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EDITORIAL

It is unethical to incarcerate people with disabling mental disorders. Is it also unlawful?

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My orthopaedic surgeon suggests a unicompartmental knee replacement: a detailed look at the long-term outcomes of a single surgeon's practice

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Obreniokibo I Amiesimaka, Kristina Aluzaite, Rhiannon Braund, Michael Schultz

Given the constraints to accessing medications for inflammatory bowel disease (IBD) therapy in Aotearoa New Zealand, medication adherence is of utmost importance. Electronic health records (EHRs) are widely used in medication adherence (MA) assessment. Poor adherence in patients with IBD can lead to worse disease outcomes and increased health costs. We explored the suitability of Southern New Zealand EHRs for estimating adherence and its relationship with corticosteroid dispensings (indicating negative disease outcomes). Findings reveal that useful EHR data is available in Southern New Zealand, but needs optimisation for adherence assessments; so, we recommend avenues for achieving this via better data collection (of needed variables), formatting and presentation. Accessible data indicates that IBD patients' adherence is high, but further research is needed to confirm this and better understand the association between adherence and disease outcomes.

Auckland Regional Cleft Palate Service: service accessibility and speech outcomes

Melanie Street, Anna Miles

Children born in Auckland and Northland with a cleft palate will receive surgery, specialist therapy, dental and psychology services as well as outpatient appointments in a South Auckland hospital. This paper looks at how far people have to travel, what speech therapy is offered and what their speech outcomes are like at 5 years old. Consideration is given to speech outcomes of Maaori in particular, as Maaori have one of the highest incidences of cleft in the world and more Maaori live in rural areas than non-Maaori.

Falls at home: hospital admissions in a health region of Aotearoa New Zealand

Ciaran Simpkins, Ishani B Soysa, Grant Christey

Fall related injuries (FRIs) are a significant source of trauma burden in New Zealand. Falls are the leading cause of unintentional injury at home among children aged ≤4 years and adults aged 20–64 years, and nearly a third of self-reported injuries, requiring medical attention, occur at home in New Zealand. FRIs also have a significant economic impact on the New Zealand health system and beyond through the Accident Compensation Corporation. Evidence reporting on the descriptive epidemiology of patients admitted following injury from falls across all ages and all severities is limited. This study examines the characteristics of patients of all age groups and severities that were admitted to New Zealand hospitals following falls at home over a prolonged period (11 years).

Mā te Whakarongo—a qualitative study exploring the impact of middle ear disease on New Zealand Māori

Lance Buckthought, Jeannine Stairmand, Rebecca Garland

The study aimed to explore the impact of middle ear disease on the lives of New Zealand Māori through a series of semi-structured interviews. The study demonstrates that living with middle ear disease presents many challenges and disadvantages for Māori, and the importance of early detection and referral to specialist care.

The deconstruction of chronic orofacial pain and a hiding inhibition pathway of orofacial pain: the trigeminal proprioceptive mesencephalic periaqueductal gray pathway

Ajith Polonowita, Li Mei, Guangzhao Guan

It is important to understand the neurophysiology of chronic pain in order to diagnose and manage chronic orofacial pain. Modification of pain information may take place in several ascending or descending pathways. The trigeminal proprioceptive mesencephalic periaqueductal gray pathway may be considered as the mechanism of action of occlusal appliance in managing orofacial pain related to TMD. The factors such as sleep, psychological disease (e.g., anxiety/depression), hormonal and other factors not yet identified might be under the influence of genetics. The interplay between genes is still an active area of research in understanding chronic pain.

Patient experience surveys are vital in the twenty-first century: let's put some myths to rest

Catherine Gerard, Inga O'Brien, Carl Shuker, Jo Swanson, Richard Hamblin

Critics of the use of patient-reported experience data from surveys have been using tired arguments that don't stack up. Aotearoa New Zealand's patient experience survey is one of the biggest and most robust surveys in the country and allows us to hear directly from patients about what did and did not happen during their healthcare encounters. This allows us to hear the patient voice and act on it to develop a truly patient-centred healthcare system. Use of this data is an ethical requirement, and now a legal requirement under the new Code of Expectations for Consumer Engagement appended to the Pae Ora (Healthy Futures) Act 2022. Critics need to stop trotting out tired, unsubstantiated arguments and start engaging with this large repository of robust data.

Discordant thyroid function tests—beware of albumin variants

Bobby Li, Marianne Elston, Jordyn Moore, Stephen du Toit, Howard Potter, Indika Ranasinghe, Chris Florkowski

Some people have uncommon variants of albumin, the most abundant protein in blood plasma, which binds to some thyroid hormones more than usual. This can cause thyroid function tests to be inaccurate and may make monitoring more challenging. This can be investigated with liaison with the laboratory. We believe we describe the first known case of this condition in an Afghan patient.

It is unethical to incarcerate people with disabling mental disorders. Is it also unlawful?

Erik Monasterio

"Morales y luces son nuestras primeras necesidades" ("Morals and lights are our first necessities") – Simon Bolivar

A Royal Commission of Inquiry is underway. This is investigating abuses to tamariki, rangatahi and adults in State and faith-based care in Aotearoa New Zealand between 1950–1999. Commissioners have established a vision for the Inquiry. They want to ensure that both the outcome of the Inquiry and the process for engaging communities and survivors will transform the way care is provided to the most vulnerable people in our communities.[1] The full report is not expected until 2024. However, a case study report, *Beautiful Children,* which investigated the Lake Alice Child and Adolescent Psychiatric Unit, has been published.[1] This catalogues extensive and disturbing human rights abuses. An editorial review comments "*the Lake Alice story is also a story of a toxic culture, systems failures, staff complicity, institutional racism, and a litany of failings by State agencies*."[2] In decades to come, can we expect an Inquiry into the human rights abuses in our prisons of the 2020s, much like the current Royal Commission of Inquiry? Are we willing to allow history to repeat itself?

After 23 years of work as a forensic psychiatrist, I spent the last 7 as Clinical Director and Forensic Director of Area Mental Health Services (DAMHS) in Canterbury. Despite trying as hard and creatively as I could, systemic factors meant I could not discharge that leadership responsibility to a reasonable standard. I therefore felt no option other than to resign. This article explains those systemic factors which led to my decision. I do so in the hope of advocating for people suffering from severe and disabling mental illnesses, who are excluded from care and living in the most precarious and traumatic of conditions—whether in prison or at the margins of our community. I seek to bring attention to what stood in the way of me helping care for this group of people, leading me to be unable to meet my responsibility as Forensic DAMHS. If these impediments are not well understood and addressed, the human rights of patients will continue to be breached. There will be further loss of staff and deeper entrenchment of the current crisis, with increased risks to patients and the community.

Human rights breaches

In March 2020, all Clinical Directors and DAMHS of Aotearoa New Zealand Forensic Services took the unprecedented step of publishing an editorial in the *New Zealand Medical Journal*, which highlighted human rights violations of acutely mentally ill people in our prisons.[3] This followed a previous publication warning of a looming health crisis in prisons.[4] It gave a united opinion of an expanding mental health crisis in which prisons were being used to contain patients with severe mental health problems who needed immediate care. In particular, the editorial sought to draw urgent attention to the failure of successive governments to address the need to care for and protect some of our society's most vulnerable people, echoing the failures that led to the current Royal Commission of Inquiry.

Widely cited data from two key Aotearoa New Zealand prison studies show the high and increasing prevalence of substance use and mental health disorders (including severe psychotic disorders) among people in prison.[6,7] Prisoners' health is public health, and improving their health outcomes is central to reducing health inequalities and improving public health.[5]

The 2020 editorial followed a failure to make any headway with the Director-General of Health, the Ministry of Health and the Health Minister. Together with other Forensic Clinical Directors, I met with former Health Minister David Clark in December 2018. We presented data on the frequent use of solitary confinement cells in prison to contain acutely mentally disordered prisoners

who could not be transferred to hospital because there was not a bed available. Regional forensic services consistently reported data to the Ministry of Health on waiting lists for urgent admissions, including for patients detained in Intervention and Support Units (ISU) in prisons. The median waiting time for transfer to a Forensic bed was 4 weeks,[3,8] meaning more than half of people waited over a month for care that would have been immediately available had they been in the community.

Detention in ISU involves 23-hour per day solitary lockdown, at times without access to natural light and fresh air. There is a wide body of research that shows that detention in solitary confinement is harmful, disproportionally so for those who suffer from mental illness. It is associated with a range of serious cognitive impairments, severe emotional distress and exacerbation of symptoms. There is an increased risk for self-harm, suicide and future impaired functioning.[8,9] Those so detained were often too unwell to accept medications, engage in talking therapies or participate in therapeutic activities. Often, they could not even maintain basic self-care.

The editorial made clear that detention in ISUs, in the opinion of the authors, the Ombudsman Office and the Human Rights Commission, constituted Human Rights abuses and breached national and international agreements on the minimal standard of care for prisoners.[3,8,9] A recent finding by the European Court on Human Rights found that use of solitary confinement, as occurs in the ISUs, constitutes a Human Rights breach: *"The Court considers that prolonged solitary confinement entails an inherent risk of harmful effects on any person's mental health, irrespective of the material or other conditions surrounding it"* (para. 140).[10]

The situation at the time of the 2020 editorial was intolerable. Now it is far worse. Instead of increased psychiatrist staff and bed numbers, across many of the regional forensic and general adult services there has been unprecedented increase in psychiatric staff departure and vacancies. The Canterbury forensic service alone has lost over half its senior medical staff within 3 years. At the same time, hospital management reduced the service's acute inpatient bed capacity by 20% without consultation with the remaining senior medical staff, and seemingly with no understanding of the impact of this decision on the rights of mentally ill people in prison.

He Ara Oranga report

The incoming Labour-led Government in 2017 brought optimism to beleaguered specialist mental health services. This Government set itself the goal of understanding and addressing the mental health crisis, underscored by *"the politics of kindness"*.[11] Their approach was to commission the *"once-in-a-generation"* He Ara Oranga report.[12] The incorrect assumption before commencing the Inquiry was that specialist mental health services were already available for the 3% of people with the most severe mental health needs (page 8).[12] The Inquiry therefore largely ignored this group and promoted extension of services to up to 20% of the population with mild to moderate mental illness and distress, within 5 years of the inquiry.[12] However, between 2016 and 2020, Aotearoa New Zealand was ranked thirty-second out of 38 OECD countries for the number of hospital psychiatric beds. Aotearoa New Zealand reported 31 beds per 100,000 population, which was far below the OECD average (69 beds per 100,000 population) and well below the minimally required number (50 beds per 100,000 population).[13] Also, the consistent advice and data on waiting lists' numbers for urgent hospital admissions and the use of prison beds (including ISUs) to contain the acutely mentally disordered before the Inquiry was unambiguous. Submissions made on behalf of forensic mental health services went unheeded. There has therefore been no increase in acute mental health beds for the past 7 years.[14] Moreover, consistent loss of specialist mental health staff and erosion of morale has lowered the capacity to treat those who are most severely and chronically ill. A recent investigative report has found that despite the $2 billion investment following the Inquiry, there has been little tangible benefit in the general population, and the situation for those with serious mental illness is no better.[15] Experts have argued there is an urgent need to re-focus: the limited resources must be targeted towards those with serious mental illness, in areas with the highest levels of deprivation.[13,16]

Unlawful detention of people with "mental disorders" in prison?

There are increasing numbers of people with mental illnesses detained in Aotearoa New Zealand prisons, with overrepresentation of Māori and Pasifika. Over 90% of the prison population have a lifetime diagnosis of a mental health or substance

use disorder, with 61% prevalence within the past year.[6,7] The prevalence of most psychiatric disorders, and in particular of psychotic symptoms, is far higher in prison than in the general community.[6] The risk of imprisonment after inpatient discharge has increased in the past decade, with nearly 1% of people entering prison within 28 days.[17] Much of this increase is made up of men of Māori or Pacific ethnicity who present with aggression in the context of substance use and psychotic disorders.

The *Mental Health (Compulsory Assessment and Treatment) Act 1992* (*MHA*) provides the legal framework to protect and care for those who fulfil the criteria for a "mental disorder" and who, as a consequence, are at risk to themselves or others or have significantly diminished capacity for self-care. The *MHA* has inbuilt legal safeguards. These include access to independent legal advice, review by a District Inspector, prompt review by a Judge, access to a psychiatric second opinion and a requirement that patients who have capacity to do so can give consent for treatment. Part 3 of the *MHA* also explicitly states that *"this Act shall bind the Crown"*. The intention of the *MHA* is to ensure treatment and protection for those with qualifying mental disorders who cannot be treated in a less restrictive manner. Failure to exercise the *MHA* deprives mentally unwell people of those legal rights which are enshrined within the *Act*. As the *MHA "binds"* the Crown, for people who are acutely mentally disordered appearing before a criminal court or who are detained in custody, it is legally unjustifiable not to resort to the use of *MHA*, if instead those people will be excluded from care and will suffer serious harm.

The longstanding lack of psychiatric inpatient beds has now normalised the use of ISUs. The situation will continue into the future unless urgent steps are taken. For health and corrections staff, working in an environment where human rights are breached and from which there is no obvious relief causes moral injury.[18] It breaches the basic principle we have all vowed to adhere to—*"primum non nocere"*. For health training institutions, as are all Regional Forensic Services, the exposure to and seeming acceptance of such practices for trainees on placement models unethical practices. For those in leadership positions the conflict is more serious. The Forensic DAMHS are appointed by the Director-General of Health (Ministry of Health) to be responsible for the adequate management of the *MHA*, and the persistence of this practice makes it impossible to discharge this duty to a minimally acceptable standard. Disturbingly, this situation parallels the abuses that occurred in Lake Alice Hospital, and it should not be ignored.[1,2]

Specialist mental health courts

Mental health courts are specialised courts that offer an alternative (or diversion) to standard prosecution for people with mental health problems who are charged with an offence. These courts are available across a number of states in Australia, but they have not been introduced in Aotearoa New Zealand. The courts have been shown to achieve successful outcomes, particularly for lower risk offenders.[19] The advantage of these courts is that they identify offenders with immediate mental health needs early and can divert to psychiatric care, rather than resorting to incarceration.[19,20] This, in turn, permits forensic specialist mental health services to focus limited resources on the treatment of higher risk offenders with mental illness.

Proposed solutions

1. **Re-focus on serious mental illness:** there are insufficient resources to provide adequate care for those with serious mental illness. Unless it is to be accepted that prisons will be utilised to contain the mentally ill, inpatient psychiatric beds will need to be increased to at least 50 beds/100,000 population (the OECD average is 69) with commensurate staff resources to manage this.[13,16]
2. **Clarify legal parameters of the MHA:** it is the author's opinion that the discretionary application of the *MHA*, in situations when not to invoke its use leads to exclusion from care and serious harm, is not only inconsistent with the spirit of the *MHA* but may also be unlawful (pursuant to Part 3, "*This Act shall bind the Crown*"). Putting this matter to the court will resolve this question and determine whether an amendment to the *MHA* is required to protect the seriously mentally disordered in the criminal courts.
3. **Urgently introduce specialist mental health courts in Aotearoa New Zealand.**
4. **Drug courts:** drug courts have been successfully piloted in Aotearoa New Zealand since 2012 but have not yet been rolled out throughout the country. They

are more widely available in Australia and provide more flexible sentencing options and diversion to treatment for people who have offended, including alternatives to prison.[20]

5. **Quantify the extent of staff loss and talk to staff:** in my experience working across various regions of Aotearoa New Zealand, there have been unprecedented departures of specialist mental health staff, including forensic psychiatrists. Issues raised in this editorial are likely to contribute to this. It is imperative to quantify and acknowledge staff shortages, and in particular to determine the reason for the staff loss. Without understanding the reason for staff losses, measures to counter this will not be able to be instituted. A recent editorial in the *Journal* has cautioned that the healthcare workforce is the foundation of Aotearoa New Zealand's mental health system and is on the brink of collapse, requiring urgent action.[22]

Conclusion

This editorial highlights pressing challenges which preclude Forensic Clinical Directors and DAMHS in Aotearoa New Zealand from discharging their ethical and legal duties. Aotearoa New Zealand is failing to provide a minimal standard of care for seriously mentally ill people. There is a disproportionally negative impact on Māori and their whānau, in breach of the Crown's duty to protect as part of its Te Tiriti obligations. The impact on Pasifika is also dire.

It is not morally acceptable or legally defensible to utilise prisons to deal with this health crisis. The Royal Commission of Inquiry into Abuse in Care shows we must front up to and learn from our past mistakes. It is not too late to tackle this problem head on. Doing so now may prevent the need for a future Inquiry into the mistreatment of mentally unwell people in our prisons.

COMPETING INTERESTS
Nil.

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REFERENCES
1. Abuse in Care – Royal Commission of Inquiry. Beautiful children Te Uiui o te Manga Tamariki me te Rangatahi ki Lake Alice/Inquiry into the Lake Alice Child and Adolescent Unit [Internet]. Wellington (NZ): Royal Commission of Inquiry; 2022 [cited 2023 Dec]. Available from: www.abuseincare.org.nz/our-progress/reports/inquiry-into-the-lake-alice-child-and-adolescent-unit/.
2. Every-Palmer S, Sutherland O. Abuses in psychiatric care: The shameful story of the Lake Alice Child and Adolescent unit in Aotearoa New Zealand. Aust N Z J Psychiatry. 2023;57(9):1193-1197. doi: 10.1177/00048674231193381.
3. Monasterio E, Every-Palmer S, Norris J, et al. Mentally ill people in our prisons are suffering human rights violations. N Z Med J. 2020;133(1511):9-13.
4. Foulds JA, Monasterio E. A public health catastrophe looms: The Australian and New Zealand prison crisis. Aust N Z J Psychiatry. 2018;52(11):1019-1020. doi: 10.1177/0004867418802902.
5. Kinner SA, Young JT. Understanding and improving the health of people who experience incarceration: an overview and synthesis. Epidemiol Rev. 2018;40(1):4-11. doi: 10.1093/epirev/mxx018.
6. Indig D, Gear C, Wilhelm K. Comorbid substance use disorders and mental health disorders among New Zealand prisoners [Internet]. Wellington (NZ): New Zealand Department of Corrections; 2016 [cited 2023 Dec 11]. Available from: https://www.corrections.govt.nz/__data/assets/pdf_file/0013/13603/Comorbid_substance_use_disorders_and_mental_health_disorders_among_NZ_prisoners_June_2016_final.pdf.
7. Brinded PM, Simpson AI, Laidlaw TM, et al. Prevalence of psychiatric disorders in New Zealand prisons: a national study. Aust N Z J Psychiatry. 2001;35(2):166-73. doi: 10.1046/j.1440-1614.2001.00885.x.
8. Sharon Shalev. Thinking Outside the Box? A Review of Seclusion and Restraint Practices in New Zealand [Internet]. Auckland (NZ): Human Rights Commission; 2017 [cited 2023 Dec 11]. Available from: https://papers.ssrn.com/sol3/papers.cfm?abstract_id=2961332.
9. Lamusse T. Solitary confinement in New Zealand prisons. ESRA. 2018:1-37. doi: 10.13140/RG.2.2.27336.47365.
10. Case of Schmidt and Šmigol v. Estonia (2023) 3501/20 45907/20 43128/21. Judgment (Merits and Just Satisfaction). Court (Third Section). Available from: https://hudoc.echr.coe.int/?i=001-229386.
11. Elliott P. Politics of Kindness: Exploring the political discourse of Kindness articulated by Ardern in New Zealand [master's thesis]. Hamilton (NZ): The University of Waikato; 2020.
12. Patterson R, Durie M, Disley B, Tiatia-Seath S. He Ara Oranga Report of the Government Inquiry into Mental Health and Addiction [Internet]. Government Inquiry into Mental Health and Addiction; 2018 [cited 2023 Dec 15]. Available from: https://mentalhealth.inquiry.govt.nz/assets/Summary-reports/He-Ara-Oranga.pdf.
13. Mulder RT, Bastiampillai T, Jorm A, Allison S. New Zealand's mental health crisis, He Ara Oranga and the future. N Z Med J. 2022;135(1548):89-95.
14. Lynch J. New Zealand has same number of acute mental health beds as when Labour came to power. Newshub [Internet]. 2022 Nov 22 [cited 2023 Dec 15]. Available from: https://www.newshub.co.nz/home/politics/2022/11/new-zealand-has-same-number-of-acute-mental-health-beds-as-when-labour-came-to-power.amp.html.
15. Bradley A. What ever happened to the $2 billion that was poured into mental healthcare? RNZ [Internet]. 2023 Dec 11 [cited 2023 Dec 15]. Available from: https://www.rnz.co.nz/programmes/in-depth-special-projects/story/2018918734/what-ever-happened-to-the-dollar2-billion-that-was-poured-into-mental-healthcare.
16. Allison S, Bastiampillai T, Castle D, et al. The He Ara Oranga Report: What's wrong with 'Big Psychiatry' in New Zealand? Aust N Z J Psychiatry. 2019;53(8):724-726. doi: 10.1177/0004867419848840.
17. Skipworth J, Garrett N, Pillai K, et al. Imprisonment following discharge from mental health units: A developing trend in New Zealand. Front Psychiatry. 2023;14:1038803. doi: 10.3389/fpsyt.2023.1038803.

18. Griffin BJ, Purcell N, Burkman K, et al. Moral injury: An integrative review. J Trauma Stress. 2019;32(3):350-362. doi: 10.1002/jts.22362.
19. Lim L, Day A. Mental health diversion courts: A two year recidivism study of a South Australian mental health court program. Behav Sci Law. 2014;32(4):539-51. doi: 10.1002/bsl.2126.
20. Davidson F, Heffernan E, Greenberg D, et al. Mental health and criminal charges: variation in diversion pathways in Australia. Psychiatr Psychol Law. 2017;24(6):888-898. doi: 10.1080/13218719.2017.1327305.
21. Mitchell O, Wilson DB, Eggers A, MacKenzie DL. Assessing the effectiveness of drug courts on recidivism: A meta-analytic review of traditional and non-traditional drug courts. J Crim Justice. 2012;40(1):60-71. doi: 10.1016/j.jcrimjus.2011.11.009.
22. Foulds JA, Beaglehole B, Mulder RT. Time for action, not words: the urgent rebuilding of New Zealand's mental health workforce. N Z Med J. 2023;136(1576):8-10.

My orthopaedic surgeon suggests a unicompartmental knee replacement: a detailed look at the long-term outcomes of a single surgeon's practice

Samuel J Lynskey, Christopher M Frampton, Timothy G Lynskey

ABSTRACT

AIM: This single surgeon case series with up to 29-year follow-up evaluated the survival of the Oxford unicompartmental knee replacement (UKR) for isolated medial compartment osteoarthritis.

METHOD: Four hundred and four knees in 330 patients were followed for between 12 days to 29 years, with an average of 13 years. Kaplan–Meier survival estimates were calculated using revision for total knee replacement and re-operation for any reason over 5-year intervals. Revision and reoperation rates per 100 component years were used to compare subsets, cemented and uncemented prostheses and Phase 2 and Phase 3 instrumentation.

RESULTS: Of 404 UKRs, 292 (72%) were cemented, 96 (24%) uncemented and 16 hybrid (4%); 137 (34%) were undertaken using Phase 2 instrumentation and 267 (66%) Phase 3 instrumentation. Estimated revision-free implant survival at 20 years was 78%, and estimated reoperation-free survival at 20 years was 72%.

CONCLUSION: Unicompartmental knee replacement is a very successful procedure, with 78% of knees remaining revision-free at 20 years. Progression of lateral compartment arthritis was the most common cause for revision, affecting 60% of revision cases. There was no significant difference in the revision or reoperation rate between cemented and uncemented prostheses or Phase 2 and minimally invasive Phase 3 instrumentation.

Unicompartmental knee replacement (UKR) has been a popular treatment since Ahlbäck discovered that knees with clinical osteoarthritis have isolated medial compartment degeneration.[1] Benefits of UKR include a shorter operative time, reduced hospital stay, lower blood loss, greater post-operative range of movement and higher activity level at the time of discharge than in total knee replacement (TKR).[2] In addition, long-term benefits include preservation of bone stock, lower morbidity, higher functional activity and a subjective feeling of a "normal knee" due to cruciate ligament preservation.[3] UKR is associated with higher revision rates compared to TKR; however, head-to-head, UKR performs better across multiple domains, and so patients must be encouraged to consider these factors.[4] Further, for those patients with predisposing comorbidities, a lower risk of thromboembolic and major cardiac events in UKR as compared to TKR is likely to be material and should also be discussed as part of the consent process.[5] Nevertheless, patients often elect to undergo joint replacement procedures based upon the recommendation of family members or friends. This mode of decision making, though influential, should not substitute for time spent in ensuring an understanding of the nature, benefits and risks of the procedure, as well as viable alternatives; and, in the case of UKR, what the options are if and when the prosthesis fails. As clinicians we are responsible for justifying our decisions and actions, and in order to enter into a model of shared decision making, the patient must be provided with transparent, robust and understandable information.[6] To do this accurately, the clinician must have a comprehensive understanding of these factors enabled by contemporaneous knowledge of the evidence as well as participation in self-audit.

UKR has been in use since 1970; however, there are few long-term case studies published.[5,7] This study is a retrospective single-surgeon case series of prospectively acquired data over a 31-year follow-up period. The primary aim was to review

and report the survivorship of the Oxford UKR for isolated medial compartment osteoarthritis of the knee, as determined by revision to TKR or re-operation for any reason. The secondary aims were to compare the subsets of Phase 2 with Phase 3 instrumentation, as well as cemented with uncemented prostheses. Finally, we aimed to investigate and discuss causes of failure and report long-term functional scores associated with UKR.

Methods

Included patients were those who underwent UKR with the Oxford prosthesis for isolated medial compartment osteoarthritis who satisfied the criteria according to the prosthetic designers.[8] Specifically, patients with fewer than 15 degrees flexion contracture, 100 degrees of flexion on the operating table, a correctable varus deformity in 20 degrees of knee flexion, a clinically or radiologically intact anterior cruciate ligament, full thickness cartilage loss in the anterior aspect of the medial compartment, but preserved posteriorly, and a lateral joint space of 5 millimetres or more on a valgus stress X-ray taken in 20 degrees of knee flexion were included.

Primary outcome measures

Survival of the prosthesis was determined by identifying patients who underwent revision to TKR. These patients were captured through representation to the primary surgeon and by screening the New Zealand Joint Registry. Revision-free estimates were calculated for 5-year follow-up intervals using Kaplan–Meier estimates. Reoperation for any reason was determined by screening local hospital records and via telephone communication with the patient. Reoperation-free estimates were calculated for 5-year follow-up intervals using Kaplan–Meier estimates. The time to revision and reoperation was used to calculate a revision rate per 100 component years (py) with 95% confidence intervals (95% CI) using a Poisson approximation.

Secondary outcome measures

Revision rates were used to compare the subsets, cemented and uncemented knees, and Phase 2 and Phase 3 instrumentation using Log-Rank tests. A two-tailed p-value less than 0.05 was taken to indicate a statistically significant difference between groups.

The chief investigator kept a database from 1 February 1991, the first surgery, until 28 November 2022, the last audit. The database was password protected and compliant with governing local ethics review requirements. Functional scores were determined using the Oxford Knee Score (OKS) obtained from the New Zealand Joint Registry. From the year 2000 onwards, all patients undergoing UKR surgery in New Zealand were posted a form asking them to complete an OKS. All those who responded were posted a further form at 5-yearly intervals out to a maximum of 15 years post-operation. Final follow-up was determined to be either the date the patient was deceased, underwent reoperation for any reason or the date of final review, specifically 28 November 2022. Additional information, including demographic data, were obtained from patient records, theatre logbooks, the New Zealand Joint Registry and the National Patient Database.

This audit was approved by the Taranaki District Health Board Ethics Committee and local iwi.

Results

The total number of UKRs undertaken over the study period was 404 in 330 patients. No cases were lost to follow-up. One hundred and sixty-seven patients who underwent replacement were deceased at an average of 11.5 years, and 166 were still alive at an average of 17.3 years. The average prosthetic follow-up was 13 years (12 days–29 years). See Table 1.

Thirty-nine UKR patients survived over 20 years and 16 over 25 years, with the two longest survivors reaching 29 years.

The Kaplan–Meier estimate of survival using revision to TKR as the primary end point at 15 years was 84%, and at 20 years 78%. Fifty-one UKRs required revision to TKR, of which 60% were for progression of lateral compartment osteoarthritis. In this group four UKRs had a previous high tibial osteotomy (HTO) and two UKRs in one patient developed haemochromatosis. One patient on warfarin suffered recurrent haemarthroses, one developed rheumatoid arthritis and in one the medial compartment was overstuffed. Four knees became symptomatic of patellofemoral osteoarthritis and were revised to TKR for anterior knee pain. Four patients had revision to TKR following significant trauma. Two were twisting injuries resulting in anterior cruciate ligament (ACL) rupture and two suffered falls, the first with a delayed presentation of a lateral tibial plateau fracture and the second from a mountain bike with posterior cruciate and medial collateral ligament injury.

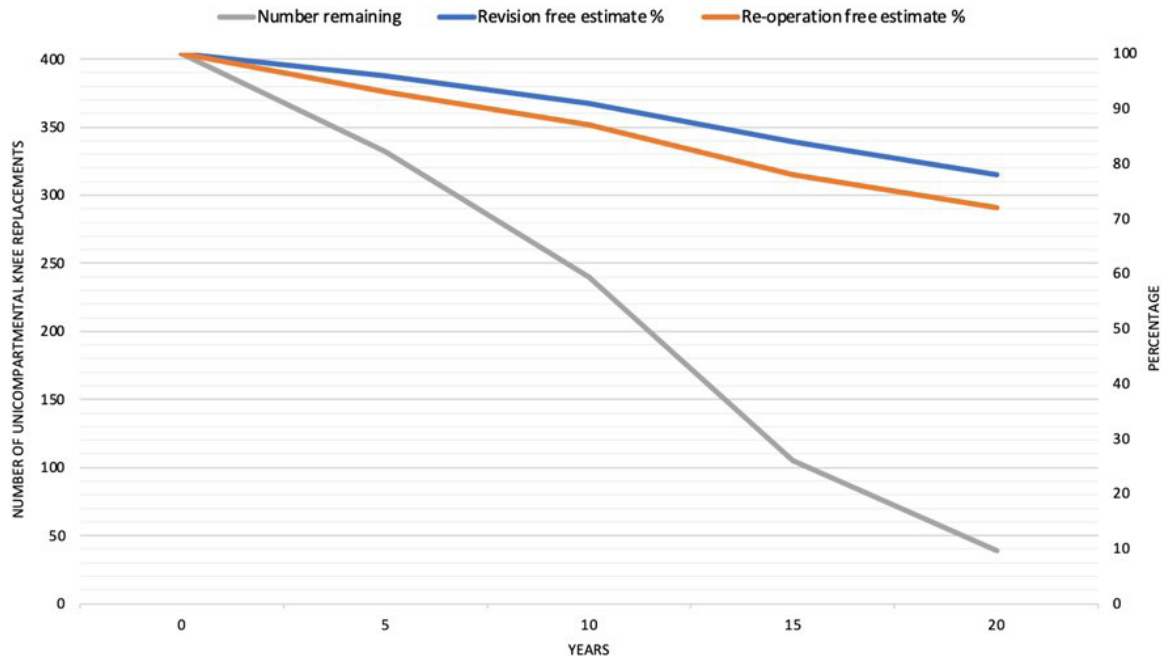
Table 1: Procedural breakdown and follow-up data, including revision, prosthetic fixation and instrumentation type.

