these functions to neurological insult.[4,26–27] However, this was not supported.

Though caution is needed given the small size of the sample, our findings do support the effectiveness of the existing programme in preventing cognitive deficits. For HPA participants, we did not find any evidence of deficits warranting active management, and for early treated PKU participants, we observed a normal cognitive profile.

In regards behaviour, while group mean scores did not differ significantly, the proportion of "Very Elevated" scores was much higher in the PKU group, occurring at a rate nearly eight times that of the normal population, especially with regards to externalising behaviour and anxiety. Executive behaviour difficulties were also more commonly self-reported by the PKU group, including significantly higher rates of problems shifting set compared to controls and poorer emotional regulation compared to both HPA and controls. Higher Phe levels were linked with conduct disordered behaviour, attention and self-monitoring difficulties, upsetting thoughts, and poorer school-related QoL.

Few studies have assessed QoL in HPA, whilst studies of PKU participants have produced contradictory, yet modest results. Though not significantly different, QoL was poorest amongst PKU participants and best amongst controls and was likely related to the burden of dietary treatment.[28–29] The weighing and monitoring of dietary protein is time intensive, and difficulty explaining the diet to others limits supports for parents as well as child social inclusion (e.g., invitations to sleepovers). Further, complaints related to the acrid taste of formula, hunger, food refusal, and temptation to eat restricted foods increase parent–child conflict, guilt and emotional strain.

Strengths and limitations

This is the first study to assess New Zealanders diagnosed with HPA. Assessment was broad, including cognition, executive abilities, behaviour and QoL, and spanned both self and parent reports. This study adds to the literature on QoL assessed using the PKU-QoL. Data from the PKU-QoL elucidated the link between strict dietary adherence and parental burden.

As with any rare disease, the biggest challenge was sample size. Small sample size greatly compromised power and increased the risk of type I error. Use of the CBRS, which has a minimum administration age (11 years), further reduced sample size. Males were not represented in this study (only two males were diagnosed with HPA within the period). Gender has not been found to impact outcomes in HPA.[24–25]

Clinical implications

While the findings suggest cognition is preserved for both HPA and PKU participants within the current model of care, further examination of the impact of PKU, and especially PKU treatment, on behavioural and QoL outcomes is warranted.

With PKU, it is recognised that there is no "one size fits all" target Phe level. While there are international guidelines re recommended Phe levels, due to the complexity and difficulty of the diet these may not be easily achieved in some patients. The younger developing brain is more vulnerable than the older brain, and so a more intensive diet is desired for younger children to ensure Phe levels remain in a safe range. However, Phe targets may be slightly relaxed for older children, especially those who struggle with adherence due to the adverse impacts of this on individual and family QoL.

With HPA, less rigorous follow-up including monitoring of Phe levels and clinical review may be indicated, without routine treatment as per PKU. Formalising a pathway for routine monitoring would also benefit identification and appropriate treatment of women at risk of maternal HPA.[31]

Conclusion

At this stage, there is not sufficient evidence to warrant dietary treatment of HPA except during pregnancy but there is also insufficient evidence to safely exclude the presence of cognitive impairment. Establishing conclusive, evidence-based recommendations for the management of HPA will likely require an international consensus statement regarding HPA as well as collaborative research strategies that pool resources and provide shared knowledge platforms.[16]

COMPETING INTERESTS
Nil

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Documented incontinence after stroke: a secondary analysis of a cohort study. Reducing Ethnic and Geographic Inequities to Optimise New Zealand Stroke Care (REGIONS Care)

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ABSTRACT

AIM: To estimate the prevalence of incontinence after stroke in Aotearoa New Zealand overall and by ethnicity, the associations between incontinence and subsequent mortality and living in residential care, and to estimate the health utilities in relation to continence.

METHOD: Secondary analysis of data from a prospective (1 May to 31 July 2018) cohort study (REGIONS Care study) of patients with a confirmed stroke admitted to New Zealand hospitals. Logistic and linear regression were used, and multivariate models were adjusted for age, sex, ethnicity, and stroke severity. The association between living in residential care, incontinence, and mobility was also assessed.

RESULTS: There were 320/2,377 (13.5%) patients with documented incontinence during hospitalisation after stroke. Incontinence was not associated with ethnicity but was associated with increased mortality/living in residential care, at discharge, three, six and twelve months after stroke. Stroke survivors with independent mobility were more likely to live in residential care if incontinent. Health utility scores were lower at three, six and twelve months for those with incontinence after stroke.

CONCLUSION: This study likely underestimated incontinence prevalence after stroke, although incontinence was associated with increased mortality and probability of living in residential care.

Incontinence after stroke is a common problem. Additionally, urinary incontinence and faecal incontinence are both associated with more severe stroke.[1] Urinary incontinence after stroke may be caused by the direct effects of the cerebral lesion on neurological bladder control physiology, but may also be caused by motor (mobility and dexterity), cognitive (apraxia and agnosia), visual, and language effects of the stroke that adversely affect toileting.[2] The reported prevalence of urinary and faecal incontinence after stroke varies by study but it is likely that more than half of patients may experience urinary incontinence,[3,4] and up to 40% have faecal incontinence in the acute stage after stroke.[5] Urinary incontinence after stroke is associated with poor functional outcomes,[3,6] lower quality of life,[7–10] more social isolation and depression,[10] greater likelihood of living in residential care,[4,6,11] and greater mortality.[4,11,12] There is evidence that faecal incontinence after stroke is associated with increased mortality,[5,13] and likelihood of living in residential care.[13] This contrasts with a New Zealand study of those without stroke, which reported that in older adults urinary but not faecal incontinence was a risk factor for admission to residential care.[14]

In Aotearoa New Zealand, two studies report on incontinence after stroke. A 1986 study reported the prevalence of urinary incontinence after stroke was high and depended on the time of assessment after stroke. In that study 60%, 42% and 29% of survivors after stroke, had urinary incontinence one, four and 12 weeks after stroke, respectively.[15] The Auckland Stroke Outcomes (ASTRO) study reported that 27% of stroke survivors had "bladder control problems" five years after stroke.[16] The prevalence of incontinence for different ethnic groups after stroke is unclear. The ASTRO study reported that there was no statistical evidence of a difference in bladder control problems after stroke in relation to ethnicity, although a higher proportion, 36% of Pacific peoples, reported problems with bladder control, compared to 27% of participants overall. The number of Pacific peoples in the study was small (n=30), and the study likely lacked statistical power to definitively assess whether a meaningful difference in urinary incontinence prevalence in relation to ethnicity was present or not.

The Australian and New Zealand Clinical Guidelines for Stroke Management recommend that all patients with stroke who may have continence problems should have a "structured functional assessment", and those with incontinence should have a continence management plan implemented.[17] A recent survey of stroke service provision in New Zealand reported that 74% of hospitals that routinely treat patients with acute stroke had a protocol or guideline in place to manage urinary incontinence,[18] and 59% had one for managing faecal incontinence (unpublished data, 2020).[18] In Australia, a 2019 acute stroke services clinical audit reported that although 32% of patients had urinary incontinence documented, only 37% of these patients had a documented continence management plan.[19]

There is a lack of contemporary data about the prevalence and management of incontinence after stroke in New Zealand to inform planning and provision of services aimed to enable people with stroke to live well, for longer, in the community. This study is a secondary analysis of data collected for the Reducing Ethnic and Geographic Inequalities to Optimise New Zealand Stroke (REGIONS) Care study.[20] The current study aims were to: 1) estimate the prevalence of incontinence after stroke; 2) explore if the prevalence of incontinence after stroke was related to ethnicity including after adjustment for other important predictors of incontinence such as age and stroke severity; 3) estimate the strength of association, adjusted for important predictors of incontinence, between mortality and/or living in residential care with incontinence; 4) assess whether mobility status is an effect modifier to the association between living in residential care and continence; and 5) estimate the health utilities in patients with and without incontinence.

Methods

REGIONS Care is a nationwide study assessing the impact of hospital geographic location and ethnicity on stroke outcomes and access to best practice care. Full study methods and primary analyses for REGIONS Care are described elsewhere.[20–22]

In brief, REGIONS Care was a prospective, observational cohort study of all patients with a confirmed diagnosis of stroke who were admitted to the 28 hospitals that treat patients with acute stroke between 1 May and 31 July 2018. After this date, data collection continued until hospitals had achieved a minimum sample size of 150 (for stroke clot retrieval centres) or 100 for other centres, or until 31 October 2018—whichever occurred first. Data (baseline characteristics; acute treatment, investigations, and rehabilitation) up to and including the three months post stroke follow-up were collected by hospital staff. As registry-based data collection this process was considered audit and did not require individual patient consent. Patients were then contacted and were invited to consent to extended follow-ups at six and twelve months, data linkage with health administrative data, focus groups and surveys (Central Health and Disability Ethics Committee, reference 17/CEN/164).

Selected participant baseline characteristics, and possible predictors of incontinence (any type) after stroke were extracted from the full data set. These included: age, sex, ethnicity, stroke risk factors, primary diagnosis, pre-morbid level of function, pre-stroke living situation, and stroke severity at time of hospital admission using the six simple variables (SSV) model (a composite of age, living alone pre-stroke, independence in activities of daily living pre-stroke, Glasgow Coma Scale verbal component, arm power, and ability to walk unaided at admission).[23] Patient outcomes at discharge were death and discharge destination (home or residential care), and at three, six and twelve months, outcomes were death or living in residential care which is a composite measure for a poor outcome that encompasses both severe functional deficits and mortality. The composite measure is needed because a high proportion of those with stroke die, and so an outcome that just depends on residential care status will have missing data. As a potential effect modifier, mobility status (independent or dependent), was determined at each time point from the relevant modified Rankin Scale (mRS) score. The dichotomisation (mRS 0 to 3; mRS 4 or 5) was made to capture different levels of disability likely to influence the functional aspects of toileting, particularly walking to the toilet independently. People with a mRS score of 4 or 5 require external assistance for walking or are bedridden respectively. The EuroQol EQ-5D-3L quality of life measure was used to estimate Health Utility Scores which are scaled as a score from zero, representing a quality of life that is equivalent to death, to one, representing perfect health.[24]

Incontinence-related variables in the REGIONS Care dataset were any documented incontinence (any type), documented continence management

plan, use of an indwelling catheter (IDC), urinary tract infection (UTI) and constipation post stroke, during acute care and before discharge; all these were collected as yes/no responses. If an IDC was used, a reason could be stated. Data collection did not distinguish between urinary or faecal incontinence, or existence of both. Incontinence data were collected in a non-compulsory data field related to stroke complications; incontinence may have been documented in clinical notes but inconsistently in study data.

Continuous variables are summarised by mean and standard deviation (SD), and median and interquartile (IQR) range, as appropriate. Counts are summarised by proportions and appropriate confidence intervals for a proportion. Logistic regression examines associations with dichotomous outcome variables and linear regression with continuous outcome variables. Multivariate models were adjusted for age, sex, ethnicity, and stroke severity as confounders and the association between living in residential care and incontinence main effects and mobility assessed by appropriate interaction terms. Stata/IC 16.0 was used for analysis.

The Health Research Council of New Zealand (HRC 17/037) funded the REGIONS Care study and the study received ethics approval from the Central Region Health and Disability Ethics Committee (17CEN164).

Results

The baseline characteristics of patients are summarised in Table 1. There were 2,379 patients with stroke admitted to hospital during the study period, of whom 320 had documented incontinence and 2057 did not; two did not have recorded data about incontinence. The mean (SD) age was 75 (13.7) years; 1,219/2,379 (51.2%) were male; and 2,052/2,343 (87.6%) with a recorded stroke type had ischemic stroke. Baseline characteristics of patients with and without incontinence (any type) are shown in Table 1.

The overall prevalence of incontinence problems and related issues are summarised in Table 2.

The prevalence of incontinence and related variables by ethnicity is summarised in Appendix 1. There was no evidence that the proportion of those with incontinence was associated with ethnicity in univariate (P=0.61) or multivariate analysis adjusting for age, sex and stroke severity, P=0.58. The univariate and multivariate adjusted odds ratios (OR) are shown in Table 3 with NZ European ethnicity as the reference level.

Patients with documented incontinence were more likely to die (52/320 (16.3%)) or be discharged to residential care (118 (36.9%)) than those with no documented incontinence (9.6% and 11.8% respectively). Table 4 shows the summary data, unadjusted and adjusted (for age, sex, and stroke severity) odds ratios for the individual outcomes at discharge, and the composite outcomes at the subsequent time points, in relation to documented incontinence.

Disability on discharge, for those who were discharged alive, was an effect modifier for the risk of entering residential care. In a univariate analysis 247/360 (68.6%) of those discharged to residential care had a mRS of 4 or 5 (includes lacking independent mobility) compared to 159/1,749 (9.1%) of those discharged to the community, OR (95% CI) 18.5 (13.9–24.7), P<0.001. For the same group of those discharged alive, 118 (32.8%) of those discharged to residential care had incontinence compared to 149 (8.5%) of those discharged to the community; OR (95% CI) 1.87 (1.32–2.65), P<0.001. However, the P-value for the interaction between disability and incontinence in relation to discharge status was statistically significant (P<0.001) indicating that a person with less disability was more likely to be discharged to residential care if incontinent. Table 5 shows the proportions of those with incontinence amongst those with more versus less disability discharged to residential care or the community. In those with less disability, incontinence was strongly related to discharge to residential care; but there was no association in those with more disability.

The mean (SD) health utility scores for patients with documented incontinence were 0.52 (0.28) at three months, 0.55 (0.28) at six months and 0.53 (0.27) at 12 months; compared to 0.71 (0.23) at three months, 0.71 (0.24) at six months and 0.70 (0.25) at 12 months for patients without documented incontinence. The differences in mean health utility score by continence status in the univariate modelling was -0.19 (95% CI -0.23 to -0.15) at three months, -0.16 (95% CI -0.21 to -0.10) at six months, and -0.17 (95% CI -0.24 to -0.12) at 12 months. In the multivariate model (adjusted for age, sex, ethnicity and stroke severity), the differences were -0.11 (-0.15 to -0.07), -0.07 (-0.13 to -0.02), and -0.10 (-0.16 to -0.04) respectively. For patients who reported no problem on each of the five EQ-5D-3L dimensions, thus scoring a health utility score of 1 (meaning full health), the mean (SD) EQ-VAS scores at three, six and twelve months were 82.3 (13.7), 82.5 (15.5) and 84.2 (11.8) respectively.

Table 1: Baseline characteristics of patients with and without documented incontinence.

	Documented incontinence N=320 n (%)	**No documented incontinence N=2,057 n (%)**	**p value**
Age (years)			
<40	1 (0.3)	37 (1.8)	<0.001
40–49	7 (2.2)	73 (3.6)	
50–59	17 (5.3)	201 (9.8)	
60–69	36 (11.3)	365 (17.7)	
70–79	77 (24.1)	504 (24.5)	
80–89	117 (36.7)	636 (30.9)	
≥90	64 (20.1)	241 (11.7)	
Sex (male)	155 (48.4)	1,062 (51.6)	0.29
Ethnicity			
NZ European	258 (80.6)	1,563 (76.0)	0.35
Māori	27 (8.4)	246 (12.0)	
Pacific peoples	14 (4.4)	100 (4.9)	
Asian	13 (4.1)	102 (5.0)	
Other	8 (2.5)	46 (2.2)	
Type of stroke			
ICH	69 (21.6)	222 (11.0)	<0.001
Ischaemic	251 (78.4)	1,801 (89.0)	
Stroke severity (SSV)			
3 (least severe)	18 (5.6)	793 (38.6)	<0.001
2	78 (24.4)	496 (24.1)	
1	108 (33.8)	368 (17.9)	
0 (most severe)	116 (36.3)	399 (19.4)	
Independent pre-stroke (mRS 0–2)*	235 (74.1)	1,805 (88.7)	<0.001
Living situation pre-stroke (at home)*	275 (85.9)	1,897 (92.3)	0.001
Employed pre-stroke*	30 (9.4)	435 (21.3)	<0.001
Comorbidities			
Hypertension*	258 (81.1)	1,437 (70.3)	<0.001
Diabetes*	98 (30.9)	473 (23.2)	0.003
Dyslipidaemia*	114 (36.0)	884 (43.6)	0.01
Atrial fibrillation*	134 (42.1)	673 (33.1)	0.002
Smoking*	31 (7.8)	256 (12.6)	0.15
Prior stroke*	93 (29.3)	422 (20.7)	0.001
Urban hospital	209 (65.3)	1,220 (59.1)	0.04
Length of hospital stay (days) median (IQR)	19.3 (8.1 to 34.4)	5.1 (2.4 to 14.2)	<0.0001

Notes: *Missing values: stroke severity N=1; independent pre-stroke N=25 (documented incontinence N=3); living situation N=1; employed N=17 (documented incontinence N=2); hypertension N=14 (documented incontinence N=2); diabetes N=19 (documented incontinence N=3); dyslipidaemia N=34 (documented incontinence N=3); atrial fibrillation N=22 (documented incontinence N=2); smoking N=29 (documented incontinence N=3); prior stroke N=22 (documented incontinence N=2).
Abbreviations: SSV = six simple variable model; ICH = intracerebral haemorrhage.

Table 2: Prevalence of documented incontinence and related variables in the total stroke cohort.

	n/N (%)	95% CI
Documented incontinence	320/2,377 (13.5)	12.1 to 14.9
Documented continence plan*	221/309 (71.5)	66.1 to 76.5
Indwelling catheter (IDC)		
Reason for IDC**	217/2,377 (9.1)	8.0 to 10.4
Urinary retention	25/196 (12.8)	8.4 to 18.3
Pre-existing catheter	78/196 (39.8)	32.9 to 47.0
Urinary incontinence	34/196 (17.3)	12.3 to 23.4
Need for accurate fluid balance monitoring	23/196 (11.7)	7.6 to 17.1
Critical skin care	6/196 (3.1)	1.1 to 6.5
No reason documented	30/196 (15.3)	10.6 to 21.1
Urinary tract infection	102/2,377 (4.3)	3.5 to 5.2
Constipation	171/2,377 (7.2)	6.2 to 8.3

Notes: * Missing: N=11; ** Missing: N=21.

Table 3: Odds ratio for documented incontinence compared to NZ European ethnicity.

Ethnicity	Documented incontinence n/N (%)	OR (95% CI)	
		Univariate	Multivariate
NZ European	258/1,821 (14.2)	Reference	Reference
Māori	27/246 (9.9)	0.75 (0.79–1.14)	0.74 (0.47–1.18)
Pacific peoples	14/114 (12.3)	0.85 (0.48–1.51)	0.97 (0.53–1.80)
Asian	13/115 (11.3)	0.77 (0.43–1.40)	0.84 (0.45–1.56)
Other	8/54 (14.8)	1.05 (0.49–2.26)	1.15 (0.52–2.54)

Table 4: Association of documented incontinence with death or discharge to residential care and time.

	n/N (%)		OR (95% CI)	
	Documented incontinence	**No documented incontinence**	**Univariate**	**Multivariate***
At discharge Deceased or living in residential care	170/320 (53.1)	442/2,057 (21.5)	4.14 (3.25–5.28) P<0.001	2.17 (1.62–2.91) P<0.001
3 months Deceased or living in residential care	155/241 (64.3)	448/1,597 (28.1)	4.62 (3.48–6.15) P<0.001	2.50 (1.76–3.55) P<0.001
6 months Deceased or living in residential care	127/185 (68.7)	421/1,231 (34.2)	4.21 (3.02–5.87) P<0.001	2.00 (1.34–3.00) P=0.001
12 months Deceased or living in residential care	136/187 (72.7)	440/1,224 (36.0)	4.75 (3.37–6.69) P<0.001	2.47 (1.64–3.74) P<0.001

Notes: *Adjusted for age, sex, ethnicity, stroke severity using SSV model.

Table 5: Residential care status in relation to documented incontinence and disability at discharge in those discharged alive.

Documented incontinence		**OR (95% CI) for residential care**	**P**
Moderate or severe disability (mRS 4 or 5)			
Residential care	Community		
93/247 (37.7%)	57/159 (35.8%)	1.08 (0.71–1.64)	0.71
No, no significant, or slight disability (mRS 0–3)			
Residential care	Community		
25/113 (22.1%)	92/1,590 (5.8%)	4.63 (2.83–7.56)	<0.001

Discussion

In this secondary analysis of documented incontinence (or not) in a large cohort of hospital admission for stroke we found a much lower incontinence prevalence than reported in previous New Zealand[15,16] or more recent international research.[4,5] Documented incontinence included any type or severity (i.e., any frequency or volume of leakage) at any stage in acute care before discharge. It is difficult to explain why there is a markedly reduced prevalence of post stroke incontinence in New Zealand compared to international data. It seems unlikely this reflects pathophysiological differences or acute care. It seems more likely that this difference reflects a lower rate of identifying patients with incontinence or at least inconsistent documentation of incontinence in the inpatient record,[25] rather than being a true reflection of the prevalence of incontinence post stroke in New Zealand. When incontinence was documented, nearly three quarters of patients had a continence management plan documented too. The data collection did not allow investigation of whether incontinence, or a continence plan, was more common if the incontinence was more severe, or the person had double (urinary and faecal) incontinence. If impact on nursing is greater, e.g., bed changes, or use of containment products, it is possible that documentation is more likely. The low reported prevalence may thus be focussed on the proportion of stroke survivors with severe incontinence.

Stroke guidelines recommend that all patients with suspected incontinence are assessed.[14] The low prevalence of documented incontinence in this cohort, compared to international prevalence reports, raises concerns that not all patients with incontinence were identified and the possibility that some may have experienced unmet needs in relation to continence management. This may represent an important area for ongoing service improvement efforts, such as those found to be useful for increasing guideline adherence in other areas of stroke care[26] and incontinence care[25,27] in medical and older persons wards.

We found that those with documented incontinence had increased risk of being discharged to residential care, and increased risk of being deceased or living in residential care three, six and twelve months after stroke. These findings align with previous research reporting that incontinence after stroke is associated with poorer outcomes.[4,6,11,12] Disability was an effect modifier for the association between incontinence and living in residential care. Thus, incontinence increased the risk of moving into residential care for those stroke survivors with less disability, but not for those with moderate or severe disability. It is possible those with greater dependency are already more likely to discharge to residential care. The Sentinel Stroke National Audit in the UK found that a mRS of 4 or 5 at discharge was strongly associated with first time residential placement.[28] In contrast to our findings Dutta et al, in a subgroup analysis of stroke patients with an mRS of 4 or 5, found incontinent patients had nearly five times the odds of discharge to residential care.[28]

There was no association of documented incontinence with ethnicity. A previous study, The Auckland Stroke Outcomes (ASTRO) study, did find a higher prevalence of incontinence in Pacific peoples compared to the sample population overall. Two qualitative studies, both about women's perspectives on urinary incontinence, have specifically sought experiences of Māori and Pacific women. Lennan et al. (1999)[29] sought to understand the impacts of urinary incontinence on psychological, social and medical wellbeing—the study included eight Māori, nine Pacific, and 17 Pākehā women—and found that Māori and Pacific women were more likely to say there were barriers to help seeking, including language barriers and trust and confidentiality issues. Having a trust relationship and being assured of confidentiality were the most important attributes of the doctor for Māori and Pacific women. Tui-Samoa et al. (2022)[30] carried out a single focus group with 10 Pasifika women, not all of whom had incontinence. The findings were consistent with those from Lennan et al., with the addition of understanding that culturally safe care may vary according to the woman's generation. As continence services in New Zealand are limited and inequitably distributed,[31] further study of possible inequity in access, experience and outcomes for incontinence care after stroke may be warranted.

Participants with documented incontinence had consistently reduced health utility scores (closer to zero) than those without documented incontinence at three, six-and twelve months post stroke. The mean differences (after adjusting for age, sex, ethnicity and stroke severity) were consistent with minimal clinically important differences in EuroQoL utilities reported in stroke research.[32] This suggests additional and potentially important clinical impact, attributable to incontinence, on health-related quality of life after stroke.

The strength of this analysis is that the data came from a large prospective cohort of patients with stroke. There are several limitations. First, the REGIONS Care study was not designed to specifically investigate incontinence after stroke. Although documentation of incontinence (yes/no) was not a compulsory data collection field, the data were missing for only two of the 2,377 participants. Second, there is the lack of detail about incontinence meaning it was not possible to estimate prevalence of urinary, faecal, or double incontinence, or pre-existing incontinence symptoms, how severe incontinence symptoms were, and the influence of any of these on the associations we examined. Third, the ceiling effects of the EQ-5D-3L, may have influenced the estimated health utility scores. Most patients who reported no difficulties across any of the five dimensions of the questionnaire, thus scoring a 1 for the health utility score, corresponding to "full health", did not score their overall health at 100 on the EQ-VAS, with mean scores ranging from 82.3 to 84.2 at three and twelve months respectively. The five level version of the EQ-5D may have increased the sensitivity and reduced some of the observed ceiling effects in the three level version.

The low prevalence of documented incontinence after stroke compared to international estimates may indicate that incontinence is not reliably identified, which means some stroke survivors may have unmet continence management needs. Given the data limitations outlined above, a more detailed investigation of post stroke incontinence is needed, distinguishing urinary and faecal incontinence, estimating severity using standardised and internationally validated continence assessment instruments, and to clarify if incontinence is new or pre-existing. A better description of care received, and unmet needs would also clarify the effect of incontinence on stroke survivors and their carers. These data are needed to understand the scale of the need continence services are trying to meet and make reasonable estimates of need with population ageing. However, collecting these data may also require attention to the many barriers health professionals face in identifying and managing incontinence.

Consistent with existing literature we found an association between continence problems and increased mortality and living in residential care. The finding that stroke survivors who have none to slight disability, and incontinence, were more likely to live in residential care than those who were moderately or severely disabled is counter-intuitive; the expectation being that those with more disability and continence problems are more likely to need residential care on discharge. This may possibly reflect the nature of the data collection, and is worth exploration as effective continence management in those with less disability may enable continued community living.

Conclusion

The prevalence of documented in-hospital post stroke incontinence in New Zealand is either much less than international estimates or was, more likely, underestimated in this secondary analysis of REGIONS Care data. Prevalence might not be associated with ethnicity, although more detailed incontinence data—such as type and severity—are needed to investigate the association more closely. Documented incontinence was associated with increased mortality and probability of living in residential care at discharge, three, six and twelve months. As might be expected, those with documented incontinence have reduced health utility scores compared to those without documented incontinence.

COMPETING INTERESTS
Nil.

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Appendix 1: Prevalence of continence and related variables by ethnicity.

	n/N (%) 95% CI				
	NZ European	**Māori**	**Pacific peoples**	**Asian**	**Other**
Documented incontinence	258/1821 (14.2) 12.6–15.9	27/246 (9.9) 7.3–15.6	14/114 (12.3) 6.9–19.7	13/115 (11.3) 6.2–18.6	8/54 (14.8) 6.6–27.1
Documented continence plan*	181/249 (72.7) 0.7–0.8	17/25 (68.0) 0.5–0.9	7/14 (50.0) 0.2–0.8	11/13 (84.6) 0.7–0.8	5/8 (62.5) 0.3–0.9
Indwelling catheter (IDC)	**175/1,821 (9.6) 0.08–0.11**	**20/273 (7.3) 0.0–0.1**	**7/114 (6.1) 0.0–0.1**	**6/115 (5.2) 0.08–0.11**	**9/54 (16.7) 0.08–0.11**
Reason for IDC**					
Urinary retention	21/159 (13.2) 0.1–0.2	3/18 (16.7) 0.1–0.4	1/6 (16.7) 0.0–0.8	0/6 (0.0)	0/7 (0.0)
Pre-existing catheter	68/259 (42.8) 0.35–0.51	4/18 (22.2) 0.08–0.49	1/6 (16.7) 0.01–0.77	1/6 (16.7) 0.01–0.77	4/7 (57.1) 0.17–0.90
Urinary incontinence	24/159 (15.1) 0.10–0.22	4/18 (22.2) 0.08–0.49	2/6 (33.3) 0.05–0.82	2/6 (33.3) 0.05–0.82	2/7 (28.6) 0.05–0.76
Need for accurate fluid balance monitoring	16/159 (10.1) 0.06–0.16	4/18 (22.2) 0.08–0.49	1/6 (16.7) 0.01–0.77	2/6 (33.3) 0.05–0.82	0/7 (0.0)
Critical skin care	4/159 (2.5) 0.01–0.07	1/18 (5.6) 0.01–0.34	0/6 (0.0)	0/6 (0.0)	1/7 (14.3)
No reason documented	26/159 (16.4) 0.11–0.23	2/18 (11.1) 0.03–0.38	1/6 (16.7) 0.01–0.77	1/6 (16.7) 0.01–0.77	0.01–0.70 0/7 (0.0)
Urinary tract infection	77/1,821 (4.2) 0.03–0.05	11/273 (4.0) 0.02–0.07	6/114 (5.3) 0.02–0.11	5/115 (4.4) 0.03–0.05	3/54 (5.6) 0.01–0.15
Constipation	128/1,821 (7.0) 0.06–0.08	16/273 (5.9) 0.03–0.09	8/114 (7.0) 0.06–0.08	12/115 (10.4) 0.06–0.08	7/54 (13.0) 0.05–0.25

Notes: *Missing: European N=9; Māori N=2; **Missing: European N=16; Māori: N=2; Pacific N=1; Other N=2.

We still don't count: the under-counting and under-representation of Māori in health and disability sector data

Ricci B Harris, Sarah-Jane Paine, June Atkinson, Bridget Robson, Paula T King, Jennifer Randle, Anja Mizdrak, Melissa McLeod

ABSTRACT

AIM: To examine ethnicity data quality; in particular, the representation and potential under-counting of Māori in health and disability sector data, as well as implications for inequities.

METHODS: Māori and non-Māori ethnicity data are analysed at: 1) a population aggregate level across multiple 2018 datasets (Estimated Resident Population, Census Usually Resident Population, Health Service User (HSU) population and Primary Health Organisation (PHO) enrolments); and 2) an individual level for those linked in PHO and 2018 Census datasets. Ethnicity is drawn from the National Health Index (NHI) in health datasets and variations by age and gender are explored.

RESULTS: Aggregate analyses show that Māori are considerably under-represented in HSU and PHO data. In linked analysis Māori were under-counted on the NHI by 16%. Under-representation in data and under-counting occur across both genders but are more pronounced for Māori men with variations by age.

CONCLUSION: High quality ethnicity data are fundamental for understanding and monitoring Māori health and health inequities as well as in the provision of targeted services and interventions that are responsive to Māori aspirations and needs. The continued under-counting of Māori in health and disability sector data is a breach of Te Tiriti o Waitangi and must be addressed with urgency.

> *"Being counted is an acknowledgement of both existence and value. It means that one matters. It is the hallmark of Treaty promise (Orange 1987:257). History will judge our commitment to the Treaty in part by our ability to ensure that people are counted, that disparities are acknowledged and appropriate policies are put in place, especially those which eliminate disparities between Māori and non-Māori, a solemn commitment by the Treaty of Waitangi."*[1]

Ethnicity data matter. Longstanding and significant inequities exist between Māori and non-Māori across most health indicators including life expectancy, health determinants, health outcomes and healthcare. These inequities are rooted in processes of colonisation and colonialism, supported by a system of racism.[2] They are a breach of the Te Tiriti o Waitangi and Māori Indigenous rights.[2] Furthermore, the lack of urgency towards their elimination, particularly by Crown agencies, further reflects the disregard with which Māori and Māori health are held.

Ethnicity data have been collected for decades in health and population datasets; however, the quality, consistency and completeness continue to be problematic, particularly for Māori.[3] This has often been reflected in the systematic under-counting of Māori in datasets including the National Health Index (NHI),[4,5] in mortality datasets,[6,7] and others.[8–13] This then impacts on the ability to estimate health prevalence, rates, inequities and progress (or not) over time. Accurate ethnicity data are also critical to correctly identify individuals eligible for healthcare interventions. In New Zealand, an individual's ethnicity determines the age of their eligibility for cardiovascular risk assessment[14] and diabetes screening,[15] sore throat management,[16] eligibility for diabetes treatments,[17] and the proposed age extension to the Bowel Cancer Screening Programme.[18]

Māori health advocates have long argued for high quality ethnicity data in health as a Māori health right;[1,19,20] Māori have a "right to be counted"[2] and "Māori have the right to monitor the Crown and to evaluate Crown action and inaction".[21] While protocols for the standardised collection of ethnicity data in the health and disability sector[22] have existed for nearly 20 years, their implementation seems yet to be fully realised, with ongoing concerns regarding the under-counting of Māori in

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health and disability sector data. This study examines ethnicity data quality and its implications for Māori health and inequities. We focus on NHI ethnicity as a consistent variable across a range of health datasets. The NHI provides a unique identifier for each person who receives healthcare with associated identifying information including ethnicity.[4] NHI ethnicity is the most up-to-date record of an individual's ethnicity based on their most recent health encounters. We seek to examine in detail NHI ethnicity data quality (particularly under-count) for Māori by comparing aggregate level data for Māori and non-Māori across a range of population and health datasets, and by comparing Māori and non-Māori NHI ethnicity among individuals enrolled with a primary health organisation (PHO) with their linked self-identified ethnicity from the 2018 Census. We also examine differences by age and gender.

Methods

We are guided by kaupapa Māori research methods, particularly kaupapa Māori epidemiology that includes: 1) the right to monitor the Crown; 2) the right to be counted; 3) the right to have a powerful voice; and 4) the right to name racism and colonialism as fundamental causes of inequities for Māori.[23] Our kaupapa Māori epidemiological approach supports the use of a Māori/non-Māori analysis as a reflection of Te Tiriti.

Study design

This is a descriptive study that compares the recording of Māori and non-Māori ethnicity across a range of datasets and in two main ways:

In these analyses, Māori are defined as anyone who has their ethnicity recorded as Māori, either

1. Population aggregate analysis
2. Individual linked analysis

alone, or in combination with another ethnic group(s).[22] Non-Māori are all other people in the dataset (including those with missing ethnicity). Gender could only be examined by the binary categories of male and female due to dataset limitations. The study was approved by the University of Otago Human Ethics Committee (HD 20/079).

1. Population aggregate analysis

At an aggregate level, total population data are compared for Māori and non-Māori across a range of population and health datasets to compare both counts and proportions of Māori and non-Māori. The datasets include the 2018 Estimated Resident Population (ERP); 2018 Census Usually Resident Population (Census); 2018 Health Service User (HSU) population; and 2018 Primary Health Organisation (PHO) population. Appendix 1 provides further details on the datasets. All data are estimated at a single point in time and as close as possible to the same point in time (30 June 2018) for comparability.

The two health datasets (HSU and PHO) are compared with official population estimates (ERP) to examine potential differential under-representation of Māori compared to non-Māori in health data (either from differential access to care or under-counting of Māori). We compare the total numbers and proportions of Māori and non-Māori in each dataset overall, and by age and gender. This aggregate analysis provides an indication of potential under-counting of Māori if proportions of Māori in health data are lower than non-Māori compared to official population data. However, the contribution of differential access to care cannot be teased out from potential under-counting.

2. Individual linked analysis

In order to examine the misclassification of ethnicity and potential systematic under-counting of Māori on NHI data, we undertook a linked analysis at the level of individuals. To be eligible for inclusion, people had to be enrolled in a PHO (as at 1 April 2018), have self-completed the ethnicity question in the 2018 Census, linked in the NZ Integrated Data Infrastructure (IDI) spine and to Ministry of Health (MoH) NHI data.

Analyses were undertaken in the Statistics NZ (Stats NZ) datalab using the September 2021 IDI Refresh (available from 27 October 2021). Data were linked between the IDI spine and MoH.[24] 2018 Census data were restricted to only those who had self-identified their ethnicity in this Census i.e., ethnicity supplemented from other administrative data were not used (Appendix 1). Ethnicity data from the 2018 Census were considered the standard against which PHO NHI ethnicity data were compared. This is in line with the Ethnicity Data Protocols for the Health Sector and the Census, whereby a standard question should be used and people should self-identify their own ethnicity whenever possible, with guidance on instances where this may not be possible, e.g., young children, people who are incapacitated. [22] Those with missing NHI ethnicity (75,243 individuals; 2%) were categorised as non-Māori.

Among linked individuals, two main measures were used to examine data quality. First, we cal-

culated the proportion whose ethnicity was the same in both datasets using the Māori/non-Māori population denominators from the Census (proportion matched). This provides a measure of misclassification. Secondly, the net under-count of Māori on the NHI was estimated by 1 minus (the number of people classified as Māori on the NHI as a percentage of those classified as Māori on the Census). This indicates the extent to which the misclassification is differential.

Results

Population aggregate results

Table 1 shows the population counts across population and health datasets with numbers (and proportions) for Māori and non-Māori. Of the four datasets examined, the ERP has the most people as well as the highest proportion of Māori (16.7%). The total numbers in the HSU dataset are closest to the ERP, although lower for Māori and (correspondingly) slightly higher for non-Māori. The proportion of Māori in the HSU is lower than the ERP at 15.5%. The PHO dataset has the least people and the lowest proportion of Māori (14.6%).

Figure 1 shows the number of people recorded as Māori and non-Māori in the datasets by 5-year age group. The ERP has the highest number of Māori across almost all ages. The exception is the youngest age group where ERP and HSU numbers are similar. PHO data are lowest for Māori across all ages. Differences between datasets are less obvious with increasing age.

For non-Māori, PHO and Census data have lower counts across most ages compared to ERP. For PHO data, this is more obvious for adolescents and young adults. The HSU data has higher numbers of non-Māori than ERP in children, lower numbers in young adults and similar numbers among adults and older people.

Compared to the ERP, there are fewer Māori males and females in every age group in the Census, HSU (except 0–4 years) and PHO datasets and the percentage differences are disproportionately larger than for non-Māori (Figure 2). The percentage differences for males tend to be disproportionately larger than for females in the datasets (Census, HSU, PHO) compared to the ERP, for both Māori and non-Māori. Within the HSU data, Māori males are under-represented across most age groups. For non-Māori, there is under-representation of young males and over-representation of females in multiple age groups.

Individual linked results

3,498,789 people on the PHO dataset were linked to a 2018 Census record. This equates to 78% linkage of people on the PHO dataset. Linkage varied by ethnic grouping (based on NHI ethnicity on the PHO dataset), with 64% linkage for Māori and 80% for non-Māori. Within the PHO linked sample, 491,928 (14.1%) of the population were Māori (using Census ethnicity data), compared to 414,846 (11.9%) using NHI data (Table 2). Both of these proportions are lower than the total population percentage of Māori in the ERP of 16.7% (Table 1).

Of the 491,928 individuals who identified as Māori on the Census, 386,976 (78.7%) were also recorded as Māori on the NHI (Table 2). Of the 3 million individuals categorised as non-Māori ethnicity from the Census, 2,978,991 (99.1%) were categorised as non-Māori on the NHI. The high match of NHI and Census ethnicity for non-Māori was seen across all age groups.

For Māori (on the Census) there is some variation by age, with the youngest age group (0–4 years) having a higher match (approx. 90%) and a lower level of match in those aged 20–24 years (Figure 3). The level of match is lower for Māori males than Māori females, and it is more apparent in adults over 20 years old where matching for Māori males remains consistently low.

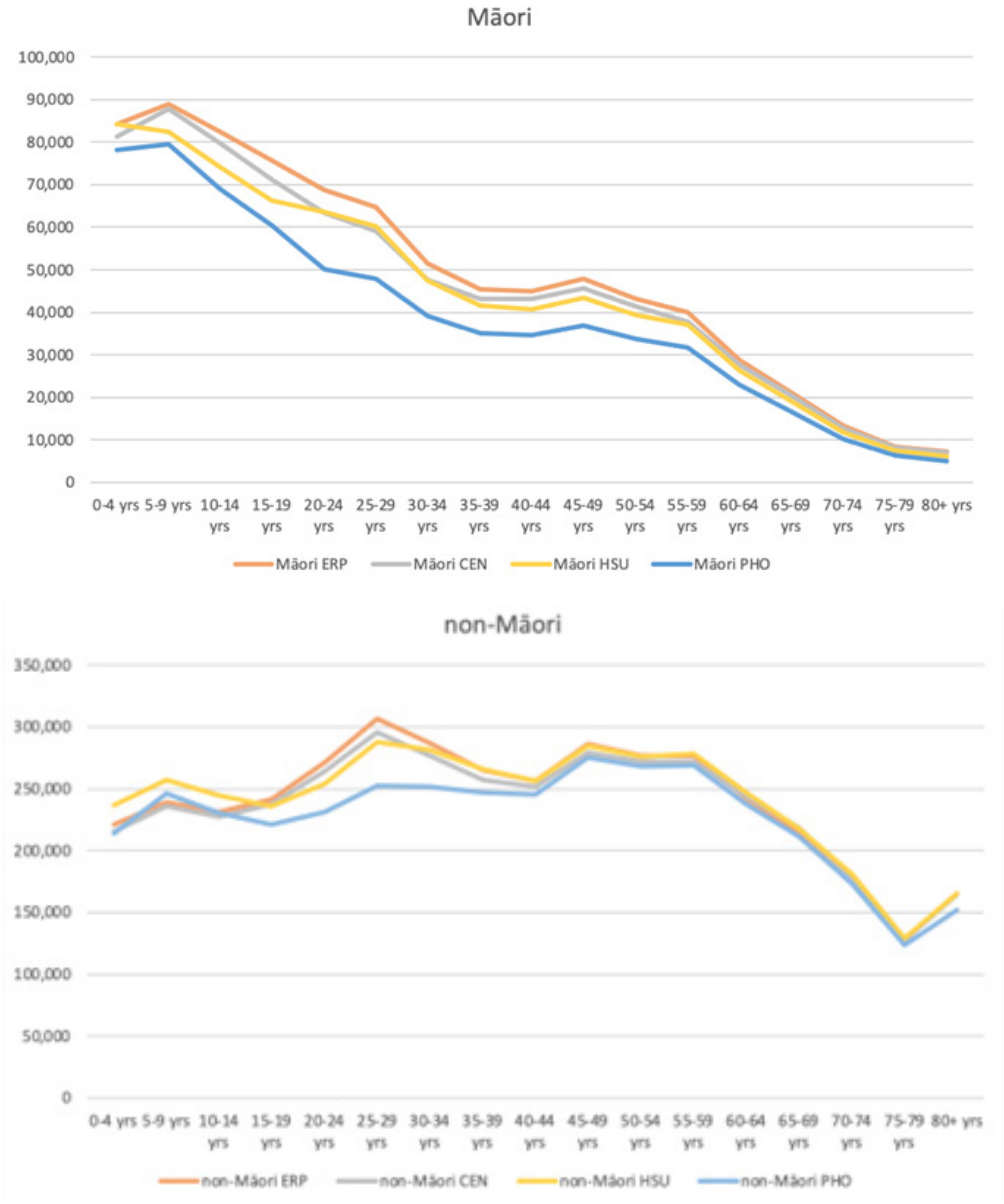
Figure 4 examines the net under-counting of Māori ethnicity on the NHI by age and gender. Net under-count tells us overall how much the NHI data under-counts Māori compared to the 2018 Census (assuming the Census is the "correct" classification). The net under-count captures the proportion of Māori missed from the NHI (i.e., those identified as Māori on the Census but not on NHI) minus non-Māori incorrectly classified as Māori in NHI (those identified as non-Māori on the Census but Māori on NHI).

The net under-count in the NHI for Māori is 15.7% overall. The net under-count for 0–4-year-olds is less than 5%, but increases to between 13–23% for all other age groups. When this is examined separately by gender, the net under-count for Māori males is higher than Māori females from 20 years on, and is greater than 20% in most age groups (20–59 years). For Māori women, there is more variation in net under-count by age, with peaks in the under-count in the age groups 20–24 years (22.7% under-count) 40–44 years (18.4% under-count), and 80+ years (19.2% under-count).

Table 1: Population numbers and proportions for 2018 ERP, 2018 Census, 2018 HSU, 2018 PHO.

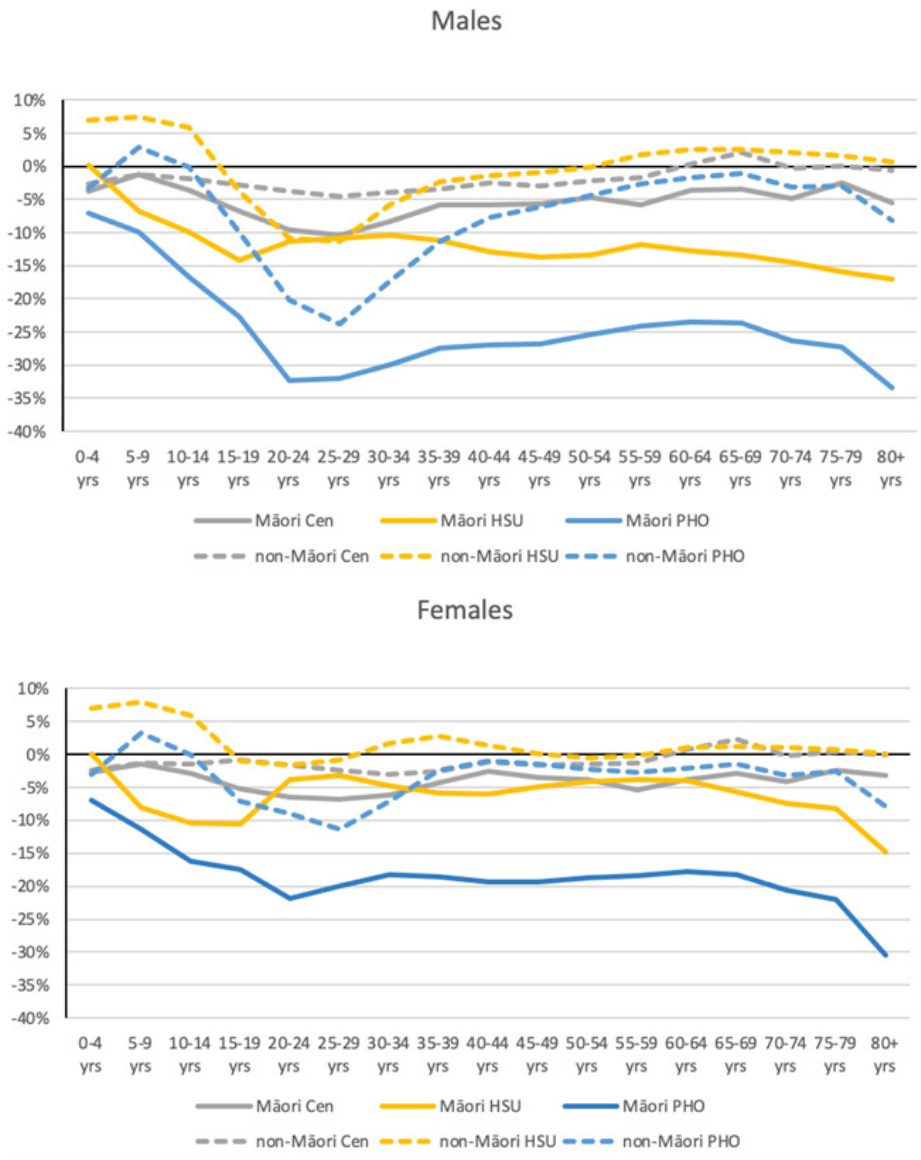
	ERP n	%	Census n	%	HSU (NHI eth) n	%	PHO (NHI eth) n	%
Māori	816,500	16.7	777,186	16.2	751,612	15.5	657,264	14.6
Non-Māori	4,084,100	83.3	4,016,172	83.8	4,097,682	84.5	3,848,193	85.4
Total	4,900,600	100.0	4,793,358	100.0	4,849,294	100.0	4,505,457	100.0

Figure 1: Numbers of Māori and non-Māori by age group 2018 ERP, 2018 Census, 2018 HSU, 2018 PHO datasets.