Procedures	404
Patients	330
Lost to follow-up	0
Male/female %	57/43%
Average age at operation	70.8 (range: 51–94)
Average follow-up	13 years (range: 12 days–29 years)
Alive	166 (41.1%)
Deceased	167 (41.3%)
Revision to total knee	51 (12.6%)
Other reoperation	20 (5%)
Cemented prostheses	292 (72%)
Uncemented	92 (24%)
Hybrid (2 reverse)	16 (4%)
Phase 2	137 (34%)
Phase 3	267 (66%)

Table 2: Reasons leading to revision to total knee replacement.

	N=51	**Average time to revision (years)**	**Range of time (years)**
Progression of lateral compartment arthritis	31	10	1.3–24
Loosening tibial component	8	10.3	1–16
Progression patellofemoral arthritis	4	8	1–14
Significant traumatic injury to knee	4	5.2	3.3–16.6
Heterotopic ossification with stiffness	1	10	
Deep infection	1	0.7	
Chronic regional pain syndrome, stiffness	1	13	
Pain without identifiable cause	1	4.5	

Figure 1: 5-yearly Kaplan–Meier revision and reoperation survival estimates.



Of the cases that went on to have TKR, 17 were revised using primary total knee components; 28 required a stemmed tibia; five required both stemmed tibia and femur; and only one patient required a stemmed femur alone. See Table 2.

Estimated reoperation-free survival at 15 years was 78%, and at 20 years 72% (see Figure 1).

There were 20 reoperations where a UKR had been preserved. The most common cause was a worn or fractured mobile bearing (see Table 3). One was size 3, and the remainder size 4. Medical records were available for all patients and showed no further surgery was undertaken in any of the 20 UKRs until death or the final review date. Of note, the survival time after re-operation is shown but was not used in any of the calculations of survival or reoperation rate.

Mobile bearing instability *without* dislocation was seen in two uncemented UKRs. The mobile bearing dislocation rate in the cemented UKRs was 0.3%, and in the uncemented UKRs it was significantly higher at 3% (p=0.05). One patient suffered bilateral recurrent mobile bearing dislocation and was treated by conversion to a fixed bearing prosthesis, Oxford-M. No obvious cause for dislocation was found.

One patient suffered mobile bearing impingement causing discomfort and a click in full extension due to bone regrowth over the anterior aspect of a cemented femoral component, which required bone resection. There were two cases of trauma where the UKR was retained; a fall causing a tibial plateau fracture, which was internally fixed, and direct trauma to the knee as a result of a cow causing an anterior cruciate ligament rupture, which was revised to the Oxford-M prosthesis.

The revision rate per 100 component years for all UKRs with 4,653 component years was 1.10 (range 0.82–1.44) and the reoperation rate was 1.52 (range 1.19–1.92). There was no significant difference in the revision rate between cemented and uncemented prostheses or between Phase 2 and Phase 3 instrumentation as shown in Table 4. Revision for patellofemoral arthritis occurred in four cases in our series (1%). Bearing wear requiring replacement was noted in six UKRs, all of which were cemented.

Returned OKS showed 91% excellent or good scores at 15 years (see Table 5).

Table 3: Reason for reoperation, other than conversion to total knee replacement.

Reason for reoperation	Procedure	N=20	Time to reoperation	Further surgery	Survival after reoperation
Worn or fractured mobile bearing	Replace bearing	6	12 (10–15) years	No	12 (4–17) years
Mobile bearing instability	Insert larger bearing	2	1 & 6 years	No	7 & 10 years
Isolated post-operative mobile bearing dislocation	Replace bearing	2	5 & 6 weeks	No	2 & 13 years
Recurrent mobile bearing dislocation	Convert to Oxford-M	2 in one patient	0.5 & 4.7 years	No	9 years
Anterior mobile bearing impingement	Arthroscopic debridement	1	2 years	No	13 years
Loose femoral component	Re-cement	1	4 years	No	12 years
Medial ligament injury, bearing instability	Insert larger bearing	1	2.5 years	No	7 years
Impinging osteophyte cruciate footprint of tibia	Excise osteophyte	1	2 years	No	13 years
Cement loose body	Remove loose body	1	1.4 years	No	14 years
Irritation overhang tibial component, 4mm	Revise tibia size A to AA	1	16 weeks	No	10 years
Traumatic anterior cruciate ligament rupture	Convert to Oxford-M	1	16 weeks	No	10 years
Traumatic proximal tibial fracture	Internal fixation	1	3 weeks	No	10 years

Table 4: Revision and reoperation rates per 100 component years.

	Total number of UKRs	Sum of component years	Number of UKRs revised	Rate/100 component years	Lower 95% CI	Upper 95% CI	p-value
Revision							
All UKRs	404	4,653	51	1.10	0.82	1.44	
Reoperation							
All UKRs	404	4,653	71	1.53	1.19	1.92	
Revision							
Cemented	294	3,849	44	1.14	0.83	1.53	0.93
Un-cemented	102	756	7	0.93	0.37	1.91	
Reoperation							
Cemented	294	3,849	55	1.43	1.08	1.86	0.06
Un-cemented	102	756	16	2.12	1.21	3.44	
Revision							
Phase 2	137	1,906	22	1.15	0.72	1.75	0.99
Phase 3	277	2,748	29	1.06	0.71	1.52	
Reoperation							
Phase 2	137	1,906	29	1.52	1.02	2.18	0.92
Phase 3	267	2,748	42	1.53	1.10	2.07	

Table 5: Returned Oxford Knee Scores (OKS).

	6 months	5 years	10 years	15 years
Number returned	208	115	67	11
Average score	40	41	42	41
Excellent	47%	64%	64%	36%
Good	34%	23%	28%	55%
Fair	13%	6%	3%	9%
Poor	5%	5%	3%	0%

OKS: excellent >41; good 34–41; fair 27–33; poor <27.[9]

Discussion

This single-surgeon series with long-term follow-up reports the survivorship of the Oxford UKR for isolated medial compartment osteoarthritis of the knee, as determined by revision to TKR or reoperation for any reason. Further, we compared subsets of Phase 2 with Phase 3 instrumentation and cemented with uncemented prostheses. We also described causes of failure and reported long-term functional scores associated with UKR.

When patients reach the stage of considering joint replacement surgery for isolated medial compartmental osteoarthritis, the possible options include UKR, TKR or HTO. In general, the chief investigator reserves HTO for patients under 50 because of less predictable and durable results and a slower recovery and, when necessary, a more difficult revision to TKR.[10] The benefit of UKR over TKR in a matched cohort of patients is clear—with less pain, higher activity level, longer discomfort-free walking time, greater satisfaction with functional activities and higher probability of reporting "satisfied" or "very satisfied" up to 1 year post-operatively.[11] Our study adds to the weight of evidence supporting "excellent" or "good" knee function in 91% of respondents, with 15-year follow-up. Patients undergoing TKR also have a higher risk of complications including myocardial infarction, thromboembolism and stroke, plus higher readmission rates and mortality; such that if 100 patients receiving TKR underwent a UKR instead, there would be one less death and three more reoperations in the first 4 years post-operatively.[12] The decision then for surgeons to offer, *and for patients to consent to*, UKR for all its benefits must be balanced against the *known greater* revision and reoperation rate.[4,12]

Long-term survival of Oxford medial compartment replacement for unicompartmental osteoarthritis in our series is similar to the New Zealand Joint Registry for all UKR, which shows a survival for reoperation at 15 and 20 years of 83% and 76% respectively.[13] Compared with previous reports regarding the Marmor UKR—a prosthesis previously used at our institution—the Oxford showed a higher earlier revision rate with survival at 10 years of 91% compared with 95%, but a similar survival at 15 years and more UKRs surviving beyond 20 years.[14] Similar to the Marmor UKR, in our series the most common cause for revision was for progression of disease involving the lateral compartment followed by loosening of the tibial component.[15]

With time and experience there have been modifications to the original indications for the Oxford UKR.[8] Previous HTO and bone loss with grooves on the lateral side of the patellofemoral joint are now contraindications to UKR.[16] In our review we have chosen not to exclude patients with prior HTO or pre-existing patellofemoral joint osteoarthritis in the analysis, which in part may explain marginal differences in revision rates compared with other case series; nonetheless, these are comparable to those reported in the New Zealand Joint Registry for the Oxford UKR. We recommend therefore that patients presenting with prior HTO or patellofemoral joint disease not necessarily be excluded from UKR, but rather be carefully evaluated on a case-by-case basis.

Regarding our secondary aim, we found no significant difference in the revision rate between cemented and uncemented UKRs. This finding is discordant with a meta-analysis reporting significantly better survival in the uncemented Oxford UKR; however, these data were limited by a relatively shorter follow-up period of 18.3 months (ranging from 18.3 months to 7.6 years).[17] Our reoperation rate per 100 component years for cemented prostheses was 1.43 (95% CI 1.08–1.86), and was consistent with the New Zealand Joint Registry figure of 1.37 (CI 1.27–1.48). However, the uncemented prostheses rate of 2.12 (CI 1.21–3.44) did not achieve overlap with New Zealand Joint Registry figure of 0.87 (CI 0.77–0.97).[13] Nine of the 20 UKRs that underwent reoperation were uncemented and included three cases involving significant trauma at 3 weeks, 16 weeks and 2.4 years, and two additional cases of mobile bearing dislocation at 5 weeks and 24 weeks post-operatively. The relatively small total number of uncemented UKRs combined with the number of early reoperations in this series undoubtedly contributed to the higher revision rate. Our finding of a statistically significantly lower mobile bearing dislocation rate in the cemented UKRs (0.3%) compared with uncemented UKRs (3%) (p=0.05) is consistent with other reports.[18] We question whether these bearing-related problems were indicative of a learning curve for uncemented UKRs, tibial component subsidence over time or some other factor. We question if the intraoperative selection of bearing thickness should take into account possible future subsidence. Whether tibial component subsidence is associated with early or late mobile bearing dislocation, however, remains to be elucidated and warrants further investigation.

The introduction of Phase 3 instrumentation facilitated minimally invasive surgery that has been reported to be associated with suboptimal implant positioning and a higher revision rate.[19] Of note, however, we found no difference in the revision rate with the adoption of Phase 3 instrumentation.

Surgeon volume has been shown to influence revision rate. The ideal percentage of knee replacements that are deemed eligible for UKR has been estimated to be between 6 and 50%.[20] The chief investigator's percentage would be approaching the higher figure but, as in other series, this would reflect referral pattern rather than extended indications.[16] An excellent or good OKS of 91% at 15 years in our series compares favourably with Joint Registry (84%) and other studies (79%).[13,16]

In the event of complications necessitating further surgery and where the indications for UKR still exist we have preferred to preserve the UKR. Contrary to the Oxford Group, in cases of progression of arthritis we preferred to revise to a TKR despite the UKR components still being well fixed.[16] The exception to this is in a younger patient with recurrent mobile bearing dislocation or anterior cruciate ligament rupture through injury where literature supports conversion to fixed bearing UKR designs.[21] The most commonly reported reason for reoperation for any reason was symptomatic progression of arthritis, which occurred in 35 knees (49%). This finding is consistent with literature and occurs most commonly in the lateral compartment.[16,22,23] Reported progression of patellofemoral arthritis is common, but revision for this problem is low and reported as 3%, as compared to 1% in our study.[18] Reoperation for mobile bearing exchange occurred in only 1.5% of patients in our series, all of which were cemented prostheses and were either 3 or 4mm as recommended by the designers.[16] Further, these mobile bearings did not demonstrate macroscopic evidence of impingement on retrieval, suggesting alternate causes for failure other than overstuffing or impingement.

The primary industry in our region is farming and workplace trauma as a cause for revision in our series is noteworthy. We could not find a similar incidence of injuries in other comparable studies.[16,22,23] WorkSafe New Zealand statistics show farm assistants, labourers and agricultural workers are more likely to suffer injuries at work than other self-employed people, with the majority being soft tissue injuries and 6% being fractures or dislocations.[24]

Strengths of this study include a mean length of follow-up of 13 years with no loss to follow-up. Additionally, we have detailed and discussed the reasons for revision or reoperation in all but one case that had pain without an identifiable cause and was not improved by revision.

Limitations of this series includes the absence of radiological, limb alignment and range-of-motion assessment data. Further, adjustment for comorbid disease was not assessed in determining revision and reoperation rates. There is also a possibility of reporting bias with respect to our OKS, given the diminishing response rate over time. Many of our UKRs survived well into the third decade; however, we did experience a number of bearing-related complications and we therefore suggest that a prospective randomly controlled trial of fixed and mobile bearing UKRs would be valuable.

This study serves as a tool for personal audit and adds to the body of literature regarding URK demonstrating excellent long-term results. Although many factors should be taken into account when considering joint replacement including patient, technical and rehabilitative factors, we feel that this paper will enable clinicians, as it has our practice, to give patients presenting with medial compartment osteoarthritis of the knee comprehensive, long-term information necessary to make an informed decision.

Conclusions

Unicompartmental knee replacement is a very successful procedure that has undergone a number of changes over the years to improve patient outcomes and enable less invasive implantation. Patients can be advised their UKR has a 78% chance of surviving 15 years and a 72% chance of surviving 20 years without further surgery. The chances of excellent or good knee function are 91%. If further surgery is necessary, there is a 28% chance of leaving a functioning UKR intact and a 72% chance of needing conversion to a total knee. The most common reason for further surgery leaving the UKR intact has been to replace a worn or dislocated meniscal bearing, which has an overall chance of 1.5%. The most common cause of needing to convert the UKR to a TKR has been progression of arthritis in the remainder of the knee. Traumatic injury to the UKR can result in the need for further surgery and the chances of this are 2%. The risks of infection and perioperative mortality within the first 2 weeks are both 0.25%. Over time there have been changes to instrumentation and prosthesis design to allow minimally invasive surgery, which has not resulted in an increase in complications.

COMPETING INTERESTS
These authors have no relevant financial or non-financial interests to disclose.

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REFERENCES
1. Ahlbäck S. Osteoarthrosis of the knee A radiographic investigation. Acta Radiol Diagn (Stockh). 1968:Suppl 277:7-72.
2. Wiik AV, Manning V, Strachan RK, et al. Unicompartmental knee arthroplasty enables near normal gait at higher speeds, unlike total knee arthroplasty. J Arthroplasty. 2013;28(9 Suppl):176-8. doi: 10.1016/j.arth.2013.07.036.
3. Hooper N, Snell D, Hooper G, et al. The five-year radiological results of the uncemented Oxford medial compartment knee arthroplasty. Bone Joint J. 2015;97-B(10):1358-63. doi: 10.1302/0301-620X.97B10.35668.
4. Di Martino A, Bordini B, Barile F, et al. Unicompartmental knee arthroplasty has higher revisions than total knee arthroplasty at long term follow-up: a registry study on 6453 prostheses. Knee Surg Sports Traumatol Arthrosc. 2021 Oct;29(10):3323-3329. doi: 10.1007/s00167-020-06184-1.
5. Wilson HA, Middleton R, Abram SGF, et al. Patient relevant outcomes of unicompartmental versus total knee replacement: systematic review and meta-analysis. BMJ. 2019;364:l352. doi: https://doi.org/10.1136/bmj.l352.
6. Bretthauer M, Kalager M. What is my risk, doctor? How to convey disease risk and treatment effects. BMJ. 2023;381:e075289. doi: 10.1136/bmj-2022-075289.
7. Argenson JN, Parratte S. Unicompartmental Knee Replacement: a European perspective. 5th ed. Churchill Livingstone; 2011.
8. Goodfellow JW, Tibrewal SB, Sherman KP, O'Connor JJ. Unicompartmental Oxford Meniscal knee arthroplasty. J Arthroplasty. 1987;2(1):1-9. doi: 10.1016/s0883-5403(87)80025-6.
9. Kalairajah Y, Azurza K, Hulme C, et al. Health outcome measures in the evaluation of total hip arthroplasties--a comparison between the Harris hip score and the Oxford hip score. J Arthroplasty. 2005;20(8):1037-41. doi: 10.1016/j.arth.2005.04.017.
10. Weale AE, Newman JH. Unicompartmental arthroplasty and high tibial osteotomy for osteoarthrosis of the knee. A comparative study with a 12- to 17-year follow-up period. Clin Orthop Relat Res. 1994;(302):134-7.
11. Jansen K, Beckert M, Deckard ER, et al. Satisfaction and Functional Outcomes in Unicompartmental Compared with Total Knee Arthroplasty: Radiographically Matched Cohort Analysis. JB JS Open Access. 2020;5(3):e20.00051. doi: 10.2106/JBJS.OA.20.00051.
12. Liddle AD, Judge A, Pandit H, Murray DW. Adverse outcomes after total and unicompartmental knee replacement in 101,330 matched patients: a study of data from the National Joint Registry for England and Wales. Lancet. 2014;384(9952):1437-45. doi: 10.1016/S0140-6736(14)60419-0.
13. McKie J, Hobbs T, Frampton C, et al. Twenty-two year report: January 1999 to December 2020 [Internet]. The New Zealand Joint Registry. 2021 Dec [cited 2023 Jun 1]. Available from: https://www.nzoa.org.nz/sites/default/files/NZJR_22_Year_Report_Final.pdf.
14. Yang S, Hadlow S. Unicompartmental knee arthroplasty: is it durable? N Z Med J. 2003;116(1183):U627.
15. Squire MW, Callaghan JJ, Goetz DD, et al. Unicompartmental knee replacement. A minimum 15 Year followup study. Clin Orthop Relat Res. 1999;(367):61-72.
16. Pandit H, Hamilton T, Jenkins C, et al. The clinical outcome of minimally invasive

Phase 3 Oxford unicompartmental knee arthroplasty: a 15-year follow-up of 1000 UKAs. Bone Joint J. 2015;97-B(11):1493-500. doi: 10.1302/0301-620X.97B11.35634.
17. Ma J, Yan Y, Wang W, et al. Lower early revision rates after uncemented Oxford Unicompartmental Knee Arthroplasty (UKA) than cemented Oxford UKA: A meta-analysis. Orthop Traumatol Surg Res. 2021;107(3):102802. doi: 10.1016/j.otsr.2021.102802.
18. Foran JR, Brown NM, Della Valle CJ, et al. Long-term survivorship and failure modes of unicompartmental knee arthroplasty. Clin Orthop Relat Res. 2013;471(1):102-8. doi: 10.1007/s11999-012-2517-y.
19. Müller PE, Pellengahr C, Witt M, et al. Influence of minimally invasive surgery on implant positioning and the functional outcome for medial unicompartmental knee arthroplasty. J Athroplasty. 2004;19(3):296-301. doi: 10.1016/j.arth.2003.09.013.
20. Willis-Owen CA, Brust K, Alsop H, et al. Unicondylar knee arthroplasty in the UK National Health Service: an analysis of candidacy, outcome and cost efficacy. Knee. 2009;16(6):473-8. doi: 10.1016/j.knee.2009.04.006.
21. Cartier P, Sanouiller JL, Grelsamer RP. Unicompartmental knee arthroplasty surgery. 10-year minimum follow-up period. J Arthroplasty. 1996;11(7):782-8. doi: 10.1016/s0883-5403(96)80177-x.
22. Mohammad HR, Strickland L, Hamilton TW, Murray DW. Long-term outcomes of over 8,000 medial Oxford Phase 3 Unicompartmental Knees—a systematic review. Acta Orthop. 2018;89(1):101-7. doi: 10.1080/17453674.2017.1367577.
23. Price AJ, Svard U. A second decade lifetable survival analysis of the Oxford unicompartmental knee arthroplasty. Clin Orthop and Relat Res. 2011;469(1):174-9. doi: 10.1007/s11999-010-1506-2.
24. Te Kāwanatanga o Aotearoa – New Zealand Government. WorkSafe New Zealand [Internet]. 2020 [cited 2023 1 Jul]. Available from: https://www.govt.nz/organisations/worksafe-new-zealand/.

Using electronic health records in analysing medication adherence in southern New Zealand patients with inflammatory bowel diseases

Obreniokibo I Amiesimaka, Kristina Aluzaite, Rhiannon Braund, Michael Schultz

ABSTRACT

AIMS: Electronic health records (EHRs) are widely used in medication adherence (MA) assessment. Poor adherence in patients with inflammatory bowel diseases (IBD) can lead to worse disease outcomes and increased health costs. This study explores the suitability of southern New Zealand EHRs for estimating adherence, and the relationship between adherence and corticosteroid dispensings (indicating negative disease outcomes).

METHODS: Medication dispensing EHR data of former Southern District Health Board IBD patients were analysed to estimate 3-year adherence, using daily polypharmacy possession ratio. The correlation with the number of corticosteroid dispensings was investigated.

RESULTS: Of 248/1,290 (19%) consenting patients, only 108/248 (44%) had sufficient data available (46%/54% Crohn's disease/ulcerative colitis; 57% female; 89.8%/0.9% NZ European/Māori; mean 5.1 corticosteroid dispensings).

Mean adherence was 83.2% (95% confidence interval [CI] 80.0–86.4; standard deviation [SD]:16.7), with 69% of patients having MA ≥80% (good adherence). Median adherence was 13% higher for males versus females (96% vs 83%; p=0.0001). There was no correlation between adherence and the number of corticosteroid dispensings (Pearson's r=0.11; p>0.05). These findings should be considered with caution as the data were not obtained from all pharmacies and the quantum/nature of missing data is unknown.

CONCLUSIONS: The patients' adherence seems high, with no correlation with corticosteroid dispensings demonstrated. Useful EHR data are available but need optimisation for adherence assessments.

Inflammatory bowel diseases (IBD) are chronic diseases consisting of Crohn's disease (CD), ulcerative colitis (UC) and IBD-unclassified (IBD-U) and are typified by intestinal inflammation, and often by extra-intestinal manifestations.[1–4] A life-long medication regimen is central to IBD therapy alongside dietary/surgical/other interventions, besides ongoing monitoring for disease progression and complications.[1–4] Medication adherence (MA) is defined as "*the process by which patients take their medication as prescribed*" and comprises initiation, implementation and persistence/discontinuation (starting, continuing and ending a regimen as recommended).[5] Poor adherence is associated with negative disease outcomes in IBD including higher morbidity and mortality, and disability and health costs, alongside flare-ups and complications requiring therapy escalation (involving surgery and/or recurring corticosteroid use).[6] Adherence levels of patients with IBD have previously been reported as 31.1%[7] and 22.9%–77%[6,8,9] in southern Aotearoa New Zealand and abroad.

With an estimated yearly growth rate of 5.6%, IBD burden in Aotearoa New Zealand is poised to double by 2028 (to over 40,000) from 20,792 patients in 2016, with combined direct and indirect annual costs of NZ$245 million.[2] In Aotearoa New Zealand, access to advanced IBD therapy is restricted as several medications (e.g., biologics/Janus kinase inhibitors) available in other countries are not publicly funded.[10] Consequently, ensuring sufficient adherence is important for helping Aotearoa New Zealand IBD patients derive maximum benefits from available medications. Also, monitoring adherence can identify patients with low adherence levels for targeted interventions. A literature search identified only one study that has assessed adherence in Aotearoa New Zealand (Southern District Health Board [SDHB]) patients with IBD;[7] using self-reported survey tools, this study found that a third of patients had poor adherence.

Electronic health records (EHRs) including claims databases, commonly collected primary/secondary health databases, disease registries and others[11] often contain readily available, detailed,

cross-linked and population-wide data and are widely employed in assessing adherence[12,13] as they could provide more objective evaluations of adherence than self-reported measures. There are several publicly/privately owned EHRs used in different parts of the Aotearoa New Zealand health system, with some holding directly input data and others collating data from other EHRs. Individual patients' data are linked via a unique National Health Index (NHI) code across the databases, which include prescription, dispensing, hospitalisation and clinical information, among others. An NHI number is allocated to an individual at birth (or at first time accessing healthcare). This number is used at every healthcare event that allows comprehensive healthcare utilisation to be possible.

While nationwide databases exist, including hospitalisation data (both events and discharge information) and medication dispensing (claims) data,[14,15] the development of subscription-based EHRs that pool centrally held data with primary care subscribers (i.e., general practices and pharmacies) allow subscribers the ability to see a patient's journey more holistically and reduces some of the problems that occur with transition of care. The utilisation of shared EHRs could be useful in assessing patients' adherence. Therefore, the objective of this study was to determine whether the available subscription-based EHRs used by primary care practitioners in southern Aotearoa New Zealand provided suitable and sufficient information for adherence research by conducting the steps of calculating the adherence levels of local IBD patients and investigating the association between adherence and corticosteroid prescriptions (a proxy for negative health outcomes as corticosteroids are often used for managing IBD flare-ups[1,3]).

Methods

Study region

Until 30 June 2022, the SDHB was the regional health authority for the Otago and Southland Regions of Aotearoa New Zealand. Now superseded by the national Te Whatu Ora – Health New Zealand service, the SDHB patients still reside and/or receive healthcare within these regions, which are the settings for this study.

Study design

This was a retrospective longitudinal population-wide study, utilising dispensing data of IBD patients within the study region from 1 January 2015 to 31 December 2019, curated from a patient management database (PMD; detailed below) and used to calculate adherence levels. The adherence levels were calculated using a daily polypharmacy possession ratio[13] (DPPR; details provided below) for the first 3 years from the earliest dispensing (*DispDx*) of select chronic ongoing medications of interest for patients with at least 3 years of dispensings. The association between DPPR and the number of corticosteroid dispensings was also evaluated.

Ethics approval

Ethics approval was granted by the University of Otago Human Ethics Committee (Health) (H21/171).

Participant recruitment

The research population comprises patients (≥18yrs) with IBD residing in the area of the former SDHB. Patients hospitalised in the SDHB with an IBD International Statistical Classification of Diseases and Related Health Problems, 10th Revision (ICD-10)[16] code between 1 January 2015 and 31 December 2019 (see Appendix Table 1) were identified from SDHB coding records. The following three patient management databases used in the SDHB, EpiSoft (Episoft Australia),[17] Crohn's and Colitis Care (CCCare) (Crohn's Colitis Cure Australia)[18] and Health Connect South (HCS) (Orion Health New Zealand),[19] were used *solely* to *confirm* the IBD diagnoses and addresses of the identified patients. These three databases are used in the SDHB and contain patients' demographic data alongside disease information, clinic appointments, care plans, clinical reports/letters and more. Patients with a confirmed diagnosis of IBD were invited to participate (and provide informed consent) via post and email.

Data curation

The pharmacy dispensing data for specific medications of interest (chronic ongoing IBD medicines, namely: thiopurines—azathioprine and 6-mercaptopurine (6-MP); methotrexate; 5-aminosalicylic acid therapeutics—sulfasalazine and mesalazine, and adalimumab; alongside corticosteroids—budesonide, prednisone and hydrocortisone acetate) were curated from the PMD for consenting patients. The PMD is owned by a public-private partnership. The PMD contains health information, by NHI, for patients within the South Island of Aotearoa New Zealand, including on medications dispensed and

prescribed at community pharmacies and healthcare providers, e.g., general practices (GPs), who subscribe to the network.

The following data were extracted and curated manually per patient from the PMD: medications dispensed (name and formulation); administration instructions (wherefrom the dose, frequency and daily dose were derived); quantity dispensed and the date of dispensing. The days' supply (DS) is the number of days with the daily dose available to the patient; DS was calculated as the quantity dispensed/daily dose. These variables were used to calculate MA to chronic IBD medications, chosen because they are self-administered, are commonly used for IBD therapy in Aotearoa New Zealand and are intended for continuous long-term use. Similar data were curated on the number of concurrent corticosteroid dispensings as, although not meant for consistent use, they are often used for managing IBD flare-ups.[1,3]

The consolidated dataset compiled contained patients' dispensing data derived from the PMD and their demographic data from HCS.

Data analysis

DPPR is used to calculate adherence for multiple-medicine regimens, alongside single-medicine regimens, and has been used in calculating adherence via secondary databases e.g., EHRs.[12,13,20] This equation shows the calculation:

$$DPPR = \frac{\sum(medication\ availability\ score\ per\ interval \times days\ per\ interval)}{observation\ window}$$

The numerator for the DPPR equation is the cumulative medication availability score per interval multiplied by the days per interval, and the denominator is the days in the observation window (OW). When a patient had an overlapping DS of the same medication, which occurs when refills are collected before current DS is exhausted, the second DS is considered to have started at the end of the first DS. A gap between two consecutive DS for a medication greater than 180 days was taken to indicate the discontinuation of that medicine. The "CMA_polypharmacy" function in the AdhereR package version 0.8.1[21] in R was used in computing the DPPR. The Appendix (Appendix Figure 1 and associated text) shows an illustration of the DPPR calculation and more details of the data analysis. Appendix Table 1 shows more detail of the ICD-10 codes and medications used as part of the analysis.

Participants are described by counts and proportions expressed as percentages. The association between DPPR scores and sex, IBD type and age group used a Wilcoxon test. The association between DPPR scores and corticosteroid dispensing was summarised by a product moment correlation coefficient. Appendix Table 2 shows more detail of the analyses conducted.

Results

Study sample

The flow of participants through the study is shown in Figure 1.

There were 108 participants who were included in the final analysis of the DPPR. This is 8% of the 1,290 IBD patients who were invited to participate in the study, although 31.2% (402/1,290) patients initially responded to the invitations, and 19% (248/1,290) consented to the study (Figure 1).

The demographics (Table 1) show that 46% (n=50) of the study population had CD and 54% (n=58) had UC, 43% (n=46) were male, 89.8% (n=97) were NZ European, 4.6% (n=5) were Other European and 0.9% (n=1) were Māori, while the median (interquartile range [IQR]) age at first dispensing was 52 years (39–62yrs; Table 1).