Note: Data in Appendix 2.

Figure 2: Percentage difference between Census, HSU and PHO datasets compared to ERP for Māori and non-Māori by age group and gender (as a percentage of the difference between Census, HSU and PHO datasets compared to ERP).

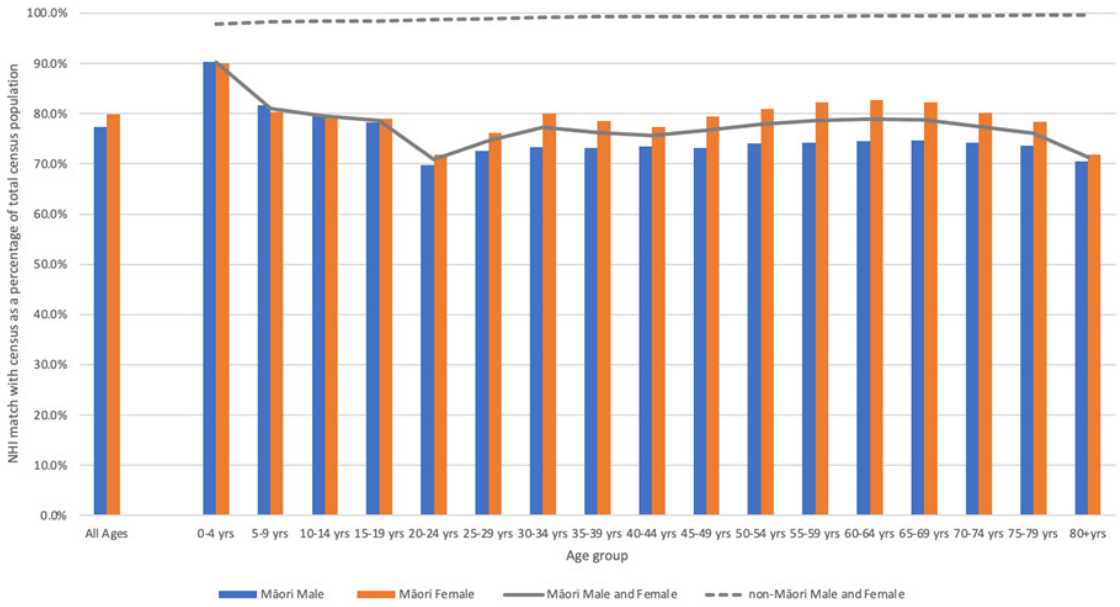


Note: Data points are in Appendix 3, derived from data in Appendix 2.

Table 2: Māori and non-Māori recording of ethnicity in 2018 Census (self-completed only) compared with NHI.

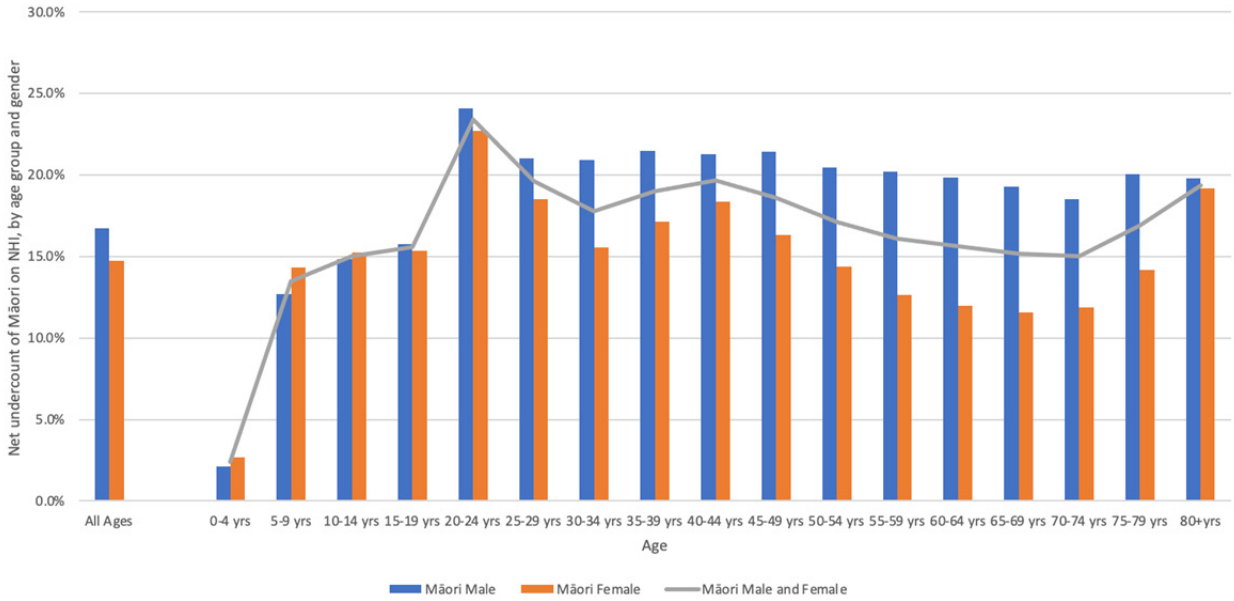
		Linked Census		
		Māori	**Non-Māori**	**Total**
NHI	Māori	386,976	27,870	414,846
	Non-Māori	104,952	2,978,991	3,083,943
	Total	491,928	3,006,861	3,498,789

Figure 3: Percentage of individuals that have matching 2018 Census (self-completed data only) and NHI ethnicity, by age group and gender.



Note: Data in Appendix 4a and 4b.

Figure 4: Net under-count of Māori on NHI compared to 2018 Census (self-completed data only), by age group and gender.



Note: Data in Appendix 4a and 4b.

Discussion

High quality ethnicity data are fundamental for understanding and monitoring Māori health and health inequities as well as in the provision of targeted services and interventions that are responsive to Māori aspirations and needs. This study shows that Māori are under-represented in health datasets (HSU and PHO), compared to official population numbers (ERP), and are systematically under-counted in NHI ethnicity data. At a population aggregate level, all four datasets had different total population and Māori numbers, reflecting differences in their composition. Additionally, there were differences in the proportion of Māori in each dataset ranging from 16.7% on the ERP to 14.6% on the PHO dataset. There were lower numbers of Māori in the HSU and PHO datasets compared to the ERP across almost all ages. This under-representation of Māori in health data is likely to be due to under-counting of Māori in health datasets as well as ongoing differential access to healthcare. This was demonstrated in the linked analysis of individuals whereby NHI ethnicity (from PHO data) was more likely to misclassify Māori as non-Māori (than non-Māori as Māori), resulting in a 16% net under-count of Māori on the NHI (with variation by age and gender).

It is clear that the health and disability sector continues to fail in its responsibility to achieve expected standards of ethnicity data collection,[22] with important implications for our ability to monitor Māori health and equity, and to target health services. In the monitoring of Māori health and equity, the under-counting of Māori in the NHI can lead to a numerator/denominator bias when rates using population data from other sources such as ERP are used, as is common practice. This will lead to underestimation of the Māori rate for any health variable of interest. In contrast, the impact of any misclassification for non-Māori will be negligible because of the much larger population. This can make *inequities* look better or worse than they really are, depending on the health variable concerned.

The HSU population is being used as one method to try and minimise the numerator/denominator bias by using it as the population denominator.[4] The 2020 HSU has been used recently in the estimation of COVID-19 vaccination coverage.[25] However, this may allow the inclusion of people in the numerator that may not be in the denominator (e.g., people who have been vaccinated but had otherwise not had a health interaction in the period of the HSU). This can potentially overestimate rates. Our study shows that the HSU should not be used to estimate actual numbers of Māori eligible for services (e.g. the number of Māori to be vaccinated), as it significantly under-represents Māori.

Other methods have also been used to mitigate the numerator/denominator bias for Māori.[26] These include the calculation of ratios to adjust for Māori under-count[6,10,12] that use similar methods as here to estimate the net under-counting of Māori. More recently, administrative datasets are being linked to create new populations in efforts to improve population estimates by ethnicity, e.g., the experimental Administrative Population Census derived from data in the IDI.[27] However, these still rely on the underlying quality of ethnicity data in the linked sources. It is important that Māori have the opportunity to self-identify their ethnicity. This is in line with Indigenous rights, as an expression of rangatiratanga and Māori rights to determine our own identities.[20]

There are implications with using ethnicity data to determine an individual's eligibility for health and disability services. A number of health services already prioritise Māori in the assessment of risk or service delivery.[14–18] Using the example of bowel cancer screening, where a lower age (from 50 years) of invitation for Māori has been proposed, a large number of Māori who are eligible would be missed if invitations to screening drew on NHI ethnicity. A smaller number of non-Māori would also be mistakenly invited.

The under-counting of Māori in health data is not new and has been examined in various settings over time. Particularly relevant to our study are other analyses that have linked individual health data with Census data. Stats NZ linked 2013 Census data to a combined NHI dataset and found Māori were under-counted by 21%.[5] More recently, the MoH has undertaken similar analyses linking people from the 2018 Census to 2019 NHI data and shown a 16% net under-count of Māori in the NHI compared to Census.[4] Our analysis also shows a 16% net under-count of Māori on the NHI from the PHO database compared to individuals' corresponding Census ethnicity.

There are a number of limitations that should be considered in the interpretation of our findings. Firstly, our analysis of linked data is a subset of Māori in the 2018 Census and 2018 PHO datasets with lower linkage for Māori (based on NHI ethnicity). We cannot determine if the level of

mismatch and under-counting of those included in this analysis is the same as those not included. While we examined data by gender and age, we were unable to explore ethnicity data for other important groups, for example those who are gender diverse and/or tāngata whaikaha. Secondly, it is possible that people self-report their ethnicity differently, in different settings, and that they can change their ethnicity.[28] However, there is also evidence from health settings that ethnicity data protocols are not always adhered to,[29] e.g., guessing someone's ethnicity based on name or appearance, disagreeing with patient's self-identified ethnicity, non-standardised forms and data recording. In addition, there is evidence of improvements in ethnicity data collection in some health-related datasets, e.g., mortality,[6,7] cancer registrations,[10] and examples where there is no (or minor) net under-count of Māori.[9,30–32] These findings would suggest that (non)adherence to ethnicity data protocols are the likely drivers of the systematic under-counting of Māori. This is a complex issue to tease out in detail, and we have chosen to focus on implications for Māori using a non-Māori comparator. However, we acknowledge that the non-Māori group is made up of multiple ethnicities combined, and that the quality of ethnicity data for specific ethnic groups, and the mismatch of ethnicity within this large grouping are not examined. In particular, similar data issues and implications are likely to be present for Pacific peoples.[4,5] We have also not looked at all potential measures of data quality here such as multiple ethnicities, missing data, and consistency with health and disability sector standards.[5,33]

The ongoing systematic under-counting of Māori in health data is a reflection that Māori are not valued and that Māori health and the elimination of inequities are not the priorities they are claimed to be. The ongoing unjust and inequitable healthcare experiences and outcomes faced by Māori require the whole of the health and disability sector to commit to the collection of high-quality ethnicity data, in order to understand inequities; to deliver equitable services to Māori; to act to address inequity; and to monitor progress on eliminating inequities. This commitment needs to come from the highest levels and be attached to accountability mechanisms and ongoing quality assessments.[34] Quality ethnicity data are fundamental to the elimination of Māori health inequities. The under-counting of Māori remains a breach of Te Tiriti o Waitangi and Indigenous rights. The ongoing acceptance of poor-quality ethnicity data for Māori and the inadequate progress towards high quality ethnicity data in the health and disability sector is itself evidence of racism as "*inaction in the face of need*".[35]

COMPETING INTERESTS
The authors have no conflicts of interest or financial disclosures.

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Stats NZ disclaimer regarding IDI data: The results in this paper are not official statistics, they have been created for research purposes from the Integrated Data Infrastructure (IDI) managed by Statistics New Zealand. The opinions, findings, recommendations and conclusions expressed in this paper are those of the author(s) not Statistics NZ or Ministry of Health. Access to the anonymised data used in this study was provided by Statistics NZ in accordance with security and confidentiality provisions of the Statistics Act 1975. Only people authorised by the Statistics Act 1975 are allowed to see data about a particular person, household, business or organisation and the results in this paper have been confidentialised to protect these groups from identification. Careful consideration has been given to the privacy, security and confidentiality issues associated with using administrative and survey data in the IDI. Further detail can be found in the Privacy impact assessment for the Integrated Data Infrastructure available from www.stats.govt.nz.

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Appendices

Appendix 1: Description of datasets used in population aggregate analysis.

Dataset	Description	Ethnicity	Date and source of datasets	References
2018 Estimated Resident Population (ERP) at 30 June 2018	The ERP is considered the official data for population estimates. The ERP "is an estimate of the number of people who usually live in New Zealand at a given date." It is "based on the 2018 Census usually resident population count, updated for residents missed or counted more than once by the census (net census under-count); residents temporarily overseas on census night; and births, deaths, and net migration between census night and the date of the estimate." (https://www.stats.govt.nz/indicators/population-of-nz)	"The Māori ethnic group ERP at 30 June of each census year is based on the respective census counts, with adjustments for census non-response (people who did not complete census forms), net census under-count (people who were missed in the census), residents temporarily overseas on census night (RTO), and estimated population change between census date and mid-year." (https://www.stats.govt.nz/methods/maori-ethnic-group-population-estimates-200618-methods-and-results)	Downloaded 5.11.2021 from Stats NZ	https://www.stats.govt.nz/methods/population-statistics-user-guide#explanations-of-selected
2018 Census Usually Resident Population (Census) at 30 June 2018	"This is a count of all people who usually live in New Zealand, or in an area of New Zealand, and are present in New Zealand on a given census night." (https://www.stats.govt.nz/methods/population-statistics-user-guide#explanations-of-selected) It does not include NZ citizens who are temporarily overseas or temporary visitors to NZ. Because of lower-than-expected participation in the 2018 Census, administrative data were used to identify people who should have been counted and 2013 Census and other administrative data used to fill in details about them, including ethnicity. (https://www.stats.govt.nz/methods/2018-census-how-we-combined-administrative-data-and-census-forms-data-to-create-the-census-dataset)	Ethnicity is sourced, in order of priority, from: self-identified ethnicity from the 2018 Census (84.4%); admin enumerated from the 2013 Census (8.2%); admin enumerated from other administrative data sources (6.2%) e.g., birth registrations and health data. (https://datainfoplus.stats.govt.nz/Item/nz.govt.stats/7079024d-6231-4fc4-824f-dd8515d33141/)	Downloaded 9.11.2021 from Stats NZ	https://www.stats.govt.nz/methods/2018-census-how-we-combined-administrative-data-and-census-forms-data-to-create-the-census-dataset https://datainfoplus.stats.govt.nz/Item/nz.govt.stats/858c2267-92e1-4eb2-a0c8-4cef372d24fd
2018 Health Service User (HSU) population at 30 June 2018	The HSU includes people who have received health services in a given year. This includes people who are enrolled in a PHO and/or have received a health service. (Cleary 2021, p.10) "The HSU is not a total population count, it doesn't contain information on people who don't use health services." (Cleary, p.5)	Latest NHI* up to July 2021. *Since 2015, the National Health Index (or NHI) can be updated by multiple health services (Cleary 2021), NHI gets overwritten as latest ethnicity and could not be obtained for the dates of the datasets	Dataset obtained 21.10.2021 from MoH	Cleary L. Using ethnicity data in health statistics. Wellington: Ministry of Health; 2021. https://www.health.govt.nz/covid-19-novel-coronavirus/covid-19-data-and-statistics/covid-19-vaccine-data

Appendix 1 (continued): Description of datasets used in population aggregate analysis.

Dataset	Description	Ethnicity	Date and source of datasets	References
2018 Primary Health Organisation (PHO) database at 1 April 2018	People enrolled with a PHO. To enrol in a PHO people must be eligible for publicly funded healthcare in New Zealand and live or intend to live long-term in NZ.	Latest NHI* up to July 2021	Data available in datalab from 27.10.2021 (September 2021 IDI Refresh "20211020")	https://www.health.govt.nz/system/files/documents/pages/eligibility-direction-2011.pdf https://tas.health.nz/assets/Primary-psaap-u14/Enrolment-Requirements-for-Contracted-Providers-and-PHOs-Version-4.1.pdf https://www.health.govt.nz/new-zealand-health-system/eligibility-publicly-funded-health-services/resources-service-providers-check-eligibility/eligibility-enrol-primary-health-organisation

Note: HSU and PHO data may contain some people who have gone overseas and are not included in the Census URP.
Abbreviation: MoH = Ministry of Health.

Appendix 2: Data for Figure 1–numbers of Māori and non-Māori by age group and gender, 2018 Estimated Resident Population (ERP), 2018 Census Usually Resident Population (CEN), 2018 Health Service User (HSU), 2018 Primary Health Organisation (PHO) datasets.

		Māori				non-Māori			
		ERP	**CEN**	**HSU**	**PHO**	**ERP**	**CEN**	**HSU**	**PHO**
Males	All Ages	406,200	383,688	364,783	316,416	2,024,000	1,980,627	2,006,989	1,864,548
	0–4 yrs	43,360	41,715	43,430	40,302	113,460	110,448	121,379	109,824
	5–9 yrs	45,800	45,246	42,705	41,247	122,630	121,050	131,697	126,087
	10–14 yrs	42,460	40,914	38,251	35,403	118,220	116,010	125,254	118,026
	15–19 yrs	38,930	36,294	33,412	30,099	124,170	120,624	119,485	111,780
	20–24 yrs	34,990	31,641	31,028	23,685	141,810	136,596	126,421	113,187
	25–29 yrs	32,170	28,827	28,692	21,855	157,170	149,928	139,457	119,850
	30–34 yrs	24,790	22,719	22,206	17,379	142,410	136,737	134,195	117,642
	35–39 yrs	21,940	20,658	19,474	15,918	130,990	126,534	127,860	116,088
	40–44 yrs	21,860	20,586	19,028	15,981	125,440	122,352	123,773	115,788
	45–49 yrs	23,160	21,834	19,978	16,947	139,460	135,351	138,163	130,929
	50–54 yrs	21,020	20,022	18,206	15,693	135,340	132,480	135,189	129,303
	55–59 yrs	19,100	18,003	16,849	14,505	134,720	132,321	137,038	131,106
	60–64 yrs	13,630	13,131	11,886	10,440	117,960	118,377	121,052	116,031
	65–69 yrs	10,060	9,717	8,708	7,677	105,030	107,271	107,692	103,875
	70–74 yrs	6,210	5,913	5,309	4,578	86,760	86,538	88,506	84,021
	75–79 yrs	3,870	3,774	3,255	2,814	59,940	59,955	60,901	58,149
	80+ yrs	2,850	2,694	2,366	1,899	68,450	68,052	68,927	62,850

Appendix 2 (continued): Data for Figure 1–numbers of Māori and non-Māori by age group and gender, 2018 Estimated Resident Population (ERP), 2018 Census Usually Resident Population (CEN), 2018 Health Service User (HSU), 2018 Primary Health Organisation (PHO) datasets.

		Māori				non-Māori			
		ERP	CEN	HSU	PHO	ERP	CEN	HSU	PHO
Females	All ages	410,300	393,489	386,770	340,779	2,060,200	2,035,557	2,090,025	1,982,736
	0–4 yrs	40,750	39,624	40,781	37,932	107,460	104,874	114,919	104,232
	5–9 yrs	43,240	42,636	39,780	38,313	116,240	114,627	125,439	120,000
	10–14 yrs	40,070	38,886	35,894	33,576	112,760	111,060	119,376	112,692
	15–19 yrs	36,770	34,866	32,902	30,324	117,480	116,523	116,368	109,179
	20–24 yrs	33,830	31,629	32,542	26,460	129,450	127,332	127,258	117,711
	25–29 yrs	32,440	30,228	31,423	25,980	149,410	145,782	148,251	132,540
	30–34 yrs	26,580	24,918	25,324	21,723	144,450	140,073	146,804	134,190
	35–39 yrs	23,420	22,407	22,047	19,071	133,720	130,317	137,510	130,440
	40–44 yrs	23,140	22,548	21,756	18,651	130,740	129,246	132,477	129,432
	45–49 yrs	24,650	23,781	23,434	19,866	146,310	143,943	146,421	144,057
	50–54 yrs	22,160	21,309	21,231	18,027	141,660	139,647	140,975	138,441
	55–59 yrs	20,990	19,848	20,186	17,130	141,620	139,815	141,415	137,697
	60–64 yrs	15,140	14,559	14,535	12,450	124,210	125,112	125,457	121,524
	65–69 yrs	11,050	10,728	10,428	9,027	108,950	111,435	110,185	107,268
	70–74 yrs	7,080	6,792	6,555	5,616	91,610	91,416	92,549	88,644
	75–79 yrs	4,570	4,458	4,195	3,567	67,500	67,791	67,948	65,763
	80+ yrs	4,410	4,272	3,757	3,069	96,590	96,570	96,673	88,920

Appendix 2 (continued): Data for Figure 1–numbers of Māori and non-Māori by age group and gender, 2018 Estimated Resident Population (ERP), 2018 Census Usually Resident Population (CEN), 2018 Health Service User (HSU), 2018 Primary Health Organisation (PHO) datasets.