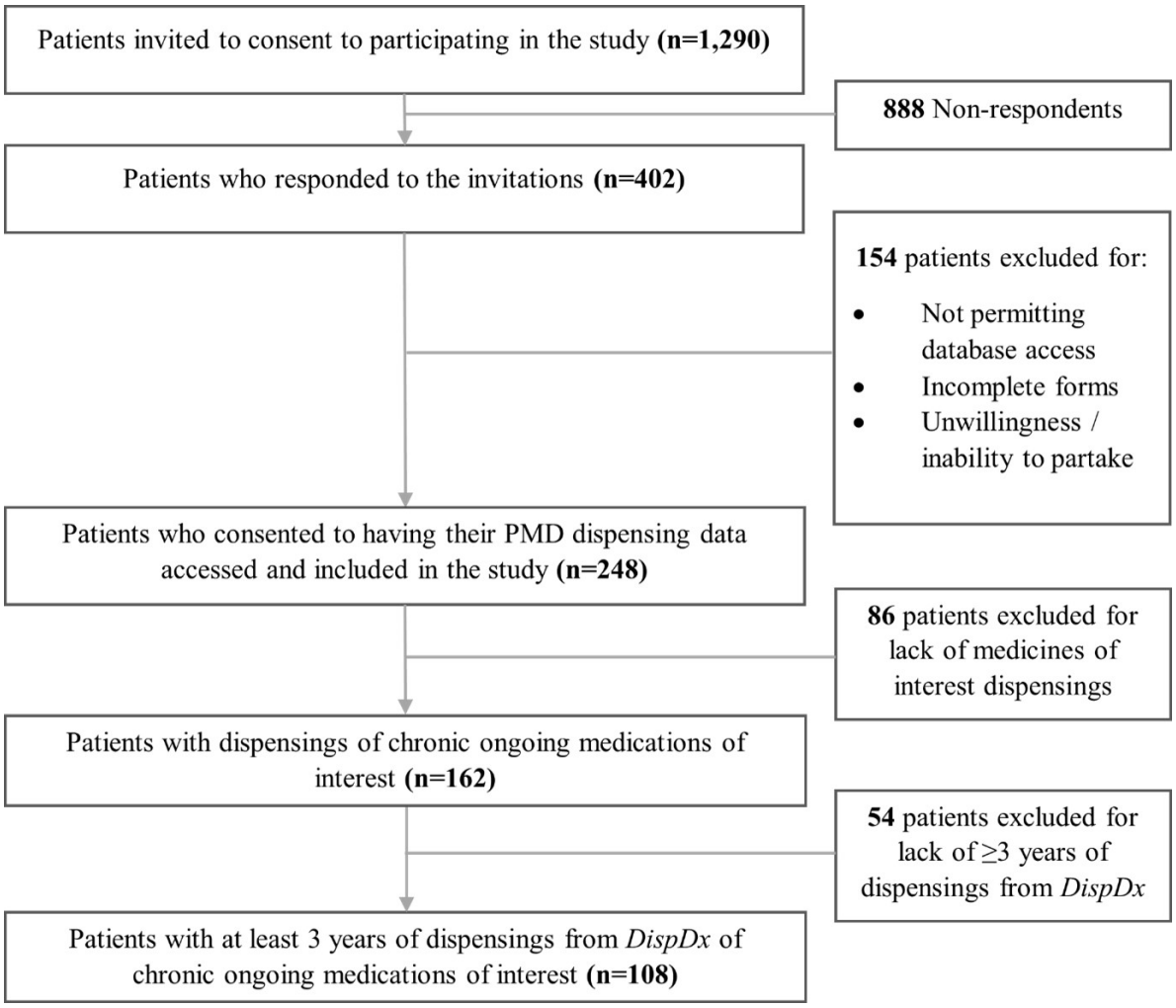
Each patient had mean (standard deviation [SD]) 35.5 (28.3) chronic medicine dispensings. Forty-six percent (n=50) of patients had corticosteroid dispensings; of these, each had mean 5.1 (7.1) corticosteroid dispensings. Regarding chronic medication type, 18.5% (n=20) of patients used three to four, 36.1% (n=39) of patients used two and 45.4% (n=49) of patients used one during the period. Mesalazine, mercaptopurine, methotrexate, sulfasalazine, adalimumab and azathioprine were used by 70% (n=76), 11% (n=12), 7% (n=8), 5% (n=5), 16% (n=17) and 37% (n=40) of patients, respectively (Table 1).

Indicative DPPR value and relationship with clinical outcomes

The mean DPPR (%) estimate for the study population was 83.2% (95% confidence interval [CI] 80.0–86.4; SD 16.7), with 69% of patients having a DPPR at or above the ≥80% threshold of good adherence.[6] The median DPPR (%) scores were 13% higher for males versus females (96% vs 83%; p=0.0001), 1% higher for UC versus CD (88% vs 87%; p=0.2307) and 6% higher for ≥65 year olds versus those <65 years (92% vs 86%; p=0.3736).

There was no statistically significant

Figure 1: Patient flow diagram.



Abbreviations: DispDx = date of earliest dispensing of a chronic medication of interest; PMD = patient management database.

Table 1: Characteristics of patients and dispensing events in the study population dataset.

Patient characteristics	**n/108 (%)**		
Total patients	108		
Patients with steroid dispensings	50 (46)		
Patients per IBD type			
CD	50 (46)		
UC	58 (54)		
Sex			
Female	62 (57)		
Male	46 (43)		
Prioritised ethnicity			
NZ European	97 (89.8)		
Other European	5 (4.6)		
NZ Māori	1 (0.9)		
Others	5 (4.6)		
Number of chronic medications			
3–4	20 (18.5)		
2	39 (36.1)		
1	49 (45.4)		
Medication			
Mesalazine	76 (70)		
Azathioprine	40 (37)		
Adalimumab	17 (16)		
Mercaptopurine	12 (11)		
Methotrexate	8 (7)		
Sulfasalazine	5 (5)		
	Mean (SD)	**Median (IQR)**	**Min–Max**
Age at first dispensing	50.3 (16.2)	52 (38.5–62)	14–81
Chronic medicine dispensings per patient	35.5 (28.3)	34 (19–39)	7–218
Corticosteroid dispensings per patient*	5.1 (7.1)	2 (1.25–5.75)	1–42

CD = Crohn's disease; UC = ulcerative colitis; n = number of patients.
* For the 50 patients with corticosteroid dispensings.

(Pearson's r= 0.11; p>0.05) correlation or association between DPPR and the number of corticosteroid dispensings per patient in the concurrent period (see Appendix Table 2).

Discussion

This study investigated the availability of EHRs for computing long-term MA of IBD patients residing in the area of the former SDHB in Aotearoa New Zealand. We excluded 35% (n=86/248) of patients' data as they had no dispensings of our chronic medications of interest recorded (Figure 1); data collation challenges would have contributed to this. The PMD used is a patient management system with data collected recording patients' health histories to aid clinical decision making, but these were not intended for adherence assessments. Hence, we suggest ways by which it might be improved for evaluating adherence.

Accurate adherence assessment requires EHRs that record all relevant details (including time/quantity/medicine type etc.) of patients' medications for all dispensings and prescriptions.[22] Currently, the dispensing and prescription data is automatically pooled from community pharmacy and healthcare provider/prescriber (e.g., GPs) systems respectively, for (nearly) all such vendors in the South Island of Aotearoa New Zealand, including the former SDHB region. This automatically excludes dispensings from other Aotearoa New Zealand regions, highlighting the need for a nation-wide database. All vendors need to be connected to the database network with newly opened vendors onboarded timeously. Dispensing and prescription data from public hospitals or hospital pharmacies are not collected, with limited contributions from private sector healthcare providers. Furthermore, the paucity of prescription data precluded the matching of prescriptions with dispensings (to highlight missing dispensings) and the assessment of *initiation* in adherence (primary non-adherence, i.e., a patient's failure to begin a medication regimen).[5] Linking prescriptions with the dispensing(s) in the EHRs—perhaps via a unique code—would allow better study and tracking of adherence by researchers and clinicians. As it is unclear what proportion of patients the foregoing affected, the quantum and nature of missing data are unknown; hence, the findings stated above should be treated with caution as they are for the patient sample with data available. Consequently, this valuable resource, containing the bulk of the needed data, needs optimisation for more reliable adherence assessments. Seemingly recognising this, the database disclaims assurance of the accuracy/completeness/reliability of the data notwithstanding best efforts in data collation.

Calculating adherence also requires EHR data presentation in an accessible format. The DS—essential for computing adherence—for each dispensing should ideally be contained in a consistent (numeric) format to limit room for misinterpretation and human error. Presently, DS is not provided and must be manually calculated by dividing the stated medication quantity dispensed by the daily dose, which itself is derived from the statement-formatted administration instructions available. These instructions/statements are sometimes non-specific or describe tapering/incremental doses requiring complex calculations to deduce the DS. Besides, the database should ideally allow for searches of data by specific dates and the time point of prescriber-advised medication discontinuations should be highlighted.

Although patients can purposely have their data excluded from the database, there was no indication of this for this study's participants and the overall number of patients opting off is thought to be infinitesimal. Moreover, there appears to have been a material increase in dispensing and prescription recordings on the database since 2020 (after our study period), driven by the adoption of digital health services since the start of the COVID-19 pandemic; the impact of this on adherence evaluation requires serious assessment.

For the 108 consenting patients with available data, their 3-year mean DPPR,[13] a new adherence measure, was high at 83.2%. Moreover, 69% of patients had a DPPR ≥80%, the general threshold of good adherence.[6] We consider these results to be indicative only and they should be considered with caution due to data quality factors discussed above. To our knowledge, no other studies have evaluated adherence in patients with IBD taking multiple concurrent medications (polypharmacy) by calculating DPPR. Other studies (cited below) have calculated adherence to single-medicine regimens using other adherence measures, e.g., medication possession ratio (MPR) and/or proportion of days covered (PDC) etc. Studies investigating IBD patients' polypharmacy adherence[23] commonly use self-reported surveys, which yield more subjective adherence estimates than those using EHRs. Moreover, the medicines for which adherence is calculated differ between studies.

These heterogeneities limit comparison of adherence results.

Using measures including MPR/PDC, 38–77%[6,24] of patients with IBD have been reported as adherent (MA ≥80%) to biologic/anti-tumor necrosis factor (anti-TNF) medicines. Likewise, 22.9%[9] and 60%[25] of IBD patients have been classed as non-adherent to non-biologic medicines e.g., thiopurines, 5-aminosalicylic acid (5-ASA) drugs etc. Other studies from southern Aotearoa New Zealand,[7] the Netherlands,[24] Australia and the United Kingdom,[8] using self-reported surveys, report that 31.1% of IBD patients had "below medium" adherence, 76% had medium and low adherence and 28.7% were non-adherent. Findings of other publications report associations between female gender and lower IBD adherence;[26] these align with our results showing that male patients had a statistically significantly higher DPPR than female patients.

Forty-six percent of patients had corticosteroid dispensings in the 3-year period, which compares to United States CD patients, 40% of whom used corticosteroids within a year from starting infliximab use.[27] In the present study, the correlation between DPPR and the number of concurrent corticosteroid dispensings (a clinical outcome indicator, as corticosteroids are used for managing IBD flare-ups[1,3]) was not statistically significant. Further research could consider if DPPR predicts future steroid dispensings, although literature posits that chronic disease DPPR does not predict future hospitalisations,[12,20] another indicator of clinical outcomes. There is, however, little consensus about the appropriate lag period parameters for determining when adherence might impact the outcomes.[12,20] We counted the corticosteroid dispensings within the same period as DPPR was calculated to maximise patient inclusion, as using a lag period would have meant excluding patients with shorter durations of total data contribution. Furthermore, as a heterogenous mix of medicines of interest (each therapeutic may have different effects on disease outcomes and adherence per medication may be different[28]) and study participants (of varying ages, disease statuses/severities etc.) were included, these could have also contributed to this outcome. Moreover, corticosteroids could have been prescribed for comorbidities, rather than for IBD. The lack of readily available data precluded analysis of these factors.

The insights gained from using the SDHB EHRs in this study align broadly with those addressed in literature.[11,29] Although adherence can be calculated from the SDHB PMD, the aforementioned issues limit the reliability of the results. Therefore, to be best optimised for adherence computation, the database should pool real-time dispensing and prescription data from all primary to tertiary healthcare providers/vendors across the public and private health sectors. Also, the data should be provided with the DS calculated and presented in a numeric format with the database being searchable by date.

Nonetheless, we recognise that EHRs of dispensing/prescription data, no matter how complete or accurate, are secondary databases and might not necessarily reflect patients' medication-taking behaviours. For instance, patients might increase or decrease their dosage depending on their IBD disease activity, as some prescribers recommend due to its linkage with better health outcomes in diverse chronic diseases.[30] Notwithstanding, EHRs like those available in the SDHB are a valuable resource for adherence assessment both for research and patient support purposes. Moreover, patients who did not consent to partaking in this study may have been aware of their non-adherence, which could have contributed to the high adherence we found among the consenting patients.

Dispensing/prescription datasets exist in the now-defunct SDHB region, the PMD especially, which indicate that adherence for IBD patients is high. Nonetheless, optimisation in data collation is needed to improve the data quality for more accurate adherence calculations.

COMPETING INTERESTS
The authors have no conflicts of interest to declare.
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REFERENCES
1. Gastroenterological Society of Australia (GESA). Clinical Update for General Practitioners and Physicians: Inflammatory Bowel Disease [Internet]. GESA; 2018 [cited 2023 Dec 11]. Available from: https://www.gesa.org.au/public/13/files/Education%20%26%20Resources/Clinical%20Practice%20Resources/IBD/2018_IBD_Clinical_Update_May_update.pdf.
2. Kahui S, Snively S, Ternent M. Reducing the Growing Burden of Inflammatory Bowel Disease in New Zealand [Internet]. Wellington: Crohn's and Colitis New Zealand Charitable Trust; 2017 Nov [cited 2023 Dec 11]. Available from: https://issuu.com/crohnsandcolitisnz/docs/271017_master_formatted_bod_report_.
3. Eliadou E, Day AS, Thompson-Fawcett MW, et al. New Zealand Society of Gastroenterology Guidelines for the Management of Refractory Ulcerative Colitis. N Z Med J. 2015;128(1423):63-76.
4. Matsuoka K, Kobayashi T, Ueno F, et al. Evidence-based clinical practice guidelines for inflammatory bowel disease. J Gastroenterol. 2018;53(3):305-53. doi: 10.1007/s00535-018-1439-1.
5. Dima AL, Allemann SS, Dunbar-Jacob J, et al. TEOS: A framework for constructing operational definitions of medication adherence based on Timelines-Events-Objectives-Sources. Br J Clin Pharmacol. 2021;87(6):2521-33. doi: 10.1111/bcp.14659.
6. Khan S, Rupniewska E, Neighbors M, et al. Real-world evidence on adherence, persistence, switching and dose escalation with biologics in adult inflammatory bowel disease in the United States: A systematic review. J Clin Pharm Ther. 2019;44(4):495-507. doi: 10.1111/jcpt.12830.
7. Aluzaite K, Braund R, Seeley L, et al. Adherence to Inflammatory Bowel Disease Medications in Southern New Zealand. Crohns Colitis 360. 2021;3(3):otab056. doi: 10.1093/crocol/otab056.
8. Selinger CP, Eaden J, Jones DB, et al. Modifiable factors associated with nonadherence to maintenance medication for inflammatory bowel disease. Inflamm Bowel Dis. 2013;19(10):2199-206. doi: 10.1097/MIB.0b013e31829ed8a6.
9. Selinger CP, Ochieng AO, George V, Leong RW. The Accuracy of Adherence Self-report Scales in Patients on Thiopurines for Inflammatory Bowel Disease: A Comparison With Drug Metabolite Levels and Medication Possession Ratios. Inflamm Bowel Dis. 2019;25(5):919-24. doi: 10.1093/ibd/izy309.
10. Amiesimaka O, Braund R, Aluzaite K, Schultz M. Constraints on medication-based inflammatory bowel disease therapy in Aotearoa New Zealand - why medication adherence is important. N Z Med J. 2023;136(1574):82-9.
11. Farmer R, Mathur R, Bhaskaran K, et al. Promises

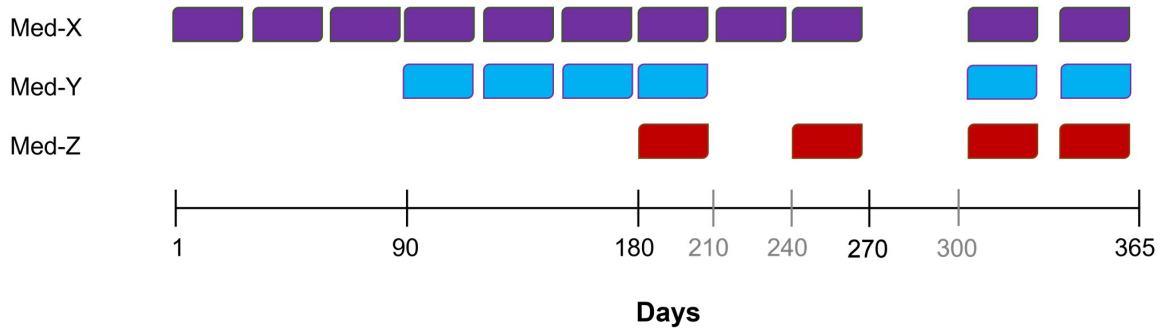
and pitfalls of electronic health record analysis. Diabetologia. 2018;61(6):1241-8. doi: 10.1007/s00125-017-4518-6.
12. Franchi C, Ludergnani M, Merlino L, et al. Multiple Medication Adherence and Related Outcomes in Community-Dwelling Older People on Chronic Polypharmacy: A Retrospective Cohort Study on Administrative Claims Data. Int J Environ Res Public Health. 2022;19(9):5692. doi: 10.3390/ijerph19095692.
13. Arnet I, Greenland M, Knuiman MW, et al. Operationalization and validation of a novel method to calculate adherence to polypharmacy with refill data from the Australian pharmaceutical benefits scheme (PBS) database. Clin Epidemiol. 2018;10:1181-94. doi: 10.2147/CLEP.S153496.
14. Te Whatu Ora – Health New Zealand. National Minimum Dataset (hospital events) [Internet]. 2019 [cited 2023 Dec 11]. Available from: https://www.tewhatuora.govt.nz/our-health-system/data-and-statistics/nz-health-statistics/national-collections-and-surveys/collections/national-minimum-dataset-hospital-events.
15. Te Whatu Ora – Health New Zealand. Pharmaceutical Collection [Internet]. 2022 [cited 2023 Dec 11]. Available from: https://www.tewhatuora.govt.nz/our-health-system/data-and-statistics/nz-health-statistics/national-collections-and-surveys/collections/pharmaceutical-collection.
16. The International Statistical Classification of Diseases and Related Health Problems 10th Revision (ICD-10)-2014-WHO Version for ;2014 [Internet]. World Health Organization; 2014 [cited 2020 Aug 19]. Available from: https://icd.who.int/browse10/2014/en#/K50.0.
17. EpiSoft. Our Story [Internet]. Sydney, Australia: EpiSoft; 2019 [cited 2023 Dec 11]. Available from: https://episofthealth.com/our-story/.
18. Crohn's Colitis Cure. About Crohn's Colitis Care [Internet]. Pyrmont, Australia: CCCure; 2022 [cited 2023 Dec 11]. Available from: https://c-c-cure.org/cccare/.
19. Orion Health. South Island health system closes information gaps [Internet]. Auckland: Orion Health; 2017 Feb 13 [cited 2023 Dec 11]. Available from: https://orionhealth.com/nz/media-releases/south-island-health-system-closes-information-gaps/.
20. Zongo A, Grégoire JP, Moisan J, Guénette L. Measuring adherence to oral antidiabetic multi-drug treatment: Comparative validity of prescription claims-based adherence measures against hospitalization. Res Social Adm Pharm. 2019;15(6):738-43. doi: 10.1016/j.sapharm.2018.09.005.
21. Dima AL, Dediu D, Allemann S. Package 'AdhereR'. 2022 [cited 2023 Dec 11]. Available from: https://cran.r-project.org/web/packages/AdhereR/AdhereR.pdf.
22. Dima AL, Dediu D, Allemann S. AdhereR: Adherence to Medications [Internet]. 2022 [cited 2023 Dec 11]. Available from: https://cran.r-project.org/web/packages/AdhereR/vignettes/AdhereR-overview.html.
23. Pednekar PP, Ágh T, Malmenäs M, et al. Methods for Measuring Multiple Medication Adherence: A Systematic Review-Report of the ISPOR Medication Adherence and Persistence Special Interest Group. Value Health. 2019;22(2):139-56. doi: 10.1016/j.jval.2018.08.006.
24. van der Have M, Oldenburg B, Kaptein AA, et al. Non-adherence to Anti-TNF Therapy is Associated with Illness Perceptions and Clinical Outcomes in Outpatients with Inflammatory Bowel Disease: Results from a Prospective Multicentre Study. J Crohns Colitis. 2016;10(5):549-55. doi: 10.1093/ecco-jcc/jjw002.
25. Samson CM, Mager D, Frazee S, Yu F. Remission in Pediatric Inflammatory Bowel Disease Correlates With Prescription Refill Adherence Rates. J Pediatr Gastroenterol Nutr. 2017;64(4):575-9. doi: 10.1097/MPG.0000000000001304.
26. Fidder HH, Singendonk MM, van der Have M, et al. Low rates of adherence for tumor necrosis factor-alpha inhibitors in Crohn's disease and rheumatoid arthritis: results of a systematic review. World J Gastroenterol. 2013;19(27):4344-50. doi: 10.3748/wjg.v19.i27.4344.
27. Carter CT, Waters HC, Smith DB. Effect of a continuous measure of adherence with infliximab maintenance treatment on inpatient outcomes in Crohn's disease. Patient Prefer Adherence. 2012;6:417-26. doi: 10.2147/PPA.S31115.
28. Tomar SK, Kedia S, Singh N, et al. Higher education, professional occupation, and upper socioeconomic status are associated with lower adherence to medications in patients with inflammatory bowel disease. JGH Open. 2019;3(4):302-9. doi: 10.1002/jgh3.12160.
29. Park LG, Howie-Esquivel J, Dracup K. Electronic measurement of medication adherence. West J Nurs Res. 2015;37(1):28-49. doi: 10.1177/0193945914524492.
30. Dineen-Griffin S, Garcia-Cardenas V, Williams K, Benrimoj SI. Helping patients help themselves: A systematic review of self-management support strategies in primary health care practice. PLoS One. 2019;14(8):e0220116. doi: 10.1371/journal.pone.0220116.

Appendix

Appendix Table 1: ICD-10 codes and medications of interest used for patient selection.

ICD-10 IBD Code		**Description**
K500		Crohn's disease of small intestine
K501		Crohn's disease of large intestine
K508		Other Crohn's disease
K509		Crohn's disease, unspecified
K510		Ulcerative (chronic) pancolitis
K512		Ulcerative (chronic) proctitis
K513		Ulcerative (chronic) rectosigmoiditis
K514		Inflammatory polyps
K515		Left-sided colitis
K518		Other ulcerative colitis
K519		Ulcerative colitis, unspecified
K523		Indeterminate colitis
Chronic medications used in IBD therapy[1,2]		
1.		Methotrexate
2.	Thiopurines	Azathioprine
3.		6-Mercaptopurine (6-MP)
4.		Adalimumab (Humira)
5.	5-Aminosalicylic acid (5-ASA) drugs	Sulfasalazine
6.		Mesalazine
Corticosteroid medicines		
7.		Budesonide
8.		Prednisone
9.		Hydrocortisone acetate

Appendix Figure 1: Illustrative pattern of dispensings for a patient with three medications dispensed (Med-X, Y, Z). Each box represents days' supply (DS) for that medication. Intervals are marked by the stated days. Medicines were assumed to be prescribed for continuous use from initiation until the end of the total period.



$$DPPR = \frac{([1/1 \times 90]_{\text{days 1-90}} + [2/2 \times 90]_{\text{days 91-180}} + [3/3 \times 30]_{\text{days 181-210}} + [1/3 \times 30]_{\text{days 211-240}} + [2/3 \times 30]_{\text{days 241-270}} + [0/3 \times 30]_{\text{days 271-300}} + [3/3 \times 65]_{\text{days 301-365}})}{365} = 0.84 \times 100 = 84\%$$

Calculating daily polypharmacy possession ratio (DPPR): an illustration

The illustrative patient, in Appendix Figure 1 above, was first dispensed Med-X, Med-Y and Med-Z on days 1, 91 and 181; therefore, the availability score for the 181–365 interval could be 3/3 (if all medications were available), 2/3 (if just two medicines were available), 1/3 (if one medication was available) or 0/3 (if no medicine was available). Likewise, between days 91 and 181, the possible daily values were 2/2, 1/2 or 0/2; and for days 1–91 the possible scores were 1/1 or 0/1.

Analysis methods and software

The "CMA_polypharmacy" function in the AdhereR package version 0.8.1[3] in R was used in computing the DPPR. Furthermore, the results of the correlation analysis between average DPPR and the number of corticosteroid dispensings in the concurrent period per patient was validated using negative binomial regression (used to account for over-dispersion in the count variable).[3] All data preparation and analyses were conducted using Python 3.9.14,[4] R (v4.2.2-win).[5]

Analysis results

The strength of the association between DPPR and the number of corticosteroid dispensings was estimated using a negative binomial model (Appendix Table 2). The Pseudo R^2 (0.004) represents a very low model fit, validating the lack of a correlation, and strongly indicates that more explanatory variables are needed to better understand the association. Data on such explanatory variables (e.g., comorbidities, disease severity, urban/rural residence among several others), some of which would be confounders, was not readily available to include in the model. Besides, the data quality considerations detailed in the discussion could also have impacted this outcome. Hence, confounding could have caused the relationship to be under- or over-estimated, or even completely reversed (Simpson's paradox). Future research should consider obtaining and including relevant covariates in multivariable models for better goodness of fit, perhaps after DPPR is re-computed with higher-quality data.

Appendix Table 2: Pearson's correlation and relationship between DPPR and number of corticosteroid dispensings using negative binomial regression analysis.

Daily polypharmacy possession ratio (DPPR %) values	
Mean (SD)	83.2% (16.7)
Minimum	28.0%
Q1	76.0%
Median	88.1%
Q3	96.7%
Maximum	100.0%
DPPR and number of steroid dispensings	
Pearson's r	0.11
Cor p-value	0.45
IRR (95% CI)	2.04 (0.48–8.04)
P-value	>0.05
Pseudo R^2	0.004

Abbreviations: SD = standard deviation; IRR = incidence rate ratio; 95% CI = 95% confidence interval; Cor = correlation

REFERENCES

1. Gastroenterological Society of Australia (GESA). Clinical Update for General Practitioners and Physicians: Inflammatory Bowel Disease [Internet]. GESA; 2018 [cited 2023 Dec 11]. Available from: https://www.gesa.org.au/public/13/files/Education%20%26%20Resources/Clinical%20Practice%20Resources/IBD/2018_IBD_Clinical_Update_May_update.pdf.
2. Kahui S, Snively S, Ternent M. Reducing the Growing Burden of Inflammatory Bowel Disease in New Zealand [Internet]. Wellington: Crohn's and Colitis New Zealand Charitable Trust; 2017 Nov [cited 2023 Dec 11]. Available from: https://issuu.com/crohnsandcolitisnz/docs/271017_master_formatted_bod_report_.
3. Dima AL, Dediu D, Allemann S. Package 'AdhereR'. 2022 [cited 2023 Dec 11]. Available from: https://cran.r-project.org/web/packages/AdhereR/AdhereR.pdf.
4. Python Software Foundation. Python 3.9.14 documentation [Internet] 2022 [cited]. Available from: https://docs.python.org/3.9/.
5. R Core Team. R: A language and environment for statistical computing. Vienna, Austria: R Foundation for Statistical Computing; 2022 [cited 2023 Dec 11]. Available from: https://www.R-project.org/.

Auckland Regional Cleft Palate Service: service accessibility and speech outcomes

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ABSTRACT

AIM: This study explored speech outcomes for five-year-olds born with cleft palate residing in the Auckland Regional Cleft Palate Service catchment, with a focus on ethnicity and geographical influences on service accessibility and speech outcomes.

METHOD: A retrospective audit of children born with cleft palate between 2013–2016 was conducted (n=89), including secondary surgeries, ethnicity, distances travelled, services offered and attended. Speech outcomes at five years were compared to international benchmarking.

RESULTS: Seventy-nine children were included; 10 were lost to follow-up before their 5 years review. At 5 years, 30% children presented with moderate to severe velopharyngeal incompetence; 30% had residual cleft speech characteristics which warranted speech therapy intervention. There was no significant difference in speech outcomes for Māori vs non-Māori. Attendance at clinic was not significantly associated with distance from centre. However, Māori had significantly lower attendance at clinic appointments and were significantly more likely to be referred to community services.

CONCLUSION: Auckland Regional speech outcomes are better than the national average, yet still do not compare favourably with international benchmarking. While speech outcomes were not associated with ethnicity, attendance at specialist clinic appointments was lower in Māori. Community services were offered, but attendance at, and satisfaction with, these appointments is unknown. Further work is needed to reach all children regionally in a timely manner to ensure Aotearoa New Zealand speech outcomes meet international standards.

When a child is born with an orofacial cleft and lives between Cape Reinga in the far North and Mercer in the middle of the North Island, they fall under the care of the Auckland Regional Cleft Palate Service, based in South Auckland, Aotearoa New Zealand. The Auckland Regional Cleft Service consists of a multidisciplinary team (MDT) with a Clinical Nurse Specialist (CNS), Plastic Surgeons, Orthodontists, Health Psychologists, Otorhinolaryngologists (ORL) and Speech-language Therapists (SLT). Each profession plays a key role at differing times in the cleft journey. The CNS is a constant point of contact from diagnosis through to point of discharge. It is the CNS who provides support and education to whānau and links in with other community support services, where needed, to support attendance and engagement. All surgical procedures are carried out at Middlemore Hospital, or Starship Children's Hospital for tamariki with additional medical needs. Outpatient appointments are held at Middlemore Hospital or off-site in a community outpatient facility in South Auckland. Given the catchment area for this service, twice-yearly clinics are also held in Whangārei Hospital, Northland. Tamariki may access additional speech therapy through community child development services (<3 years old), at school (Ministry of Education funded, ~5 years+) and/or privately.

From the point of diagnosis, whānau meet with the MDT. Tamariki with a cleft lip will undergo surgery for lip repair (+/- hard palate repair) at 4–6 months old, and a cleft palate repair takes place at approximately 9–12 months. Internationally, it is accepted that the goal is to have "normal" speech by 5 years, with an acknowledgement that 20–30% may require secondary surgical speech procedures before 5 years for reduced or absent velopharyngeal closure—referred to as velopharyngeal incompetence (VPI) or fistula.[1–5] They may require grommet insertion, alveolar bone grafting at 9–11 years and possibly orthognathic surgery once growth is complete. In addition, many tamariki born with a cleft palate +/- cleft lip will require speech therapy intervention, orthodontic treatment and psychological support. There are internationally recognised psychosocial impacts

on whānau, especially those with lower household incomes.[6–8] Psychosocial wellbeing and positive engagement with services has been strongly linked with early diagnosis and counselling, establishing a close relationship with the MDT and social supports.[7]

Māori and orofacial cleft

Māori have highest incidence of cleft palate in the world. The incidence of cleft palate in Māori is over twice that of European (1.54 vs 0.73 per 1,000 live births).[9] As a result, the overall incidence of orofacial cleft in Aotearoa New Zealand, over a 10 year period, was found to be 1.79 per 1,000 live births.[10] Māori experience disparities in outcomes compared to the rest of the population across nearly all areas of health, due to inequity in determinants of health, including access to quality healthcare.[11] Māori have on average the poorest health status of any ethnic group in Aotearoa New Zealand.[12] Manatū Hauora – Ministry of Health have identified health equity and cultural safety as two strategic areas for review.[13] For many Māori, the existing public health system can be experienced as hostile and alienating. The large number of health professionals involved and the lack of relationships with health professionals have been cited as a barrier to whānau engagement.[14] When we also consider the large geographical area that the Auckland Regional Cleft Service covers, it is noteworthy that Māori are more likely to be living in small urban areas (15% of the Māori population) and rural areas (18%), compared with the total population (10% and 16% respectively),[15] potentially resulting in long travel times and disruption to the ability of whānau to access services.

Measuring speech outcomes

Orofacial cleft outcomes are often measured objectively by velopharyngeal function, speech, dental occlusion and general facial appearance.[16–17] Parent-reported measures are common and are considered valid measures of facial appearance, psychosocial function and speech.[18] There are other functional measures that are important to consider such as hearing status, success in both social and educational contexts[19] and psychosocial wellbeing of the whole whānau.[7] One of the key outcome measures for tamariki with orofacial cleft is speech. The velopharyngeal mechanism serves to separate the oral cavity from nasal cavity during speech and to allow an adequate build-up of intra-oral pressure for speech and efficient oral transit of food/fluid. VPI can be graded by perceptual speech analysis as mild, moderate, severe or profound.[20–21] VPI can result in nasal regurgitation when eating/drinking, hypernasal resonance, nasal air emission and weakened pressure consonants during speech. Reduced or absent velopharyngeal closure can have a significant impact on speech and is only remediable through surgery. Active cleft speech characteristics (CSCs) refer to "alternative articulatory gestures which function in place of intended consonants".[22] These are patterns of misarticulation that the child makes in order to compensate for the structural deficit caused by cleft palate. They are remediable through speech therapy intervention. Early intervention and education to whānau on how to model early, anterior pressure consonants can have a positive impact in reducing the extent of speech therapy later on.[23]

In addition to reporting VPI and CSCs, SLTs also rate speech outcomes in the functional parameters of intelligibility and acceptability. Having speech that is clear, easy to understand and sounds the same as one's peers is a key factor in social–emotional wellbeing, communicative confidence and educational achievement.[24] Children with orofacial cleft are at increased risk of developmental speech and language disorders compared with their peers[25] and, therefore, speech and language development must be assessed and treated alongside cleft speech. For children with ongoing poor hearing status, limited phonetic capacity and reduced communicative confidence can also impact on educational achievement and again, requires specific speech and audiology therapies.

Morrison et al.[26] recently published Aotearoa New Zealand's first speech outcomes for children with orofacial cleft at 5–10 years of age. This study looked at speech outcomes across the nation and included five cleft centres. Results were broken down by age and cleft diagnosis. No account for ethnicity or location was considered. This paper showed that further speech and/or surgical intervention was required in 85% with cleft lip and palate, 65% with cleft palate and 26% with cleft lip. With this complexity of MDT involvement in the early years for tamariki with orofacial cleft, it is important to audit service outcomes in Aotearoa New Zealand. In this study, we explored the speech outcomes for five-year-olds born with cleft palate who live in the catchment area of the Auckland Regional Cleft Palate Service, with a

Table 1: Primary diagnosis by ethnicity.

	Māori	European	South East Asian	Other Asian	Pasifika	Other	Total
Cleft palate	17 (77% of total for Māori)	19 (57% of total for European)	4 (40%)	2 (50%)	4 (66%)	2 (50%)	**48**
Unilateral Cleft Lip and Palate	2	10	5	2	2	1	**22**
Bilateral Cleft Lip and Palate	3	4	1	-	-	1	**9**
Total	22 (28%)	33 (42%)	10 (13%)	4 (5%)	6 (7%)	4 (5%)	**79**

Table 2: Distance (km) travelled to access service.

Travel distance from home to Middlemore	Total cohort (N=79)	Māori (n=22)	non-Māori (n=57)
0–20 km	37 (47%)	9	28
21–50 km	22 (28%)	6	16
51–100 km	7 (9%)	1	6
101–299 km	10 (13%)	3	7
>300km	3 (3%)	3	0

Table 3: Speech therapy services offered and attended.

	Total cohort (N=79)	Māori (n=22)	non-Māori (n=57)
Speech therapy attended through Auckland Cleft Service	44 (56%)	11 (25%)	33 (75%)
Number of sessions	Median 9.3, range 1–23		
Attendance		Average 63% attendance rate 3 (27%) children had 100% attendance	Average 95% attendance rate 28 (85%) of children had 100% attendance
Additional speech therapy in the community	46 (58%)	18 (82%)	27 (49%)

Table 4: VPI and CSC characteristics for Māori and non-Māori at 5 years plus distance from cleft centre.

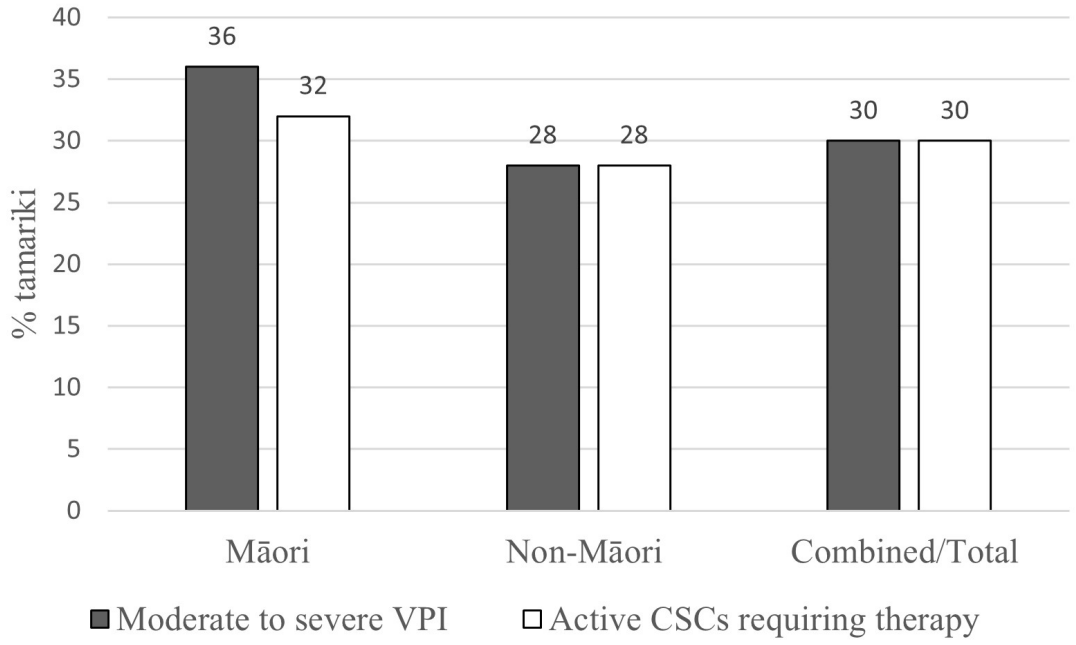
Speech outcome		Māori (n=22)	non-Māori (n=57)	Total	Kms
VF dysfunction*	Normal to Mild (0–1) Monitor Only	7	25	32	<20 km
		3	8	11	21–50 km
		1	5	6	51–100 km
		0	3	3	101–299 km
		3	0	3	300+ km
	Moderate (3–5) Require investigation and probable surgery	2	2	4	<20 km
		2	4	6	21–50 km
		0	1	1	51–100 km
		2	0	2	101–299 km
				0	300+ km
	Severe (6–7) Require investigation and likely surgery	0	3	3	<20 km
		1	4	5	21–50 km
				0	51–100 km
		1	2	3	101–299 km
		0	0	0	300+ km
Presence of cleft speech characteristics (CSC)	Normal speech (anterior or development errors) Monitor only	8	25	33	<20 km
		5	14	19	21–50 km
		1	5	6	51–100 km
		0	4	4	101–299 km
		3	0	3	300+ km
	Glottal/pharyngeal articulation/Active Nasal Fricatives Severe CSCs require therapy	1	1	2	<20 km
		0	1	1	21–50 km
		0	0	0	51–100 km
		2	0	2	101–299 km
		0	0	0	300+ km

Table 4 (continued): VPI and CSC characteristics for Māori and non-Māori at 5 years plus distance from cleft centre.

Presence of cleft speech characteristics (CSC)	**Backing to velar or uvular place** **Moderate CSCs Require therapy**	0	2	2	<20 km
		1	1	2	21–50 km
		0	1	1	51–100 km
		1	3	4	101–299 km
		0	0	0	300+ km

* Rhinocleft composite score (0= no impairment; 7=severe impairment).
** Anterior CSCs (dentalisation, lateralisation, palatalization) were not included here as they may not require ongoing therapy intervention. In cases where there was the presence of both backing to velar/uvular and glottal/pharyngeal or Active nasal fricatives, this was only counted once.

Figure 1: Speech outcomes for Māori and non-Māori (%).



focus on ethnicity and geographical factors that may influence service accessibility. We ask the research questions: what are the outcomes for tamariki in our regional service, does ethnicity or distance from Auckland impact on outcomes, and how does this compare to international standards?

Methods

This retrospective clinical file audit received ethical approval through The University of Auckland Human Patients Ethics Committee (012601). We collated data from all tamariki who received primary surgical repair through the Auckland Regional Cleft Service between 2013–2016 (inclusively). All tamariki included in this study had a cleft palate +/- cleft lip and continued in the service until they received their five-year review. Those with a cleft lip only (i.e., an intact palate) were excluded. Tamariki with an additional diagnosis of syndrome, sequence or developmental disability were included, provided their expressive language skills were sufficient to undertake standardised speech assessment. Information

was collected on residential address and primary ethnicity from the health board Clinical Portal. Information on timing and technique for surgical procedures was collected from health records. Any secondary surgeries, including fistula repair or speech surgery for VPI, and speech therapy appointments were also recorded. Surgical procedures such as grommets or dental extractions were not included.

At the time, primary surgical palate repair was carried out by one of four cleft surgeons. Since then, the Auckland Regional Cleft Palate service has reduced the number of surgeons carrying out primary cleft surgery, but there have been no significant changes in surgical techniques, timing of initial repair or care pathway within the MDT. Speech assessments were carried out by two SLTs, both with over 15 years' experience of working with cleft palate speech using the Rhinocleft assessment.[21] Speech reviews were scheduled at 5 years of age.

The service routinely reports the following speech parameters:

1. The degree of VPI using a validated ordinal rating scale—the Rhinocleft® Perceptual Cleft Palate Speech Assessment[20]—rated as absent, mild, moderate or severe. This is a cumulative score that reflects perceptual ratings in the areas of hypernasality, audible nasal air emission and weak or nasalised pressure consonants. Absent or mild VPI do not warrant further surgery.
2. The presence of active CSCs such as glottal articulation, pharyngeal articulation, backing to velar or uvular place of articulation.

Data were collated in Excel and explored through descriptive statistics and graphing. Differences between Māori and non-Māori and distance from centre were explored using Chi-squared test ($p<.05$).

Results

Eight-nine tamariki received primary surgical repair between 2013–2016. Ten were excluded as they had no follow-up at 5 years of age; seven had moved out of the area and three were lost to follow-up. The average age of primary repair was 12 months (range 6–15). Speech reviews, while scheduled at 5 years of age, in practice occurred between 4 years and 6 months and 7 years (mean age 5 years and 4 months). Primary diagnosis by ethnicity is presented in Table 1. Thirteen children had additional medical diagnosis including (but not limited to) 22q11 deletion syndrome, global developmental delay and Pierre Robin sequence.

Table 2 displays the distance that families travelled from their home to Middlemore Hospital, with an average of 59km (range 4–350km). Where attendance at clinic appointments proved difficult, clinical documentation indicated that the Cleft CNS maintained regular contact with both the families and community support services. Where needed, and if parents consented, children were offered community-based SLT and Health Psychology outside of the cleft MDT. Specific numbers of appointments and attendance rates for these external community services were not available. There was no significant correlation between ethnicity and distance to travel to clinic ($X^2= 5.36$, $p=.25$); however, 27% of Māori lived 100km+ away compared with only 12% of non-Māori.

Speech therapy services

Speech therapy with the cleft palate specialist SLT was offered to 100% of Māori and 70% of non-Māori who lived within 50km of the cleft service. Children received a range of speech therapy services from both the Auckland Regional Cleft Centre and community (Table 3). Specialist clinic attendance was significantly poorer for Māori (63%) than non-Māori (95%) ($X^2=6.43$, $p<.01$) (Table 3), but was not significantly associated with distance to travel to clinic. Māori were significantly more likely to be referred to community services (82%) than non-Māori (49%) ($X^2=12.56$, $p<.01$).

Speech outcomes

Thirty percent of tamariki, at the age of 5 years, had velopharyngeal insufficiency (VPI) that was deemed clinically to warrant further investigation with possible secondary speech surgery (Figure 1). Thirty-eight percent underwent secondary surgeries for velopharyngeal insufficiency or fistula repair before the age of 5 years. Some tamariki underwent more than one secondary procedure, with 59 secondary speech surgery procedures across 34 tamariki before 5 years of age. Thirty-six percent of Māori children (n=8) had residual moderate to severe VPI at 5 years, compared to 28% non-Māori (n=16) ($X^2=0.52$, $p=.47$) (Table 4; Figure 1).

Thirty percent of tamariki, at the age of 5 years, had residual cleft speech characteristics (CSCs) which warranted speech therapy intervention, including 32% of Māori (n=10) compared to 28% of

non-Māori (n=19) (X^2=*0.32*, p=.32) (Table 4; Figure 1). There was no significant correlation between VF dysfunction severity and domicile distance from the cleft centre (X^2= *8.87*, p=.06).

Discussion

Māori make up 26% of the Counties Manukau population and 28% of the cleft palate population within the Auckland Regional Cleft Service. There was no significant correlation between ethnicity and distance to travel to clinic; however, more Māori lived 100km+ away from the service than non-Māori. Equality of access to services and equity of outcomes are intertwined. Lost days of paid work and travel costs for multiple surgical and outpatient appointments, plus the large distances that some whānau have to travel are to be considered, not only in regard to the burden of care, but also on outcomes of speech, education and psychosocial wellbeing. It is pleasing to see that, when it comes to speech outcomes for five-year-olds with orofacial cleft, there was no significant difference across ethnicities or geographical location. As a whole cohort, Auckland Regional Cleft Service speech outcomes are better than the national reported outcomes for Aotearoa New Zealand, where 85% required further speech and/or surgical intervention at age 5 years.[26] However, internationally, it is widely accepted and reported that 20% of children born with orofacial cleft may have persistent speech disorder at the age of 5 years[2–4] in comparison to our 30% (VPI) and 30% (CSCs).

The building of relationships is an important and recurring theme in the literature around health equity for Māori. This can be difficult in a hospital setting where staff turnover is high. In our cleft team, the team is small, and staffing has remained stable for many years. Whānau have an allocated Clinical Nurse Specialist. She is their key point of contact along their journey with hospital appointments, surgery planning and post-operative follow-up. This key contact person builds relationships, provides stability and gives whānau time. This may contribute to successful engagement and attendance at cleft clinic appointments. The CNS and the Health psychologist on the team have a strong focus on patient engagement, equity of access to services and whānau wellbeing, and engage regularly with community services to reduce inequity for those unable to attend appointments in Auckland.

However, attendance was not high for all children. A barrier for many whānau may be practical—financial restraints, transport and organising leave/childcare. These can all impact on the ability of whānau to attend appointments.[14,27] Our health model allocates appointments within business hours, to which families are expected to attend. The establishment of the outreach clinic in Northland may contribute somewhat to facilitating travel burden and attendance at clinics. There may be more that can be done to improve flexibility of appointments and service provision models that can further enhance attendance on a regular basis. Since the COVID-19 pandemic, health professionals have been making more use of telehealth, which has opened up more opportunities for whānau to connect with the cleft team remotely and, in some cases, access remote speech therapy. Anecdotally, it has been noted that phoning or texting a parent following a non-attendance at clinic (for MDT clinics or speech therapy) creates better engagement and an increased attendance rate than merely sending another appointment letter.

While specialist SLT appointments were offered to 100% of Māori, attendance at the cleft service was lower for Māori compared with non-Māori. This did not, however, correlate with a significantly greater rates of VPI or CSCs in Māori. This suggests that Māori are accessing SLT through the community services available in Aotearoa New Zealand. If this is the case, then Te Whatu Ora – Health New Zealand needs to be focussing their support into these regional centres. Tamariki can access additional SLT through community-based Te Whatu Ora – Health New Zealand child development services (<3 years old), at school (Ministry of Education funded, ~5 years+) and/or privately. If Māori tamariki are accessing these services more often than the cleft service, the specialist SLTs in the cleft service need to focus education and training to these regional SLTs to ensure all tamariki get the SLT they need for best outcomes.

Limitations and recommendations

This is a retrospective audit and lacks depth of analysis to truly understand whānau engagement and satisfaction with the service. This audit provides a crude review of the service offering validated speech outcomes at 5 years old and provides a first step to build on for future research. There is an absence of a routine validated parent questionnaire in the service, and this would be an excellent addition to track parent concern and engagement. Hearing is not routinely assessed and therefore we were unable to correlate hearing status to speech

outcomes in this cohort. This is a consideration for future research. There is well established inequality in oral health in Māori compared with non-Māori. It was outside of the scope of this study to audit dental outcomes and tamariki perceptions of appearance, but this would be useful additional data to inform the way that our services are structured nationally, as well as informing the development of a clinical care pathway with health outcomes for Māori at the forefront of our care planning.

We currently use medical model measures to report outcomes. While these quantify our practice and help us measure, they are not the only measures of successful outcomes. We need to consider and work to improve outcomes that are important to whānau. It is imperative that, early on in the relationship we establish what a "good outcome" looks like for them. Parameters such as psychosocial wellbeing, educational achievement, being understood, acceptance and engagement in community activities may be important outcome measures for our patients and should be considered.

Kaupapa Māori research in this area is critically needed to explore attendance, whānau experience and satisfaction with services. Research should explore how we can prioritise Māori participation and success at all levels. It is important to recognise what we are doing well and to consider how we can continually improve our practice. We must all make greater efforts to recognise and respond to tikanga Māori practices.

Examples for consideration and future service development opportunities are considered below:

- Build rapport and warmth of interactions.[14]
- Acknowledge and consider all people's cultural and spiritual practice.
- Evaluate more flexibility in appointment times and structures to prevent the practical barriers of attending appointments.
- Proactively develop policies to improve Māori participation and success at all levels.
- Develop an assessment of cleft speech that uses te reo Māori.
- Establish regular and consistent use of parent-reported outcome measures and patient goal setting.

Conclusions

Auckland Regional Cleft Service must continue to strive for improvements in speech outcomes, both in terms of surgical and speech therapy needs at 5 years old. Consideration must be given, when planning services, to ensure equitable access to services, cultural support and whānau engagement in the process. While speech outcomes did not differ based on ethnicity, attendance at specialist outpatient appointments was lower in Māori. Community services were offered but attendance rates to these appointments is unknown. If we do not provide the best care at the best time for our tamariki, we risk losing them from our service and, therefore, risk not allowing them to reach their best possible outcomes. It is imperative that, in order to achieve the best objective and functional outcomes, whānau are engaged in their treatment, trust their MDT and are able to access timely and effective treatment.

COMPETING INTERESTS
Nil.

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REFERENCES
1. Sell D, Mildinhall S, Albery L, et al. The Cleft Care UK study. Part 4: perceptual speech outcomes. Orthod Craniofac Res. 2015;18(Suppl 2):36-46. doi: 10.1111/ocr.12112.
2. Hall C, Golding-Kushner KJ. Long-term follow-up of 500 patients after palate repair performed after 18 months of age. Paper presented at: Sixth International Congress on Cleft Palate and Related Craniofacial Anomalies; 1989; Jerusalem, Israel.
3. Sell D, Grunwell P, Mildinhall S, et al. Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. Cleft Palate Craniofac J. 2001;38(1):30-7. doi: 10.1597/1545-1569_2001_038_0030_clapci_2.0.co_2.
4. Brunnegård K, Lohmander A. A cross-sectional study of speech in 10-year-old children with cleft palate: results and issues of rater reliability. Cleft Palate Craniofac J. 2007;44(1):33-44. doi: 10.1597/05-164.
5. Britton L, Albery L, Bowden M, et al. A cross-sectional cohort study of speech in five-year-olds with cleft palate + lip to support development of national audit standards: benchmarking speech standards in the United Kingdom. Cleft Palate Craniofac J. 2014;51(4):431-51. doi: 10.1597/13-121.
6. Grollemund B, Dissaux C, Gavelle P et al. The impact of having a baby with cleft lip and palate on parents and on parent-baby relationship: the first French prospective multicentre study. BMC Pediatr. 2020;20(1):230. doi: 10.1186/s12887-020-02118-5.
7. Namdar P, Pourasghar M, Lal Alizadeh F, Shiva A. Anxiety, Depression, and Quality of Life in Caregivers of Children with Cleft Lip and Palate: A Systematic Review. Iran J Psychiatry Behav Sci. 2022;16(2):e113591. doi: 10.5812/ijpbs-113591.
8. Nidey N, Moreno Uribe LM, Marazita MM, Wehby GL. Psychosocial well-being of parents of children with oral clefts. Child Care Health Dev. 2016;42(1):42-50. doi: 10.1111/cch.12276.
9. Smit R, Fowler PV. Non-syndromal orofacial clefts in the Canterbury/West Coast region during the 2000-2009 period. N Z Dent J. 2010;106(4):129-131.
10. Thompson J, Stone P, Sanders M, et al. The incidence of Orofacial Cleft in live births in New Zealand. N Z Med J.2016;129(1440):64-71.
11. Ministry of Health. Tatau Kahukura: Māori Health Chart Book 2015. 3rd ed. Wellington (NZ): Ministry of Health; 2015.
12. Ministry of Justice. Hauora: Report on Stage One of the Health Services and Outcomes Kaupapa Inquiry [Internet]. Wellington (NZ): Ministry of Justice; 2023 [cited 2021 Oct]. Available from: https://forms.justice.govt.nz/search/Documents/WT/wt_DOC_195476216/Hauora%202023%20W.pdf.
13. Manatū Hauora – Ministry of Health. The Guide to He Korowai Oranga – Māori Health Strategy [Internet]. Wellington (NZ): Manatū Hauora – Ministry of Health; 2015 [cited 2021 Oct]. Available from: https://www.health.govt.nz/our-work/populations/maori-health/he-korowai-oranga/pae-ora-healthy-futures.
14. Graham R, Masters-Awatere B. Experiences of Māori or Aotearoa New Zealand's public health system: a systematic review of two decades of published qualitative research. Aust N Z J Public Health. 2020;44(3). doi: 10.1111/1753-6405.12971.
15. Environmental Health Intelligence New Zealand. Urban–rural profile [Internet]. Wellington (NZ): Environmental Health Intelligence New Zealand; 2022 [cited 2021 Oct]. Available from: https://ehinz.ac.nz/indicators/population-vulnerability/urbanrural-profile/.
16. Allori AC, Kelley T, Meara JG, et al. A Standard Set of Outcome Measures for the Comprehensive Appraisal of Cleft Care. Cleft Palate Craniofac J. 2017;54:540-554. doi: 10.1597/15-292.
17. Fowler PV, Wholley C, Perry JG, Thompson JMD. Craniofacial morphology and soft tissue profile outcomes for complete unilateral and complete bilateral cleft lip and palate in New Zealand. Orthod Craniofac Res. 2019;22(3):139-146. doi: 10.1111/ocr.12294.
18. Apon I, van Leeuwen N, Allori AC, et al. Rasch Analysis of Patient- and Parent-Reported Outcome Measures in the International Consortium for Health Outcomes Measurement Standard Set for Cleft Lip

and Palate. Value Health. 2021;24(3):404-412. doi: 10.1016/j.jval.2020.10.019.
19. Constantin J, Wehby GL. Academic outcomes of children with orofacial clefts: A review of the literature and recommendations for future research. Oral Dis. 2022;28(5):1387-1399. doi: 10.1111/odi.14137.
20. Fitzsimons D. The Rhinocleft®: A Comprehensive Perceptual Assessment of Velopharyngeal Function for Children [dissertation]. Sydney (AU): The University of Sydney; 2018
21. Sell D, Harding A, Grunwell P. GOS.SP.ASS.'98: an assessment for speech disorders associated with cleft palate and/or velopharyngeal dysfunction (revised). Int J Lang Commun Disord. 1999;34(1):17-33. doi: 10.1080/136828299247595.
22. Harding A, Grunwell P. Active versus passive cleft-type speech characteristics: implications for surgery and therapy. Int J Lang Commun Disord. 2011;30(S1):242-247. doi: 10.1111/j.1460-6984.1995.tb01679.x.
23. Harding-Bell A. Case Studies in Cleft Palate Speech: Data analysis and principled intervention. Havant (UK): J&R Press; 2019.
24. Law J, Dennis JA, Charlton JJV. Speech and language therapy interventions for children with primary speech and/or language disorders. Cochrane Database Syst Rev. 2017;2017(1):CD012490. doi: 10.1002/14651858.CD012490.
25. Lancaster HS, Lie KM, Chow JC, et al. Early Speech and Language Development in Children With Nonsyndromic Cleft Lip and/or Palate: A Meta-Analysis. J Speech Lang Hear Res. 2019;63(1):14-31. doi: 10.1044/2019_JSLHR-19-00162.
26. Morrison MM, Mason NT, Forde BL, et al. Speech Outcomes of a National Cohort of Children with Orofacial Cleft at 5 and 10 Years of age. Cleft Palate Craniofac J. 2022;59(11):1400-1412. doi:10.1177/10556656211044939.
27. Craig E, Reddington A, Adams J, et al. Te Ohonga Ake: The Health of Māori Children and Young People with Chronic Conditions and Disabilities in New Zealand: Series Two [Internet]. Dunedin (NZ): Te Ohonga Ake series for the Ministry of Health; 2014 [cited 2021 Oct]. Available from: https://www.health.govt.nz/publication/te-ohonga-ake-health-maori-children-and-young-people-chronic-conditions-and-disabilities-new-zealand.

Falls at home: hospital admissions in a health region of Aotearoa New Zealand

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ABSTRACT

AIM: To report on the descriptive epidemiology and costs of trauma admissions to the Te Manawa Taki Trauma System (TMT) hospitals in Aotearoa New Zealand following falls at home.

METHODS: A retrospective, observational study was conducted using data from the TMT trauma registry to identify patients of all ages who presented following falls at home from 2012 to 2022. This study reports on incidence of Fall Related Injuries (FRIs) that occurred at home with regard to age, gender, ethnicity, Injury Severity Score (ISS), injury characteristics and direct cost to TMT facilities.

RESULTS: Searches identified 13,142 events to the TMT trauma system following falls at home. Most events were classified as non-major trauma. There were statistically significant relationships between gender, ethnicity and district, and ISS category. There were two distinctive age band incidence peaks: ≤9 years and 60+ years. Males were more likely to sustain major trauma. The most common cause was fall on the same level from slipping, tripping and stumbling. The average length of stay per event was 5.5 days. The average cost per event was NZ$9,792.

CONCLUSIONS: The study has identified the demography, injury types, risk factors and outcomes for FRIs that occurred in the TMT region of Aotearoa New Zealand. The volumes and costs of injury represent a significant burden on the health system, individuals and communities. More detailed understanding of causative factors will allow targeting of prevention strategies to address highrisk activities and demographic groups.

Trauma is a significant public health concern, and is recognised as a major cause of death and disability across the globe[1,2] and in Aotearoa New Zealand. Fall-related injuries (FRIs) are a significant source of this trauma burden in Aotearoa New Zealand and globally.[3–6] Equitable use of resources for the prevention and treatment of fall injuries depends on the use of reliable and representative information on incidence rates and risks across ethnicities and demographic groups. FRIs are of a particular importance in the context of Aotearoa New Zealand. Annual reports from the New Zealand Trauma Registry indicate that in 2021/2022, falls were the most common cause of trauma presentation in patients with ISS >12; falls were responsible for 33% of major trauma presentations nationally.[7] Furthermore, data from the Global Burden of Disease project suggests Aotearoa New Zealand has a relatively high burden of disease (as measured in disability adjusted life years) compared to an average of other high-income countries.[8]

The most common location for injury hospitalisations is home, which is second to roads as the location for injury mortality.[9–13] An analysis of people aged 25–64 years in a Scandinavian study found that 44% of unintentional home injury requiring medical treatment were due to falls.[10] Falls are the leading cause of unintentional injury at home among children aged ≤4 years and adults aged 20–64 years in Aotearoa New Zealand.[6,14,15] Additionally, nearly a third of self-reported injuries requiring medical attention occur at home in Aotearoa New Zealand.[16]

FRIs also have a significant economic impact on the Aotearoa New Zealand health system and beyond through the Accident Compensation Corporation (ACC).[17] The ACC is a no-fault injury compensation scheme operating nationwide that covers the costs of medical treatment and rehabilitation.[17] For the 2012/2013 to 2021/2022 financial years, there were 3,855,876 new claims lodged as a result of FRIs at home, which translated to active costs in excess of $4 billion.[18] In 2010, 18% of the total social and economic costs of all injuries in Aotearoa New Zealand were attributable to falls, highlighting the importance of prevention.[19] The total economic and social cost per fall injury was estimated to be NZ$1,735 (2008 prices).[19] It is therefore important to characterise the pattern

of injury and determine what scope there is for preventative measures to reduce the burden of injury.

The Te Manawa Taki (TMT) Trauma Registry collects standardised data on trauma patients of all age groups and severities, providing a unique data source for studying the patterns of FRIs that result in hospitalisation. The TMT region covers a geographically large and diverse area and is demographically similar to Aotearoa New Zealand as a whole.[20–22]

The volume of literature surrounding falls in the home environment is largely focussed on falls in the elderly. Evidence reporting on the descriptive epidemiology of patients admitted following injury from falls across all ages and all severities is limited. However, it is possible using the comprehensive data included in the Te Manawa Taki Trauma Registry (TMTTR) to define at-risk groups. To our knowledge, this is the first study examining the characteristics of patients of all age groups and severities that were admitted to Aotearoa New Zealand hospitals following falls at home over a prolonged period (11 years). This study will quantify the frequency of trauma presentations following FRIs by age, sex, ethnicity and district. Additionally, this study aims to assess associations between injury severity and cause of fall, alcohol use, time of fall, length of hospital stays and cost. By characterising the incidence and clinical characteristics of FRIs at home resulting in hospital inpatient treatment in the TMT region, this study aims to identify areas for targeted intervention and further research.

Methods

Study design

A retrospective analysis of trauma data from the TMTTR was performed to identify patients of all ages that were admitted to TMT hospitals following falls at home during the 11-year period from 1 January 2012 to 31 December 2022. The TMT region is formed by five health districts (Bay of Plenty, Lakes, Tairāwhiti, Taranaki and Waikato) with a combined population of approximately 1 million people. To qualify for inclusion in the trauma registry, patients must have been admitted to an inpatient TMT bed within 7 days of injury, or died in hospital after injury. The registry exclusion criteria was as follows: patients seen and discharged from the emergency department; injuries directly attributable to documented pathological processes (osteoporosis, metastatic disease, etc.) or pre-existing medical conditions (Parkinson's, etc.); peri-prosthetic fractures; exertional injuries; hanging, drowning or foreign bodies without anatomical injury; poisoning. Cases of falls in which the patient died at the scene were not captured in this study. Data entry into the TMTTR occurs prospectively during each admission. Data-points are coded by Abbreviated Injury Scale (AIS) Scores and ICD-10-AM, and all data underwent extensive quality checking before entry into the TMTTR.[23,24] Analysis for the study was done as part of a routine health audit; therefore, ethics approval was deemed unnecessary. This study was registered with Waikato Hospital Ethics: RD023047.

Data collection and analysis

The variables of interest were extracted from the TMTTR: age, sex, ethnicity (as listed under the patients National Health Index number), ICD place of injury code, ISS, inpatient costs. The ICD place of injury code "home" was used to identify falls that had occurred in a home environment ("home" includes driveway to home, garage, garden to home, yard to home and swimming pool in private house/garden). Duplicate entries (e.g., cross-hospital transfers of the same patient on the same admission) were identified and removed. Individual registry entries were audited in to confirm record accuracy. AIS scores are used by the registry to quantify injury patterns and severity by ranking injuries in each anatomical region from 1 (minor) and 6 (non-survivable).[25] The AIS is then used to assign each patient an Injury Severity Score (ISS), allowing cases to be stratified into major trauma (ISS >12) and non-major trauma (ISS <13). Population estimates were taken from the Statistics New Zealand demographic database.[26]

Statistical analysis was conducted using RStudio Version 3.6.1.[27] Records with missing data were excluded from the analysis. Data are presented as count with percentage or mean with standard deviation (SD). Chi-squared tests were used to assess associations between injury severity and age, sex, ethnicity, district, height of fall and positive blood alcohol level (BAL). A p-value of <0.05 was considered significant.