		Māori				non-Māori			
		ERP	CEN	HSU	PHO	ERP	CEN	HSU	PHO
All genders	All ages	816,500	777,186	751,612	657,264	4,084,100	4,016,172	4,097,682	3,848,193
	0–4 yrs	84,110	81,336	84,217	78,240	220,920	215,325	236,335	214,083
	5–9 yrs	89,030	87,882	82,487	79,563	238,880	235,677	257,170	246,102
	10–14 yrs	82,530	79,803	74,155	68,991	230,980	227,070	244,682	230,766
	15–19 yrs	75,700	71,160	66,327	60,435	241,650	237,147	235,946	221,046
	20–24 yrs	68,810	63,267	63,581	50,160	271,270	263,931	253,784	231,120
	25–29 yrs	64,610	59,058	60,119	47,835	306,580	295,707	287,806	252,507
	30–34 yrs	51,370	47,640	47,532	39,099	286,850	276,807	281,061	251,904
	35–39 yrs	45,360	43,065	41,523	34,998	264,710	256,851	265,405	246,567
	40–44 yrs	45,000	43,134	40,784	34,635	256,180	251,595	256,285	245,262
	45–49 yrs	47,810	45,615	43,414	36,813	285,770	279,294	284,608	275,034
	50–54 yrs	43,180	41,331	39,441	33,723	277,000	272,127	276,180	267,780
	55–59 yrs	40,100	37,851	37,036	31,638	276,340	272,136	278,474	268,839
	60–64 yrs	28,770	27,690	26,422	22,890	242,170	243,486	246,526	237,591
	65–69 yrs	21,110	20,445	19,137	16,704	213,990	218,706	217,895	211,173
	70–74 yrs	13,290	12,705	11,864	10,194	178,380	177,951	181,066	172,683
	75–79 yrs	8,440	8,232	7,450	6,381	127,430	127,749	128,853	123,933
	80+ yrs	7,250	6,972	6,123	4,962	165,050	164,607	165,606	151,806

Appendix 3: Data for Figure 2–percentage difference between 2018 Census Usually Resident Population (Cen), 2018 Health Service User (HSU) and 2018 Primary Health Organisation (PHO) datasets compared to 2018 Estimated Resident Population (ERP) for Māori and non-Māori by age group and gender (as a percent of the difference between Census, HSU and PHO datasets compared to ERP). Calculated from data in Appendix 2.

		Māori				non-Māori			
		ERP	**Cen**	**HSU**	**PHO**	**ERP**	**Cen**	**HSU**	**PHO**
Males	All Ages	0.00%	-5.54%	-10.20%	-22.10%	0.00%	-2.14%	-0.84%	-7.88%
	0–4 yrs	0.00%	-3.79%	0.16%	-7.05%	0.00%	-2.65%	6.98%	-3.20%
	5–9 yrs	0.00%	-1.21%	-6.76%	-9.94%	0.00%	-1.29%	7.39%	2.82%
	10–14 yrs	0.00%	-3.64%	-9.91%	-16.62%	0.00%	-1.87%	5.95%	-0.16%
	15–19 yrs	0.00%	-6.77%	-14.17%	-22.68%	0.00%	-2.86%	-3.77%	-9.98%
	20–24 yrs	0.00%	-9.57%	-11.32%	-32.31%	0.00%	-3.68%	-10.85%	-20.18%
	25–29 yrs	0.00%	-10.39%	-10.81%	-32.06%	0.00%	-4.61%	-11.27%	-23.74%
	30–34 yrs	0.00%	-8.35%	-10.42%	-29.90%	0.00%	-3.98%	-5.77%	-17.39%
	35–39 yrs	0.00%	-5.84%	-11.24%	-27.45%	0.00%	-3.40%	-2.39%	-11.38%
	40–44 yrs	0.00%	-5.83%	-12.96%	-26.89%	0.00%	-2.46%	-1.33%	-7.69%
	45–49 yrs	0.00%	-5.73%	-13.74%	-26.83%	0.00%	-2.95%	-0.93%	-6.12%
	50–54 yrs	0.00%	-4.75%	-13.39%	-25.34%	0.00%	-2.11%	-0.11%	-4.46%
	55–59 yrs	0.00%	-5.74%	-11.79%	-24.06%	0.00%	-1.78%	1.72%	-2.68%
	60–64 yrs	0.00%	-3.66%	-12.80%	-23.40%	0.00%	0.35%	2.62%	-1.64%
	65–69 yrs	0.00%	-3.41%	-13.44%	-23.69%	0.00%	2.13%	2.53%	-1.10%
	70–74 yrs	0.00%	-4.78%	-14.51%	-26.28%	0.00%	-0.26%	2.01%	-3.16%
	75–79 yrs	0.00%	-2.48%	-15.89%	-27.29%	0.00%	0.03%	1.60%	-2.99%
	80+ yrs	0.00%	-5.47%	-16.98%	-33.37%	0.00%	-0.58%	0.70%	-8.18%

Appendix 3 (continued): Data for Figure 2–percentage difference between 2018 Census Usually Resident Population (Cen), 2018 Health Service User (HSU) and 2018 Primary Health Organisation (PHO) datasets compared to 2018 Estimated Resident Population (ERP) for Māori and non-Māori by age group and gender (as a percent of the difference between Census, HSU and PHO datasets compared to ERP). Calculated from data in Appendix 2.

		Māori				non-Māori			
		ERP	**Cen**	**HSU**	**PHO**	**ERP**	**Cen**	**HSU**	**PHO**
Females	All ages	0.00%	-4.10%	-5.73%	-16.94%	0.00%	-1.20%	1.45%	-3.76%
	0–4 yrs	0.00%	-2.76%	0.08%	-6.92%	0.00%	-2.41%	6.94%	-3.00%
	5–9 yrs	0.00%	-1.40%	-8.00%	-11.39%	0.00%	-1.39%	7.91%	3.23%
	10–14 yrs	0.00%	-2.95%	-10.42%	-16.21%	0.00%	-1.51%	5.87%	-0.06%
	15–19 yrs	0.00%	-5.18%	-10.52%	-17.53%	0.00%	-0.81%	-0.95%	-7.07%
	20–24 yrs	0.00%	-6.51%	-3.81%	-21.79%	0.00%	-1.64%	-1.69%	-9.07%
	25–29 yrs	0.00%	-6.82%	-3.14%	-19.91%	0.00%	-2.43%	-0.78%	-11.29%
	30–34 yrs	0.00%	-6.25%	-4.73%	-18.27%	0.00%	-3.03%	1.63%	-7.10%
	35–39 yrs	0.00%	-4.33%	-5.86%	-18.57%	0.00%	-2.54%	2.83%	-2.45%
	40–44 yrs	0.00%	-2.56%	-5.98%	-19.40%	0.00%	-1.14%	1.33%	-1.00%
	45–49 yrs	0.00%	-3.53%	-4.93%	-19.41%	0.00%	-1.62%	0.08%	-1.54%
	50–54 yrs	0.00%	-3.84%	-4.19%	-18.65%	0.00%	-1.42%	-0.48%	-2.27%
	55–59 yrs	0.00%	-5.44%	-3.83%	-18.39%	0.00%	-1.27%	-0.14%	-2.77%
	60–64 yrs	0.00%	-3.84%	-4.00%	-17.77%	0.00%	0.73%	1.00%	-2.16%
	65–69 yrs	0.00%	-2.91%	-5.63%	-18.31%	0.00%	2.28%	1.13%	-1.54%
	70–74 yrs	0.00%	-4.07%	-7.42%	-20.68%	0.00%	-0.21%	1.02%	-3.24%
	75–79 yrs	0.00%	-2.45%	-8.21%	-21.95%	0.00%	0.43%	0.66%	-2.57%
	80+ yrs	0.00%	-3.13%	-14.81%	-30.41%	0.00%	-0.02%	0.09%	-7.94%

Appendix 3 (continued): Data for Figure 2–percentage difference between 2018 Census Usually Resident Population (Cen), 2018 Health Service User (HSU) and 2018 Primary Health Organisation (PHO) datasets compared to 2018 Estimated Resident Population (ERP) for Māori and non-Māori by age group and gender (as a percent of the difference between Census, HSU and PHO datasets compared to ERP). Calculated from data in Appendix 2.

		Māori				non-Māori			
		ERP	**Cen**	**HSU**	**PHO**	**ERP**	**Cen**	**HSU**	**PHO**
All genders	All ages	0.00%	-4.81%	-7.95%	-19.50%	0.00%	-1.66%	0.33%	-5.78%
	0–4 yrs	0.00%	-3.30%	0.13%	-6.98%	0.00%	-2.53%	6.98%	-3.09%
	5–9 yrs	0.00%	-1.29%	-7.35%	-10.63%	0.00%	-1.34%	7.66%	3.02%
	10–14 yrs	0.00%	-3.30%	-10.15%	-16.40%	0.00%	-1.69%	5.93%	-0.09%
	15–19 yrs	0.00%	-6.00%	-12.38%	-20.17%	0.00%	-1.86%	-2.36%	-8.53%
	20–24 yrs	0.00%	-8.06%	-7.60%	-27.10%	0.00%	-2.71%	-6.45%	-14.80%
	25–29 yrs	0.00%	-8.59%	-6.95%	-25.96%	0.00%	-3.55%	-6.12%	-17.64%
	30–34 yrs	0.00%	-7.26%	-7.47%	-23.89%	0.00%	-3.50%	-2.02%	-12.18%
	35–39 yrs	0.00%	-5.06%	-8.46%	-22.84%	0.00%	-2.97%	0.26%	-6.85%
	40–44 yrs	0.00%	-4.15%	-9.37%	-23.03%	0.00%	-1.79%	0.04%	-4.26%
	45–49 yrs	0.00%	-4.59%	-9.19%	-23.00%	0.00%	-2.27%	-0.41%	-3.76%
	50–54 yrs	0.00%	-4.28%	-8.66%	-21.90%	0.00%	-1.76%	-0.30%	-3.33%
	55–59 yrs	0.00%	-5.61%	-7.64%	-21.10%	0.00%	-1.52%	0.77%	-2.71%
	60–64 yrs	0.00%	-3.75%	-8.16%	-20.44%	0.00%	0.54%	1.80%	-1.89%
	65–69 yrs	0.00%	-3.15%	-9.35%	-20.87%	0.00%	2.20%	1.82%	-1.32%
	70–74 yrs	0.00%	-4.40%	-10.73%	-23.30%	0.00%	-0.24%	1.51%	-3.19%
	75–79 yrs	0.00%	-2.46%	-11.73%	-24.40%	0.00%	0.25%	1.12%	-2.74%
	80+ yrs	0.00%	-3.83%	-15.54%	-31.56%	0.00%	-0.27%	0.34%	-8.02%

Appendix 4a: Individually linked data for Māori on 2018 Census (self-completed ethnicity only) and their ethnicity grouping (Māori or non-Māori) on NHI, by gender and age group, for the PHO population in 2018.

Gender	Age (years)	Māori on census but non-Māori on NHI	Māori on census and NHI	Total Māori on Census	Number with missing NHI ethnicity data	Percentage of Māori in census identified as Māori in NHI (proportion matched)	Percentage of Māori in census, categorised as non-Māori in NHI	NET under-count*
All genders	All ages	104,952	386,976	491,928	12,297	78.7%	21.3%	15.7%
	0–4	4,749	43,575	48,324	12	90.2%	9.8%	2.4%
	5–9	10,881	46,308	57,189	27	81.0%	19.0%	13.5%
	10–14	10,974	42,471	53,445	54	79.5%	20.5%	15.0%
	15–19	9,501	34,965	44,466	498	78.6%	21.4%	15.6%
	20–24	10,263	24,972	35,235	2,754	70.9%	29.1%	23.4%
	25–29	8,301	24,399	32,700	1,716	74.6%	25.4%	19.6%
	30–34	6,441	21,837	28,278	969	77.2%	22.8%	17.8%
	35–39	6,408	20,613	27,021	945	76.3%	23.7%	19.0%
	40–44	6,783	21,117	27,900	966	75.7%	24.3%	19.7%
	45–49	6,969	22,989	29,958	1,029	76.7%	23.3%	18.6%
	50–54	6,006	21,120	27,126	888	77.9%	22.1%	17.1%
	55–59	5,577	20,502	26,079	780	78.6%	21.4%	16.1%
	60–64	4,110	15,363	19,473	624	78.9%	21.1%	15.6%
	65–69	3,165	11,733	14,898	447	78.8%	21.2%	15.2%
	70–74	2,112	7,224	9,336	288	77.4%	22.6%	15.0%
	75–79	1,398	4,452	5,850	171	76.1%	23.9%	16.9%
	80+	1,335	3,327	4,662	147	71.4%	28.6%	19.4%

Appendix 4a (continued): Individually linked data for Māori on 2018 Census (self-completed ethnicity only) and their ethnicity grouping (Māori or non-Māori) on NHI, by gender and age group, for the PHO population in 2018.

Gender	Age (years)	Māori on census but non-Māori on NHI	Māori on census and NHI	Total Māori on Census	Number with missing NHI ethnicity data	Percentage of Māori in census identified as Māori in NHI (proportion matched)	Percentage of Māori in census, categorised as non-Māori in NHI	NET under-count*
Female	All ages	52,788	209,295	262,083	7,287	79.9%	20.1%	14.7%
	0–4	2,355	21,207	23,562	6	90.0%	10.0%	2.7%
	5–9	5,490	22,359	27,849	12	80.3%	19.7%	14.3%
	10–14	5,409	20,640	26,049	27	79.2%	20.8%	15.2%
	15–19	4,719	17,697	22,416	246	78.9%	21.1%	15.4%
	20–24	5,481	13,986	19,467	1,587	71.8%	28.2%	22.7%
	25–29	4,530	14,430	18,960	1,080	76.1%	23.9%	18.5%
	30–34	3,342	13,374	16,716	603	80.0%	20.0%	15.6%
	35–39	3,333	12,258	15,591	582	78.6%	21.4%	17.1%
	40–44	3,603	12,354	15,957	630	77.4%	22.6%	18.4%
	45–49	3,459	13,410	16,869	660	79.5%	20.5%	16.3%
	50–54	2,859	12,144	15,003	528	80.9%	19.1%	14.4%
	55–59	2,529	11,733	14,262	405	82.3%	17.7%	12.6%
	60–64	1,815	8,706	10,521	345	82.7%	17.3%	12.0%
	65–69	1,398	6,522	7,920	246	82.3%	17.7%	11.6%
	70–74	984	3,990	4,974	147	80.2%	19.8%	11.9%
	75–79	672	2,436	3,108	90	78.4%	21.6%	14.2%
	80+	807	2,055	2,862	93	71.8%	28.2%	19.2%

Appendix 4a (continued): Individually linked data for Māori on 2018 Census (self-completed ethnicity only) and their ethnicity grouping (Māori or non-Māori) on NHI, by gender and age group, for the PHO population in 2018.

Gender	Age (years)	Māori on census but non-Māori on NHI	Māori on census and NHI	Total Māori on Census	Number with missing NHI ethnicity data	Percentage of Māori in census identified as Māori in NHI (proportion matched)	Percentage of Māori in census, categorised as non-Māori in NHI	NET under-count*
Male	All ages	52,086	177,642	229,728	4,956	77.3%	22.7%	16.7%
	0–4	2,391	22,368	24,759	3	90.3%	9.7%	2.1%
	5–9	5,394	23,946	29,340	18	81.6%	18.4%	12.7%
	10–14	5,562	21,822	27,384	27	79.7%	20.3%	14.8%
	15–19	4,779	17,262	22,041	249	78.3%	21.7%	15.8%
	20–24	4,758	10,980	15,738	1143	69.8%	30.2%	24.1%
	25–29	3,756	9,966	13,722	627	72.6%	27.4%	21.0%
	30–34	3,087	8,466	11,553	357	73.3%	26.7%	20.9%
	35–39	3,069	8,349	11,418	360	73.1%	26.9%	21.5%
	40–44	3,168	8,766	11,934	327	73.5%	26.5%	21.3%
	45–49	3,501	9,579	13,080	363	73.2%	26.8%	21.4%
	50–54	3,144	8,979	12,123	360	74.1%	25.9%	20.5%
	55–59	3,039	8,772	11,811	366	74.3%	25.7%	20.2%
	60–64	2,283	6,660	8,943	270	74.5%	25.5%	19.8%
	65–69	1,767	5,208	6,975	201	74.7%	25.3%	19.3%
	70–74	1,125	3,237	4,362	138	74.2%	25.8%	18.5%
	75–79	723	2,016	2,739	81	73.6%	26.4%	20.0%
	80+	531	1,272	1,803	63	70.5%	29.5%	19.8%

Note: *Data from Appendix 4b i.e., non-Māori on the Census but Māori on the NHI, is used in the calculation of NET-under-count.

Appendix 4b: Individually linked data for non-Māori on 2018 census (self-completed ethnicity only) and their ethnicity grouping (Māori or non-Māori) on NHI, by gender and age group, for the PHO population in 2018.

Gender	Age (years)	Non-Māori on census and NHI	Non-Māori on census but Māori on NHI	Total non-Māori on Census	Number with missing NHI ethnicity data	Percentage of non-Māori in census identified as non-Māori in NHI (proportion matched)	Percentage of non-Māori in census, categorised as Māori in NHI
All genders	All ages	2,978,991	27,870	3,006,861	62,946	99.1%	0.9%
	0–4	162,075	3,591	165,666	33	97.8%	2.2%
	5–9	186,813	3,168	189,981	87	98.3%	1.7%
	10–14	180,366	2,940	183,306	225	98.4%	1.6%
	15–19	169,521	2,580	172,101	1,308	98.5%	1.5%
	20–24	151,947	2,028	153,975	7,059	98.7%	1.3%
	25–29	164,262	1,896	166,158	5,421	98.9%	1.1%
	30–34	181,002	1,413	182,415	3,408	99.2%	0.8%
	35–39	187,431	1,278	188,709	3,408	99.3%	0.7%
	40–44	192,267	1,299	193,566	4,758	99.3%	0.7%
	45–49	218,850	1,407	220,257	6,279	99.4%	0.6%
	50–54	214,122	1,365	215,487	5,709	99.4%	0.6%
	55–59	217,872	1,377	219,249	5,565	99.4%	0.6%
	60–64	195,201	1,068	196,269	4,965	99.5%	0.5%
	65–69	177,339	906	178,245	4,641	99.5%	0.5%
	70–74	146,805	711	147,516	3,891	99.5%	0.5%
	75–79	105,306	408	105,714	2,814	99.6%	0.4%
	80+	127,818	432	128,250	3,384	99.7%	0.3%

Appendix 4b (continued): Individually linked data for non-Māori on 2018 census (self-completed ethnicity only) and their ethnicity grouping (Māori or non-Māori) on NHI, by gender and age group, for the PHO population in 2018.

Gender	Age (years)	Non-Māori on census and NHI	Non-Māori on census but Māori on NHI	Total non-Māori on Census	Number with missing NHI ethnicity data	Percentage of non-Māori in census identified as non-Māori in NHI (proportion matched)	Percentage of non-Māori in census, categorised as Māori in NHI
Female	All ages	1,557,147	14,160	1,571,307	36,099	99.1%	0.9%
	0–4	78,885	1,725	80,610	9	97.9%	2.1%
	5–9	90,915	1,506	92,421	39	98.4%	1.6%
	10–14	88,305	1,440	89,745	111	98.4%	1.6%
	15–19	84,177	1,275	85,452	624	98.5%	1.5%
	20–24	79,734	1,059	80,793	3,819	98.7%	1.3%
	25–29	89,781	1,023	90,804	3,108	98.9%	1.1%
	30–34	99,615	741	100,356	1,938	99.3%	0.7%
	35–39	102,003	660	102,663	2,004	99.4%	0.6%
	40–44	104,280	672	104,952	3,183	99.4%	0.6%
	45–49	117,813	711	118,524	4,233	99.4%	0.6%
	50–54	113,082	702	113,784	3,453	99.4%	0.6%
	55–59	113,502	726	114,228	2,997	99.4%	0.6%
	60–64	100,710	555	101,265	2,577	99.5%	0.5%
	65–69	90,276	483	90,759	2,451	99.5%	0.5%
	70–74	74,928	393	75,321	2,034	99.5%	0.5%
	75–79	55,317	231	55,548	1,509	99.6%	0.4%
	80+	73,815	258	74,073	1,998	99.7%	0.3%

Appendix 4b (continued): Individually linked data for non-Māori on 2018 census (self-completed ethnicity only) and their ethnicity grouping (Māori or non-Māori) on NHI, by gender and age group, for the PHO population in 2018.

Gender	Age (years)	Non-Māori on census and NHI	Non-Māori on census but Māori on NHI	Total non-Māori on Census	Number with missing NHI ethnicity data	Percentage of non-Māori in census identified as non-Māori in NHI (proportion matched)	Percentage of non-Māori in census, categorised as Māori in NHI
Male	All ages	1,421,331	13,707	1,435,038	26,664	99.0%	1.0%
	0–4	83,172	1,866	85,038	18	97.8%	2.2%
	5–9	95,880	1,665	97,545	42	98.3%	1.7%
	10–14	92,019	1,500	93,519	111	98.4%	1.6%
	15–19	85,284	1,305	86,589	681	98.5%	1.5%
	20–24	72,102	972	73,074	3,177	98.7%	1.3%
	25–29	74,433	876	75,309	2,295	98.8%	1.2%
	30–34	81,354	672	82,026	1,461	99.2%	0.8%
	35–39	85,401	618	86,019	1,395	99.3%	0.7%
	40–44	87,963	627	88,590	1,563	99.3%	0.7%
	45–49	101,010	696	101,706	2,031	99.3%	0.7%
	50–54	101,016	663	101,679	2,241	99.3%	0.7%
	55–59	104,340	654	104,994	2,553	99.4%	0.6%
	60–64	94,470	510	94,980	2,379	99.5%	0.5%
	65–69	87,039	423	87,462	2,178	99.5%	0.5%
	70–74	71,868	318	72,186	1,851	99.6%	0.4%
	75–79	49,980	174	50,154	1,302	99.7%	0.3%
	80+	53,997	174	54,171	1,383	99.7%	0.3%

Mental health inequities for Māori youth: a population-level study of mental health service data

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ABSTRACT

AIM: To examine specialist mental health service, hospital discharge, and pharmaceutical dispensing data for emotional conditions (anxiety, depression), substance use, and self-harm for Māori compared to non-Māori/non-Pasifika (NMNP) youth.

METHODS: A novel population-level case identification method using New Zealand's Integrated Data Infrastructure for 232,845 Māori and 627,891 NMNP aged 10–24 years. Descriptive statistics on mental health conditions were generated and stratified by Māori/NMNP. Unadjusted and adjusted risk ratios (RRs) of mental health conditions were generated using generalised linear regression.

RESULTS: Māori were less likely to be identified for anxiety (ARR=0.88; 95% CI 0.85–0.90) or depression (ARR=0.92; 95% CI 0.90–0.95) than NMNP. They were more likely to be identified for substance problems (ARR)=2.66; 95% CI 2.60–2.71) and self-harm (ARR=1.56; 95% CI 1.50–1.63). Māori living in high deprivation areas were significantly more likely to be identified for substance problems, but less likely for emotional conditions, than Māori in least deprived areas.

CONCLUSION: Despite known high levels of mental health concerns for rangatahi Māori, administrative data suggests significant under-reporting, assessment, and treatment of emotional conditions relative to NMNP. These differences were exacerbated by deprivation. Māori were more likely to be referred to services for externalised symptoms of distress (substance use and self-harm).