Results

Searches identified 13,142 events related to falls at home. This includes both intentional and unintentional FRIs. This represented nearly 51.5% of the total falls-related trauma events recorded in

Table 1: Demographics of patients admitted to Te Manawa Taki hospitals following fall-related injuries that occurred at home between 2012 and 2022.

	Major n (%)	**Non-major n (%)**	**Total n (%)**	**Statistical test (major vs non-major)**
Total	704 (5.4)	12,437 (94.6)	13,142 (100)	
Gender				
Female	289 (3.9)	7,183 (96.1)	7,472 (100)	χ^2 = 75.11, p<0.001
Male	415 (7.3)	5,254 (92.7)	5,669 (100)	RR=0.53 (CI: 0.46–0.61)
Ethnicity				
Māori	110 (3.7)	2,829 (96.3)	2,939 (100)	χ^2 =19.17, p<0.001
non-Māori	594 (5.8)	9,594 (94.2)	10,188 (100)	RR=0.64 (CI: 0.53–0.78)
Te Whatu Ora – Health New Zealand District				
Waikato	366 (6.7)	5,107 (93.3)	5,473 (100)	χ^2 =26.83, p<0.001
Bay of Plenty	234 (5.8)	3,795 (94.2)	4,029 (100)	
Lakes	57 (3.6)	1,526 (96.4)	1,583 (100)	
Taranaki	82 (5.2)	1,497 (94.8)	1,579 (100)	
Tairāwhiti	34 (4.2)	772 (95.8)	806 (100)	

the registry during the study period. Of the 13,142 events, 12,437 (94.4%) events were classified as non-major trauma events.

Across all age bands, 56.9% (n=7,472) of falls involved females, and 43.1% involved males (Table 1). There were statistically significant relationships between gender, ethnicity and district, and having a major or non-major admitted trauma. Nearly 42% of all injuries occurred in the Waikato district and the majority of them fell while engaged in other activity. Among ≤9 years olds, 46.7% were female and 53.3% were male. However, from aged 40 years onward the proportion of falls at home among females increases and remains significantly higher (63.0%) than the proportion of males (37.0%) (p<0.001). However, across all age bands, males accounted for 58.9% of all major trauma (ISS >12) due to falls at home. Overall, females were around half as likely to present as major trauma (Risk Ratio [RR] = 0.53, CI: 0.46–0.61) in comparison to males.

There are two distinct age peaks among trauma events due to falls at home: children aged ≤9 years, and a wide peak among adults aged 60 years and over (Figure 1). Falls at home resulting in major trauma (ISS >12) are strongly skewed towards older age groups, with a peak in the aged over 60. Among children aged ≤9 years, 98.8% (2,479) of falls at home were non-major trauma (ISS <13) events (2012–2022).

Across all ages, Māori accounted for 22.3% (n=2,939) of trauma events due to falls at home; however, this was highly skewed towards the ≤9 years. In the ≤9 years, Māori represented 42.9% of falls at home trauma. Above the aged 60 years, non-Māori represented 91.5% of falls at home trauma.

During the study period, the annual incidence of falls at home increased steadily from a minimum of 902 events in 2012 to a maximum of 1,456 events in 2022. A breakdown of events over time is demonstrated in Table 2. A minority (n=704, 5.4%) of events due to falls at home have been major trauma (ISS >12), with 94.6% (n=12,437) of cases classed as non-major trauma (ISS <13).

As shown by the heat map in Figure 2, falls

at home were most common on weekends, particularly in the 15–64 age group. The older age groups (65+) had less variation by day of week than younger age groups. Falls at home were most common during the day (8 am–4 pm) and least common in the early morning (midnight–4 am). In the ≤14 age group, admissions typically occurred between 4 pm to 8 pm. Within the 15–64 age group, the admissions were commonly on Saturday night between 7 pm to midnight.

Figure 1: Trauma events due to falls at home by age group, gender and severity (2012–2022, n=13,142).

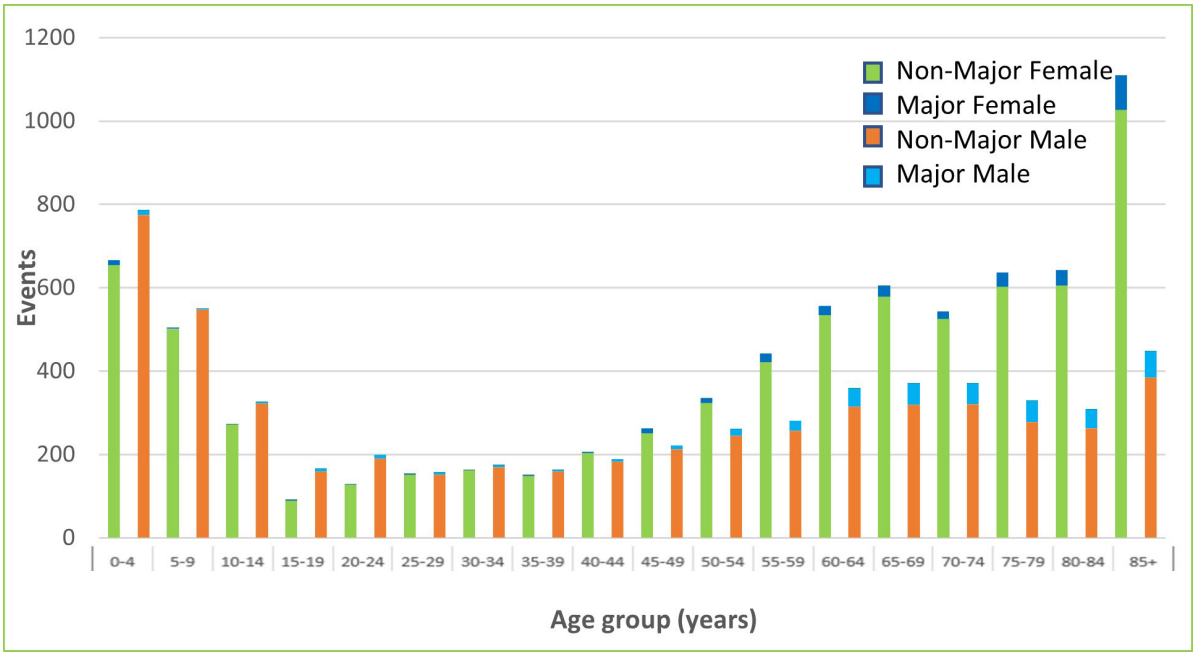

Table 2: Annual trauma events due to falls at home by severity (2012–2022, n=13,142).

Year	Number of events (n=13,142)		
	Major trauma	Non-major trauma	Total
2012	32	870	902
2013	38	963	1,001
2014	45	1,042	1,087
2015	40	1,094	1,134
2016	59	1,198	1,257
2017	47	1,068	1,115
2018	62	1,090	1,152
2019	61	1,161	1,222
2020	84	1,296	1,380
2021	101	1,334	1,435
2022	135	1,321	1,456

The three most common causes of falls at home were as follows: fall on same level from slipping, tripping, and stumbling (W01) (n=4,547, 34.6%); fall on and from stairs and steps (W10) (n=1,683, 12.8%); other fall on same level (W18) (n=1,383, 10.5%). A detailed breakdown of all fall causes is shown in Table 3.

There were 13,011 events where the height of fall was known. Falls from <1m were the most common, accounting for 9,621 (73.9%) events. Falls from 1–5m were present in 3,296 events (25.3%), and falls from >5m were the most uncommon (n=94, 0.7%). Across all age groups, falls from <1m remained the most common height of fall. The number of fall events for each height of fall split by age and subsequent injury severity is shown in Table 4. There was a significant association between height of fall and injury severity (χ^2=219.6, p<0.001). The proportion of major trauma events compared to non-major trauma appeared to increase with height of fall.

There was a large variation in the incidence of injuries to the ISS coded body regions. In falls at home the most commonly injured region was the face (n=8,814). The number of events involving other body regions were as follows: external (n=3,735); head and neck (n=2,157); abdominal and pelvic contents (n=1,026); extremities and pelvic girdle (n=509); chest (n=426).

There was a total of 152 deaths in hospital following fall at home events, including all "medical" deaths primarily due to medical causes rather than the injuries sustained. When medical deaths were excluded, this figure was reduced to 97 deaths. Of these, 58 deaths were due to central nervous system failure, 17 due to multi organ failure, 11 due to other, 7 due to hemorrhage and 4 were unknown. The global case fatality rate (CFR) for all FRI events was 0.74% (n=97 [died] vs n=13,045 [survived]). Older adults (65+ years) had the highest CFR of 1.57% (n=83 [died] vs 5,283 [survived]), with 15–64 year olds shown to have a CFR of 0.3% (n=14 [died] vs 4,654 [survived]); no children (≤14 years) died following fall at home events. Major trauma events had a CFR of 9.93% (n=57 [died] vs n=574 [survived]) compared to a CFR of 0.32% (n=40 [died] vs n=12,471 [survived]) in non-major trauma.

The mean length of stay (LOS) per event was 5.5 (SD 6.9) days. Admissions were typically longer in major trauma events, with an average LOS of 8.7 (SD 8.7) days compared to 5.3 (SD 6.8)

Figure 2: A heat map demonstrating the number of events for each time across the week, split by age group.

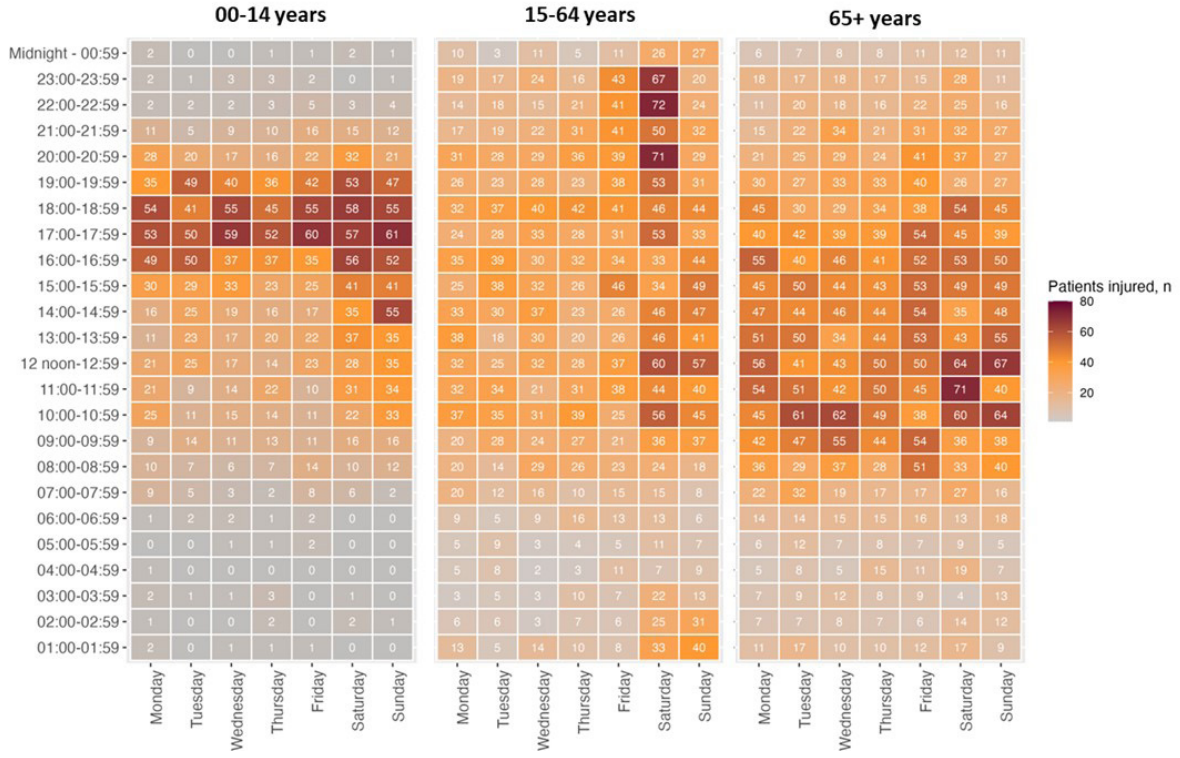

Table 3: The number of events by the Cause of Injury (total n=13,142).

Cause of Injury	n=13,142 n (%)
W01: Fall on same level from slipping, tripping and stumbling	4,547 (34.6)
W10: Fall on and from stairs and steps	1,683 (12.8)
W18: Other fall on same level	1,383 (10.5)
W13: Fall from, out of or through building or structure	930 (7.1)
W09: Fall involving playground equipment	821 (6.2)
W11: Fall on and from ladder	778 (5.9)
W06: Fall involving bed	451 (3.4)
W07: Fall involving chair	445 (3.4)
W19: Unspecified fall	439 (3.3)
W17: Other fall from one level to another	351 (2.7)
W02: Fall involving ice-skates, skis, roller-skates or skateboards	296 (2.3)
W14: Fall from tree	282 (2.1)
W08: Fall involving other furniture	281 (2.1)
W04: Fall while being carried or supported by other persons	147 (1.1)
W15: Fall from cliff	90 (0.7)
W03: Other fall on same level due to collision with, or pushing by, another person	72 (0.5)
W12: Fall on and from scaffolding	50 (0.4)
W05: Fall involving wheelchair	39 (0.3)
W16: Diving or jumping into water causing injury other than drowning or submersion	38 (0.3)
W00: Fall on same level involving ice and snow	19 (0.1)

days for non-major events. Average LOS appeared to increase with age categories from 1.8 (SD 1.9) days in the ≤14 year age group to 5.2 (SD 6.2) days for 14–64 year olds, and 7.8 (SD 8.3) days in the 65+ year age group.

During 2022, trauma events due to falls at home cost approximately NZ$24.3 million directly to TMT facilities. Over the study period, the average cost across all events was NZ$9,792 per event. Costs appeared to increase with both age and severity. Patients over 65 years incurred the greatest cost per event with an average of NZ$11,834, compared to NZ$10,681 for the 15–64 year aged bracket and NZ$4,930 for those up to age 14. The average cost for major trauma events (ISS >12) was NZ$18,322, compared to an average of NZ$9,362 for an event where the ISS was <13.

There were 260 patients whose BAL were recorded. BAL is only tested for in major trauma patients aged 16 and over that receive a trauma call on arrival (within 6 hours of first arrival). Of these 260 patients, 8.1% (n=21) had a positive BAL (EtOH/BAC 2mmol/L or more) on admission. A positive BAL was significantly (χ^2=8.40, p=0.0037)

Table 4: The number of major and non-major events split by height of fall and age group.

	All age groups			≤14 age group			15–64 age group			65+ age group		
Height of Fall	Major	Non-major	Total	Major	Non-major	Total	Major	Non-major	Total	Major	Non-major	Total
Fall <1m	314	9,253	9,621	15	1,883	1,898	65	3,231	3,296	234	4,139	4,427
Fall 1–5m	289	3,007	3,296	19	1,120	1,139	128	1,141	1,269	142	746	888
Fall >5m	20	74	94	1	21	22	9	38	47	10	15	25
Total	623	12,334	13,011	35	3,024	3,059	202	4,410	4,612	386	4,900	5,340

more common in 15–64 year olds, with positive findings in 13.9% (n=15) of events compared to a rate of 3.9% (n=6) in the 65+ age group.

Discussion

This is the first study to report the descriptive epidemiology of patients admitted to hospital with a fall-related injury occurring at home, across all ages and all injury severities within a health region in Aotearoa New Zealand. Considering the significant biopsychosocial and economic impacts of FRIs, identifying at-risk populations and risk factors for at-home falls allows for better targeting of injury prevention strategies.

The study includes 13,142 events related to falls at home, with only 5.4% meeting severity criteria for major trauma (n=704) and 94.6% (n=12,437) classified as non-major trauma. There were statistically significant relationships between gender, ethnicity and district, and ISS. As expected, fall height was significantly associated with severity of injury ($p<0.001$), with falls from >5m proportionately more likely to present as major trauma.

The two most common causes of falls (slipping and tripping, and falls involving steps) accounted for 47.4% of all falls at home. This aligns with previous work suggesting structural hazards for falls are common in Aotearoa New Zealand homes.[28] Stairs without handrails were shown to be present in 54% of homes, which was identified as a specific risk factor for falls at home. Interventions specifically targeting structural hazards within the home may therefore have a marked impact on the volume of FRI events given the disproportionate ratio of events due to these causes.

The gender analysis shows that across all age bands, 56.9% (7,472) of falls at home trauma events involved females and were around half as likely to present as major trauma (RR=0.53, CI: 0.46–0.61) in comparison to males. From aged 40 years onward, the proportion of falls at home among females increases and remains significantly higher (63.0%) than the proportion of males (37.0%) ($p<0.001$). However, across all age bands, males accounted for 58.9% of all major trauma (ISS >12). Either this may possibly reflect a greater proportion of females surviving into older age, or differences in time spent at home between genders, as well as other factors.

The age group analysis of the study shows two distinct age peaks among trauma events due to falls at home, children aged ≤9 years, and a wide peak among adults aged 60 years and over. Of these admissions, there was a peak of major trauma in 60+ year age group. As a result, older adults are more likely to have worse outcomes than in younger aged groups.[29,30] Among children aged ≤9 years, 98.8% (2,479) of falls were non-major trauma (ISS <13) events.

Analysis of trauma events by ethnicity showed that though Māori accounted for 22.3% (n=2,939) of falls, this was highly skewed towards the ≤9 years (42.9%). This result is consistent with the literature, which identified the Māori population as being at greater risk of injuries at home, with disproportionate skew towards ≤9 years.[31] Keall et al. specifically targeted this population in a randomised control trial, which evaluated the impact of low-cost home modifications, specifically designed to reduce the risk of falls in Māori households.[32,33] They estimated an annual reduction in FRI events of 31% associated with the intervention. Although not specific to the age group we have identified here, the study demonstrates the potential

for successful targeted intervention to reduce the risk of FRIs in an at-risk population.

The results also show that falls at home were most common on weekends. This effect was most significant in the 15–64 age group, and may be attributed to working age adults spending greater periods of time at home during this time period. It may be beneficial to consider initiation of fall prevention initiatives at an earlier age than has generally been the case.

Interestingly, we identified a significant relationship (p=0.0037) between positive BAL and age group within the major trauma subgroup. Positive BAL in major trauma were shown to be significantly more common in those aged 15–64 years. This would be in keeping with previous evidence (originating from Aotearoa New Zealand and Australia) to suggest alcohol consumption increases both the likelihood and severity of injuries in the home setting, especially in younger, working age adults.[15,34,35] Considering that alcohol-related presentations to the emergency department predominantly occur at weekends,[36] this appears to be consistent with the weekend admission relationship for working age adults. Consequently, alcohol specific interventions may be useful to reduce FRIs in the 15–64 age group. In contrast to the 15–64 years age group, there is no evidence to show an association between alcohol consumption and falls in the older age group (≥65 years). Given that BAL was only collected in major trauma events, our assessment of the relationship between alcohol consumption and FRIs is limited. However, future studies may provide in-depth analysis of the relationship between alcohol consumption and FRIs of all severities, and the nature of the association between alcohol and falls in older age groups.

The study is unique in that it utilises a continuously collected dataset that represents hospitalised patients of all ages and injury severity within an Aotearoa New Zealand health region, including estimates of hospital cost. The TMT region is also representative of Aotearoa New Zealand as a whole in terms of demography.[20–22] This study does not represent a population sample of all FRI that occurred at home because it does not include non-admitted persons, nor pre-hospital deaths from injuries. Notwithstanding these limitations, this study provides an up-to-date overview of patients admitted following FRI at home that can now be used for targeted interventions and health service planning.

Conclusion

Males and older age groups predominate in falls at home, resulting in significant cost and public health burden. Most events were classified as non-major (ISS <13) trauma. However, the significant relationships between gender, ethnicity and district and injury severity may suggest that some groups are more at risk of major trauma events. While the ≤9 years of Māori presenting with FRIs is much higher than the other age groups, additional research is required to better understand the causative reasons. Further work should also extend to in-depth analysis of the causation of FRIs occurring at home within the at-risk groups to help reduce this significant and potentially preventable burden on the health system, patients and the community.

COMPETING INTERESTS
The authors declare no competing interests.

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REFERENCES
1. Vos T, Lim SS, Abbafati C, et al. Global burden of 369 diseases and injuries in 204 countries and territories, 1990–2019: a systematic analysis for the Global Burden of Disease Study 2019. Lancet. 2020;396(10258):1204-22. doi: 10.1016/S0140-6736(20)30925-9.
2. Rossiter ND. "Trauma-the forgotten pandemic?". Int Orthop. 2022;46(1):3-11. doi: 10.1007/s00264-021-05213-z.
3. Chandran A, Hyder AA, Peek-Asa C. The global burden of unintentional injuries and an agenda for progress. Epidemiol Rev. 2010;32(1):110-20. doi: 10.1093/epirev/mxq009.
4. Haagsma JA, Graetz N, Bolliger I, et al. The global burden of injury: incidence, mortality, disability-adjusted life years and time trends from the Global Burden of Disease study 2013. Inj Prev. 2016;22(1):3-18. doi: 10.1136/injuryprev-2015-041616.
5. O'Leary K, Kool B, Christey G. Characteristics of older adults hospitalised following trauma in the Midland region of New Zealand. N Z Med J. 2017;130(1463):45-53.
6. Safekids Aotearoa. Child Unintentional Deaths and Injuries in New Zealand, and Prevention Strategies [Internet]. Auckland (NZ): Safekids Aotearoa; 2015 [cited 2023 Jun 22]. Available from: https://media.starship.org.nz/download-pdf-safekids-injury-prevention-strategies%3E%3E/Safekids_Aotearoa_Databook_CIP_NZ_and_Prevention_Strategiesv3.pdf.
7. Te Hononga Whētuki ā-Motu National Trauma Network. Annual Report 2021/22: New Zealand Trauma Registry & National Trauma Network [Internet]. Wellington (NZ): Te Hononga Whētuki ā-Motu National Trauma Network; 2022 [cited 2023 Jun 22]. Available from: https://www.majortrauma.nz/assets/Annual-reports/NZMT/NZMT2021-2022.pdf.
8. IHME. New Zealand: Institute for Health Metrics and Evaluation [Internet]. Seattle (US): The Institute for Health Metrics and Evaluation; 2019 [cited 2023 Jun 22]. Available from: https://www.healthdata.org/new-zealand.
9. Driscoll TR, Mitchell RJ, Hendrie AL, et al. Unintentional fatal injuries arising from unpaid work at home. Inj Prev. 2003;9(1):15-9. doi: 10.1136/ip.9.1.15.
10. Kopjar B, Wickizer TM. Home injuries among adults in Stavanger, Norway. Am J Public Health. 1996;86(3):400-4. doi: 10.2105/ajph.86.3.400.
11. Langley JD, McLoughlin E. Injury mortality and morbidity in New Zealand. Accid Anal Prev. 1989;21(3):243-54. doi: 10.1016/0001-4575(89)90015-8.
12. Runyan CW, Casteel C, Perkis D, et al. Unintentional injuries in the home in the United States Part I: mortality. Am J Prev Med. 2005;28(1):73-9. doi: 10.1016/j.amepre.2004.09.010.
13. Routley V, Valuri J. Home Injuries. Hazard. 1993;14:1-20.
14. Kool B, Ameratunga S, Robinson E, Jackson R. Hospitalisations and deaths due to unintentional falls at home among working-aged New Zealanders. Injury. 2007;38(5):570-5. doi: 10.1016/j.injury.2006.10.014.
15. Thornley S, Kool B, Marshall RJ, Ameratunga S. Alcohol intake, marijuana use, and sleep deprivation on the risk of falls occurring at home among young and middle-aged adults: a case-crossover study. N Z Med J. 2014;127(1406):32-8.
16. Coggan C, Hooper R, Adams B. Self-reported injury rates in New Zealand. N Z Med J. 2002;115(1161):U167.
17. Accident Compensation Corporation. About ACC [Internet]. NZ: ACC; 2023 [cited 2023 Jun 22].

Available from: https://www.acc.co.nz/about-us/.
18. ACC Analytics & Reporting. Dataset for publication - "Falls" [Internet]. Wellington (NZ): Data.govt.nz; 2022. Available from: https://catalogue.data.govt.nz/dataset/fall-data.
19. Wren J, Barrell K. Five-year Evaluation: The costs of injury in New Zealand and methods for prioritising resource allocation [Internet]. Wellington (NZ): New Zealand Injury Prevention Secretariat, ACC; 2010 [cited 2023 Jun 22]. Available from: https://thehub.swa.govt.nz/assets/documents/costs_injury_New-Zealand_methods_prioritising_allcoation.pdf.
20. Feigin VL, Theadom A, Barker-Collo S, et al. Incidence of traumatic brain injury in New Zealand: a population-based study. Lancet Neurol. 2013;12(1):53-64. doi: 10.1016/S1474-4422(12)70262-4.
21. Wood A, Duijff JW, Christey GR. Quad bike injuries in Waikato, New Zealand: an institutional review from 2007–2011. ANZ J Surg. 2013;83(4):206-10. doi: 10.1111/ans.12106.
22. Jones AR, Smith A, Christey G. Equine-related injuries requiring hospitalisation in the Midland Region of New Zealand: a continuous five-year review. N Z Med J. 2018;131(1483):50-8.
23. National Centre for Classification in Health. International Classification of Disease (ICD-10-AM). 6th ed. Sydney (AU): Australian Institute of Health and Welfare; 2006.
24. Palmer CS, Gabbe BJ, Cameron PA. Defining major trauma using the 2008 Abbreviated Injury Scale. Injury. 2016;47(1):109-15. doi: 10.1016/j.injury.2015.07.003.
25. Baker SP, O'Neill B, Haddon W Jr, Long WB. The injury severity score: a method for describing patients with multiple injuries and evaluating emergency care. J Trauma. 1974;14(3):187-96.
26. Statistics New Zealand. Population estimates at 30 June 2018-20 (2020 boundaries) [Internet]. Wellington (NZ): Statistics New Zealand; 2018 [cited 2023 Jun 22]. Available from: http://nzdotstat.stats.govt.nz/wbos/#.
27. Team R. RStudio: Integrated Development for R [Internet]. Boston, MA (US): RStudio; 2020 [cited 2023 Jun 22]. Available from: http://www.rstudio.com/.
28. Kool B, Ameratunga S, Lee M, et al. Prevalence of risk and protective factors for falls in the home environment in a population-based survey of young and middle-aged adult New Zealanders. Aust N Z J Public Health. 2010;34(1):63-6. doi: 10.1111/j.1753-6405.2010.00475.x.
29. Keller JM, Sciadini MF, Sinclair E, O'Toole RV. Geriatric trauma: demographics, injuries, and mortality. J Orthop Trauma. 2012;26(9):e161-5. doi: 10.1097/BOT.0b013e3182324460.
30. Abey-Nesbit R, Schluter PJ, Wilkinson TJ, et al. Risk factors for injuries in New Zealand older adults with complex needs: a national population retrospective study. BMC Geriatr. 2021;21(1):630. doi: 10.1186/s12877-021-02576-1.
31. Kool B, Chelimo C, Robinson E, Ameratunga S. Deaths and hospital admissions as a result of home injuries among young and middle-aged New Zealand adults. N Z Med J. 2011;124(1347):16-26.
32. Keall M, Tupara H, Pierse N, et al. Study Protocol of a Randomized Controlled Trial of Home Modification to Prevent Home Fall Injuries in Houses with Māori Occupants. Methods Protoc. 2020;3(4):71. doi: 10.3390/mps3040071.
33. Keall MD, Tupara H, Pierse N, et al. Home modifications to prevent home fall injuries in houses with Māori occupants (MHIPI): a randomised controlled trial. Lancet Public Health. 2021;6(9):e631-e640. doi: 10.1016/S2468-2667(21)00135-3.
34. Thornley S, Kool B, Robinson E, et al. Alcohol and risk of admission to hospital for unintentional cutting or piercing injuries at home: a population-based case-crossover study. BMC Public Health. 2011;11:852. doi: 10.1186/1471-2458-11-852.
35. Woods C, Jones R, Usher K. The impact of unintentional alcohol-related falls on emergency departments. Australas Emerg Care. 2019;22(1):22-27. doi: 10.1016/j.auec.2018.11.001.
36. Stewart R, Das M, Ardagh M, et al. The impact of alcohol-related presentations on a New Zealand hospital emergency department. N Z Med J. 2014;127(1401):23-39.

Mā te Whakarongo—a qualitative study exploring the impact of middle ear disease on New Zealand Māori

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ABSTRACT

WHĀINGA (AIM): The study aimed to explore the impact of middle ear disease on the lives of New Zealand Māori. Ear disease is common, yet there is a paucity of research into the effect it has on people's lives, particularly indigenous populations.

TIKANGA (METHOD): The study used Kaupapa Māori-based qualitative methodology and involved a series of seven semi-structured interviews with Māori adults living with middle ear disease.

OTINGA (RESULTS): All participants felt there were delays in recognition and treatment of their ear condition and that there were barriers to accessing healthcare. The ear condition prevented participation in cultural and recreational activities, particularly those involving water. The associated hearing loss affected education and employment opportunities, and together with ear discharge, resulted in social isolation and disconnection from Te Ao Māori (the Māori world). Overall, the condition impacted negatively on mental and spiritual wellbeing. Participants felt that funding for hearing aids, earlier recognition and treatment of the condition and healthcare staff with a better understanding of Te Ao Māori could reduce the morbidity associated with middle ear disease.

WHAKAPAUNGA (CONCLUSION): The study demonstrates that living with middle ear disease presents many challenges and disadvantages for Māori and the importance of early detection and referral to specialist care.

Observational studies suggest that New Zealand Māori have higher rates of otitis media than non-Māori and similar findings have been demonstrated in other indigenous populations around the world.[1–4] Although causality remains uncertain, healthcare access and socio-economic factors resulting from colonisation are likely to have an important role in the higher rates of middle ear disease in New Zealand Māori.[4]

Previous studies regarding the symptom burden of chronic otitis media (COM) have used questionnaires developed from a physician perspective.[5–9] These questionnaires were developed by clinicians experienced in treating middle ear disease, translating experiences (qualitative) into numbers (quantitative). In doing so, judgements are made weighting the symptoms and experiences within the questionnaire. These judgements are informed by their own worldviews and paradigms. There is a paucity of qualitative research related to middle ear disease in any population. Specifically, no studies have considered the effects on quality of life for adult Māori patients living with middle ear disease.

We hypothesise that middle ear disease affects Māori adults in a variety of physical, psycho-social and cultural domains, and affects education and work opportunities. The study aims to explore this impact.

Method

Reflexive statement

The first author (LB) is a Māori otolaryngology trainee working with RG at the time of the study. During training, LB noted that Māori are disproportionately impacted by middle ear disease and recognised the need for further research in this area.[1,3,4] The senior author (RG) is a mid-career Pākehā consultant otolaryngologist. A confluence of critical reflection on outcome measures in middle ear disease and cultural safety (RG) led to questioning of paradigms in reporting of otology symptoms. JS is a Māori health and Kaupapa Māori Researcher who specialises in cancer and chronic conditions. Discussions between LB, RG and JS informed study design, analysis and reporting.

Study design

A qualitative study design with thematic analysis[10–12] using a Kaupapa Māori lens[13] was chosen to gather experiences of Māori adults with middle ear disease using patient interviews. Qualitative research explores in depth the views

of a small number of participants, exploring the lived experience in detail. By analysing the language used and the types of experiences that are common in the group interviewed (thematic analysis), a picture of not only symptoms but the impact of those symptoms can be developed.[10–12] The study was approved by the Health and Disability Ethics Committee (20/NTB/28).

Sampling

To be included in the study participants needed to self-identify as New Zealand Māori, be over 16 years of age and have current clinical follow-up for middle ear disease through the Capital & Coast District Health Board (DHB), Hutt Valley DHB or Wairarapa DHB otolaryngology service.

Potential participants were identified using purposeful sampling[10] by the two otolaryngology clinicians involved in the study. Initial invitation was by a clinical nurse uninvolved in the research, and those interested were provided with an information sheet and invited to be interviewed. Interviews were undertaken at a place and time convenient for the participant. Whānau attendance at interviews was encouraged. A written consent form was completed on the day of the interview.

A semi-structured interview schedule was designed based on the researchers' previous clinical experience with input from Māori. Interviews were conducted between April 2021 and December 2021 by a Māori clinical researcher (otolaryngology registrar). The interview process followed Kaupapa Māori-based principles, acknowledging the importance of te reo Māori (the Māori language) and tikanga (Māori culture) where appropriate, with an emphasis on whakawhanaungatanga (building relationships), and all participants were provided with a koha (gift) of grocery vouchers.[13–15] Five participants were interviewed face-to-face and two by phone. All interviews were audio recorded.

The audio-recordings were transcribed verbatim,[16] with researcher LB checking for correctness of te reo Māori words. The transcripts were analysed drawing off Braun and Clarke reflexive thematic analysis.[17] Researchers familiarised themselves with the data, generated codes utilising NVivo (version 10 software) and developed and revised themes and sub-themes.[16] Participants were offered their transcripts to review and are referred to below as P1–7. Recruitment stopped when the interview transcripts no longer revealed new themes.[11] This was cross-checked with the clinicians' own extensive experience interviewing patients to clarify if any important themes may be absent.

Results

Participants were seven Māori adults residing in the Wellington region but with whakapapa (genealogy) to iwi (tribes) around Aotearoa New Zealand. Two additional individuals were invited but declined to participate. Six of the participants have experienced middle ear disease since childhood, with one having symptom onset in adulthood. All participants had undergone previous surgical intervention for their ear pathology and have ongoing symptoms requiring otolaryngology input. For participant demographics, see Table 1.

The data analysis revealed living with chronic middle ear disease impacts significantly on Māori patients and their whānau. Participants' quotations are presented in Table 2 and briefly in the text. Themes align with the Te Whare Tapa Whā model described by Sir Mason Durie.[18] These themes are taha tinana (physical wellbeing), taha hinengaro (mental wellbeing), taha wairua (spiritual wellbeing) and taha whānau (family wellbeing). Additionally, there were themes involving interface with the healthcare system and the impact of the symptoms on education and work opportunities which relate to the Te Pae Māhutonga model, also described by Sir Durie.[19]

Taha tinana (physical wellbeing)

Participants reported that the physical symptoms left them feeling socially isolated, with particular emphasis on the smell from ear discharge (taringa pirau) and the hearing loss (taringa turi).

Hearing loss impaired communication, particularly in large groups, in the presence of background noise or on the phone, which is very isolating and mentally exhausting.

> *"I get mentally tired at the end of the day too because my head is trying so hard to listen to people and I just get brain fade." – P3*

Participants described their suppositions about how others see them. They were often worried about appearing rude when not answering a question they had not heard or would feel unintelligent when repeating things that had already been said.

Table 1: Participant demographics.

	Age (years)	Sex
Participant 1	52	M
Participant 2	59	M
Participant 3	41	F
Participant 4	50	M
Participant 5	25	F
Participant 6	35	M
Participant 7	64	F

Table 2: Participant quotations.

Category	Quote
Tinana	
Taringa turi	*"I can't hear them, and I get anxious about it [phone] ringing, like I can see it ringing there but I don't want to pick it up just in case it's something really important." – P3* *"If we're in a big social gathering and a big conversation is going on and everyone's talking, I can't participate. I can't hear what's going on properly." – P4*
Taringa pirau	*"My infection was terrible in the sense of the smell. You could smell it. A lot of it was more embarrassing when I was at school." – P2* *"I was that dirty little Māori child. Yeah. I had the cold sores, had the discharging ear. You know, I don't think anybody really wanted to get close to me." – P7* *"When you see that discharging, everybody goes, oh what's wrong with your ear? And then it will dry up and leave like a resin on the outside." – P2* *"Definitely had a lot of discharge when I was a kid. I was just, I was ostracised, I think. And I never…. I didn't make friends." – P7*
Hinengaro	*"And to lose all of those things it was actually, I was right down in my lowest, I bloody broke into tears because we were back where we started." – P4* *"It makes me feel like a bit of an idiot when I know I'm not an idiot, I know what I'm talking about." – P7* *"Personal things at home like the fire, we've got a beautiful big fireplace, to hear that wood crackling, the pinecones, you smell them but don't hear them crackle no more." – P2*
Wairua	
Te reo	*"And learning reo as well kind of gets difficult. Especially when I can't wrap my hearing around what they're actually saying; it might be 'na' instead of 'nga'." – P5*

Table 2 (continued): Participant quotations.

Wai	*"I've always liked providing for the family and stuff, but it's just the love of the deep. I went two years without going for a snorkel." – P4* *"I always watch the boys go diving, I just hold the rope when they go down. So that's as much as I get." – P6* *"I love swimming and diving and I can't go diving because of the hole in my eardrum. I just get middle ear infections all the time." – P1* *"I would just beg to go swimming and of course then I'd end up with earaches crying my eyes out." – P5*
Whānau	*"Being intimate with your partner, you don't hear certain things. And you're like oh what's wrong with her? And she has said something to you, but you didn't hear her. It can cause an argument." – P2* *"Definitely always felt like I was on the outside. Definitely didn't feel like I was included in anything. I was really very much on the outside of everything." – P7*
Interface with health system	*"We fell off the radar for a little bit and tried to keep up with all the doctors and stuff and all the records throughout everywhere." – P6* *"Na, you fullas have actually been quite good lately. Just that when I didn't have any doctors or follow-up from my childhood to now was quite difficult." – P6* *"Yeah, it never, never got picked up. They were in the system and the system just let them down every single time." – P7* *"I spent like maybe eight months going to my GP before they decided to even refer me." – P3* *"I kind of just wish it [surgery] was done earlier." – P5* *"We can come here to as many appointments at the hospital as you want, but, you know, what would really benefit him would be one hearing aid that was decent, you know, it's crazy." – P1* *"Whakawhanaungatanga [connecting to each other] is really important. And they don't even realise that it is important, you know what I mean?" – P7* *"From the time that I was referred, I feel like the service that I've had has been just outstanding … when it comes to anything hospital related, I found that people know my history. You know, people … know what questions to ask me." – P3*

> *"People treated me a bit differently because of the fact that they thought I was, you know, dumb." – P7*

One of the most common and distressing manifestations of middle ear disease is the recurrent episodes of taringa pirau. Participants all had a similar experience of being subject to bullying in childhood, reduced attendance at school and avoidance of social situations in later years.

> *"I'd be around people who were like, what's that smell. And I knew it was my ears." – P5*

Taha hinengaro (mental wellbeing)

Participants reported avoiding social situations as they are unable to participate in conversation. To counter the embarrassment and isolation, one participant relied heavily on alcohol at social events. Some participants found they became completely dependent on a partner or support

person to communicate on their behalf.

"People think I'm anti-social because I don't go to the Christmas party and it's just no good for me. I just sit there, and I get drunk." – P4

Some participants reported becoming depressed and anxious from the culmination of the associated symptoms, the social and cultural restrictions and the chronic nature of the condition.

"I felt like I was losing myself and that's why I was so upset, you know." – P4

Taha wairua (spiritual wellbeing)

Several participants felt that their middle ear pathology and subsequent hearing loss made it difficult to learn or prevented them from learning te reo Māori, which impacted on their sense of cultural identity and ability to access Te Ao Māori.

"Then I was ignorant to the Māori language and ignorant to the Māori people." – P2

Participants felt their condition prevented them from taking on certain cultural responsibilities such as kaikaranga or kaikōrero (welcoming and speaking on the marae) and dictated what position they held within their iwi, hapū (subtribe) and marae. Furthermore, as noted by one of the participants, kapa haka relies heavily on one's ability to learn via listening.

"Na I just stay back with the crowd; I don't get up and talk or anything." – P6

"Do you think that might have been different?" – I

"Yeah definitely. If I knew how to talk, then I would be able to say something. Cos I'm not someone to sit back. But there's other ways around it, you just go into the kitchen and just help out there." – P6

"So, it changes what position you have on the marae?" – I

"Yeah." – P6

For Māori, wai (water) forms the basis of many traditional cultural practices. Iwi, hapū and marae have intimate connections with certain bodies of water that represent their whakapapa. To be immersed in that water is a connection to their tūpuna (ancestors). Participants felt that one of the most significant consequences of suffering from middle ear disease was the inability to connect with water.

"We literally had a huge connection with the water. It was like everything you know." – P7

Several participants reported an inability to gather kaimoana (seafood) because of their ear disease. This is often an important social and cultural activity, a way of spending time with friends and providing kai (food) for whānau and events such as tangihanga (funeral). Furthermore, not being able to provide kaimoana results in the added cost of having to purchase kai. Simple daily activities like having a bath or shower were a challenge.

"I mean, you can't go diving with the boys who go out. Yeah, can't go get kaimoana and all that sort of stuff. For like the tangi the other week and you can't go with them." – P1

"I do my best to keep my ear dry. Cos, I know that's what causes the infections. So, what I always do when I'm having a shower or washing my hair is I just fold the top half of my ear over my ear." – P1

The inability to swim is also a barrier to physical activity and can contribute to weight gain and general decline in physical and mental wellbeing.

"I missed out on all the fun of going swimming and all that sort of stuff. And also, the extra exercise I could get from swimming." – P1

Some participants felt their wairua had been tampered with following surgery on the head or after having hair removed.

"You know, your head has been played with now and that to me blew me away. Having somebody go inside your head when you didn't really have to." – P2

"It was just the fact that they cut my hair ... that was my main problem, because I woke up and what was sitting on my chest was my hair." – P5

Taha whānau (family wellbeing)

The condition and particularly the resulting hearing loss was reported as a cause of strain on relationships.

"And they are the same too, though they can't hear either. So, we're all yelling at each other. So, what ends up happening is we don't, we just don't because it's just too much effort. But there's other ways we interact and it's like through kapa haka and singing and stuff." – P3

Some patients have previously said they are unable to sleep in the same bed as their partner when their ear is discharging due to the smell. There was a significant impact on interpersonal relationships, deepening the feelings of isolation. One participant found that her hearing loss and otorrhea made it more difficult living away from her iwi and whānau support networks as it was more challenging to communicate and develop connections with local rōpū (groups), such as kapa haka.

Interface with health system

Consistent with findings in other medical research in Aotearoa New Zealand, all participants felt that there were multiple barriers to accessing treatment for their middle ear condition and several reported experiences of being subject to racism.[20,21]

"We often got treatment like, well, you know, what do you expect, you are fat and black." – P1

"It's because I look Pākehā that I've been able to get more or better accessibility." – P7

Participants recalled challenges navigating the health system due to location changes and an inflexible disconnected system creating challenges for continuity of care with a chronic health condition. Lack of funding for hearing aids was a common complaint.

Participants reported the effects of a lack of connection with individual healthcare providers (whakawhanaungatanga), which was detrimental to the therapeutic relationship. Some felt they were not listened to or taken seriously, leading to a delay in referral. Conversely, positive relationships in healthcare settings made a big difference.

Education and work opportunities

The chronic nature of the condition combined with symptoms of hearing loss and ear discharge impacted on participants' ability to benefit from education. Some recalled the embarrassment of the smell made them reluctant to attend school. This also affected work opportunities and work relationships.

Discussion

The results of this study demonstrate that middle ear disease has a profound impact on the quality of life of adult Māori, in addition to far reaching effects for their whānau. The implications are devastating and complex, ranging from physical manifestations that cause social anxiety and withdrawal from culture and community, to impacts on mental health and wairua that result in disconnection from one's identity.

Of the physical symptoms experienced, hearing loss and otorrhea had the most profound impact on quality of life. The mental exhaustion associated with hearing loss has been previously described.[22] Hearing loss and otorrhea frequently resulted in social isolation, embarrassment and discrimination. A study of a Nepalese population suffering from middle ear disease showed that all individuals experienced and feared the stigma associated with their ear condition.[9] The invisible disability of hearing loss combined with the social stigma of the discharge can mean this suffering is hidden. This potentially limits collective health advocacy of individuals experiencing this condition.

While most research quality of life tools designed for middle ear disease have otorrhea and hearing loss as data points, this study shows the shocking depth and breadth of the symptom impact, often with far reaching consequences from childhood to adulthood. Many physicians might describe ear discharge as a minor annoyance, especially if intermittent, not reflecting the patient's experience. Therefore, both in clinical practice and in research, more significance needs to be given to this symptom. The sense of isolation, challenges connecting with others and shameful feelings about symptoms influenced the

mental wellbeing of patients. It was common to experience the sense of losing one's identity as a result of the condition, as individuals were often unable to engage in recreational, social and cultural activities that previously defined them.

Several patients felt that their condition had either prevented them from or made it very difficult to learn te reo Māori, and in some situations deterred them from taking on cultural responsibilities and privileges such as gathering kaimoana for tangihanga and speaking on the marae. This can cause a loss of right that may result in abdication of important responsibilities to other family members and subsequent implications for that individual's tamariki.

In Te Ao Māori, water is the essence of life and represents the blood of Papatūānuku (the earth mother).[23] Patients with middle ear disease who expose their ears to water risk infection of the mucosa, resulting in purulent and malodorous discharge. Most participants reported that the inability to enter and connect with water was one of the most significant consequences of middle ear disease.

Certain aspects of middle ear disease and the treatment were noted to have deeper influences on the individual's wairua. Surgery on the head and cutting of hair resulted in a violation of tapu (sacred/prohibited) for some individuals. It is important to consider carefully what to tell Māori patients about upcoming treatment such as surgery and what to expect in the postoperative period. Cultural safety training, including developing local hospital policies, e.g., tikanga guidelines, will be important to foster a safe place for Māori in all wellbeing domains.[24,25]

The higher rates of middle ear disease in Māori are likely related in part to long reaching effects from colonisation.[21] The results suggest that middle ear disease can result in further disconnection from Te Ao Māori, poorer educational and work opportunities and subsequently lower socio-economic status and poorer health outcomes. Exclusion from schooling when ears are leaking and isolation from peers due to hearing loss can have far reaching long-term impacts on affected individuals. Furthermore, the ear condition often resulted in strain with personal or work-based relationships.

The inequities that exist in access to healthcare for Māori due to colonisation, marginalisation and institutional racism are widely known. The culturally unsafe environment of our healthcare systems contributes to the significant disparities in health outcomes for Māori.[21] Participants reported challenges with the health system interface, providing further evidence of these inequities. While the study participants proved resilient and were able to access care, often through perseverance, there are without doubt many Māori in the community with active middle ear disease that have been unable to navigate the healthcare system to access treatment.

Funding models for hearing aids leave many with out-of-pocket significant expense, or worse still, no access to unaffordable devices. As childhood middle ear disease is not considered permanent, some adults no longer had access to fully funded hearing aids, especially if they were not able to access them as children. These system issues represent an example of institutional racism which is not responsive to the needs of Māori.

Culturally safe practice should inform shared decision-making and advocacy for policy improvements. Clinicians and health systems that understand the significance of symptoms of common ear disorders can be better placed to prioritise these appropriately. This may include earlier referrals, higher triage scores for specialist services/surgeries and types of operations that support the goals of the patient, such as return to water-based activities. Understanding the personal and cultural importance of these goals can inform practice.

A strength of the study is it provides detailed information on the impact of middle ear disease for Māori and the health inequities that exist across multiple domains. The study used a Māori cultural lens to consider wider wellbeing (hauora), enabling the voice of Māori in a common condition. This research is part of an effort to re-imagine the evidence infrastructure used to inform policy and decision making by including patient experiences. Prior to this study there was a paucity of qualitative information available regarding the experiences of Māori with middle ear disease.

Due to the lack of qualitative research in middle ear disease, comparisons with other groups were not possible in this study. We might infer there is likely to be some overlap with other groups; however, the implications on status within hapu and marae, spiritual connection with water and violation of tapu during surgery may well represent true differences. Further research into other ethnic and social groups with middle ear disease would be interesting.

We acknowledge that the researchers have prior clinical and personal experience that has influenced the formation of methodology and thematic analysis.

We interviewed only a small number of Māori, all with different backgrounds, whakapapa and connection to Te Ao Māori, and caution is needed in generalising these findings to all Māori. However, the study provides valuable insights on the impacts of hearing loss on the lives of indigenous and New Zealand adults, populations understudied in this area. We acknowledge those not included in the study, whose experiences may be different, including those with unmet need and those who have completed treatment.

Qualitative research has an important role in informing quantitative research to measure appropriate variables. By nature, it uses small sample sizes to gather the depth and significance of experiences. This informs quantitative methods such as surveys and disease-specific quality of life tools giving importance to the patient voice.[10,11,12] This research is particularly relevant with Māori whose experiences may differ from the majority of clinicians, policy makers and researchers.

COMPETING INTERESTS
Nil.

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REFERENCES
1. Giles M, Asher I. Prevalence and natural history of otitis media with perforation in Māori school children. J Laryngol Otol. 1991;105:257-260. doi: 10.1017/s0022215100115555.
2. Coleman A, Wood A, Bialasiewicz S, et al. The unsolved problem of otitis media in indigenous populations: a systematic review of upper respiratory and middle ear microbiology in indigenous children with otitis media. Microbiome. 2018;6(1):199. doi: 10.1186/s40168-018-0577-2.
3. Thompson PAP. Occurrence of hearing loss & middle ear dysfunction among primary school children in the Christchurch area [master's thesis]. Christchurch (NZ): University of Canterbury; 2008.
4. McCallum J, Craig L, Whittaker I, Baxter J. Ethnic differences in acute hospitalisations for otitis media and elective hospitalisations for ventilation tubes in New Zealand children aged 0-14 years. N Z Med J. 2015;128(1416):10-20.
5. Baumann I, Gerendas B, Plinkert PK, Praetorius M. General and disease-specific quality of life in patients with chronic suppurative otitis media--a prospective study. Health Qual Life Outcomes. 2011;9:48. doi: 10.1186/1477-7525-9-48.
6. Indius JH, Alqaderi SK, Kjeldsen AD, Heidemann CH. Middle ear disease in Danish toddlers attending nursery day-care – Applicability of OM-6, disease specific quality of life and predictors for middle ear symptoms. Int J Paediatr Otorhinolaryngol. 2018;110:130-134. doi: 10.1016/j.ijporl.2018.04.031.
7. Jung KH, Cho YS, Hong SH, et al. Quality-of-life assessment after primary and revision ear surgery using the chronic ear survey. Arch Otolaryngol Head and Neck Surg. 2010;136(4):358-365. doi: 10.1001/archoto.2010.24.
8. Nadol JB Jr, Staecker H, Gilklich RE. Outcomes assessment for chronic otitis media: the Chronic Ear Survey: Laryngoscope. 2000;110(3 Pt 3):32-35. doi: 10.1097/00005537-200003002-00009.
9. Skilton MK, Poole N, Metcalfe CW, et al. The impact of ear disease and hearing impairment on the lives of Nepali patients in Pokhara: a qualitative study. Int Health. 2016;8(2):101-107. doi: 10.1093/inthealth/ihv052.
10. Clarke V, Braun V. Successful Qualitative Research: A Practical Guide for Beginners. California (US): SAGE Publications; 2013.
11. Eakin JM. Educating critical qualitative health researchers in the land of the randomized controlled trial. Qualitative Inquiry. 2015;22(2):107-118. doi: 10.1177/1077800415617207.
12. Busetto L, Wick W, Gumbinger C. How to use and assess qualitative research methods. Neurol Res Pract. 2020;2:14. doi: 10.1186/s42466-020-00059-z.
13. Curtis, E. Indigenous positioning in health research: The importance of Kaupapa Māori theory-informed practice. AlterNative. 2016;12:396-410. doi: 10.20507/AlterNative.2016.12.4.5.
14. Cram F, Phillips H, Tipene-Matua B, et al. A 'Parallel Process'? Beginning a constructive conversation about a Māori methodology. J Bioeth Inq. 2004;1(1):14-19. doi: 10.1007/BF02448902.
15. Jones B, Ingham T, Davies C, Cram F. Whānau Tuatahi: Māori community partnership research using a

Kaupapa Māori methodology. MAI Review. 2010;3.
16. QSR International Pty Ltd. NVivo. Burlington, MA (US): QSR International; 2020.
17. Clarke V, Braun V. Teaching thematic analysis: Overcoming challenges and developing strategies for effective learning. Psychologist. 2013;26:120-123.
18. Rochford T. Whare Tapa Whā: A Māori model of a unified theory of health. J Prim Prev. 2004;25(1):41-57. doi: 10.1023/B:JOPP.0000039938.39574.9e.
19. Durie M. (1999). Te Pae Māhutonga: a model for Māori health promotion. Health Promotion Forum of New Zealand. 49.
20. Jansen P, Bacal K, Buetow S. A comparison of Māori and non-Māori experiences of General Practice. N Z Med J. 2011;124(1330):24-9.
21. Robson B, Harris R, editors. Hauora: Māori Standards of Health IV: A study of the years 2000-2005. Wellington (NZ): Te Rōpū Rangahau Hauora a Eru Pōmare; 2007.
22. Davis H, Schlundt D, Bonnet K, et al. Understanding listening related fatigue: Perspectives of adults with hearing loss. Int J Audiol. 2021;60(6):458-468. doi: 10.1080/14992027.2020.1834631.
23. Te Rūnanga O Ngāi Tahu. Freshwater Policy [Internet]. Christchurch (NZ): Environment Canterbury Regional Council [cited 2022 Aug 2]. Available from: https://api.ecan.govt.nz/TrimPublicAPI/documents/download/2738412.
24. Lacey C, Huria T, Beckert L, et al. The Hui Process: a framework to enhance the doctor-patient relationship with Māori. N Z Med J. 2011;124(1347):72-78.
25. Pitama S, Robertson P, Cram F, et al. Meihana Model: A Clinical Assessment Framework. N Z J Psychol. 2007;36(3):118-125.

The deconstruction of chronic orofacial pain and a hiding inhibition pathway of orofacial pain: the trigeminal proprioceptive mesencephalic periaqueductal gray pathway

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ABSTRACT

Chronic orofacial pain has a significant negative impact that influences individuals' quality of life and our society. The prevalence is around 11.2% to 33.2% and remains high in females. Currently, there are two main diagnostic classification systems that are used internationally for chronic pain: the International Classification of Diseases, 11th Revision (ICD-11), which was published by the World Health Organization (WHO) in 2018, and the International Classification of Orofacial Pain, which was published by the International Association for the Study of Pain (IASP) in 2020. Deficits in ascending and descending pain modulation pathways may be involved in the chronic pain pathophysiology. A newly described "trigeminal proprioceptive mesencephalic periaqueductal gray pathway" is considered to be the mechanism of action of occlusal appliance in managing orofacial pain. The genetic basis of chronic orofacial pain is not yet fully understood, but a genetic susceptibility involving multiple genes among the peripheral nerves, brainstem and higher brain regions to regulate and suppress the transmission of pain signals, thereby modulating the perception of pain, is likely.

Pain is defined as "*an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage*", and this definition was recommended by the Subcommittee on Taxonomy and adopted by the International Association for the Study of Pain (IASP) Council in 1979,[1] but was expanded by following contextual points in 2020:[2]

- Pain is always a personal experience that is influenced to varying degrees by biological, psychological and social factors.
- Pain and nociception are different phenomena. Pain cannot be inferred solely from activity in sensory neurons.
- Through their life experiences, individuals learn the concept of pain.
- A person's report of an experience as pain should be respected.
- Although pain usually serves an adaptive role, it may have adverse effects on function and social and psychological wellbeing.
- Verbal description is only one of several behaviours to express pain; inability to communicate does not negate the possibility that a human or a nonhuman animal experiences pain.

The common feature of chronic pain is sensitisation of the neural pathways, and this could involve peripheral and/or central sensitisation. Three main mechanisms of chronic pain were described: nociceptive, neuropathic and nociplastic (Table 1).[3] By the IASP definition, nociceptive pain is a response to real or potential tissue-damaging stimuli that activates a neural pathway; neuropathic pain is caused by a lesion or disease of the somatosensory nervous system; and nociplastic pain is a type of pain that develops from the improper processing of pain signals without any obvious signs of tissue injury or distinct somatosensory system dysfunction.[4]

The term "orofacial pain" describes the pain that arises from the oral cavity, face and neck.[5] The term "chronic orofacial pain" refers to a group of painful regional disorders or conditions that have a persistent, unremitting pattern for 3 months or longer. Although there is no clear delineation of when the acute pain becomes chronic, the International Classification of Diseases and the International Classification of Orofacial Pain (ICOP) suggests any pain persisting beyond the expected healing time (more than 3 months and on at least 15 days per month) is pathological.[6,7]

The prevalence of orofacial pain has been

reported between 11.2% and 33.2%.[8–12] Females have a higher prevalence of chronic orofacial pain than males—it is reported to be twice as high in adult females compared to adult males.[8,13] Regarding race and ethnicity, study results are mixed, with some suggesting that white females showed the highest incidence, whereas others suggest there were no racial differences.[14–16] For example, a study of Jewish and Arab–Israeli patients found no differences.[17] Another study showed higher incidence in African Americans than in Asians.[18] Financial factors, cultural differences and a lack of access to care may be some of the reasons for racial disparities. [19] Indigenous peoples, according to studies, display fewer obvious pain behaviours and are reluctant to talk about the causes of their suffering, maybe because pain that weakens a person is seen as a sign of weakness.[20] In New Zealand, the pronounced under-attendance of Pacific and Asian races is evidence of ethnic differences in access to chronic pain care.[21,22] The most frequently reported orofacial pains were temporomandibular disorder (TMD), burning mouth syndrome, persistent idiopathic dentoalveolar pain (atypical odontalgia) and persistent idiopathic facial pain (atypical facial pain).[8,23] The peak age ranges vary from among different types of orofacial pain. For example, the peak age incidence for TMD is from 20 to 40 years of age.[24] Burning mouth syndrome is from around 50 to 70 years of age.[25] Persistent idiopathic dentoalveolar pain is from around 35 to 63.[26]

Classification

There are two main diagnostic classification systems that are currently used internationally for chronic pain. The International Classification of Diseases, 11th Revision (ICD-11), published by the World Health Organization in 2018, includes codes and classifications for a wide range of diseases and conditions across all medical specialities.[27]

The ICD-11 divides chronic primary pain into five subgroups: 1) chronic primary visceral pain, 2) chronic widespread pain, 3) chronic primary musculoskeletal pain, 4) chronic primary headache or orofacial pain, and 5) complex regional pain. Chronic primary orofacial pain and chronic primary TMD pains are coded within chronic primary headache or orofacial pain under this classification. However, chronic migraine, burning mouth syndrome, chronic tension-type headache, chronic cluster headache and hemicrania continua are coded within other categories. The phrase "chronic primary pain" was selected and is intended to be agnostic with regard to aetiology. It also tries to avoid the antiquated distinction between "physical" and "psychological" factors, as well as terminology that is vague or imprecise, (for example, "nonspecific").[28] Apart from chronic primary pain, the ICD-11 also indicates other chronic pain categories, such as chronic cancer-related pain, chronic post-surgical or post-traumatic pain, chronic secondary musculoskeletal pain, chronic secondary visceral pain, chronic neuropathic pain, chronic secondary headache or orofacial pain, other specified chronic pain and chronic pain (unspecified).

The ICOP, 1st edition, was developed by the IASP in 2020.[7] It is a specialised classification system specifically focussed on orofacial pain conditions, which provides a framework for the diagnosis and classification of orofacial pain disorders. It provides detailed descriptions, diagnostic criteria and classification guidelines for various types of orofacial pain disorders. It classified orofacial pain into seven groups: 1) orofacial pain attributed to disorders of dentoalveolar and anatomically related structures, 2) myofascial orofacial pain, 3) temporomandibular joint (TMJ) pain, 4) orofacial pain attributed to lesion or disease of the cranial nerves, 5) orofacial pain resembling presentations of primary headaches, 6) idiopathic orofacial pain, and 7) psychosocial assessment of patients with orofacial pain. However, "chronic pain" was described within some of the subtypes.

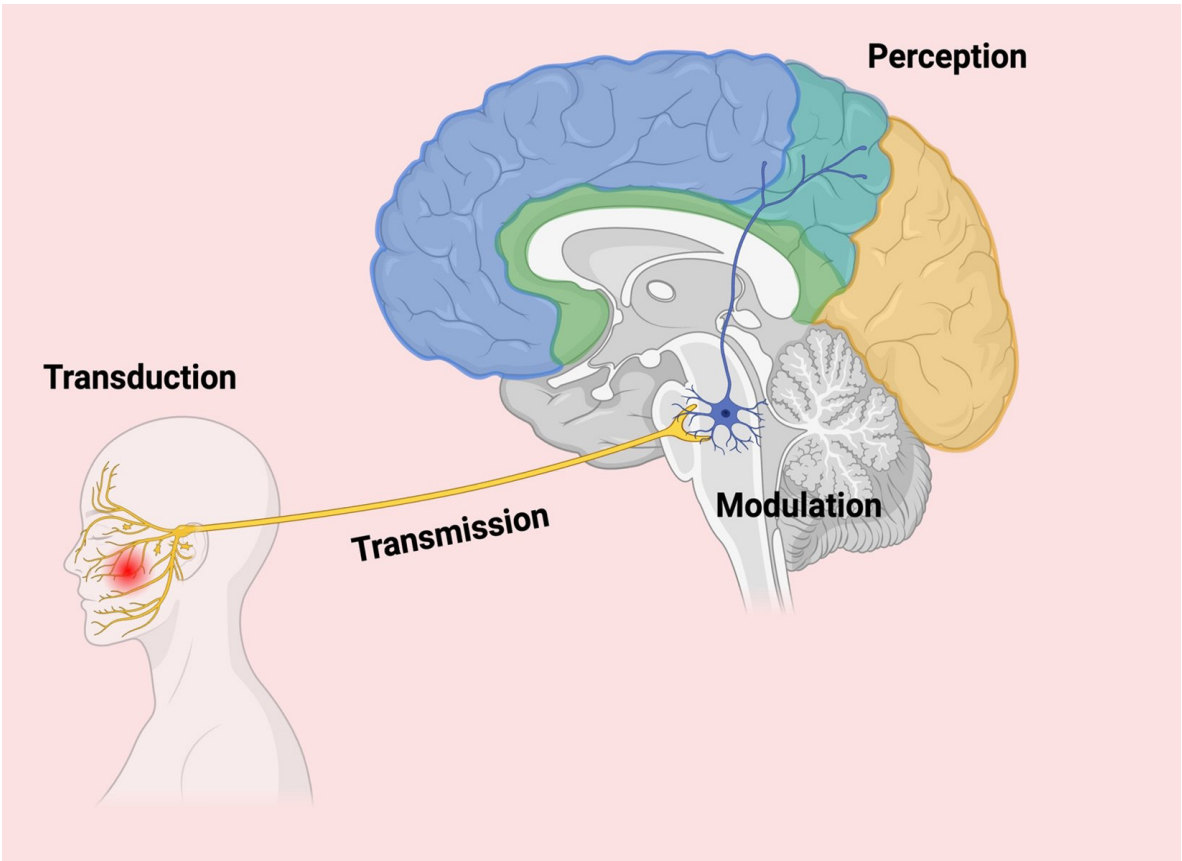
Orofacial pain pathway

In general, there are four major pain processes, which include transduction, transmission, modulation and perception (Figure 1).[29] The orofacial pain pathways might include primary afferent neurons, trigeminal ganglion, brainstem nociceptive neurons and higher brain function that controls orofacial nociception.[30] The trigeminal nerve is a sensory nerve that innervates the orofacial region. Although C, A-delta and A-beta fibres are the most common names for sensory nerve fibres (neurones), there are others, and to varying degrees they can respond to chemical, thermal and mechanical energy.[31] In general, there are three major classes of nociceptors: Aδ mechanosensitive nociceptors, Aδ mechanothermal nociceptors and polymodal nociceptors (C-fibres). The peripheral nociceptors are activated by the chemical factors from damaged tissue, such reactive oxygen species, protons, kinins, prostanoids, adenosine triphosphate, serotonin, histamine, and neurogenic

Table 1: Three main mechanisms of chronic pain.