Youth mental health distress, disorder and addictions are serious issues in Aotearoa New Zealand. Nearly a quarter (23%) of high school students report depressive symptoms, up from 13% in 2012.[1] Young adults aged 15–29 years have the highest rates of serious injuries from intentional self-harm (27.8 per 100,000 people).[2] Substance use is common, with 22% of high school students reporting binge drinking in the last four weeks, although these reported rates have been declining over the last 20 years.[3] Longitudinal studies in Aotearoa show that by age 25 years, 12.5% have met the DSM-IV criteria for cannabis dependence, and 3.6% for other drug dependences.[4] These patterns are broadly consistent with patterns in other developed countries.[5]

Rangatahi Māori—Māori youth—experience higher rates of mental health distress and addictions compared to young Pākehā—New Zealanders of European descent.[6] There are growing numbers of rangatahi Māori who report depressive symptoms (28% in 2019 vs 14% in 2012).[1] Māori are more likely to be hospitalised for intentional self-harm[7] and more likely to use substances than non-Māori.[3]

Despite higher reported rates of mental health distress, there are treatment inequities for Māori compared to Pākehā.[8,9] Lee and colleagues found that Māori adults are more likely to report "psychological distress" and be at higher risk of developing anxiety and depression, but Pākehā are more likely to report a clinical diagnosis of depression or anxiety.[9] Earlier research from Te Rau Hinengaro using diagnostic assessment of adults aged 16 years and over found that Māori had higher prevalence of anxiety, mood, major depressive, and substance use disorders compared to non-Māori/non-Pasifika (NMNP).[10] However, except for substance use disorders, these differences did not remain significant after adjustment for confounders including education and household income. Māori compared to NMNP were; however, less likely to visit services for mental health reasons. Māori youth are less likely to receive medications (e.g., antidepressants) compared with non-Māori youth.[11] Māori are more likely, however, to be admitted to hospital, readmitted after discharge, secluded and treated under the compulsory assessment and treatment protocols and in forensic services.[6] Overall, previous findings suggest that Māori are not able to access the

services that they need to be properly assessed and diagnosed. Multiple and complex determinants of mental health include poverty, discrimination and social isolation.[12] Inequities for Māori also result from the ongoing impacts of colonisation, racism, barriers to accessing services, and a lack of culturally appropriate services.[12]

Administrative data collections hold mental health information including specialist mental health and addiction services in inpatient and community settings, hospital discharge, and pharmaceutical data.[13] These datasets cannot be used to estimate population prevalence rates of all mental health conditions because they do not capture young people who report mild-to-moderate mental distress and who may be managed in primary care. The datasets, however, do contain data on whether a person had contact with a specialist mental health service or were hospitalised and were coded as having a mental health condition, or prescribed pharmaceuticals.[13] Moreover, the datasets contain whole-of-population data with large sample sizes to enable enough statistical power to undertake analyses focused on the Māori population.

The aim of the present study was to examine population-level administrative data on specialist mental health services, hospitalisations, and pharmaceutical dispensing for rangatahi Māori. We examined differences in specialist mental health service use, hospital discharges, and pharmaceutical dispensing for: anxiety, depression, emotional conditions and any emotional conditions (see descriptions in Methods section), substance use, and self-harm between rangatahi Māori and NMNP youth aged between 10–24 years of age. A companion paper by Ruhe et al. (in preparation) examines differences between Pasifika and NMNP youth.[14]

Methods

Study design

This was a national cross-sectional study using data sourced from the Integrated Data Infrastructure (IDI). The IDI is a large, population-level database containing administrative and survey data, probabilistically linked at the individual level by Statistics New Zealand (Stats NZ). Previously described in detail,[15] the IDI contains data from over 60 different data sources including health, data on people and communities, and population (e.g., Census). Strict protocols and approval processes are in place to both access IDI data and release results.

The University of Otago Human Research Ethics Committee reviewed and approved the present study as a "Minimal Risk Health Research – Audit and Audit related studies" proposal (Reference: HD17/004). Clearance for this study and access to data were also approved by Stats NZ.

Participants

The participants were a national cohort of young people (10–24 years), alive and living in Aotearoa, as of 30 June 2018. This cohort was established using an existing IDI-based method for determining the estimated resident population (ERP).[16] Individuals were included in the ERP if they had used key services in Aotearoa (e.g., health) over the preceding two years.

Measures

Mental health

Mental health measures were generated using a novel IDI-based case identification method developed by Bowden et al.[13] The method uses data from four Ministry of Health datasets:

- the programme for the integration of mental health data (PRIMHD)—specialist mental health service use data;
- the national minimum dataset (NMDS)—hospital discharge data (diagnosis on admission);
- the pharmaceutical collection (Pharms)—publicly subsidised medication dispensing data from community pharmacies;
- disability support services needs assessment data (Socrates).

The method was employed to identify anxiety, depression, emotional conditions (a composite group of indeterminant anxiety or depression based primarily on indications from pharmaceutical dispensing that either anxiety or depression can be identified but not one specifically), and any emotional conditions (combining the three aforementioned groups into one single indicator); substance (addiction and abuse) problems; and self-harm. A complete list of all the diagnosis and pharmaceutical dispensing codes used to indicate mental health conditions can be found in Bowden et al.[13] For each mental health condition, an indication was made if a young person appeared in at least one dataset with a related diagnostic or dispensing code within a five-year period between 1 July 2013 until 30 June 2018. PRIMHD data are

only available up to 30 June 2018. We chose a five-year window vs a shorter period of time in order to obtain full coverage of all datasets used.

Ethnicity

Ethnicity information was drawn from the IDI personal details dataset using the total concept approach that permits individuals to identify with multiple ethnic groups. We classified the population as Māori and Pasifika utilising the New Zealand Standard Classification 2005 V2.0.0. All other participants were classified as NMNP. For the present study only data for Māori and NMNP were analysed.

Socio-demographics

We were restricted to using current statistical standards for sex, which are female/male. Changes to the statistical standard for sex and gender identity that address issues including limited inclusiveness of intersex and transgender populations are likely in the future based on national Stats NZ consultation undertaken in 2021. Age (in years) were grouped to align with mental health case identification method: 10–14, 15–19, and 20–24 years. Area-level deprivation (NZDep2018) and urban/rural profile of residence, defined by the Urban Rural Indicator 2018, were derived from address notification data and the meshblock where the individual resides.[17,18] NZDep is a socio-economic measure of deprivation, defined at the meshblock level. NZDep scores were collapsed into quintiles, 1 representing the least deprived and 5, the most. Urban/rural profile of residence was collapsed into a 5-level categorical variable: major urban areas (populations of 100,000 or more); large urban areas (30,000–99,999); medium urban areas (10,000–29,999); small urban areas (1,000–9,999); and rural areas (<1,000). Time varying measures (age, NZDep2018, and urban/rural) were determined as of 30 June 2018.

Procedures and statistical analyses

Data were accessed from the June 2020 refresh of the IDI, extracted using SAS 7.1, and analysed using Stata MP version 15. All Stats NZ confidentiality requirements were adhered to including rounding to base three, and suppression of counts less than six. Reporting of studies conducted using Observational Routinely collected health Data (RECORD) guidelines were used to inform the reporting of analyses.[19] The data analyses and reporting were conducted in line with the Ngā Tikanga Paihere framework for IDI use.[20] The study was led by a Māori health researcher.

Descriptive statistics on the number of mental health conditions identified by data source and by sociodemographic subgroup were generated and stratified by Māori/NMNP. Unadjusted and adjusted risk ratios (RR) and associated 95% confidence intervals (CI) of mental health conditions were generated using generalised linear regression with a log link and binomial distribution. In adjusted models, sex, age, deprivation, and rurality were controlled for. Two-tailed tests (α=0.05) defined significance. In addition, an adjusted regression included a rate ratio for an interaction between ethnicity and deprivation. That interaction is displayed in Figure 1.

Results

The ERP of 10–24-year-olds for the 2017/18 fiscal year included 232,845 Māori and 627,891 NMNP (see Table 1).

Table 2 shows the number of mental health conditions identified by data source for Māori and NMNP aged 10–24 years. Overall, Pharms was the main source of identifications for emotional conditions, PRIMHD for substance problems, and NMDS the only source for self-harm.

Table 3 shows the population-based rate of mental health conditions identified by service use and pharmaceutical dispensing between 1 July 2013 until 30 June 2018. Emotional conditions and substance problems were the most commonly identified for rangatahi Māori, each accounting for a five-year prevalence of more than 8% of the population, respectively. The identification of two or more conditions for Māori was 2.8% compared to 1.9% of NMNP young people (see Table 3), while identification of three conditions was 0.5% of Māori compared to 0.3% for their NMNP peers.

Table 4 shows unadjusted and adjusted ethnicity risk ratios. Rangatahi Māori compared to their NMNP peers were significantly less likely to be identified for any emotional conditions, anxiety, depression, or emotional conditions (indeterminant anxiety or depression) as documented in the IDI dataset. However, Māori were more likely than NMNP to be identified for substance problems, self-harm, two or more conditions, and three conditions as documented in the IDI dataset.

Figure 1 shows differing patterns of mental health conditions for Māori and for NMNP by level of deprivation.

Rangatahi Māori who lived in the most socio-

Table 1: Characteristics of the participants: Estimated Residential Population (ERP) 2017/18 for Māori and non-Māori/non-Pasifika (NMNP) aged 10–24 years.

		Māori		NMNP	
		N	%	N	%
ERP	**Total**	**232,845**	**100.0**	**627,891**	**100.0**
Sex	Female	113,802	48.9	303,027	48.3
	Male	119,046	51.1	324,861	51.7
Age	10–14 years	84,888	36.5	195,702	31.2
	15–19 years	76,584	32.9	201,051	32.0
	20–24 years	71,373	30.7	231,138	36.8
Area-level deprivation	1 (least deprived)	20,169	8.7	143,484	22.9
	2	26,298	11.3	136,479	21.7
	3	34,950	15.0	132,702	21.1
	4	52,236	22.4	125,727	20.0
	5 (most deprived)	98,220	42.2	85,215	13.6
Urban/rural	Major urban (<100,000)	94,782	40.7	353,574	56.3
	Large urban (30,000–99,999)	47,832	20.5	75,180	12.0
	Medium urban (10,000–29,999)	18,813	8.1	49,008	7.8
	Small urban (1,000–9,999)	32,535	14.0	48,102	7.7
	Rural (<1000)	37,941	16.3	97,863	15.6

Table 2: Mental health conditions by administrative data source (1 July 2013–30 June 2018) for Māori (n=232,845) and non-Māori/non-Pasifika (NMNP) (n=627,891) aged 10–24 years.

Condition	PRIMHD[a]	NMDS[b]	Pharms[c]	Socrates[d]	Overall
Māori					
Any emotional[e]	5,334	2,487	17,340	90	19,788
Anxiety	2,829	1,560	3,765	81	6,906
Depression	2,967	1,419	3,081	18	6,111
Emotional conditions[f]	528	102	14,928	n/a	15,288
Substance problems	16,671	3,963	111	6	18,783
Self-harm	0	3,882	0	0	3,882
NMNP					
Any emotional	14,805	5,385	58,467	309	63,648
Anxiety	8,898	3,549	14,172	285	23,088
Depression	7,911	3,030	12,555	42	19,326
Emotional conditions	774	117	50,136	n/a	50,529
Substance problems	13,428	5,304	327	..S	17,121
Self-harm		6,465			6,462

[a]Programme for the integration of mental health data–specialist mental health service use data.
[b]National minimum dataset–hospital discharge data.
[c]Pharmaceutical collection–publicly subsidised medication dispensing.
[d]Disability support services needs assessment data.
[e]Combination of anxiety, depression, and emotional conditions[f] (a composite group of indeterminant anxiety or depression based primarily on indications from pharmaceutical dispensing that either anxiety or depression can be identified but not one specifically).

Table 3: Five-year population prevalence (1 July 2013–30 June 2018) of mental health conditions identified by specialist mental health service, hospitalisation, and pharmaceutical dispensing administrative data for Māori and non-Māori/non-Pasifika (NMNP) aged 10–24 years.

Condition	Māori				NMNP			
	N (%)				N (%)			
	Total	**10–14y**	**15–19y**	**20–24y**	**Total**	**10–14y**	**15–19y**	**20–24y**
	232,845	**84,888**	**76,584**	**71,373**	**627,891**	**195702**	**201,051**	**231,138**
Any emotional[a]	19,788 (8.5)	1,227 (1.4)	6,048 (7.9)	12,516 (17.5)	63,651 (10.1)	4,197 (2.1)	18,306 (9.1)	41,145 (17.8)
Anxiety	6,906 (3.0)	777 (0.9)	2,283 (3.0)	3,849 (5.4)	23,091 (3.7)	2,808 (1.4)	7,134 (3.5)	13,146 (5.7)
Depression	6,111 (2.6)	147 (0.2)	1,797 (2.3)	4,167 (5.8)	19,329 (3.1)	333 (0.2)	4,821 (2.4)	14,175 (6.1)
Emotional conditions[b]	15,288 (6.6)	600 (0.7)	4,635 (6.1)	10,050 (14.1)	50,532 (8.0)	2,070 (1.1)	14,814 (7.4)	33,648 (14.6)
Substance problems	18,780 (8.1)	930 (1.1)	7,935 (10.4)	9,915 (13.9)	17,124 (2.7)	465 (0.2)	5,772 (2.9)	10,884 (4.7)
Self-harm	3,882 (1.7)	162 (0.2)	1,815 (2.4)	1,902 (2.7)	6,462 (1.0)	174 (0.1)	2,751 (1.4)	3,537 (1.5)
Two or more conditionsc	6,627 (2.8)	117 (0.1)	2,352 (3.1)	4,158 (5.8)	11,619 (1.9)	159 (0.1)	4,005 (2.0)	7,455 (3.2)
All three conditionsc	1,083 (0.5)	9 (0.0)	435 (0.6)	636 (0.9)	1737 (0.3)	12 (0.0)	555 (0.3)	1,170 (0.5)

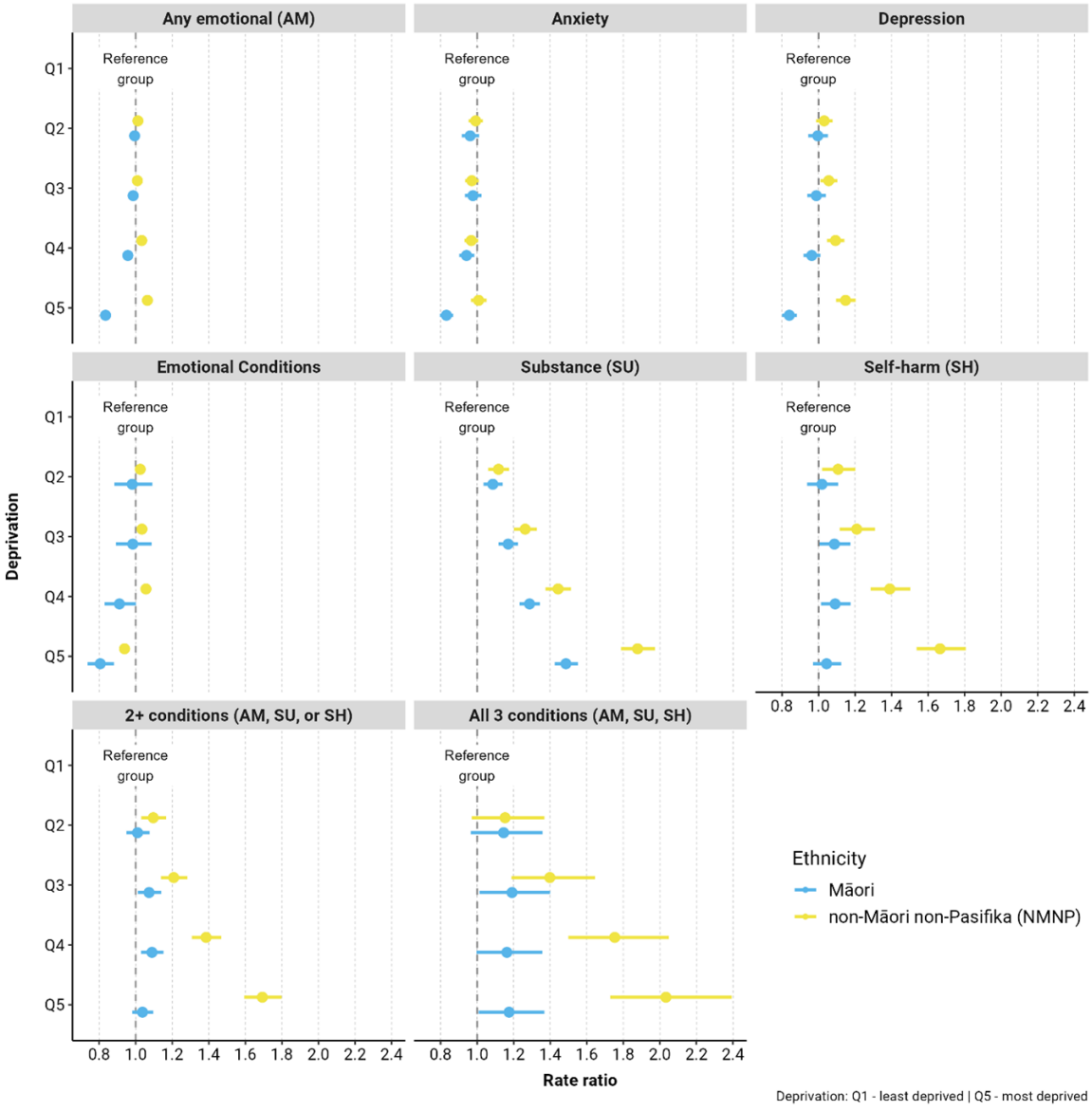
[a]Combination of anxiety, depression, and emotional conditions[b] (a composite group of indeterminant anxiety or depression based primarily on indications from pharmaceutical dispensing that either anxiety or depression can be identified but not one specifically).
[c]Any emotional conditions, substance problems, self-harm.

Table 4: Risk ratios for Māori compared to non-Māori/non-Pasifika (NMNP) aged 10–24 years by mental health condition.

	Unadjusted RR (95% CI)	**Adjusted[a] RR (95% CI)**
Any emotional[b]	0.838 (0.826, 0.851)	0.909 (0.895, 0.924)
Anxiety	0.807 (0.786, 0.828)	0.875 (0.850, 0.899)
Depression	0.852 (0.829, 0.877)	0.924 (0.897, 0.952)
Emotional conditions[c]	0.816 (0.802, 0.830)	0.887 (0.871, 0.903)
Substance	2.958 (2.899, 3.018)	2.656 (2.600, 2.714)
Self-harm	1.619 (1.557, 1.685)	1.562 (1.497, 1.630)
Two or more conditions[d]	1.539 (1.493, 1.585)	1.519 (1.471, 1.568)
All three conditions[d]	1.683 (1.560, 1.816)	1.629 (1.501, 1.767)

[a]Analyses controlled for sex, age, area-level deprivation, and rurality.
[b]Combination of anxiety, depression, and emotional conditions[c] (a composite group of indeterminant anxiety or depression based primarily on indications from pharmaceutical dispensing that either anxiety or depression can be identified but not one specifically).
[d]Any emotional conditions, substance problems, self-harm.

Figure 1: Rate ratios of mental health conditions for Māori and non-Māori/non-Pasifika (NMNP) aged 10–24 years by NZ Deprivation Index (NZDep2018).



AM=Combination of anxiety, depression, and emotional conditions (a composite group of indeterminant anxiety or depression based primarily on indications from pharmaceutical dispensing that either anxiety or depression can be identified but not one specifically).

economically deprived areas were significantly less likely to be identified as having any emotional conditions (ARR=0.84; 95% CI 0.81–0.86), anxiety (ARR=0.83; 95% CI 0.80, 0.87), depression (ARR=0.84; 95% CI 0.80, 0.88), or emotional conditions (indeterminant anxiety or depression) (ARR=0.81; 95% CI 0.74, 0.88) as documented in the IDI, than Māori living in the least deprived areas (Figure 1). Māori in the most deprived areas were more likely to be identified with substance problems (ARR=1.49; 95% CI 1.42–1.55) and all three conditions (ARR=1.18; 95% CI 1.01–1.12) than Māori living in the least deprived areas. In contrast, NMNP living in the most deprived areas were significantly more likely to be identified as having any emotional conditions (ARR=1.07; 95% CI 1.04–1.09), depression (ARR=1.15; 95% CI 1.10–1.20), self-harm (ARR=1.67; 95% CI 1.54–1.81), two or more conditions (ARR=1.70; 95% CI 1.59–1.80), and all three conditions (ARR=2.03; CI 1.73–2.39).

Discussion

Despite known high levels of mental health concerns for rangatahi Maori, administrative data suggests significant under-reporting, assessment, and treatment of emotional conditions relative to NMNP. However, Māori were more likely to be referred to services for externalised symptoms of distress like substance use and self-harm. We found that rangatahi Māori were 63% more likely than their NMNP peers to be receiving treatment for all three conditions—any emotional conditions, substance problems, and self-harm. These findings suggest that rangatahi Māori as compared to NMNP youth are less likely to be identified at earlier stages of distress until distress symptoms become more severe with explicit markers (e.g., self-harm injuries).

Rangatahi Māori were 12% and 8% less likely than NMNP youth to be identified through specialist mental health service use, hospitalisation, and pharmaceutical dispensing data for anxiety and depression, respectively. Our results align with findings showing unmet need for Māori.[9,21] Emotional conditions (anxiety, depression, and a composite group of indeterminant anxiety or depression) in this study were primarily identified through pharmaceutical dispensing data suggesting lower rates of anti-depressant/anti-anxiety dispensing for rangatahi Māori compared to their peers, as shown in previous studies.[11]

Rangatahi Māori were 56% more likely to be being admitted to hospital for self-harm than their NMNP peers. Rangatahi Māori were also significantly more likely than NMNP youth to have contact with a specialist mental health service or be admitted to hospital for substance problems. Moreover, there was a 49% higher likelihood of substance problems for rangatahi Māori living in the most deprived areas compared to Māori living in the least deprived. Alcohol misuse is associated with a range of conditions including depressive symptoms and self-harm.[22] A recent paper found that one quarter of all suicides between 2007–2020 involved acute alcohol use, with higher proportions at younger ages (28.7% for 15–24 year olds) and for Māori aged 15 years and older (32.3%).[23]

The determinants of mental health are multiple and complex. Social determinants include poverty, unemployment, unstable housing, and loss of community and communal spaces.[12] Underlying social determinant pathways are issues like exclusion through discrimination (e.g., racism, sexism, homophobia, transphobia, ableism), social isolation, trauma, adverse childhood experiences, and the stigma of having mental health issues.[12] We found that rangatahi Māori living in high deprivation areas vs those living in low deprivation areas were less likely to be identified as having any emotional condition or depression. Conversely, NMNP youth living in higher vs low deprivation areas were more likely to be identified as having any emotional condition or depression. Our analyses took into account differences in age, gender, rurality, and the population size in each deprivation quintile. We can only speculate as to other factors outside of the scope of this present study that may be affecting these comparative differences. Factors that may include differential barriers for rangatahi Māori to accessing services for emotional conditions or barriers within services including not receiving a diagnosis or treatment. Previous reports have noted that for Māori, colonisation, racism, "Western" models of health, barriers to accessing primary care, and a lack of culturally approriate services perpetuate inequities.[12] These interrelated social and economic factors will have direct and indirect impacts on mental health for rangatahi Māori with cumulative effects over their lifetimes. Further research is needed to understand the intersectionality between deprivation and ethnicity in relation to mental health inequities.

A lifecourse prevention and early intervention approach, addressing the structural determinants of health, with Māori-led services and models, are key solutions.

Māori are a young population, compared to non-Māori, with more than half aged 25 years or younger. Focusing on prevention and early intervention can reduce mental health distress before issues develop into diagnosable mental health conditions, thereby reducing the future burden of disease and related issues (e.g., poorer academic achievement, unemployment).[12] Whole-school approaches and universal skills-based programmes, starting in early childhood, can promote wellbeing, reduce stigma, and promote help-seeking.[24] There is growing evidence of the effectiveness of digital health interventions,[25,26] and youth focussed and friendly, low or no-cost, community-based integrated healthcare environments.[27]

Alcohol and drugs are serious public health concerns affecting communities, particularly higher deprivation areas. Broader environmental and community level interventions aimed at reducing alcohol availability include alcohol pricing, advertising bans, limiting alcohol outlet density, and higher minimum legal drinking age.[28] Addressing the structural determinants of the marginalised position of Māori in Aotearoa, and using a Te Tiriti o Waitangi framework with a commitment to equity by partnership with Māori to support effective policy, regulation, and service delivery of programmes appropriate for rangatahi Māori, are fundamental. More broadly, Aotearoa lacks a coordinated, integrated approach to social services to tackle the social and economic determinants of mental health.[12] The central Government is responsible for coordinating policy, practice, and investment to address social wellbeing at the environmental level including providing support for parents and whānau and increasing access to recreational areas and safer neighbourhoods.[6]

The strengths and limitations of the present study should be considered. Using IDI data enabled us to examine mental health conditions at a population level. There are few datasets that have the statistical power to produce robust findings for Māori. We were able to identify clinically relevant cases of mental health conditions and examine inequities by ethnicity and level of deprivation. We were unable to examine service use in primary care, and therefore our results do not reflect overall youth mental health need. The absence of primary care data in existing administrative datasets means limited data on treatment without medication (e.g., public or privately funded psychological therapy).[13] PRIMHD was only available up to 2018 and future research is needed to examine the long-term impacts of COVID-19 on the mental health of young people in Aotearoa.

Overall, we found inequities in the identification of rangatahi Māori with mental health conditions, particularly those living in high deprivation areas. Our findings suggest that rangatahi Māori are not able to access the services that they need to be properly assessed, diagnosed and/or prescribed anti-depressant/anti-anxiety medications, for emotional conditions (anxiety, depression). Conversely, they are more likely to be identified through contact with specialist mental health services and hospital admissions for substance problems and self-harm. These issues contribute to long-standing mental health inequities for rangatahi Māori.

Stats NZ disclaimer

These results are not official statistics. They have been created for research purposes from the Integrated Data Infrastructure (IDI) which is carefully managed by Stats NZ. For more information about the IDI please visit:

https://www.stats.govt.nz/integrated-data/.

COMPETING INTERESTS
Nil

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The past, present and future of liver cancer control for Māori

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ABSTRACT

Liver cancer is among the most commonly diagnosed and least-survivable cancers in New Zealand. There are stark disparities between the Indigenous Māori population in incidence of and mortality from liver cancer relative to non-Māori. In this review, we have summarised the key risk factors for liver cancer, and the key activities undertaken in New Zealand, over time, to control this disease, with a focus on how risk factors and interventions aimed at reducing them differentially impact Māori.

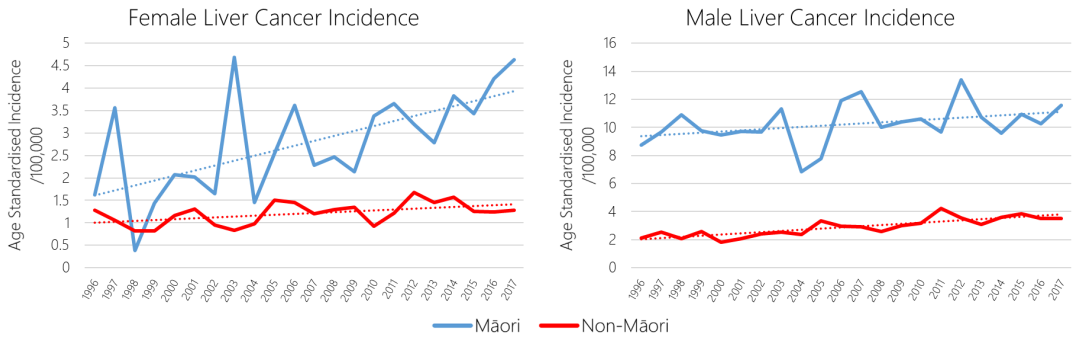
We have conducted a narrative literature review. The disproportionate burden of liver cancer experienced by Māori is primarily driven by disparities in viral exposure to hepatitis B and C between ethnic groups. Efforts to control hepatitis-associated liver cancer in New Zealand have lacked national coordination, further driving disparities in liver cancer survival between Māori and NZ Europeans.

A national primary care-based programme to detect and treat hepatitis B and C and to screen for liver cancer among high-risk patients, along with renewed effort to maximise hepatitis B vaccination rates, has the potential to substantially reduce the burden of hepatitis-associated liver cancer and address a significant health disparity between Māori and non-Māori.

Primary liver cancer (hereafter liver cancer) is the seventh most common cancer globally, with 906,000 cases occurring in 2020.[1] Liver cancer is also among the most deadly cancers, having the third highest cancer mortality rate globally (behind lung and breast cancer) and with 830,000 deaths in 2020.[1] This high mortality rate is driven by poor survival among those diagnosed with liver cancer, of whom approximately 40% will survive one year post diagnosis, and 20% will survive five years.[2] Liver cancer is curable if diagnosed early; curative treatments for early stage disease include liver resection, liver transplantation, and targeted ablation to small lesions. Unfortunately, most patients present with intermediate or advanced stage disease. Treatment for intermediate stage disease include therapies such as chemoembolisation or systemic therapies,[3] while patients who present with symptomatic advanced stage disease are generally only treated with supportive palliative care.[4] As such, the importance of early diagnosis cannot be overstated.

Liver cancer is one of the top 10 most common cancers among New Zealand's Indigenous Māori population, and one of the top 5 most common causes of Māori cancer death.[5,6] The most common causes of liver cancer are well known and preventable, and yet liver cancer incidence and mortality rates continue to increase over time for Māori (see Figure 1).[6] Māori remain substantially more likely to be diagnosed with liver cancer than non-Māori, and less likely to survive once diagnosed.[2,5]

Figure 1: Incidence of primary liver cancer in NZ for Māori and non-Māori between 1996–2017, by sex.[6]



How did we get here? What can we learn from the history of liver cancer control in New Zealand that might help to explain current liver cancer incidence and mortality rates for Māori? What is New Zealand's current approach to liver cancer control, and what future approaches might yield significant gains in this area for Māori, and therefore deserve our consideration? In this manuscript, we summarise relevant literature regarding liver cancer risk factors and prevention in New Zealand, with a focus on Māori. We then consider, where evidence exists, previous and current liver cancer prevention activities, and the extent to which these have focussed on Māori. Finally, we consider the future direction of liver cancer control for Māori in New Zealand and make some key recommendations for next steps.

Types of liver cancer

There are two primary types of liver cancer: hepatocellular carcinoma (HCC), in which a primary cancer develops within liver cancer cells (hepatocytes); and intrahepatic cholangiocarcinoma (ICC), which forms within cholangiocytes lining small intrahepatic bile ducts.[7] HCC is the most common of the two, accounting for 71% of liver cancer cases diagnosed in New Zealand.[8] While the aetiology of ICC is poorly understood, the key risk factors for HCC are well described—with the distribution of these risk factors intrinsically linked to the substantial disparity in liver cancer burden observed between Māori and non-Māori New Zealanders.[5]

There are four key categories of risk factors for HCC: viral hepatitis, alcohol-related liver disease, non-alcohol-related liver disease, and other risk factors (including aflatoxin). Chronic hepatitis B virus (Hep-B) and hepatitis C virus (Hep-C) infections are by far the most frequent cause of hepatocellular carcinoma (HCC), accounting for ~80% of HCC globally.[9] As such, the most important of the risk factors for HCC (by a substantial margin) in terms of both absolute disease burden and as a driver of inequity for Māori are the Hep-B and Hep-C viruses. For the purposes of brevity and focus, this review will focus on the prevention of hepatitis and viral hepatitis-associated HCC within New Zealand. We have presented an extended review of the additional risk factors, and their relevance to Māori, within Appendix 1.

The hepatitis B virus

Hep-B is a DNA virus that is the most prominent risk factor for the development of HCC, with approximately 55% of HCC cases worldwide occurring due to Hep-B chronic infection.[10] Those with Hep-B infection are 30–60 times more likely to develop HCC than those without Hep-B infection.[11–13]

Hep-B is one of the most common chronic infections globally, with 5% of the world's population infected (240–350 million people),[14] of which 35–87 million will die due to HCC.[15] Hep-B is not yet curable, but vaccination has successfully contributed to a reduction in the incidence of HCC,[16] although many Hep-B infected unvaccinated individuals (257 million globally in 2015) are still at risk for developing HCC.[16]

Two mechanisms of the causal pathway from Hep-B to HCC have been proposed. The first involves Hep-B inducing inflammation and cirrhosis-promoting carcinogenesis by up-regulating hepatocyte regeneration, DNA damage, and reactive oxygen species (ROS) production. The second mechanism involves Hep-B DNA incorporation into the host genome, thus affecting activation of proto-oncogenes, chromosomal instability, and transcription of pro-carcinogenic Hep-B genes.[17] It remains unclear as to which of these is the most dominant mechanism, by which chronic Hep-B infection leads to HCC.[18]

The hepatitis C virus

Hep-C, which is an RNA virus, is the second most prominent risk factor in HCC development—accounting for approximately 10–25% of all cases internationally. It is the main cause of HCC in Western countries.[16]

Chronic Hep-C infection is associated with a 20–30-fold risk of developing HCC.[19] However, due to effective treatment using direct-acting antiviral drugs, the risk of HCC attributed to Hep-C has significantly decreased.[20] The World Health Organization (WHO) launched a Global Health Sector Strategy of Viral Hepatitis 2016–2021 aimed at reducing new Hep-C infections by 90%, and at reducing deaths due to all-cause viral hepatitis by 65% by 2030;[21] however, the achievement of these goals will likely be impacted by the COVID-19 pandemic.[20]

The mechanism of Hep-C-associated HCC carcinogenesis occurs progressively over 20–30 years of chronic Hep-C infection.[22] The infection promotes a wide range of changes within the liver from lipid

accumulation within hepatocytes, impaired oxidative stress metabolism, and architectural changes associated with fibrogenesis which impacts liver function.[18] Over time, repeated cell cycles under the influence of oxidative stress induced by the virus and the host immune response leads to the accumulation of mutations in the hepatocytes, ultimately leading to the development of HCC.[23]

Prevention of viral hepatitis-associated HCC

Transmission of hepatitis B and C

Despite their preventable nature and some efforts to reduce the national burden of disease, Hep-B and Hep-C are endemic within New Zealand.[24–27] Hep-B is commonly spread through physical contact, perinatally from mother to child, and through contact with blood and other bodily fluids.[28] Similarly, Hep-C is primarily a blood borne virus which is largely transmitted parenterally through needles or other sharp instruments.[27,29] Those most at risk of contracting Hep-C are injecting drug users; people receiving blood; blood products or organs that are not screened—particularly prior to the introduction of robust screening protocols (see below); those in the prison population; and healthcare workers due to exposure to sharp instruments.[30] Screening of blood products prior to transfusion and a safe, sterile environment when using needles or other sharps are crucial to stop the transmission of Hep-B or Hep-C.[28,30]

Prevention of hepatitis B

Hepatitis B can be prevented at a population level through vaccination, with 90%+ effectiveness against chronic illness achievable via immunisation during childhood.[31] New Zealand first introduced neonatal vaccination against Hep-B in 1985, subsequently making it available for people of all ages in 1988, being one of the first countries to do so internationally.[25] There are still many New Zealanders who were born before the introduction of the neonatal vaccination programme who are unvaccinated and may already be infected, albeit unaware: as of 2018, approximately 3.2 million New Zealanders remained unvaccinated for Hep-B and from this, 93,609 are thought to have chronic Hep-B infection.[26] Additionally, Gane et al. found that 4% of children under the age of 15 were infected with chronic Hep-B.[32] Considering the effectiveness of the vaccine in delivering immunity from chronic Hep-B, it is probable that these children never received a vaccine at birth.[32]

There is also a high risk of transmission to infants from mothers who are carriers of Hep-B. To prevent transmission of Hep-B during labour, Hep-B immunoglobulin can be administered promptly within the first few hours post-delivery.[25] Antiviral treatment late in pregnancy is also recommended for women with a high viral load.[33] Furthermore, administration of the Hep-B vaccine within three days of life was shown to prevent transmission in 91–94% of cases.[25] Hep-B immunoglobulin can also be administered to anyone following exposure to bodily fluid of someone with Hep-B, to prevent infection.[34] Together, neonatal vaccination and Hep-B immunoglobulin for infants of Hep-B-positive mothers, as well as targeted Hep-B vaccination of high-risk individuals,[31] could eradicate Hep-B in New Zealand and reduce the burden of disease caused by Hep-B-associated HCC.[34]

Relevance to Māori

It is widely supported that the burden of Hep-B disease is disproportionate among Māori and Pacific peoples compared to NZ Europeans, and the high incidence of liver cancer among Māori is largely attributable to the incidence of Hep-B.[35–37] Māori also had the lowest Hep-B vaccination rates in 2018, with 333,906 Māori over the age of 30 remaining unvaccinated.[26] One of the major barriers to Māori being vaccinated is likely to be a mistrust in the healthcare system due to institutionalised racism and culturally unsafe service provision, with one in four Māori having experienced discrimination in the previous 12 months.[37] As demonstrated with COVID-19 vaccinations, Māori-led campaigns are more successful than taking a "one-size-fits-all" approach, which often results in poorer access to vaccination for Māori.[38–43]

Reducing social inequities continues to be an important, unmet area of concern.[26] Māori are over-represented in areas of higher deprivation, which is associated with increased transmission of infectious diseases.[26] Hep-B is transmitted horizontally in families, especially those living in overcrowded conditions, where just one person needs to be infected with Hep-B to spread it to other unvaccinated whānau. Horsfall et al. found that among 17,000 Hep-B patients, over 25% lived in areas of highest deprivation, and Māori and Pacific peoples had the highest median deprivation level.[26]

Prevention of hepatitis C

Unfortunately, there is no vaccine available for hepatitis C, emphasising the importance of

other preventative measures.[31] According to the New Zealand Ministry of Health's *"Action Plan on Hepatitis C Prevention"*, current preventative strategies are divided into primary and secondary categories.[27] Primary prevention strategies include: a) controlling injectable drug availability via border control and local and international police activities;[29,44] b) educating the population regarding issues such as needle exchange and the risk of spread of infectious diseases;[29] c) supporting safe injectable drug use by providing environments where injecting equipment can be used safely;[27,29] d) screening blood products to guarantee blood safety;[29,45] and e) workplace legislation to either directly or indirectly reduce the transmission of Hep-C in the workplace.[29] Secondary prevention strategies are closely linked to screening and surveillance, but also include activities such as research into risk factors, transmission and changes in disease incidence, along with providing further training and education to healthcare professionals.[29]

Relevance to Māori

Patients with a history of injecting drug use have an increased likelihood of being marginalised and stigmatised.[46] When this is combined with the high incidence of discrimination Māori also face in healthcare,[37] engaging with services such as the Needle Exchange Programme can be an arduous and difficult experience. Furthermore, patients with Hep-C-associated HCC are commonly the major income earners providing for large families in low decile areas.[48] As noted previously, Māori have a higher median deprivation level compared to non-Māori, making access to these prevention services, such as needle exchange centres and primary healthcare, increasingly difficult.[26] This is often due to a multitude of factors such as lack of transport, inability to get time off work, and childcare commitments.

Screening for hepatitis B

Since the 1980s, Hepatitis B Surface Antigen (HBsAg) has been included in antenatal and blood donor screening.[48] Initially, screening for Hep-B was done in an ad hoc way, which is not effective considering the large number of people who remained undiagnosed.[49] Estimates suggest about 50% of the approximately 100,000 people living with chronic Hep-B, and 40% of the 45,000 chronic Hep-C patients, still remain undiagnosed or have been lost to follow-up.[47] Because of concerns regarding the ramifications of undiagnosed and untreated hepatitis, in 1994 the New Zealand Hepatitis Foundation, a non-governmental organisation (NGO) based in Whakatāne campaigned for organised Hep-B screening to reduce the burden of chronic Hep-B infection, but this was not rolled out nationally at the time.[48]

In 1998, the Government funded a Hep-B screening programme, which set out to target Māori, Pacific and Asian communities in Northland and wider Auckland regions over the age of 15—those who were unlikely to be protected by the Hep-B vaccine.[32] The screening programme was executed by two different providers: 1) the Northern Region Hepatitis Consortium, which was made up of Auckland District Health Board staff, Ngāti Whātua (a tribal entity based in central west Auckland) and Māori and Pacific primary care and public health organisations; and 2) the Hepatitis Foundation. Both agencies were responsible for screening and follow-up, and the Northern Consortium also offered immunisation, counselling and surveillance services.[48] The South Island was not covered, with this approach rationalised at the time by relative differences in the Māori, Pacific and Asian populations between the North and South Islands.[48]

The Hepatitis Foundation led screening in the Northland and Auckland regions from July 1999 to 2002, and largely worked out of purpose-built screening centres and marae (traditional meeting houses). Additionally, from April 2000 to December 2002, the Northern Consortium was up and running—largely focussing on supporting practitioners along with Māori and Pacific Health providers to recruit individuals opportunistically through invitation, phone call, and local advertisement via radio, churches and marae.[48]

In terms of the screening test itself, a blood sample was taken after gaining informed consent from the participant and transported to a designated laboratory for testing. Each blood sample was analysed for HBsAg and those who were positive were further tested for e-antigen (HBeAg), along with alanine aminotransferase (ALT) and alpha-fetoprotein (AFP), which are markers of active hepatitis and HCC, respectively.[48]

This short-lived screening programme tested 177,000 people and found that 4,081 people (6%) were positive for HBsAg.[48,49] Later analysis suggested that had the programme continued and completed screening the total target population,

an estimated 80,000 people would have been identified as positive for HBsAg—with subsequent ramifications in terms of HCC surveillance and prevention.[24]

Importantly, 40% of patients diagnosed with advanced stage HCC associated with Hep-B are unaware of their Hep-B status at their initial presentation.[24] A pressing issue arising from this screening programme is ensuring that the remaining people with chronic Hep-B are receiving effective follow-up and surveillance. This has proven difficult for several reasons: largely because primary care providers struggle to access specialist services for referral of patients, and some patients are lost to follow-up or hard to trace.[48]

Relevance to Māori

When the Hep-B Screening Programme was up and running, only 27% of eligible Māori (aged over 15 years) were screened for Hep-B. Disappointingly, this was significantly below the goal of 70% coverage rate, despite engaging with Māori healthcare providers and marae.[35,48] Gane et al. suggests the poor recruitment of Māori could reflect perceptions on mass screening, which may stem from previous negative publicity around screening programmes.[32]

Surveillance of hepatitis B and C

HCC tumours are largely asymptomatic in the early stages, making them difficult to detect during this period without effective surveillance following a diagnosis of Hep-B or Hep-C.[50] When viral hepatitis-associated HCC is caught at an early stage, it is more likely to be treatable.[26,51] However, most cases of Hep-B or Hep-C-associated HCC in New Zealand are picked up at later stages where curative treatment is often not an option.[47] As such, monitoring high risk patients such as those with chronic liver disease from hepatitis is extremely important.

Ideally, all patients with chronic Hep-B infection should be recruited on to a monitoring programme (such as the programme run by the Hepatitis Foundation), to receive regular monitoring of serum ALT and Hep-B DNA. This will identify any potential need for Hep-B suppression therapy to reduce the risk of HCC.[26] For higher risk patients, six monthly imaging of the liver and analysis of AFP in viral hepatitis patients will also help detect HCC at an earlier stage and improve survival.[47]

The number of HCC cases increased substantially between 1998 (introduction of the surveillance programme) and 2017, with the most common causes of HCC in these patients being Hep-B (42%) and Hep-C (28%).[47] In support of this, Mules et al. also found the rate of Hep-B-associated HCC had increased over time in New Zealand.[24] Furthermore, those who had been previously diagnosed with Hep-B but not recruited on to a surveillance programme presented with advanced stage HCC and had poor prognoses; several factors were found to be contributing to this late presentation, including a lack of initial Hep-B diagnosis, and/or poor follow-up and surveillance.[24] In November 2019, only 19% of New Zealanders with Hep-B had been recruited onto the national monitoring programme.[26]

However, Mules et al. also noted that some patients who were receiving "optimal surveillance" still presented with advanced disease, which emphasises the limitations of our current surveillance methods.[24] Firstly, measurements of serum AFP have a low sensitivity and specificity as nearly one-third of HCC cases will have a measured AFP in the normal range, and healthy patients may also produce an elevated AFP (mostly caused by pregnancy). Secondly, the sensitivity and specificity of abdominal ultrasounds (USS) are also low, as 15% of HCC tumours may only be detected by USS when they are large, and other imaging methods such as MRI or CT may be required to differentiate HCC tumours from benign abnormalities.[24]

Relevance to Māori

It is estimated that a Māori male with chronic Hep-B infection has a 10–15% chance of developing HCC by the time they are 70 years of age.[48] Schauer et al. found that majority of patients (92%) with Hep-B-associated HCC were Māori, Pacific or Asian, with NZ European only making up 5% of cases.[47] Furthermore, Māori were largely over-represented in patients with advanced stage HCC due to viral hepatitis infection, with 45% of Māori cases from Hep-B and 23% from Hep-C.[24,47] In support of this, Mules et al. found that 143 out of 368 eligible HCC patients presenting with late stage HCC associated with Hep-B at the New Zealand Liver Transplant Unit were Māori.[24]

Crucially, Chamberlain et al. found that only around 40% of Māori who developed HCC from either Hep-B or Hep-C were on surveillance.[35] As such, there is clearly a strong need to ensure adequate access to surveillance services for Māori.[47] Māori are also over-represented in areas of higher deprivation, where barriers to medical

care and treatment are abundant, and they are more likely to experience poorer health literacy and understanding of the risks involved with viral hepatitis and HCC.[24,47,50]

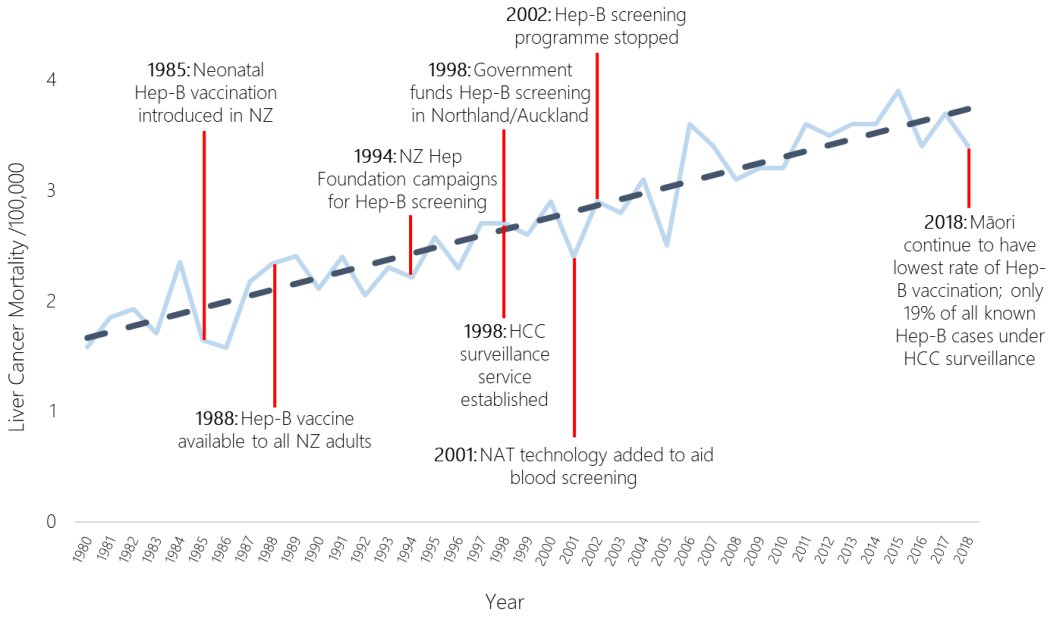
Key lessons and recommendations

This review has summarised the public health efforts aimed at detecting and treating patients with chronic hepatitis B and C in New Zealand over the last 25 years (see Figure 2). Chronic infection with Hep-B and Hep-C remains a significant health issue in New Zealand, with many patients currently undiagnosed and untreated, particularly within Māori and Pacific communities. As presented by Horsfall et al., an estimated 2 million Europeans, 333,000 Māori and 215,000 Pacific peoples in New Zealand remain unvaccinated for Hep-B, while an estimated 10,000 Europeans, 19,000 Māori and 15,000 Pacific have chronic Hep-B infection.[26] Around 40% of the 45,000 chronic Hep-C patients still remain undiagnosed or have been lost to follow-up.[47]

The neonatal Hep-B vaccination program has been successful in reducing transmission in the neonatal period through use of immunoglobulin in Hep-B positive mothers presenting for delivery;[25,26] however, unvaccinated children are most likely to become infected between the ages of 1 and 4 years.[25] In addition, there remains a significant reservoir of adults with chronic active Hep-B infections in the community who are an important cause of horizontal transmission within family groups. An additional issue is that vaccination funding ceases at 18 years of age, meaning that adults will likely need to pay for their own vaccination. While no vaccination exists for Hep-C, contemporary antiviral therapy will result in viral elimination in over 99% of affected patients.[27]

While the neonatal vaccination program has been successful, public health initiatives aimed at adults with these conditions have been episodic and undertaken without a national focus or national coordination. This is despite the conditions and their sequelae, such as HCC, being very much national healthcare issues, if not emergencies. There is now excellent data that confirms the prevalence and lethality of hepatitis and HCC in Māori and similar, although less complete, data in Pacific peoples residing in New Zealand. Addressing the disproportionate effects of these largely preventable conditions in these populations must be an important part of any health

Figure 2: Rate of liver cancer mortality per 100,000 New Zealanders between 1980 and 2018, overlaid by a chronology of actions taken to control hepatitis-associated liver cancer. Mortality data sourced from the Ministry of Health.[54]



strategy that is attempting to address issues of health equity for these populations.