	Nociceptive pain	Neuropathic pain	Nociplastic pain
Causes	Actual or potential tissue damage	Diseases or damage of the nervous system	Dysregulation of nociceptive process. No evidence of tissue or nerve damage
Examples	Toothache, infection, mucosal ulcers and trauma	Trigeminal neuralgia, post-herpetic neuralgia and diabetic neuropathy	Fibromyalgia, irritable bowel syndrome, chronic lower back pain and temporomandibular disorder
Signs and symptoms	Well-localised pain, with infrequent or no sensory deficits	Electrical-like, lancinating pain and follow dermatomal distribution. Sensory deficits (numbness and tingling) are common. Neurological weakness may present if motor nerve is affected	Diffused, widespread aching and not confined to an anatomical structure. Often associates with psychological stress
Medical management	NSAIDs, paracetamol, opioids and peripheral management	Tricyclic antidepressants, carbamazepine, gabapentinoids, SNRI and lidocaine	Tricyclic antidepressants, gabapentinoids and SNRI

Figure 1: The pathway of pain perception (created with BioRender.com).



substances, and inflammation mediators from the immunocytes, such as cytokines (IL-1beta, IL-6, IL-8, tumour necrosis factors), neurotrophins and neuropeptides, after receiving repetitive noxious stimuli from infection and inflammation.[32] This process is called **transduction**—when the chemical, thermal and/or mechanical energy is changed into electrical signals.[29] The key element in the **transmission** of nociceptive signals is the action potential.[33] The first order neuron transmits the pain signals to the trigeminal ganglia (similar to dorsal root ganglia). The pain signals then transmit to the second order neurons in the trigeminal nucleus (main sensory nucleus and spinal trigeminal nucleus) at the brainstem. The second order neurons decussate at the brainstem. The ventral post-eromedial nucleus of the contralateral thalamus is where the second order neurones' axons end (trigeminothalamic tract). The third order neuron in the thalamus then connects to the sensory cortex. Pain perception occurs at this level and could be influenced by transmission, modulation and cognitive evaluation.[34] **Modulation** is the process by which the normally functioning nervous system adjusts to changes in and around the body.[35]

Pain perception, regulation and inhibition

Based on the previous study (positron emission tomography and functional magnetic resonance imaging), six areas of the brain have been identified, and are thought to contribute to the acute pain process.[36] They are the primary somatosensory cortex, secondary somatosensory cortex, anterior cingulated cortex (ACC), insula, prefrontal cortices (PFC) and thalamus (see Table 2 and Figure 2). However, chronic pain is a complex sensory and emotional experience that includes biological, psychological and social factors. In our brain, emotions are thought to be regulated by the frontal cortex, amygdala, ACC, insula and several interconnected structures.[37] Chronic pain often engages these brain areas for cognitive and emotional processes, suggesting that this component of pain may have a distinguishing characteristic between chronic and acute pain.[38] For example, research showed that insula and PFC connectivity was increased in chronic pain studies.[39,40] Since the insula and PFC are both engaged in emotion, motivation and pain modulation, this suggests that the processing of pain may have an impact on the pain perception.[41] Changes in these centres are thought to be associated with the chronification of pain.[42] A recent study also suggested that an activated cingulate cortex (emotional and cognitive processing) insula pathway could induce and maintain nociceptive hypersensitivity in the absence of peripheral noxious stimuli. This pathway may facilitate the transition from acute to chronic pain.[43] An imaging study has demonstrated that amplification of the thalamic, insular and secondary somatosensory cortex responses has been linked to abnormal pain that is elicited by allodynia. In addition, several pathways such as ACC–amygdala, ACC–thalamus–amygdala and ACC–periaqueductal gray–rostromedial ventral medulla–spinal dorsal horn that are associated with ACC might be activated in chronic pain conditions.[44–46] These suggest that ACC plays an important role in the initiation, development and maintenance of chronic pain. In the orofacial region, several studies have found increased PFC, ACC and insula activities in chronic pain conditions.[47–50]

Table 2: Pain areas of activity in the brain.

Brain region	Activity
Primary somatosensory cortex	Sensory discrimination—determines where pain message is coming from[34,51]
Secondary somatosensory cortex	Pain intensity-related activation [52,53]
Anterior cingulated cortex (ACC)	Integration of sensory, executive, attentional, emotional and motivational components of pain and pain intensity[54,55]
Insula	Pain perception, modulation and contribution of chronification[56,57]
Prefrontal cortex (PFC)	Pain processing, modulation, induction of pain chronification[58–60]
Thalamus	Receiving, processing and transmitting to various parts of the cortex [61,62]

Figure 2: Ascending pain pathway (created with BioRender.com).

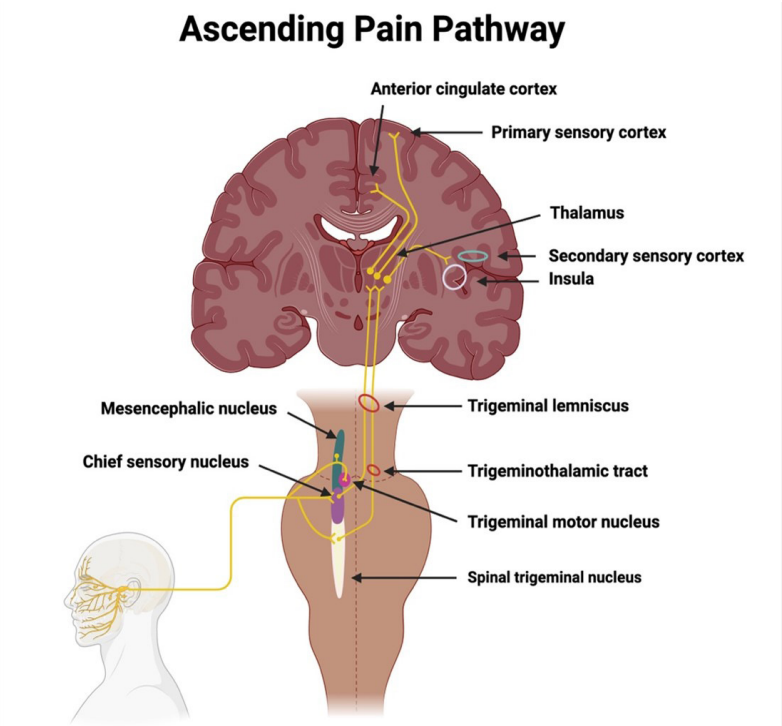


Figure 3: Descending pain pathway (created with BioRender.com).

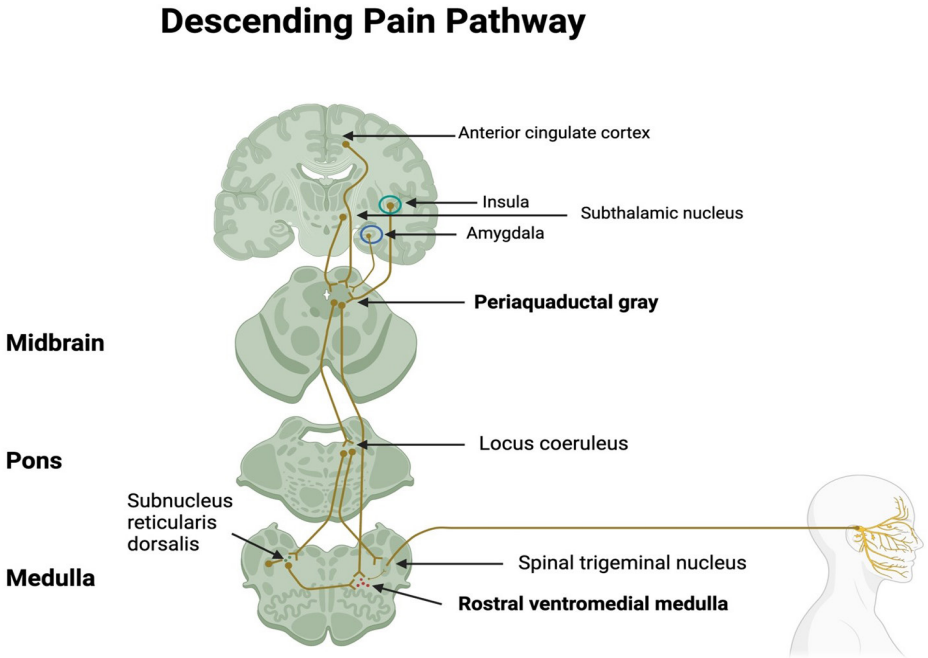
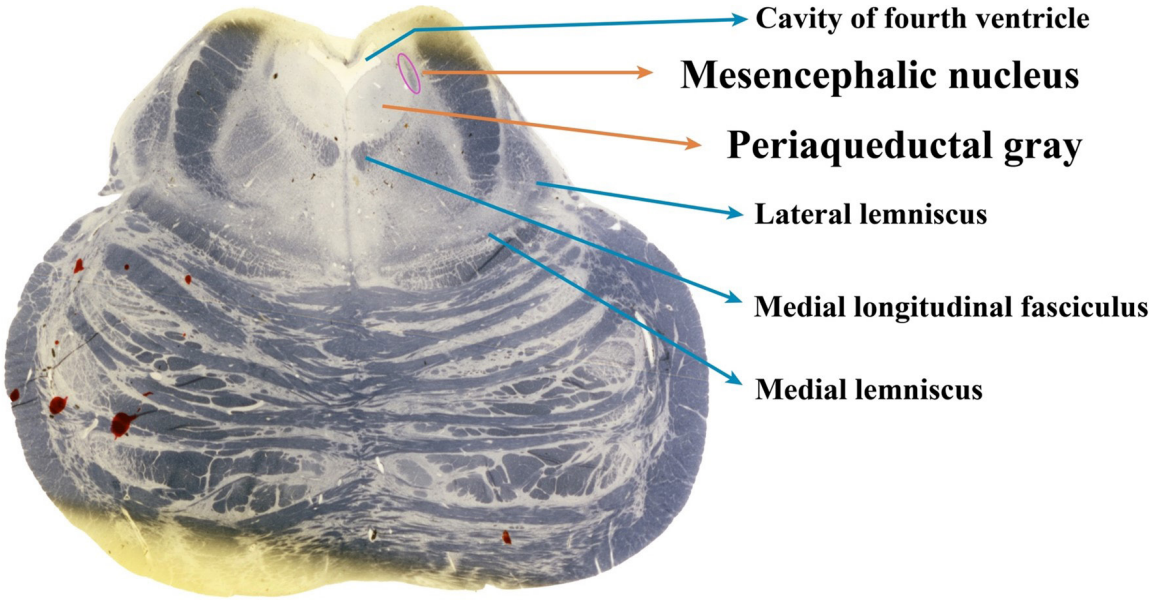


Figure 4: The closed association between mesencephalic nucleus and periaqueductal gray at the level of upper pons (from W D Trotter Anatomy Museum, University of Otago).



The pain inhibition pathway refers to the complex network of structures and processes in the body that modulate or suppress the transmission of pain signals (Figure 3). In general, there are four main components that involved pain inhibition, such as the "gate control theory" at the peripheral nervous system,[63] the presence of inhibitory interneurons at the spinal cord,[64] the pain descending pathway (periaqueductal gray and rostroventromedial medulla)[65] and pain modulation centres in the brainstem and brain such as the thalamus, amygdala and PFC.[58,66] In addition, several brainstem regions, such as periaqueductal gray matter, rostral ventromedial medulla, locus coeruleus and subnucleus reticularis dorsalis, are considered as key structures that modulate pain.[67–69] It is thought that the inhibitory and facilitatory systems of descending pain work together to maintain a baseline condition of sensory processing.[70] Several studies have shown that dysfunction of the periaqueductal gray–rostroventromedial medulla–dorsal horn/spinal trigeminal nerve pathways may lead to a pronociceptive state, eventually facilitating neurotransmission and promoting pain.[71,72] In addition, the pain inhibition pathway is associated with the emotional centres such as amygdala, insula and PFC in the brain.[73] Studies using functional MRIs have shown an increased activity in the emotion regions of the brain such as PFC, insula and cingulate, but decreased activity in the descending inhibition pathway at the brainstem in chronic pain patients.[74–77]

Dysfunction of inhibition pathway could comprise both an aberrant pain response to a non-noxious stimulus at the injury site or surrounding areas and a heightened pain response to a noxious stimulus at the injury site or surrounding regions, referred to as primary and secondary allodynia. These have been demonstrated in several chronic pain conditions such as fibromyalgia, irritable bowel syndrome, chronic lower back pain and TMD.[78–82] Moreover, the inhibition pathway might be involved with the "placebo-related" changes seen in pain management.[83] This may also relate to meditation and positivity, which have a positive effect on pain improvement, whereas catastrophising has a negative effect.[84] The epidemiological studies provided evidence of sex differences in

pain perception. An animal study suggested this could be due to the greater activation of periaqueductal gray–rostroventromedial medulla pathway in males than females.[85] However, human studies investigating sex differences in pain inhibition pathways have shown mixed results. It depends on both the experimental methodology and the modes of measurement of the effect.[86] Furthermore, chronic pain often coexists with sleep disorders, which worsen the pain. Pain perception is often affected by many forms of sleep disturbance, but it is unclear if these effects are the same for males and females.[87] Dysfunction of inhibition pathway, therefore, may lead to the development of chronic pain, thereby accompanied by cognitive deficits and aversive emotional states.

The hiding inhibition pathway of orofacial pain: the trigeminal proprioceptive mesencephalic periaqueductal gray pathway

The trigeminal nerve, also known as cranial nerve V or CN V, includes sensory and motor functions. Three branches of the nerve—the combined sensory and motor mandibular nerve (V3), the sensory maxillary nerve (V2) and the sensory ophthalmic nerve (V1)—supply the face. The sensory, also known as afferent neurones transmit of general somatic information from the face, such as pain, temperature, vibration, fine and crude touch and proprioception to the brainstem in contrast to the motor or efferent neurones, project information from the brainstem to the tensor veli palatini, tensor tympani, anterior belly of the digastric, mylohyoid and muscles of mastication, such as masseter, temporalis, lateral and medial pterygoids.[88] The trigeminal nerve is associated with three sensory nuclei (mesencephalic nucleus, the chief/principal sensory nucleus and spinal trigeminal nucleus) and one motor nucleus (trigeminal motor nucleus). The sensory fibres from V1, V2 and V3 travel via axons from pseudounipolar neurones to their cell bodies in the trigeminal ganglion. The afferent neurons then decussate at the brainstem to join the trigeminal lemniscus. The secondary neuron joins the tertiary neuron at the thalamus.

However, most of the proprioceptive afferents for the orofacial region in the trigeminal nerve are slightly different, as they have their cell bodies located in the mesencephalic trigeminal nucleus. The mesencephalic nucleus is involved with proprioception of the teeth, palate, TM, and muscles of mastication; that is, detecting of the position and controlling force and pressure of the muscles and joints.[89] The mesencephalic nucleus is situated on the anterolateral aspect of the periaqueductal gray and ascends to the height of the inferior colliculus[88] (Figure 4). Studies suggested that periaqueductal gray received input/nerve fibres from mesencephalic nucleus.[90, 91]

The occlusal appliance therapy as one of pain management modalities has been used in a number of orofacial pain conditions, such as TMD and tension-type headache.[92,93] However, the mechanism of action of occlusal appliance used for the successful treatment of orofacial pain remains unclear and controversial. There are a few concepts, which explain how occlusal appliance could help, including prevention in maximal intercuspal position,[94] even distribution of forces,[95] stabilisation of periodontal ligament proprioception,[96] relief of jaw muscle tension,[97] guidance for muscle relaxation,[98] reposition of the jaw[99] and impact on vertical dimension of occlusion.[100] Most of the mechanism appears to be associated with the activating/changing of the trigeminal mesencephalic response. As the mesencephalic nucleus is highly associated with the pain inhibition centre (periaqueductal gray), we propose that occlusal appliance improves orofacial pain by activating/facilitating the periaqueductal gray via the mesencephalic nucleus, known as **the trigeminal proprioceptive mesencephalic periaqueductal gray pathway** (TPMP). This is a significant discovery as it could direct the future management of orofacial pain. Thus, physical therapy, including occlusal appliance (activation of periaquedual gray) may be as effective as medication and surgery in managing orofacial pain, and it should be used as first line as it has fewer side effects.[101] The activation of TPMP can be confirmed by functional MRI. This may include the investigation of the activity patterns in the TPMP between normal subjects and orofacial pain patients, the association between the ascending pain pathway and TPMP and the possible link between TPMP with the higher pain process centre.

Chronic pain and genetics

Chronic overlapping pain conditions (COPCs) are a group of chronic pains that may include TMD, fibromyalgia, irritable bowel syndrome, vulvodynia, myalgic encephalomyelitis/chronic fatigue syndrome, interstitial cystitis/painful bladder syndrome, endometriosis, chronic

tension-type headache, migraine headache and chronic lower back pain.[102] There is now a considerably wider range of research that could be applied to orofacial pain thanks to the relationship between these prevalent chronic pain syndromes, as shown by the COPC. A genetically pain-susceptible person who may also have susceptibility to a high state of psychological distress may convert to a chronic pain sufferer through an exogenous trigger;[103] this has also been suggested with regard to chronic orofacial pain, such as chronic TMD.[104] This might lead to sensitisation "down-up" from periphery to central pathway.[105] Ongoing pain might then further lead to a "top-down" sensitisation, which would further wind up the pain level.[106] Understanding these mechanisms allows us to better understand the current strategies that have worked.

Research on the topic of genetic variants associated with chronic pain is still in its initial phase. The genetic basis of chronic pain is not yet fully understood. It is also known that genetic variation and changes could make a person more susceptible to becoming a chronic pain sufferer.[103] However, several genes have been identified as potential contributors to the development and modulation of chronic pain. For example, increased expression of SCN9A may affect the perception and intensity in acute pain, and the susceptibility to chronic pain.[107,108] The COMT gene encodes a protein that breaks down norepinephrine, epinephrine and dopamine. Low COMT activity may increase the risk of some of chronic conditions, such as fibromyalgia or chronic widespread pain.[109] OPRM1, a mu opioid receptor gene, together with COMT, has been linked to the initiation of chronic pain.[110] GCh1 is a pain-protective gene (responsible for the production of the neurotransmitters such as serotonin, dopamine and norepinephrine), and it could decrease the level of pain, perhaps by influence on the COMT enzyme activity.[111] TRPV1 (gene for transient receptor potential cation channel) participates in chronic pain through transcriptional and translational regulation, and also the development of nociceptive and depressive behaviours.[112] SLC6A4, ADRB2 and HTR2A may be associated with chronic widespread pain.[103] These genes only make up a small portion of the genetic components that may be involved in the aetiology of chronic pain, which is a complicated and multifaceted disorder.

Conclusion

It is important to understand the neurophysiology of chronic pain in order to diagnose and manage chronic orofacial pain. Modification (nociplasticity) of pain information may take place in several ascending or descending pathways. TPMP may be considered as the mechanism of action of occlusal appliance in managing orofacial pain related to TMD. Factors such as sleep, psychological disease (e.g., anxiety/depression), hormonal and other factors not yet identified might be under the influence of genetics. The interplay between genes is still an active area of research in understanding chronic pain.

COMPETING INTERESTS
The authors have no conflict of interest to declare.

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REFERENCES

1. Pain terms: a list with definitions and notes on usage. Recommended by the IASP Subcommittee on Taxonomy. Pain. 1979;6(3):249.
2. Raja SN, Carr DB, Cohen M, et al. The revised International Association for the Study of Pain definition of pain: concepts, challenges, and compromises. Pain. 2020;161(9):1976-1982. doi: 10.1097/j.pain.0000000000001939.
3. Cohen SP, Vase L, Hooten WM. Chronic pain: an update on burden, best practices, and new advances. Lancet. 2021;397(10289):2082-2097. doi: 10.1016/S0140-6736(21)00393-7.
4. International Association for the Study of Pain. Terminology [Internet]. 2022 [cited 2023 Nov 26]. Available from: https://www.iasp-pain.org/resources/terminology/?ItemNumber=1698.
5. Ghurye S, McMillan R. Orofacial pain - an update on diagnosis and management. Br Dent J. 2017;223(9):639-647. doi: 10.1038/sj.bdj.2017.879.
6. Treede RD, Rief W, Barke A, et al. Chronic pain as a symptom or a disease: the IASP Classification of Chronic Pain for the International Classification of Diseases (ICD-11). Pain. 2019;160(1):19-27. doi: 10.1097/j.pain.0000000000001384.
7. International Classification of Orofacial Pain, 1st edition (ICOP). Cephalalgia. 2020;40(2):129-221. doi: 10.1177/0333102419893823.
8. Derafshi R, Rezazadeh F, Ghapanchi J, et al. Prevalence of Chronic Orofacial Pain in Elderly Patients Referred to Shiraz Dental School From 2005 to 2017. Anesth Pain Med. 2019 Dec 7;9(6):e91182. doi: 10.5812/aapm.91182.
9. Horst OV, Cunha-Cruz J, Zhou L, et al. Prevalence of pain in the orofacial regions in patients visiting general dentists in the Northwest Practice-based REsearch Collaborative in Evidence-based DENtistry research network. J Am Dent Assoc. 2015;146(10):721-8.e723. doi: 10.1016/j.adaj.2015.04.001.
10. de Melo Júnior PC, Aroucha JMCNL, Arnaud M, et al. Prevalence of TMD and level of chronic pain in a group of Brazilian adolescents. PLoS One. 2019;14(2):e0205874. doi: 10.1371/journal.pone.0205874.
11. Ananthan S, Benoliel R. Chronic orofacial pain. J. Neural Transm (Vienna). 2020;127(4):575-588. doi: 10.1007/s00702-020-02157-3.
12. Leung WS, McMillan AS, Wong MC. Chronic orofacial pain in southern Chinese people: experience, associated disability, and help-seeking response. J Orofac Pain. 2008;22(4):323-330.
13. Häggman-Henrikson B, Liv P, Ilgunas A, et al. Increasing gender differences in the prevalence and chronification of orofacial pain in the population. Pain. 2020;161(8):1768-1775. doi: 10.1097/j.pain.0000000000001872.
14. Plesh O, Adams SH, Gansky SA. Racial/Ethnic and gender prevalences in reported common pains in a national sample. J Orofac Pain. 2011;25(1):25-31.
15. Riley JL 3rd, Gilbert GH, Heft MW. Orofacial pain: racial and sex differences among older adults. J Public Health Dent. 2002;62(3):132-139. doi: 10.1111/j.1752-7325.2002.tb03434.x.
16. Lipton JA, Ship JA, Larach-Robinson D. Estimated Prevalence and Distribution of Reported Orofacial Pain in the United States. J Am Dent Assoc. 1993;124(10):115-121. doi: 10.14219/jada.archive.1993.0200.
17. Yanko R, Badran Y, Leibovitz S, et al. Exploring the Effect of Ethnicity on Chronic Orofacial Pain: A Comparative Study of Jewish and Arab Israeli Patients. Healthcare (Basel). 2023;11(14):1984. doi: 10.3390/healthcare11141984.
18. Slade GD, Sanders AE, Bair E, et al. Preclinical episodes of orofacial pain symptoms and their association with health care behaviors in the OPPERA prospective cohort study. Pain. 2013;154(5):750-760. doi: 10.1016/j.pain.2013.01.014.
19. Riley JL 3rd, Gilbert GH, Heft MW. Race/ethnic differences in health care use for orofacial pain among older adults. Pain. 2002(1-2);100:119-130.

doi: 10.1016/s0304-3959(02)00256-7.
20. Mittinty MM, McNeil DW, Jamieson LM. Limited evidence to measure the impact of chronic pain on health outcomes of Indigenous people. J Psychosom Res. 2018;107:53-54. oi: 10.1016/j.jpsychores.2018.02.001.
21. Lewis GN, Upsdell A. Ethnic disparities in attendance at New Zealand's chronic pain services. N Z Med J. 2018;131(1472):21-28.
22. Lewis G, Borotkanics R, Upsdell A. Inequity in outcomes from New Zealand chronic pain services. N Z Med J. 2021;134(1533):11-20.
23. Wirz S, Ellerkmann RK, Buecheler M, et al. Management of chronic orofacial pain: a survey of general dentists in german university hospitals. Pain Med. 2010;11(3):416-424. doi: 10.1111/j.1526-4637.2010.00805.x.
24. Gauer RL, Semidey MJ. Diagnosis and treatment of temporomandibular disorders. Am Fam Physician. 2015;91(6):378-386.
25. Souza FT, Santos TP, Bernardes VF, et al. The impact of burning mouth syndrome on health-related quality of life. Health Qual Life Outcomes. 2011;9:57. doi: 10.1186/1477-7525-9-57.
26. Sanner F, Sonntag D, Hambrock N, Zehnder M. Patients with persistent idiopathic dentoalveolar pain in dental practice. Int Endod J. 2022;55(3):231-239. doi: 10.1111/iej.13664.
27. World Health Organization. ICD-11 for Mortality and Morbidity Statistics [Internet]. Geneva, Switzerland: World Health Organization; 2018 [cited 2023 Nov 26]. Available from: https://icd.who.int/browse11/l-m/en.
28. Nicholas M, Vlaeyen JWS, Rief W, et al. The IASP classification of chronic pain for ICD-11: chronic primary pain. Pain. 2019;160(1):28-37. doi: 10.1097/j.pain.0000000000001390.
29. Yam MF, Loh YC, Tan CS, et al. General Pathways of Pain Sensation and the Major Neurotransmitters Involved in Pain Regulation. Int J Mol Sci. 2018;19(8):2164. doi: 10.3390/ijms19082164.
30. Rotpenpian N, Yakkaphan P. Review of Literatures: Physiology of Orofacial Pain in Dentistry. eNeuro. 2021;8(2):ENEURO.0535-20.2021. doi: 10.1523/ENEURO.0535-20.2021.
31. Hoegh M. Pain Science in Practice: What Is Pain Neuroscience? Part 1. J Orthop Sports Phys Ther. 2022;52(4):163-165. doi: 10.2519/jospt.2022.10995.
32. Dray A. Inflammatory mediators of pain. Br J Anaesth. 1995;75(2):125-131. doi: 10.1093/bja/75.2.125.
33. Urch C. Normal Pain Transmission. Rev Pain. 2007;1(1):2-6. doi: 10.1177/204946370700100102.
34. Bushnell MC, Duncan GH, Hofbauer RK, et al. Pain perception: is there a role for primary somatosensory cortex? Proc Natl Acad Sci U S A. 1999;96(14):7705-7709. doi: 10.1073/pnas.96.14.7705.
35. Basbaum AI, Bautista DM, Scherrer G, Julius D. Cellular and molecular mechanisms of pain. Cell. 2009;139(2):267-284. doi: 10.1016/j.cell.2009.09.028.
36. Apkarian AV, Bushnell MC, Treede RD, Zubieta JK. Human brain mechanisms of pain perception and regulation in health and disease. Eur J Pain. 2005;9(4):463-484. doi: 10.1016/j.ejpain.2004.11.001.
37. Davidson RJ, Putnam KM, Larson CL. Dysfunction in the neural circuitry of emotion regulation--a possible prelude to violence. Science. 2000;289(5479):591-594. doi: 10.1126/science.289.5479.591.
38. Apkarian VA, Hashmi JA, Baliki MN. Pain and the brain: specificity and plasticity of the brain in clinical chronic pain. Pain. 2011;152(3):S49-S64. doi: 10.1016/j.pain.2010.11.010.
39. Zamorano AM, Montoya P, Cifre I, et al. Experience-dependent neuroplasticity in trained musicians modulates the effects of chronic pain on insula-based networks - A resting-state fMRI study. Neuroimage. 2019;202:116103. doi: 10.1016/j.neuroimage.2019.116103.
40. Baliki MN, Petre B, Torbey S, et al. Corticostriatal functional connectivity predicts transition to chronic back pain. Nat. Neurosci. 2012;15(8):1117-1119. doi: 10.1038/nn.3153.
41. Yang S, Chang MC. Chronic Pain: Structural and Functional Changes in Brain Structures and Associated Negative Affective States. Int J Mol Sci. 2019;20(13):3130. doi: 10.3390/ijms20133130.
42. Hashmi JA, Baliki MN, Huang L, et al. Shape shifting pain: chronification of back pain shifts brain representation from nociceptive to emotional circuits. Brain. 2013;136(Pt 9):2751-2768. doi: 10.1093/brain/awt211.
43. Tan LL, Pelzer P, Heinl C, et al. A pathway from midcingulate cortex to posterior insula gates nociceptive hypersensitivity. Nat Neurosci. 2017;20(11):1591-1601. doi: 10.1038/nn.4645.
44. Zhuo M. Neural Mechanisms Underlying Anxiety-Chronic Pain Interactions. Trends Neurosci. 2016;39(3):136-145. doi: 10.1016/j.tins.2016.01.006.
45. Moon HC, Heo WI, Kim YJ, et al. Optical inactivation of the anterior cingulate cortex modulate descending pain pathway in a rat model of trigeminal neuropathic pain created via chronic constriction injury of the infraorbital nerve. J Pain Res. 2017;10:2355-2364. doi: 10.2147/JPR.S138626.
46. Tsuda M, Koga K, Chen T, Zhuo M. Neuronal and

microglial mechanisms for neuropathic pain in the spinal dorsal horn and anterior cingulate cortex. J Neurochem. 2017;141(4):486-498. doi: 10.1111/jnc.14001.
47. Weissman-Fogel I, Moayedi M, Tenenbaum HC, et al. Abnormal cortical activity in patients with temporomandibular disorder evoked by cognitive and emotional tasks. Pain. 2011;152(2):384-396. doi: 10.1016/j.pain.2010.10.046.
48. Younger JW, Shen YF, Goddard G, Mackey SC. Chronic myofascial temporomandibular pain is associated with neural abnormalities in the trigeminal and limbic systems. Pain. 2010;149(2):222-228. doi: 10.1016/j.pain.2010.01.006.
49. Zhao YP, Ma XC, Jin Z, et al. Cerebral activation during unilateral clenching in patients with temporomandibular joint synovitis and biting pain: an functional magnetic resonance imaging study. Chin Med J (Engl). 2011;124(14):2136-2143.
50. Gerstner GE, Gracely RH, Deebajah A, et al. Posterior insular molecular changes in myofascial pain. J Dent Res. 2012;91(5):485-490. doi: 10.1177/0022034512443366.
51. Vierck CJ, Whitsel BL, Favorov OV, et al. Role of primary somatosensory cortex in the coding of pain. Pain. 2013;154(3):334-344. doi: 10.1016/j.pain.2012.10.021.
52. Timmermann L, Ploner M, Haucke K, et al. Differential coding of pain intensity in the human primary and secondary somatosensory cortex. J. Neurophysiol. 2001;86(3):1499-1503. doi: 10.1152/jn.2001.86.3.1499.
53. Lockwood PL, Iannetti GD, Haggard P. Transcranial magnetic stimulation over human secondary somatosensory cortex disrupts perception of pain intensity. Cortex. 2013;49(8):2201-2209. doi: 10.1016/j.cortex.2012.10.006.
54. Mohr C, Binkofski F, Erdmann C, et al. The anterior cingulate cortex contains distinct areas dissociating external from self-administered painful stimulation: a parametric fMRI study. Pain. 2005;114(3):347-357. doi: 10.1016/j.pain.2004.12.036.
55. Fuchs PN, Peng YB, Boyette-Davis JA, Uhelski ML. The anterior cingulate cortex and pain processing. Front Integr Neurosci. 2014;8:35. doi: 10.3389/fnint.2014.00035.
56. Labrakakis C. The Role of the Insular Cortex in Pain. Int J Mol Sci. 2023;24(6):5736. doi: 10.3390/ijms24065736.
57. Choi S, Kim K, Kwon M, et al. Modulation of Neuropathic Pain by Glial Regulation in the Insular Cortex of Rats. Front Mol Neurosci. 2022;15:815945. doi: 10.3389/fnmol.2022.815945.
58. Ong WY, Stohler CS, Herr DR. Role of the Prefrontal Cortex in Pain Processing. Mol Neurobiol. 2019;56(2):1137-1166. doi: 10.1007/s12035-018-1130-9.
59. Lorenz J, Minoshima S, Casey KL. Keeping pain out of mind: the role of the dorsolateral prefrontal cortex in pain modulation. Brain. 2003;126(Pt 5):1079-1091. doi: 10.1093/brain/awg102.
60. Shiers S, Price TJ. Molecular, circuit, and anatomical changes in the prefrontal cortex in chronic pain. Pain. 2020;161(8):1726-1729. doi: 10.1097/j.pain.0000000000001897.
61. Yen CT, Lu PL. Thalamus and pain. Acta Anaesthesiol Taiwan. 2013;51(2):73-80. doi: 10.1016/j.aat.2013.06.011.
62. Ab Aziz CB, Ahmad AH. The role of the thalamus in modulating pain. Malays J Med Sci. 2006;13(2):11-18.
63. Lin T, Gargya A, Singh H, et al. Mechanism of Peripheral Nerve Stimulation in Chronic Pain. Pain Med. 2020;21(Suppl 1):S6-S12. doi: 10.1093/pm/pnaa164.
64. Stachowski NJ, Dougherty KJ. Spinal Inhibitory Interneurons: Gatekeepers of Sensorimotor Pathways. Int J Mol Sci. 2021;22(5):2667. doi: 10.3390/ijms22052667.
65. Li C, Liu S, Lu X, Tao F. Role of Descending Dopaminergic Pathways in Pain Modulation. Curr Neuropharmacol. 2019;17(12):1176-1182. doi: 10.2174/1570159X17666190430102531.
66. Neugebauer V, Mazzitelli M, Cragg B, et al. Amygdala, neuropeptides, and chronic pain-related affective behaviors. Neuropharmacology. 2020;170:108052. doi: 10.1016/j.neuropharm.2020.108052.
67. Basbaum AI, Fields HL. Endogenous pain control systems: brainstem spinal pathways and endorphin circuitry. Annu Rev Neurosci. 1984;7:309-338. doi: 10.1146/annurev.ne.07.030184.001521.
68. Villanueva L, Bouhassira D, Le Bars D. The medullary subnucleus reticularis dorsalis (SRD) as a key link in both the transmission and modulation of pain signals. Pain. 1996;67(2-3):231-240. doi: 10.1016/0304-3959(96)03121-1.
69. Mills EP, Keay KA, Henderson LA. Brainstem Pain-Modulation Circuitry and Its Plasticity in Neuropathic Pain: Insights From Human Brain Imaging Investigations. Front Pain Res (Lausanne). 2021;2:705345. doi: 10.3389/fpain.2021.705345.
70. You HJ, Lei J, Sui MY, et al. Endogenous descending modulation: spatiotemporal effect of dynamic imbalance between descending facilitation and inhibition of nociception. J Physiol. 2010;588(Pt 21):4177-4188. doi: 10.1113/jphysiol.2010.196923.