There is a clear pathway by which we can achieve improvements in liver cancer control for all New Zealanders, and for Māori in particular. A programme to detect and treat Hep-B and Hep-C, as well as screen for HCC, could be undertaken through primary care. All enrolled patients over 18 years of age should undergo testing for Hep-B (HBsAg) and Hep-C (Hep-C Ab); those with positive results should undergo further testing including Hep-B/Hep-C viral load, liver function and platelet count. HBsAg positive patients will additionally be tested for Hep-B e-antigen and α-fetoprotein. Patients can then be started on appropriate antiviral therapy, and those with a higher risk of HCC based on the screening tests, or those with an initial positive serum α-fetoprotein, will be referred for specialist assessment at regional public hospitals and considered for HCC surveillance screening—with six monthly liver ultrasound scans and serum α-fetoprotein.[52]

The success of adequately resourced and empowered Māori and Pacific health providers in reaching their communities during the COVID-19 pandemic, and in their ongoing facilitation of vaccination and healthcare, is evidence that a nationally led, but grassroots driven, campaign would be successful.[39–42] For Māori such a program would resonate, reinforcing the importance of whānau (extended family) and the handing-on of good health from one generation to another. In 18 years (2040), or a little under one generation, New Zealand will celebrate the bicentenary of Te Tiriti o Waitangi, the founding document of our nation. If work begins now, hepatitis B and C-associated liver cancer could be largely eliminated from our population by 2040—sparing, without exaggeration, hundreds of lives each year and putting an end to a significant cause of health disparity for our Indigenous Māori population. This would be a cause for considerable celebration.

Conclusions

In this review, we have summarised the key risk factors for liver cancer, and the key activities undertaken in New Zealand to control this disease. Māori suffer a disproportionate burden of liver cancer in New Zealand, and there is evidence of systemic inadequacies in the prevention of, and outcomes from, liver cancer for our Māori population. The majority of the liver cancer burden for Māori is driven by disparities in viral exposure to hepatitis B and C; however, efforts to control hepatitis-associated liver cancer in New Zealand have been uncoordinated to date, and this remains a cause of the enduring disparity in liver cancer survival between Māori and NZ Europeans. We recommend the implementation of a national programme to detect and treat hepatitis B and C, and to screen for HCC among high-risk patients, with this programme to be delivered through primary care (including Māori health providers). Such a programme should be coupled with renewed effort to maximise hepatitis B vaccination rates, particularly for Māori. In combination, these efforts could result in the near elimination of hepatitis-associated liver cancer from Māori (and non-Māori) communities within a few generations.

COMPETING INTERESTS
Nil.

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Appendix 1: Non-hepatitis risk factors for hepatocellular carcinoma (HCC).

Alcohol-related liver disease

Alcohol and alcohol abuse is responsible for an estimated 30% of global incident cases of HCC and is expected to increase.[1–3] A meta-analysis of 19 cohort studies found a dose-response relationship between alcohol consumption and the risk of HCC.[4–6] Another meta-analysis of 19 prospective studies estimated an increased risk of 16% of HCC with alcohol consumption of three or more drinks per day, and a 22% increase with six or more drinks per day.[7,8] There is further evidence of an increased rate of HCC with alcohol consumption as low as 10g per day, relative to no consumption.[9,10] Moreover, evidence suggests that alcohol in conjunction with HBV and HCV has an additive synergistic effect on HCC risk.[1,7]

Chronic and sustained alcohol intake drives hepatocarcinogenesis by altering the architecture and functional capacity of the liver (steatosis, steatohepatitis, and cirrhosis) thereby generating a carcinogenic tissue microenvironment.[3,5] A pivotal pathophysiological factor implicated is oxidative stress, secondary to the production of ROS derived from alcohol metabolism, inflammation, and iron metabolism.[3]

Alcohol and hepatocellular carcinoma among Māori

The 2020/2021 New Zealand Health Survey shows that 20% of the population have hazardous drinking patterns (alcohol consumption that indicates a high risk of mental or physical damage).[11] Māori (33%) had a higher rate of hazardous drinking compared to NZ European/Other (21%).[11] However, the largest published study analysing the differences in patterns of alcohol consumption between Māori and non-Māori involving 6,926 Māori participants and 37,904 non-Māori demonstrated that Māori are less likely to drink alcohol, drink less often but consume a larger amount on one drinking occasion when compared with non-Māori.[12] Thus, the average alcohol consumption per day overall is similar between Māori and non-Māori [12]. These results are consistent with the 2013/14 New Zealand Health Survey which found that non-Māori were more likely to have consumed alcohol four or more times a week in the past year (RR 0.60) compared to Māori. However, of those who have consumed alcohol in the past year, Māori were more likely than non-Māori to have consumed a large amount of alcohol at least weekly (RR 1.75).[13]

A New Zealand study conducted over 24 years (1981–2004) found that 40% of individuals with HCC also had documented heavy alcohol use. Heavy alcohol use was similarly common for both Māori and non-Māori.[14] Based on available evidence, it appears unlikely that the strong disparities in liver cancer incidence observed between Māori and non-Māori in New Zealand is driven by differences in alcohol-related liver disease.

Non-alcohol-related liver disease

Non-alcohol fatty liver disease (NAFLD) is considered to be the hepatic manifestation of obesity, metabolic syndrome, and diabetes. It is the fourth most common aetiology of HCC[15–20] with the global prevalence forecasted to increase from 83 million (2015) to 101 million (2030).[21] It is becoming the most common liver disease contributing to hepatocarcinogenesis in developed countries.[22] However, it should be noted that the RR of NAFLD-related HCC is not comparable to the RR of HBV-HCC and HCV-HCC.[7]

The hallmark of NAFLD is triglyceride build-up in which sustained accumulation can lead to the development of HCC occurring as a sequential pathophysiological process; steatosis, steatohepatitis, fibrosis, cirrhosis, and eventually HCC.[16] The NAFLD-carcinogenic environment is promoted by sustained cycles of hepatocellular destruction and compensatory proliferation as a response to metabolic and oxidative toxicity, pathological inflammatory response, altered immune response, and altered endocrine/adipokine signalling.[23,24]

Non-alcohol-related liver disease and hepatocellular carcinoma among Māori

In New Zealand, the prevalence of NAFLD is 13%[25] with no existing data on the prevalence of NAFLD in Māori, or HCC attributable to NAFLD. Overall, the burden of obesity, metabolic syndrome, and diabetes is higher in Māori compared to non-Māori.[26,27] Although there is no data to demonstrate the prevalence of NAFLD induced HCC in Māori, we may infer that the increased exposure for NAFLD clinical risk factors may con-

fer an increased incidence of NAFLD-HCC among Māori compared to non-Māori.

Other causes of hepatocellular carcinoma

Aflatoxin and hepatocellular carcinoma

Aflatoxin is a mycotoxin produced by the *Aspergillus* fungus found in contaminated food such as groundnuts, tree nuts, and corn stored in damp, warm environments.[2,7] It has been estimated that 25,000–155,000 cases of HCC worldwide may be attributable to aflatoxin exposure, primarily in those regions where it is mostly found (sub-Saharan Africa, Southeast Asia and China).[28] AFB1, the most potent aflatoxin, is classed as a group 1 human carcinogen by the International Agency for Research on Cancer[29] and increases HCC risk by four-fold.[7,22] AFB1 has shown to have a synergistic association when combined with HBV, increasing HCC risk by sixty-fold compared to persons with neither risk factor.[7,30,31] There are concerns arising around the impact of climate change (driving increasing temperatures) and increased levels of aflatoxin.[28] It is expected that aflatoxin levels are to become more frequent in the future.[28,32]

A New Zealand Ministry for Primary Industries assessment determined that the overall population is exposed to low levels of aflatoxin compared to the international average. It was determined that liver cancer attributed to AFB1 would be less than 0.1 per year, thus indicating that dietary aflatoxin exposure is an insignificant contributor to HCC in New Zealand.[33] There is insufficient information available on aflatoxin exposure in Māori, although due to the insignificant AFB1 levels in New Zealand, it is unlikely that AFB1 exposure is an important contributor to the increased HCC incidence in this population.

Smoking and hepatocellular carcinoma

Tobacco smoking is responsible for 13% of global HCC cases.[9,34] The European Prospective Investigation into Cancer and Nutrition (EPIC) cohort with more than 4.4 million person-years of follow-up found that smoking has a RR of 4.55 for HCC, while a meta-analysis found an RR of 1.5 in smokers compared to non-smokers and an RR of 1.12 for previous smokers. The large RR variation can likely be explained by differences in study designs and exposure to smoking.[35,32] Smoking and tobacco exposure have also been shown to accelerate disease progression in other HCC-related diseases such as HBV and HCV infection.[35]

In New Zealand, the prevalence of adult daily smokers is 13% (2018/2019) of which 31% are Māori.[36] There is insufficient information on the prevalence of smoking and tobacco exposure attributable to hepatocarcinogenesis in Māori; however, due to increased smoking rates, we may be able to assume that smoking contributes to increased HCC prevalence in Māori. Further research is required to understand the extent to which smoking is a contributing driver of inequities in HCC incidence for Māori.

Helicobacter Pylori and hepatocellular carcinoma

Approximately 50% of the global population has been infected with the bacterium *Helicobacter Pylori* (*H. Pylori*).[37] Although principally associated with stomach cancer, several studies have found an association between *H. pylori* and HCC. A meta-analysis found the OR for the association between *H. pylori* infection and risk for HCC was 13.6.[37,38] By contrast, several high-quality studies have found a negative correlation between *H. pylori* and HCC, and evidence on the direct carcinogenic effect of *H. pylori* on hepatocytes has yet to be found.[37] Thus, as of yet, it is unclear whether *H. pylori* infections are unlikely to contribute to hepatocarcinogenesis.

Despite contrasting studies on the association of *H. pylori* infections and HCC, it is known that Māori have a greater prevalence of *H. pylori* infection (35%) compared to NZ Europeans (18%).[39] If the association of *H. pylori* induced HCC were to be proven, we can assume that it would disproportionately affect Māori compared to non-Māori.

The "other" other causes of hepatocellular carcinoma

Several studies have found an association between exposures other than those listed above and HCC. Some compounds/chemicals such as aristolochic acid,[22] areca nut (nitrosamines), betel leaves (safrole), vinyl chloride, and dichlorodiphenyltrichloroethane have been shown to increase the risk of HCC. Other studies have linked groundwater contaminants, organic solvents to hepatotoxicity.[2] However, given the insufficient body of evidence supporting these exposures and HCC, it is likely that these are not important contributors towards HCC for Māori.

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Isolated splenic hydatid disease: a rare case in rural New Zealand

David Thomas Bardsley, Gerhardus Bonnet, Michael O'Grady

Hydatid disease is an uncommon diagnosis in New Zealand. This parasitic tapeworm infection presents non-specifically and can be difficult to diagnose. In the early 1960s a national eradication strategy was implemented and by 2002, New Zealand declared "provisional freedom" from the disease.[3] This case report presents a recent instance of incidentally discovered hydatid disease in a patient presenting to primary care with back pain in the Whanganui District Health Board (DHB) and is a reminder that the diagnosis should not be forgotten in the New Zealand population.

Case report

A 55-year-old man presented to his general practitioner with back pain. Review of systems identified no other symptoms. Of note, he was a life-long farmer and had prolonged contact with canines. A lumbar X-ray revealed an incidental finding of a calcified lesion in the left upper quadrant, raising the possibility of a hydatid cyst (Figure 1). Ultrasound imaging confirmed an intrasplenic cystic structure. Serological testing for *Echinococcus granulosus* was indeterminant. Liver biochemistry and inflammatory markers were unremarkable. Retrospective review of historical chest x-rays performed up to 11 years prior to presentation showed left-sided subphrenic calcifications.

Abdominal computerised tomography (CT) imaging showed a peripherally calcified cystic lesion within the superior aspect of the spleen measuring 85x74x67mm consistent with a hydatid cyst (Figure 2). There was no other organ involvement. The patient was referred to the general surgeons and a laparoscopic splenectomy was performed electively. A large cystic structure was seen in the spleen and this was excised without perforation (Figure 3). Histology revealed sections that were compatible with an "ancient" hydatid cyst. The patient received his vaccinations as part of the splenectomy protocol.

The patient had an uneventful post-operative recovery and was discharged with no ongoing surgical follow-up.

Discussion

The incidence of hydatid disease in New Zealand is extremely low. Between 2006 and 2017, less than eight cases were reported per year. Whanganui DHB had no documented cases for the past 15 years.[4]

Hydatid disease is caused by the tapeworm, *Echinococcus granulosus*. Canines are the definitive hosts, while humans are involved as incidental hosts in the parasites lifecycle.[1,2] Usual intermediate hosts include cattle, sheep, goats and horses. The parasite causes cysts that may affect most systems of the body. Humans ingest the eggs, which migrate via the splanchnic venous drainage. Thus, the most common organs affected are the liver (50–75% of cases) and lungs (15–30%).[2,10] Splenic involvement is rare (approx. 5% of cases).[7,10]

Hydatids are often asymptomatic and can remain so for decades.[8] Symptoms typically develop with increasing cyst size. Pulmonary cysts may cause cough, dyspnoea and chest pain and hepatic cysts may result in right upper quadrant abdominal pain, nausea and malaise.[1,5]

Complications of hydatid cysts can arise from the cysts mass effects including obstruction of lymphatic and venous drainage systems. The cysts may become secondarily infected by bacteria or can rupture. Anaphylaxis following rupture has been described.[6]

There is serological testing available to aid in the diagnosis of hydatid disease. This testing typically includes IgG antibodies. The diagnostic accuracy varies depending on organ involvement. Sensitivity of serology testing in the presence of liver, lung and spleen involvement is 80–100%, 50–56% and 25–56%, respectively.[9]

Diagnosis of hydatid disease uses a combination of imaging, serology and clinical suspicion. Ultrasonography is the imaging modality of choice, but CT offers benefit for extra-hepatic lesions and anatomical mapping.

Treatment depends upon cyst classification, anatomical location and risk of complications. Antiparasitic agents, most commonly albendazole, can be definite or adjunctive therapies. Surgical removal of intact hydatid cysts or the use of the PAIR procedure (puncture, aspiration, installation

Figure 1: Abdominal X-ray in primary care; incidental finding of calcified lesion in the left upper quadrant.

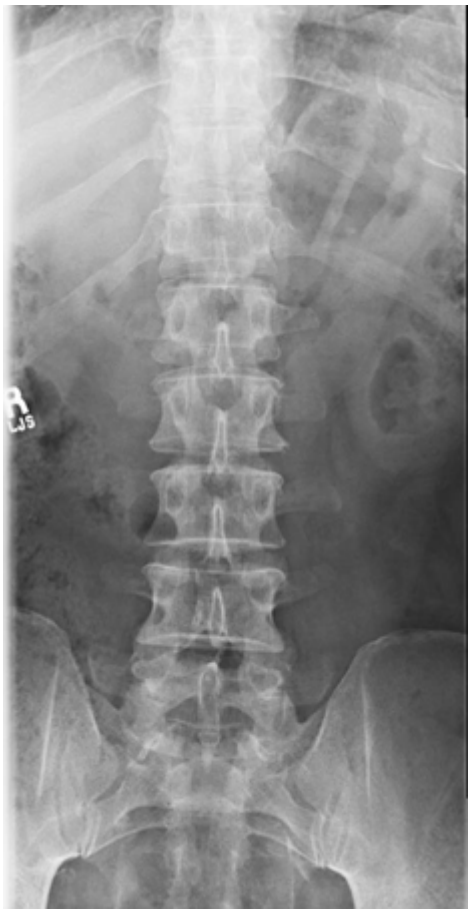


Figure 2: Axial computerised tomography of the abdomen showing a large cystic lesion in the spleen.

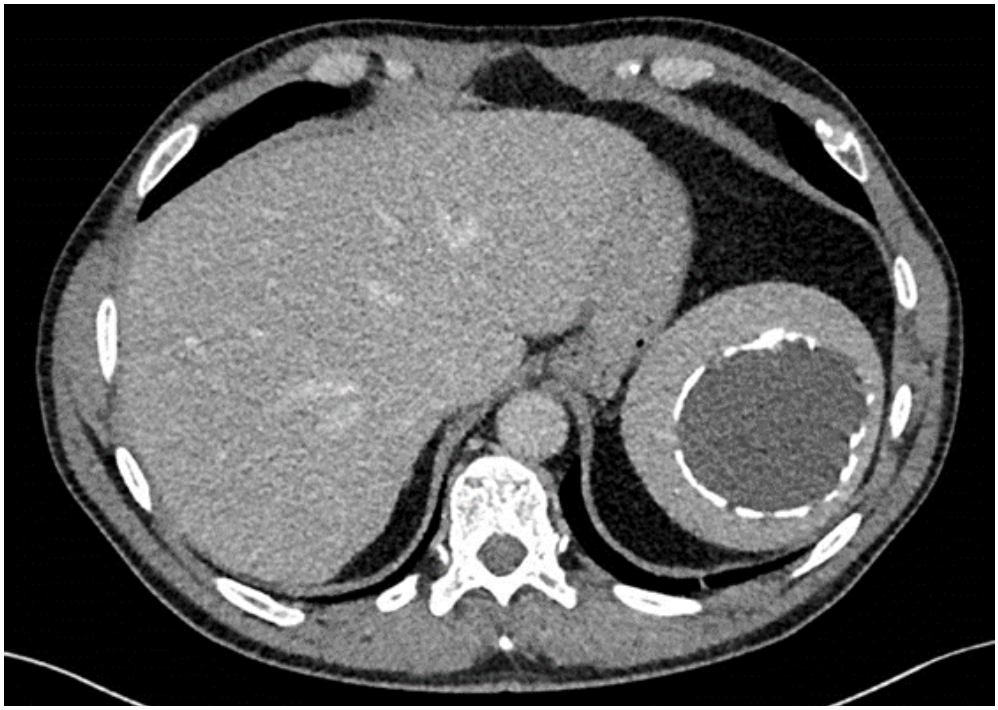
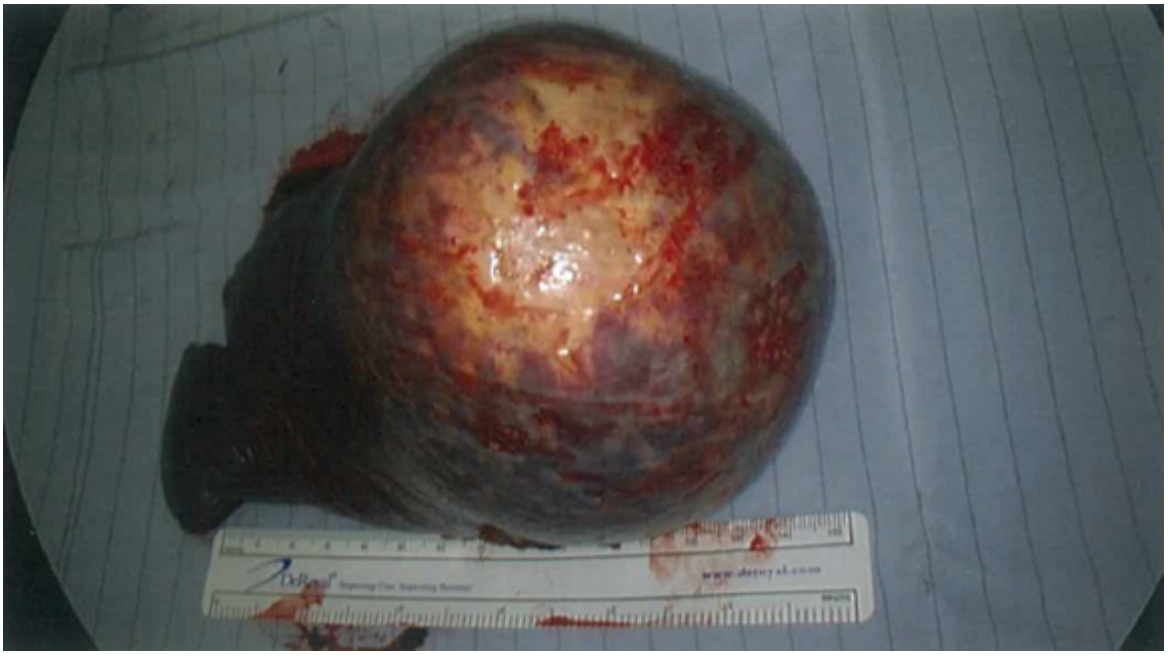


Figure 3: Surgical spleen specimen showing a large cystic lesion.



[of a protoscolicidal agent] reaspiration) are interventional treatments.

This rare case highlights the persistent possibility of hydatid disease in the New Zealand population.

Key points

- Although uncommon, cases of hydatid disease in New Zealand still exist. Cases likely relate to distant disease (if acquired in New Zealand) or brought in from areas of the world with a greater prevalence.
- The complications of hydatid disease can be life-threatening.
- Cysts can develop outside of the liver and lungs. These cysts may have a different appearance on imaging than those seen in the liver.
- Serological testing sensitivity for *E. granulosus* varies depending upon organ involvement.

COMPETING INTERESTS
Nil.

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Inadequate sun protection practices lead to sunburn among New Zealanders

Geraldine F H McLeod, Bhubaneswor Dhakal, Anthony I Reeder

New Zealand has among the highest age-standardised incidence and mortality rates for cutaneous malignant melanoma (CMM) in the world.[1] In 2018, New Zealand Cancer Registry age standardised registrations for melanoma were 42.1 and 31.8 per 100,000 for males and females respectively; 296 deaths from CMM were recorded.[2] The more numerous keratinocyte cancers are not required to be registered but accounted for an additional 204 deaths that year.[2]

The most effective way to prevent skin cancer is to reduce exposure to ultraviolet radiation (UVR) and sunburn with: body coverage (clothing, hat, sunglasses), using sunscreen and seeking shade or rescheduling activity when UVR is lower.[3] However, concurrent use of multiple sun-protection strategies is rarely reported by research participants, as many people use only one or two options when outdoors.[4] Previous research by Bleakley and colleagues[4] conceptualised three main forms of sun-protection ("Cover" using clothing or hat, "Protect" using sunscreen or sunglasses, and "Avoid" using shade or rescheduling outdoor activity) based on known mechanisms to reduce UVR exposure and sunburn. Conceptualisation of sun-protection in this manner is useful to show groups of similar strategies and their influence on sunburn. Their study showed that using one sun-protective strategy may preclude the use of other strategies, known as compensation behaviour.[4] However, their findings were limited by applying regression approaches to each sun-protection strategy individually. We hypothesise that it is possible to extend Bleakley and colleague's study[4] by employing a more sophisticated Seemingly Unrelated Structural Equation Model (SEM) to analyse the data, which allows for simultaneous testing of all sun-protection strategies against the outcome sunburn.

With the Southern Hemisphere summer bringing extreme levels of UVR between September and April,[3,5] now is the time to encourage the use of multiple sun-protection strategies among New Zealanders, particularly those with sun-sensitive skin types who sunburn readily and tan rarely.[6] The aims of this paper are to broadly replicate the study by Bleakley and colleagues[4] to report: 1) frequencies of specific sun-protection strategies ("Cover", "Protect", and "Avoid"); 2) associations of these strategies with sunburn outcomes; and 3) evidence of compensation behaviours for sunburn prevention among the New Zealand population, using data from a cross-sectional nationally representative dataset.

Methods

Study design and participants

Te Hiringa Hauora/New Zealand Health Promotion Agency Sun Exposure Survey (SES) dataset is a cross-sectional, nationally representative sampling of New Zealanders aged 13+ years, conducted during the 2016 Southern Hemisphere summer, who were outdoors for at least 15 minutes between 10am to 4pm on the day selected as the reference interview day.[7] In brief, n=2,272 people were interviewed, distributed by geographic region according to quota targets based on known population distributions.[7] For consistency with previous studies, only data for participants aged 15+ years who met the outdoor criteria were analysed (n=1,924).[8,9] Prior to weighting, sample sizes for age groups were 15–24 years 25.5%, n=491; 35–34 years 11.9%, n=229; 35–44 years 18.7%, n=359; 45–54 years 20.5%, n=395; and 55+ years 23.4%, n=450. Males 45.9%, n=884; females 54.1% n=1,040. Ethnicity sample sizes were Māori 10.6%, n=204; Pasifika 3.0%, n=58; Asian 6.0%, n=116; NZ European 79.9%, n=1520; and Other 1.4%, n=26. Due to missing data on sun protection strategies, some analyses are based on smaller samples. Ethical approval for analysing and reporting this audit-related data was obtained from the University of Otago Human Ethics Committee (HD17/039). Respondent participation was taken as informed consent.

Measures

Sunburn

Participants were asked "Did you get sunburnt? By sunburnt we mean any amount of reddening of the skin after being in the sun".

Sun-protection behaviours

Participants were asked a series of dichotomous questionnaire items about sunburn prevention behaviours they used while outdoors, grouped into three strategies based on Bleakley and colleagues:[4] Cover (with t-shirt, shorts, wide-brimmed hat); Protect (with sunglasses, sunscreen); Avoid (using shade, rescheduling outdoor activity). These strategies, conceptualised by Bleakley and colleagues,[4] were based on known mechanisms to reduce UVR exposure and sunburn/skin cancer. Each strategy was classified as 1="Yes"/0="No" reflecting whether the respondent had reported any of the associated sunburn prevention behaviours.

Covariates included self-reported sociodemographic characteristics (respondent sex, age (years), skin-type using Fitzpatrick sun-sensitivity scale I–IV,[6] ethnicity, region of residence and of outdoor activity, highest educational qualification, household income, self-assessed skin cancer risk score), duration outdoors (minutes) and concurrent climatic conditions (UVR, air temperature).

Statistical methods

Sun-protection behaviours were concatenated to show strategies used by participants. A Seemingly Unrelated SEM[10] was applied to the data to simultaneously test all sun-protection strategies against the outcome sunburn/not sunburned. The parameters were estimated using the full information likelihood method to account for indigeneity problems. Data were analysed using SAS 9.4[11] and Mplus.[12]

Figure 1: Proportion of sunburned participants who reported sun-protection behaviors (%).

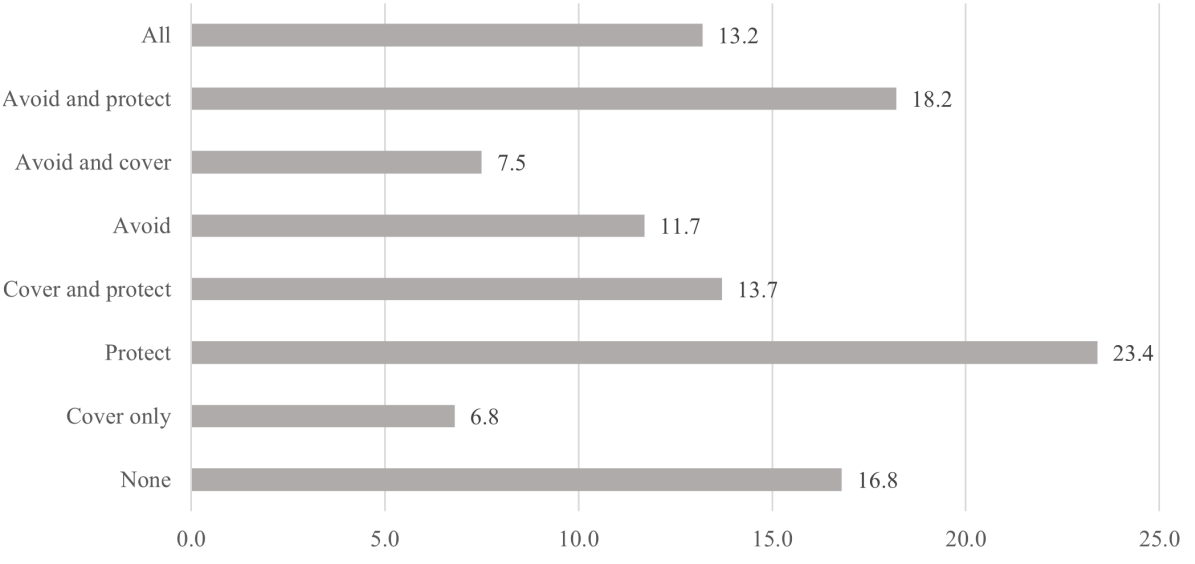

Table 1: Seemingly Unrelated Structural Equation Model (SEM) standardised model results of behaviors that may be associated with sunburn prevention.

Prevention behavior	B(SE)	p-value
Cover	-0.057 (0.031)	0.068
Protect	0.182 (0.033)	<0.001
Avoid	-0.049 (0.033)	0.140

Note: Co-variates of demographic characteristics, duration outdoors and concurrent weather conditions were included in the models (see Methods). Full model results are available upon request from the corresponding author.

Results

Sunburn was reported by 14.9% (279/1,877) of participants. Figure 1 shows the sun-protection strategies of those sunburned on the target day: 16.8% (19/113) did not use any sun-protection strategies and 13.2% (60/456) used all strategies. Most (69.7%) (1,308/1,877) behaviour was consistent with a compensation strategy of only one or two to sun-protection options. The protect strategy (sunglasses and sunscreen) yielded the most (23.4%) (52/222) sunburn cases. Examination of the SEM (see Table 1) shows no particular behavioural strategy statistically significantly prevented sunburn ($p>0.07$); sunglasses and sunscreen were positively associated with sunburn ($p<0.001$).

Discussion

The aims of this study were, firstly, to report use of specific sun-protection practices. We found that, during the Southern Hemisphere summer of 2016, 16.8% of sunburned participants did not use any sun-protection strategies. The highest proportion of sunburn cases were among those who used the Protect strategy (23.4%). Secondly, two of three SEM pathways between protective strategies and prevention of sunburn were statistically non-significant. Use of sunglasses and sunscreen was positively associated with sunburn ($p<0.001$). We concluded that these findings were consistent with the practice of compensation behaviours—in support of Bleakley and colleagues[4] in which the majority of sunburned participants used sub-optimal sun-protection. Caution is advised regarding interpretation of results as assessment of weekend sunburn prevalence may not accurately extend to findings on studies reporting annual sunburn prevalence. Further, our approach may potentially yield different estimates as our sun protection scores were calculated differently to Bleakley and colleagues (2018).[4] In addition, sun protection factor (SPF), brand or amount of sunscreen used by the participants was not recorded. Other limitations include the potential for bias from untested instrument validity among the New Zealand population. Nevertheless, the study sample was representative of the New Zealand population.

In conclusion, this study confirms that many sunburned New Zealanders did not use optimal sun-protection, consistent with the operation of compensation behaviours. Each protective strategy may be seen as an alternative rather than complementary pattern of behaviour, precluding the use of other strategies. Further investigation of compensation behaviours is warranted to provide insight into sun-protection barriers because ineffective sun-protection is problematic among populations with high skin cancer rates. Now that New Zealand is in a period of high UVR during months September to April,[3,5] there is an opportunity to reduce sunburn and consequent CMM among those with sun-sensitive skin types. This can be achieved by using multiple sun-protection strategies (body coverage, using sunscreen, seeking shade or rescheduling outdoor activity) during high UVR months.

COMPETING INTERESTS
Nil.

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Iodine-only supplements for breastfeeding women: a call to action in New Zealand

Ying Jin, Jane Coad, Sheila Skeaff, Cheryl Benn, Shao J Zhou, Louise Brough

The current New Zealand guideline recommends iodine-only supplementation (150µg/day) is needed during breastfeeding to meet increased iodine requirements, even for women with a well-balanced diet.[1] In 2009, mandatory fortification of bread with iodised salt was introduced in New Zealand[2]—this was expected to improve the iodine status of the general population, but was not sufficient to meet the increased needs of pregnant and lactating women. In 2010, a major Government initiative was launched in New Zealand to provide iodine-only supplements for all pregnant and lactating women.[1] In 2021, the Mother and Infant Nutrition Investigation (MINI), an observational longitudinal cohort study spanning the first postpartum year, recruited 87 breastfeeding mother–infant pairs in New Zealand.[3] This study found that, for women who did not take iodine supplements, both mothers and their infants were iodine deficient at three months postpartum.[4] Iodine deficiency can compromise the thyroid function of both the mother and her breastfed infant, which would be exacerbated if the woman became pregnant again soon.[5]

The MINI study reported that 46% (40/87) of breastfeeding women at three months postpartum took iodine supplements in the 24hr period prior to the data collection,[3] and throughout the first postpartum year the percentage of breastfeeding women using iodine supplements declined (to 11% at six months and 6% at 12 months).[4] A 2019 cross-sectional online survey of New Zealand breastfeeding women reported 63% (179/284) used iodine supplements within the first six months postpartum.[6] Data showed much lower usage amongst breastfeeding mothers than was expected, which implies a failure of adhering to the current iodine supplementation recommendation in New Zealand.

Adequate iodine supply is required for the optimal production of the thyroid hormones, triiodothyronine and thyroxine, which are essential for the development of the central nervous system during the first 1,000 days of a child's life.[7] It can take several weeks for absorbed iodine to be incorporated into thyroid hormones, thus it is essential to achieve adequate intrathyroidal iodine stores periconceptually is to allow for the increased thyroid hormone production during pregnancy and after parturition.[8] During lactation, iodine is required for maternal thyroid function and is secreted into breastmilk to ensure adequate iodine supply for optimal infant thyroid function and neurological development.[9] In New Zealand, women are recommended to use iodine-only supplements once pregnancy is confirmed until they cease breastfeeding completely, as well as consuming balanced diet to ensure optimal infant brain development.[1]

The cross-sectional online survey of New Zealand women reported 81% of lactating women who used iodine supplements did so because of advice given by health professionals.[6] The most frequently reported reason for not taking supplements in the MINI study was women were not specifically advised to do so by a health professional.[3] Although women may be provided with a prescription for iodine supplements by their health professional, this may be too costly for those who are already on a tight budget or require extra travel or transportation for collection of supplements from their pharmacy. A 2016–2017 New Zealand online survey reported 13% of women who received prescriptions for iodine supplementation did not collect their prescriptions,[10] however, reasons for this were not stated.

In New Zealand, six weeks after childbirth, the medical care for mothers and their infants' transfers from their lead maternity carer (Midwife or Obstetrician) to their general practitioner (GP), in conjunction with child nurses (i.e., Tamariki Ora or Whānau Āwhina). These health professionals are well-placed to provide timely and comprehensive advice to postpartum women. Strategies to reduce barriers for women to access Government-subsided iodine-only supplements are needed. For example, removing the cost of dispensing fee at pharmacy, having such free supplements available at midwife or GP clinics, so that women who require them can collect them imme-

diately, or midwives could get a supply of iodine on practitioner supply order to give to women when they attend visits. There is an urgent need for greater public awareness and information sharing through the health professionals including pharmacists in New Zealand, to promote iodine specific nutrition advice including: 1) the importance of iodine in maternal health; 2) the existing recommendations of taking iodine-only supplementation (150µg/day) for all pregnant and breastfeeding women; and 3) to encourage the routine prescription of Government-subsided iodine-only supplements, which is more affordable for all lactating women. Proactively increasing knowledge of the benefits of iodine supplementation during breastfeeding through health professionals and others who are well-placed to provide such information and support to future mothers, will raise awareness of what can be seen as an ongoing barrier to good maternal and infant health in New Zealand.

COMPETING INTERESTS
Nil.

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Aotearoa New Zealand must learn how to vaccinate children most at risk of vaccine-preventable diseases first

Cameron C Grant, Owen Sinclair, Teuila Percival

Ko a tātou tamariki, ngā taonga mo āpōpō
Our children are the treasures of tomorrow

Do we truly treasure our tamariki, our children?

If you had a treasure that required protection against the deadly vaccine-preventable pneumococcal disease, and you had a choice of two vaccines, which one would you choose? One that cost more or one that was cheaper but provided protection against fewer pneumococcal serotypes? Since we did choose this cheaper vaccine, how do you feel now that that we can see, since this choice was made, that the number of pneumococcal serotype 19A cases in young children, and the proportion of pneumococcal isolates that are penicillin-resistant, has increased?[1]

And if, in 2021, you had seen that when international borders opened briefly, respiratory viral infections quickly re-entered Aotearoa New Zealand and caused many of your treasured children to be hospitalised,[2] and caused some to die,[3] how would you plan for 2022 when borders would open more fully? Would you immediately offer your children the influenza vaccine knowing that the influenza virus would be entering Aotearoa New Zealand in 2022, and knowing that influenza infection can then lead onto severe bacterial pneumonia?[4] Or would you only offer the influenza vaccine to adults and wait for influenza to arrive, for many of your children to be hospitalised with severe illnesses caused by influenza and bacterial pathogens that influenza enables,[4,5] and then offer the influenza vaccine to your children also?

If a schedule of life-saving vaccines were required to be delivered on time to all your treasured children, whilst you were focussing on the vaccine prevention of life-threatening illness in adults, would you stop giving the schedule of life-saving vaccines to your children? How would your children feel if they knew that, in 2022, one-in-three of them were not given all of their infant vaccinations?[6] Would they feel treasured?

Knowing that Māori and Pasifika children are at increased risk of vaccine preventable diseases, how would you feel if, in 2022, only 46% of your Māori children and 62% of your Pasifika children at age six months had received all of their infant vaccinations?[6] And how would your Māori and Pasifika children feel if they knew that since 2019, while vaccine coverage at age six months had decreased by 12% for your New Zealand European children, it had decreased by 26% for your Māori children and by 19% for your Pasifika children?[7,8] Would they feel treasured?

If you knew that fewer than 3% of pregnant women in New Zealand do not intend for their children to receive vaccines,[9,10] would you be surprised that, in June 2022, only 67% of children at age six months had received their infant immunisations?[6] And, if you knew that pregnant women living in the poorest neighbourhoods had the highest intentions for their children to receive their vaccines,[11] would you be surprised if their children had the lowest coverage for infant vaccines?

If you knew that infants in other developed countries, such as the United Kingdom, Ireland and Italy, receive the meningococcal vaccine,[12] but not infants in Aotearoa New Zealand, how would you feel? You might remember that this vaccine was available in New Zealand in the past,[13] and that because we used it then, other countries now feel confident to include it in their childhood immunisation schedules now.[12] But we have decided not to.

Albert Einstein is quoted as saying: "*Insanity is doing the same thing over and over and expecting different results*". Having read the preceding paragraphs, would you now agree that Aotearoa New Zealand is systematically failing in its childhood vaccine delivery and needs to do this differently?

It is time Aotearoa New Zealand learnt how to vaccinate all its treasured children and to vaccinate those most at risk of vaccine-preventable

diseases first. As a result of COVID-19, we now have a larger and more diverse vaccination workforce and many more strategies for delivering vaccines to those who want them and really need them, but for whom the current healthcare system is incapable of delivering. We need to apply lessons learnt from the COVID-19 vaccine strategy to address the insanity of our childhood vaccine delivery system. But will we?

COMPETING INTERESTS
Nil.

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The Treatment of Psychic Disorders

NZMJ, 1922

The use of the term "functional" to describe the milder mental disorders such as hysteria, the obsessive or compulsive neuroses, anxiety states, and neurasthenia, should be discarded for two reasons.

Firstly, organic disease, *e.g.*, cancer of the stomach, causes, like any other organic disease, a disturbance of the functions of the organ concerned, and secondly, what has been called "functional" disease, is now regarded as of psychic or mental origin. The distinction that is practically of importance is that between disease which is due to organic change and disease which is of purely mental origin. The objection to calling hysteria mental disease is the odium attached to the phrase, but the use of the term "psychic" avoids this.

The treatment of these milder forms of mental disease is one of the utmost practical importance, and in this connection in this journal two questions have recently been raised. One is the value of psycho-analysis, and the other is the need for and the method of use of institutional treatment.

If all the hysteria, etc., is to be treated in a special institution it will need to be a very large one. These milder mental disorders are among the most crippling ills that man is heir to. The wage-earning capacity, or efficiency of life, is seriously impaired by them, often for prolonged periods. Expense of treatment is therefore an important consideration. At the present time the treatment they receive is often prolonged and expensive. They are isolated in private hospitals and massaged, etc., or undergo prolonged courses of medicine with a variety of tonics or of sedatives. They are sent away on holidays or to special institutions. The treatments sometimes do good, but often they do no good, and the chiropractic steps in. The success of the chiropractic is due chiefly, I take it, not to any great amount of good in that system, but to the fact that the medical profession, speaking broadly, does not understand these mild mental ailments. An attitude very commonly adopted towards these patients recalls the ancient manner in which lunatics were regarded and cruelly treated.

Associated with the question of institutional treatment of the hysteric is the question of the utility of psycho-analysis. For some 10 or 12 years I have studied this form of treatment. My practice of it I have allowed gradually to grow, as my self-analysis progressed and definite opinions on it formed, and as I felt able to undertake more of the work. Now I find I am able, without undue fatigue, to practise the treatment for five or six hours daily. I find that in the practice of a general physician there are many cases where a few treatments, one to three, produce a most beneficial result, and that in definite pronounced cases the treatment often gives success otherwise unobtainable. It is particularly effective in young people, but I have gained good results in cases over 50. In most cases my patients have lived at home and frequently have followed their ordinary avocation while under treatment.

I propose to give, as bearing on the question of institutional treatment, and on the questin of psycho-analysis, a brief account of some work done during the last six months. The report shows a fair average of results.

EPILEPSY—(1) Girl aged 21. This case had been subject to fits from 8 years of age. I saw some of the fits, and they were definitely epileptic. There were also numerous hysteroid or psychotic symptoms associated, *e.g.*, a desire to cut her throat. This girl's life was gradually being narrowed down, for she could not be allowed to go to church or out for the evening as she would take fits. She could not even be allowed to go out for short messages. She was becoming dull and doing nothing. Treatment consisted of under twenty sittings one hour each once a week. It was stopped three months ago. There have been no more fits. The girl is happy and cheerful, working at home, and practising the piano. The last I heard of her is that she was going to dances. The friends are very pleased and have thanked me profusely.

(2) A boy, aged 15½ years, had taken five fits in two months. Diagnosis hystero-epilepsy. A borderland case with elements of epilepsy, migraine, and hysteria. He had been advised to leave his work and go into the country. My treatments consisted of four sittings. There have been no more fits in five months, and he worked during treatment, and has been at work ever since.

(3) A thin, miserable, nervous woman with anxiety, tremblings, attacks in the legs like Raynaud's disease, and many other symptoms. Treatments,

eight in number, one of three hours' duration, in which I sat and listened to one of the most graphically presented tragedies I have ever heard. Result, very great improvement. One attack of shaking in the last three months. Appetite is good, general condition is much improved. She should have one or two more treatments.

(4) Woman, aged 49 years, who had, some years previously, threatened suicide, and was going the same way again. Treatment, fifteen sittings, depressions and suicidal tendencies disappeared. Patient very much better, and everybody pleased. (This patient was a sister of one whom I cured of an anxiety neurosis in six sittings the previous year. As a result of that cure her constant attendance at doctors for the prescription of tonics has ceased.) In addition to analysis I restrained the tyranny of the old mother over the patient.

(5) A young man with wet dreams—three treatments—result satisfactory. Perhaps this case has been benefitted really by suggestion.

(6) A woman, 35 years, constantly sending to the doctor for tonics, and buying patent medications. After taking two doses of a bottle it was put down the sink. Treatment, some preliminary visits, then three treatments—cure of these bad habits, and improvement in other ways, but cure of nervousness is complete.

(7) A man, aged 20, with giddy turns and a bad temper. Three treatments, cured to the satisfaction of his wife.

(8) A man with giddy turns incapacitating him from work—returned for treatment which has been applied during previous year—able to work pretty fully, but still has some giddy turns, though they are not nearly as bad as they were. In this case there was no result worth speaking of for twelve treatments (three months) and then sudden marked improvement.

(9) Woman, aged 45, at climateric. Feels inclined to hide behind a door when she meets people, trembling attacks, etc., still under treatment. Treatment, twelve sittings, feeling of shame gone, better in every way, attacks of trembling still occur, but are less frequent, and less severe.

(10) Man aged 55, to return for further treatment after having been relieved of a desire to go to into a lunatic asylum as a voluntary patient. This patient is old in his arteries, and I warned him I might get no result. Patient is glad he has undergone treatment, but result is not satisfactory, although a good deal better than I promised him. He is still under treatment.