71. Ossipov MH, Morimura K, Porreca F. Descending pain modulation and chronification of pain. Curr Opin Support Palliat Care. 2014;8(2):143-151. doi: 10.1097/SPC.0000000000000055.
72. Mills EP, Di Pietro F, Alshelh Z, et al. Brainstem Pain-Control Circuitry Connectivity in Chronic Neuropathic Pain. J Neurosci. 2018;38(2):465-473. doi: 10.1523/JNEUROSCI.1647-17.2017.
73. Hemington KS, Coulombe MA. The periaqueductal gray and descending pain modulation: why should we study them and what role do they play in chronic pain? J Neurophysiol. 2015;114(4):2080-2083. doi: 10.1152/jn.00998.2014.
74. Napadow V, LaCount L, Park K, et al. Intrinsic brain connectivity in fibromyalgia is associated with chronic pain intensity. Arthritis Rheum. 2010;62(8):2545-2555. doi: 10.1002/art.27497.
75. Seminowicz DA, Davis KD. Pain enhances functional connectivity of a brain network evoked by performance of a cognitive task. J Neurophysiol. 2007;97(5):3651-3659. doi: 10.1152/jn.01210.2006.
76. Tanasescu R, Cottam WJ, Condon L, et al. Functional reorganisation in chronic pain and neural correlates of pain sensitisation: A coordinate based meta-analysis of 266 cutaneous pain fMRI studies. Neurosci Biobehav Rev. 2016;68:120-133. doi: 10.1016/j.neubiorev.2016.04.001.
77. Li T, Zhang S, Ikeda E, Kobinata H. Functional connectivity modulations during offset analgesia in chronic pain patients: an fMRI study. Brain Imaging Behav. 2022;16(4):1794-1802. doi: 10.1007/s11682-022-00652-7.
78. Meeus M, Hermans L, Ickmans K, et al. Endogenous pain modulation in response to exercise in patients with rheumatoid arthritis, patients with chronic fatigue syndrome and comorbid fibromyalgia, and healthy controls: a double-blind randomized controlled trial. Pain Pract. 2015;15(2):98-106. doi: 10.1111/papr.12181.
79. Clauw DJ, Arnold LM, McCarberg BH. The science of fibromyalgia. Mayo Clin Proc. 2011;86(9):907-911. doi: 10.4065/mcp.2011.0206.
80. Piché M, Bouin M, Arsenault M, et al. Decreased pain inhibition in irritable bowel syndrome depends on altered descending modulation and higher-order brain processes. Neuroscience. 2011;195:166-175. doi: 10.1016/j.neuroscience.2011.08.040.
81. Monaco A, Cattaneo R, Mesin L, et al. Dysregulation of the descending pain system in temporomandibular disorders revealed by low-frequency sensory transcutaneous electrical nerve stimulation: a pupillometric study. PLoS One. 2015;10(4):e0122826. doi: 10.1371/journal.pone.0122826.
82. Yu R, Gollub RL, Spaeth R, et al. Disrupted functional connectivity of the periaqueductal gray in chronic low back pain. Neuroimage Clin. 2014;6:100-108. doi: 10.1016/j.nicl.2014.08.019.
83. Damien J, Colloca L, Bellei-Rodriguez CÉ, Marchand S. Pain Modulation: From Conditioned Pain Modulation to Placebo and Nocebo Effects in Experimental and Clinical Pain. Int Rev Neurobiol. 2018;139:255-296. doi: 10.1016/bs.irn.2018.07.024.
84. Zeidan F, Vago DR. Mindfulness meditation-based pain relief: a mechanistic account. Ann N Y Acad Sci. 2016;1373(1):114-127. doi: 10.1111/nyas.13153.
85. Loyd DR, Morgan MM, Murphy AZ. Morphine preferentially activates the periaqueductal gray-rostral ventromedial medullary pathway in the male rat: a potential mechanism for sex differences in antinociception. Neuroscience. 2007;147(2):456-468. doi: 10.1016/j.neuroscience.2007.03.053.
86. Popescu A, LeResche L, Truelove EL, Drangsholt MT. Gender differences in pain modulation by diffuse noxious inhibitory controls: a systematic review. Pain. 2010;150(2):309-318. doi: 10.1016/j.pain.2010.05.013.
87. Rouhi S, Topcu J, Egorova-Brumley N, Jordan AS. The impact of sleep disturbance on pain perception: A systematic review examining the moderating effect of sex and age. Sleep Med Rev. 2023;71:101835. doi: 10.1016/j.smrv.2023.101835.
88. Atkinson ME. Anatomy for Dental Students. 4th edition. Oxford: Oxford University Press; 2013.
89. Lazarov NE. Neurobiology of orofacial proprioception. Brain Res Rev. 2007;56(2):362-383. doi: 10.1016/j.brainresrev.2007.08.009.
90. Takahashi T, Shirasu M, Shirasu M, et al. The locus coeruleus projects to the mesencephalic trigeminal nucleus in rats. Neurosci Res. 2010;68(2):103-106. doi: 10.1016/j.neures.2010.06.012.
91. Erefah AZT, Dare NW, Oyinbo CA. A study of the Structure of Mesencephalic Trigeminal Nucleus (Mes V) in the Rabbit. J Neurol Sci. 2014;31(2):310-315.
92. Kostrzewa-Janicka J, Mierzwinska-Nastalska E, Rolski D, Szczyrek P. Occlusal stabilization splint therapy in orofacial pain and tension-type headache. Adv Exp Med Biol. 2013;788:181-188. doi: 10.1007/978-94-007-6627-3_27.
93. Orzeszek S, Waliszewska-Prosol M, Ettlin D, et al. Efficiency of occlusal splint therapy on orofacial muscle pain reduction: a systematic review. BMC Oral Health. 2023;23(1):180. doi: 10.1186/s12903-023-02897-0.
94. Ré JP, Perez C, Darmouni L, et al. The occlusal splint therapy. J Stomat Occ Med. 2009;2:82-86. https://doi.org/10.1007/s12548-009-0015-y.
95. Suganuma T, Itoh H, Ono Y, Baba K. Effect of

stabilization splint on occlusal force distribution during voluntary submaximal tooth clenching: a preliminary sleep simulation study. Cranio. 2013;31(2):100-108. doi: 10.1179/crn.2013.017.
96. Srivastava R, Jyoti B, Devi P. Oral splint for temporomandibular joint disorders with revolutionary fluid system. Dent Res J (Isfahan). 2013;10(3):307-313.
97. Albagieh H, Alomran I, Binakresh A, et al. Occlusal splints-types and effectiveness in temporomandibular disorder management. Saudi Dent J. 2023;35(1):70-79. doi: 10.1016/j.sdentj.2022.12.013.
98. Cesanelli L, Cesaretti G, Ylaitė B, et al. Occlusal Splints and Exercise Performance: A Systematic Review of Current Evidence. Int J Environ Res Public Health. 2021 Sep 30;18(19):10338. doi: 10.3390/ijerph181910338.
99. Williamson EH. Temporomandibular dysfunction and repositioning splint therapy. Prog Orthod. 2005;6(2):206-213.
100. Olthoff LW, van der Glas HW, van der Bilt A. Influence of occlusal vertical dimension on the masticatory performance during chewing with maxillary splints. J Oral Rehabil. 2007;34(8):560-565. doi: 10.1111/j.1365-2842.2007.01730.x.
101. Zhang L, Xu L, Wu D, et al. Effectiveness of exercise therapy versus occlusal splint therapy for the treatment of painful temporomandibular disorders: a systematic review and meta-analysis. Ann Palliat Med. 2021;10(6):6122-6132. doi: 10.21037/apm-21-451.
102. Maixner W, Fillingim RB, Williams DA, et al. Overlapping Chronic Pain Conditions: Implications for Diagnosis and Classification. J Pain. 2016;17(9 Suppl):T93-T107. doi: 10.1016/j.jpain.2016.06.002.
103. James S. Human pain and genetics: some basics. Br J Pain. 2013;7(4):171-178. doi: 10.1177/2049463713506408.
104. Polonowita AD, Thomson WM, Thorburn DN. Clinical efficacy of a simplified approach to managing chronic temporomandibular disorders: evidence from a 1-year case series. Oral Surg Oral Med Oral Pathol Oral Radiol. 2019;128(3):227-234. doi: 10.1016/j.oooo.2019.06.008.
105. Vierck CJ, Wong F, King CD, et al. Characteristics of sensitization associated with chronic pain conditions. Clin J Pain. 2014;30(2):119-128. doi: 10.1097/AJP.0b013e318287aac7.
106. Eller-Smith OC, Nicol AL, Christianson JA. Potential Mechanisms Underlying Centralized Pain and Emerging Therapeutic Interventions. Front Cell Neurosci. 2018;12:35. doi: 10.3389/fncel.2018.00035.
107. Yeo J, Sia AT, Sultana R, et al. Analysis of SCN9A Gene Variants for Acute and Chronic Postoperative Pain and Morphine Consumption After Total Hysterectomy. Pain Med. 2020;21(11):2642-2649. doi: 10.1093/pm/pnaa109.
108. Kurzawski M, Rut M, Dziedziejko V, et al. Common Missense Variant of SCN9A Gene Is Associated with Pain Intensity in Patients with Chronic Pain from Disc Herniation. Pain Med. 2018;19(5):1010-1014. doi: 10.1093/pm/pnx261.
109. Tammimäki A, Männistö PT. Catechol-O-methyltransferase gene polymorphism and chronic human pain: a systematic review and meta-analysis. Pharmacogenet Genomics. 2012;22(9):673-691. doi: 10.1097/FPC.0b013e3283560c46.
110. Firfirey F, Shamley D, September AV. Polymorphisms in COMT and OPRM1 Collectively Contribute to Chronic Shoulder Pain and Disability in South African Breast Cancer Survivors'. Genes (Basel). 2022;14(1):9. doi: 10.3390/genes14010009.
111. Smith SB, Reenilä I, Männistö PT, et al. Epistasis between polymorphisms in COMT, ESR1, and GCH1 influences COMT enzyme activity and pain. Pain. 2014;155(11):2390-2399. doi: 10.1016/j.pain.2014.09.009.
112. Fischer SPM, Brusco I, Brum ES, et al. Involvement of TRPV1 and the efficacy of α-spinasterol on experimental fibromyalgia symptoms in mice. Neurochem Int. 2020;134:104673. doi: 10.1016/j.neuint.2020.104673.

Patient experience surveys are vital in the twenty-first century: let's put some myths to rest

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ABSTRACT

Patient-reported data derived from surveys places patient feedback at the heart of quality improvement and health system responsiveness. Such surveys are not without critics, however, who contend that there are better ways to collect feedback. Criticisms assert that response rates are too low and measures are not robust, valid or reliable, that patient experience surveys are neither valid nor reliable for Māori and Pacific peoples and that such surveys do not contribute to improved outcomes for patients.

We debunk these myths in the context of the Te Tāhū Hauora Health Quality & Safety Commission (Te Tāhū Hauora) patient experience survey programme. We explain the centrality of a strong consumer and whānau voice in a twenty-first century health system, and that listening to and acting on this voice—including use of patient-reported data—is now a statutory requirement for health entities under the *Pae Ora (Healthy Futures) Act 2022*. We describe the different surveys in the programme and explain the differences between patient satisfaction and patient experience. We address sample size and response rates, including representativeness in the surveys of Māori and Pacific peoples' experience. We look at how survey data can be used for quality improvement and to guide us toward providing equitable, culturally safe care.

We assert that, contrary to criticisms, the programme delivers valid, reliable, relevant, systematic and practical patient experience surveys and resulting data, with guides for improvement, and that we are both legally and ethically bound to listen to and use these results to improve the healthcare we deliver.

In 2016, frustrated researchers at the RAND Corporation were moved to publish a debunking of "the most prevalent myths" riddling the critiques of patient experience survey data used to measure and improve healthcare quality.[1,2]

In the Aotearoa New Zealand context, criticisms of patient-reported experience data are widespread, and equally frustrating. Few of these criticisms are, however, published in the peer-reviewed literature, possibly for the reasons we outline below in debunking these myths—they don't hold sufficient water. However, this received wisdom has come to our attention, and tends to take the form of the following chestnuts, many common to other contexts, here deployed without evidence: that Aotearoa New Zealand's patient experience survey response rates are too low to tell us anything of use, and that consequently measures are not robust, valid or reliable. That surveys undersample and are not valid or reliable for Māori and Pacific peoples.[3] That surveys provide data, sometimes insights and opportunities, but no solutions.[4] That none of this relates to improving outcomes.

Let's put some of these myths to rest, but first: why involve patient experience and the patient voice in design and delivery of care in the first place?

A strong consumer and whānau voice are critical in a twenty-first century health system—and now a statutory requirement

The consumer health movement is not new, with its roots in the de-institutionalisation of mental health,[5,6] rise in disability rights movement[7,8] and special interest consumer groups.[9] Similarly, the calls to better understand if and how these movements, and the involvement of consumers more generally, have led to a safer and higher quality health system remains a perennial subject.[10,11,12] In a 2018 narrative review in *BMJ Quality & Safety*, 11 leaders in patient safety from the US and the UK wrote, "*overwhelming evidence indicates that collecting patient feedback and including patients as equal partners in their care supports improvement in both patient experience of care and clinical outcomes*."[13] A systematic review of 55 studies published in the *BMJ* in 2013 found "*consistent positive associations between patient*

experience, patient safety and clinical effectiveness for a wide range of disease areas, settings, outcome measures and study designs … patient experience is positively associated with clinical effectiveness and patient safety, and support the case for the inclusion of patient experience as one of the central pillars of quality in healthcare."[14] Another 2014 review found, "*better patient-reported care experiences are often associated with other aspects of health-care quality, specifically:*

- *higher levels of adherence to recommended prevention and treatment processes*
- *better clinical outcomes*
- *less unnecessary healthcare utilisation*."[15]

Yet despite evidence for the benefits of patient and public involvement in designing, planning and co-producing healthcare services, and a broadly supportive policy context in the UK and internationally, a 2016 *BMJ Quality & Safety* paper found that progress in improving consumer engagement in production of healthcare services has been "*patchy and slow and often concentrated at the lowest levels of involvement*."[16]

Te Tāhū Hauora has championed consumer and whānau engagement since its inception in 2010, and this work has now culminated in the development and embedding as secondary legislation of the Code of Expectations for Engagement with Consumers and Whānau for health entities[17] (the Code) in section 59 of the *Pae Ora (Healthy Futures) Act 2022*.[18]

The Code sets the expectations for how health entities must work with consumers, whānau and communities in the planning, design, delivery and evaluation of health services. Between 2021 and 2022, Te Tāhū Hauora alongside the Health Transition Unit developed the Code, and under the *Act* there is now a statutory requirement for health entities to give effect to it, and to report annually on how it has been applied.

Part 2.2 of the Code states that health entities must use "*lived experience, including consumer experience data to inform improvements in health services with a focus on reducing health inequities, particularly for Māori, Pacific peoples and disabled people*."[19]

For health service providers, use of patient experience survey programme data is a critical element in fulfilling this statutory expectation. Te Tāhū Hauora provides an implementation guide[20] for health entities to learn how they can implement the Code, including use of survey programme data.

Satisfaction versus experience—there is a critical difference

The first critical distinction to make in understanding patient-reported survey data is the distinction between patient *satisfaction* and patient *experience*.

Both satisfaction and experience surveys include multi-choice items.

Patient satisfaction surveys ask questions related to "How did we do?" with responses from "very poor" to "very good". However, patient experience surveys ask questions related to *what happened*, with responses such as "always" to "never". From this, we can derive actionable knowledge of *what did or did not actually happen* in a healthcare experience for large groups of people, rather than a non-actionable measure of how people felt, in a given place at a given time, for in many ways unknowable reasons.

Consider your own personal mood when confronted with the HappyOrNot™ terminal at an airport baggage carousel. You have a choice ranging between a green smiley face and a red angry face. But how much is your satisfaction with your baggage handling mediated by your prior expectation of a terrible experience, for example, or a particularly awful flight, or some fantastic news you received when you turned airport mode off on your phone at disembarkation?

Patient satisfaction responses are considered subjective and biased because satisfaction is personal and related to expectations and other unknowable influences. It is not an objective reflection of the presence or quality of a service or aspect of that service.[21] Patient satisfaction responses also tend to be overwhelmingly positive and are easy to manipulate with framing of questions.[19]

Thus, rather than a question like "How did we do?", patient experience surveys ask questions such as "In the last 12 months, was there ever a time when you wanted healthcare from a GP or nurse, but you could not get it?" A large number of responses thus gives a sense of barriers of access to care, and who experiences these. Another example, from the hospital inpatient survey, is, "Were you told what the medicine (or prescription for medicine) you left the hospital with was for?" This question reflects quality of discharge and communication, and using the experience explorer (see below), and other questions around discharge, services can easily see if they are providing the same quality of discharge information

to all people, and how well they are doing in comparison with other parts of the country.

Patient experience is a more objective construct and survey questions that address specific aspects of care can prompt specific actions to generate improvement. Patient experience is also an intrinsic element of most generally accepted frameworks of healthcare quality, including the Triple Aim, a framework developed by the Institute for Healthcare Improvement that has been adopted as a set of principles for health system reform in multiple countries and organisations worldwide,[22,23] the Quadruple Aim (building on the Triple Aim with the inclusion of workforce experience of providing care)[21] and the National Academy of Medicine's early and seminal six domains of quality developed in the US.[24] For these reasons we believe patient experience surveys to be superior to satisfaction surveys.

But what of the accusations that Aotearoa New Zealand's patient experience survey programme's response rates are too low and sample sizes are too small to make accurate judgments, and that these samples are skewed in composition due to the nature of those the survey reaches, and who chooses to respond? What of the accusation that the length of the survey causes people to give up on it?

The accusations don't hold water.

Response rates, who responds and how much is enough? Scope and scale of the Aotearoa New Zealand patient experience survey programme

Te Tāhū Hauora's patient-reported survey programme[25] is in fact the second largest government survey programme in Aotearoa New Zealand we are aware of (larger than the New Zealand Household Disability Survey and the New Zealand Health Survey, and second only to the New Zealand Census). The programme surveys the experiences of adult inpatients in hospital settings and adult primary care patients (an adult hospital outpatient survey has been more recently established). Combined, the former two surveys collected data from over 154,000 respondents between May 2022 and February 2023.

The adult hospital inpatient experience survey (established 2014) and the adult primary care patient experience survey (established 2016) were also reviewed and substantially refreshed in 2019 and 2020.[26] Cognitive testing was performed to understand and improve how patients understand and interpret questions and instructions in the survey and involved multiple in-depth interviews, with a particular focus on Māori and Pacific peoples' responses.

The survey is a major repository of data. During the early period of the pandemic many national programmes, including survey rounds, were paused as workforce focussed on the pandemic response. A one-off COVID-19 specific survey to assess impacts of the pandemic response on patients' experience of access to primary care during Level 3 restrictions was conducted,[27] and surveying began again with the refreshed instruments. The data repository for the hospital inpatient survey now contains about 35,000 responses from August 2020 (after the refresh during pandemic restrictions). The primary care patient experience survey now covers over 90% of general practices, with data from 315,000 responses since the 2020 refresh. On average, 3,000 hospital inpatients respond to the adult hospital inpatient survey every quarter, and 35,000 primary care patients respond to the adult primary care patient experience survey every quarter. Every quarter this data pool grows, keeping pace with a changing system and context.

So much for response size. But what about response rate? Booker and colleagues cite comprehensive reviews of comparable surveys in primary care internationally and found response rates vary between 10–61%.[28–31]

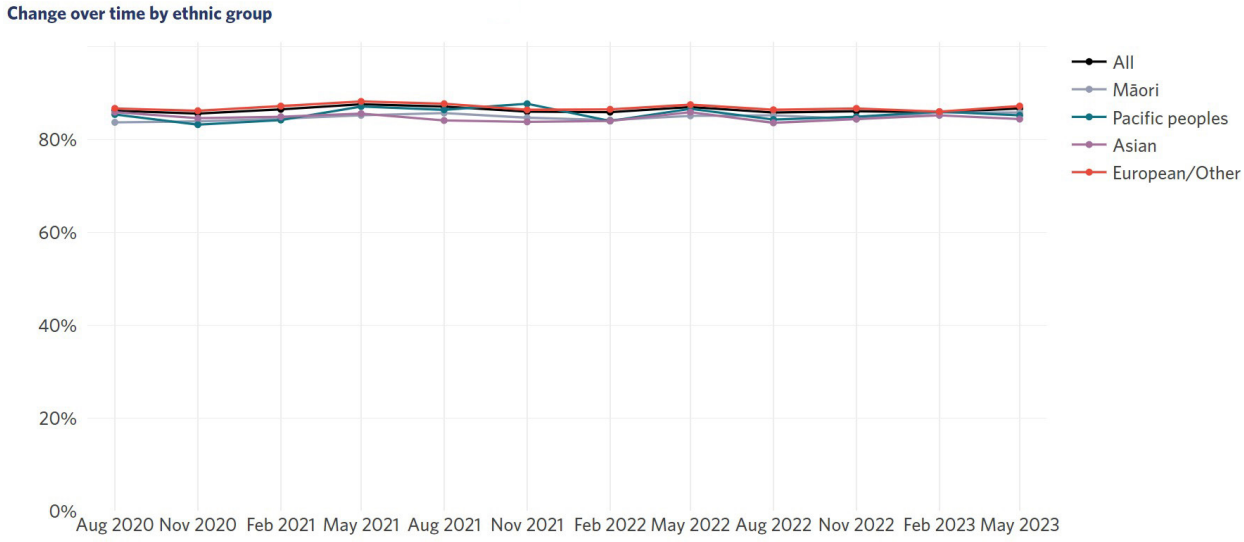
The hospital inpatient survey's response rate compares well, in the middle of the pack at 25%.

The primary care patient survey response rate, at 16–17%, is not so big, so we compensate for this by ensuring the large sample and comprehensive reach to practices (over 90%) mentioned above.

Response rates are important for three reasons, which we deal with in turn as follows:

1. Too few responders each time the survey is run can give unstable results stemming from natural variation over time alone. However, we know the survey is reliable from the consistent results we receive for the primary care survey quarter-on-quarter. As Figure 1 shows, results are consistent over time and natural variation is not creating unstable findings.
2. An unrepresentative group may be responding. We deal with this question in more detail below, but the results of focussed work in improving Māori response rates mean Māori response rates to the primary care survey now average 17%

Figure 1: The percentage of patients who reported definitely being involved in decisions about treatment and care as much as they wanted, adult primary care patient experience survey, by ethnicity, August 2020–May 2023.



annually, while non-Māori/non-Pacific response rates now average 16% annually.

3. The group who responds may be attitudinally unrepresentative compared to those who choose not to respond. In order to understand if this was the case, we conducted a peer-reviewed study published in the *New Zealand Medical Journal* in 2018.[32] We discuss the findings in more detail as follows.

Criticisms of so-called "opt-out" surveys draw attention to non-response bias, where those who choose to respond to a given survey are systematically different in some key way to those who choose not to. To understand if non-response bias existed in this large collection of survey data, a study was performed comparing the responses from a sample of initial hospital inpatient survey respondents (n=910) with the responses of a sample of non-responders who were followed up and invited again to take the survey (n=163). The study found no significant differences between the two groups, and that "responders to follow-up have similar experiences of inpatient care in New Zealand to initial responders."[31] There are of course limitations to this study, including potential bias in the follow-up responder group limiting the ability to ascertain the true extent of non-response bias.

So, there is evidence that non-response bias does not exist, and it is furthermore true that response rates *per se* are less important than representativeness—a large response rate from a group who are very different from non-responders will produced more biased results.[33] Hence the importance to the Aotearoa New Zealand patient experience survey data of Amhed et al.'s finding that "*response rates are only weakly associated with non-response bias in surveys that adhere to high standards of survey methodology*".[33–35] Our patient experience survey adheres to best-practice survey methodology promulgated by Statistics NZ and international researchers[36,37] to minimise attitudinal response bias. Measures are tested and validated, and weighting is used to adjust for patient characteristics that are likely to affect individual perceptions and expectations to enable comparison across healthcare providers.[38] Representativeness we discuss below.

The last point of contention (again unevidenced), that the length of the survey causes many respondents to give up, is also regularly monitored. The completion rate of the surveys is over 90%, suggesting the majority of those who start the survey finish it, but the programme is constantly looking to improve. Completion time, drop-outs, item non-response and patient comments about the survey are regularly monitored. This helps identify questions that people find hard to answer (issues of cognitive load and response burden), which is a more important issue than the number of questions *per se*.[39] As part of the annual review process, we also analyse responses so we can remove questions that are tightly linked to others. Another way we are reducing length is to

move questions into annual modules to be asked once a year rather than every quarter.

The primary focus of the review and a constant in the evolution of the programme has been on improving participation for Māori and Pacific peoples, including increasing their response rates. We look next at whether this has succeeded.

Representativeness in the patient experience survey of Māori and Pacific peoples' experience

Some have claimed that patient experience surveys are not valid or reliable for Māori and Pacific peoples due to low response rates. The significant work invested into improvements in this area has meant that, in the four quarterly surveys administered between early 2022 and early 2023, the historical equity gap in primary care survey response rates between Māori and non-Māori/non-Pacific peoples has disappeared.

Māori response rates now average 17% annually, while non-Māori/non-Pacific response rates now average 16% annually. Implementation of the successful improvements seen in the primary care survey are now in process for the hospital inpatient survey.

There were two key aspects of increasing participation rates: increasing the number of patients who are invited to take part, and increasing the proportion of patients who take part once invited.

The primary care survey now uses representative sampling to ensure a sufficient count of Māori and Pacific people are invited to the survey. In practice, this means a two-week sample of people identifying as being of Māori or Pacific ethnicity, and one week of all other ethnicities.

A range of other methods to increase both the numbers invited and the proportion of those who subsequently take part have addressed the entire pathway of survey response, including email address collection rates, culturally appropriate invitations, testing of the survey instruments with focus groups of Māori and Pacific peoples, and using a zero-data rated website, which allows access without using respondents' mobile data allowances.[40]

Patient experience for quality improvement

Can patient experience in fact be acted on to improve the quality of services? It is clear to most that data help identify issues and areas that can benefit from further exploration and development of insights that prompt quality improvement action, but where do providers start in using these data for quality improvement? Te Tāhū Hauora provides resources specifically designed to assist providers in using survey results for quality improvement in both adult hospital inpatient and primary care settings[41] to assist in the development of strategic, actionable insights and guide improvement.

At the national level

Results from the surveys are also currently used across the country in multiple ways. At the national level these data are used to publicly report on the quality and safety of services, for example, through the annual *Window on the quality of health care* reports published by Te Tāhū Hauora. The latest *Window* report published in June 2023[42] provided an update on effects of the pandemic on our health system and included a focus on experience of care for disabled people during pandemic-affected periods.

Survey feedback also informs policy and monitoring of how health services are performing. The interim *Health and Disability System Review* drew on patient experience survey data to highlight areas for improvement in the health system restructure process, for example in reported barriers to accessing primary care.[43] Data from the surveys where respondents report prescription cost as a barrier was used in the debate on Budget 2023's change to remove prescription costs. These insights to inform high-level policy can only be obtained through nationally consistent, robust and valid surveys.

At the local level

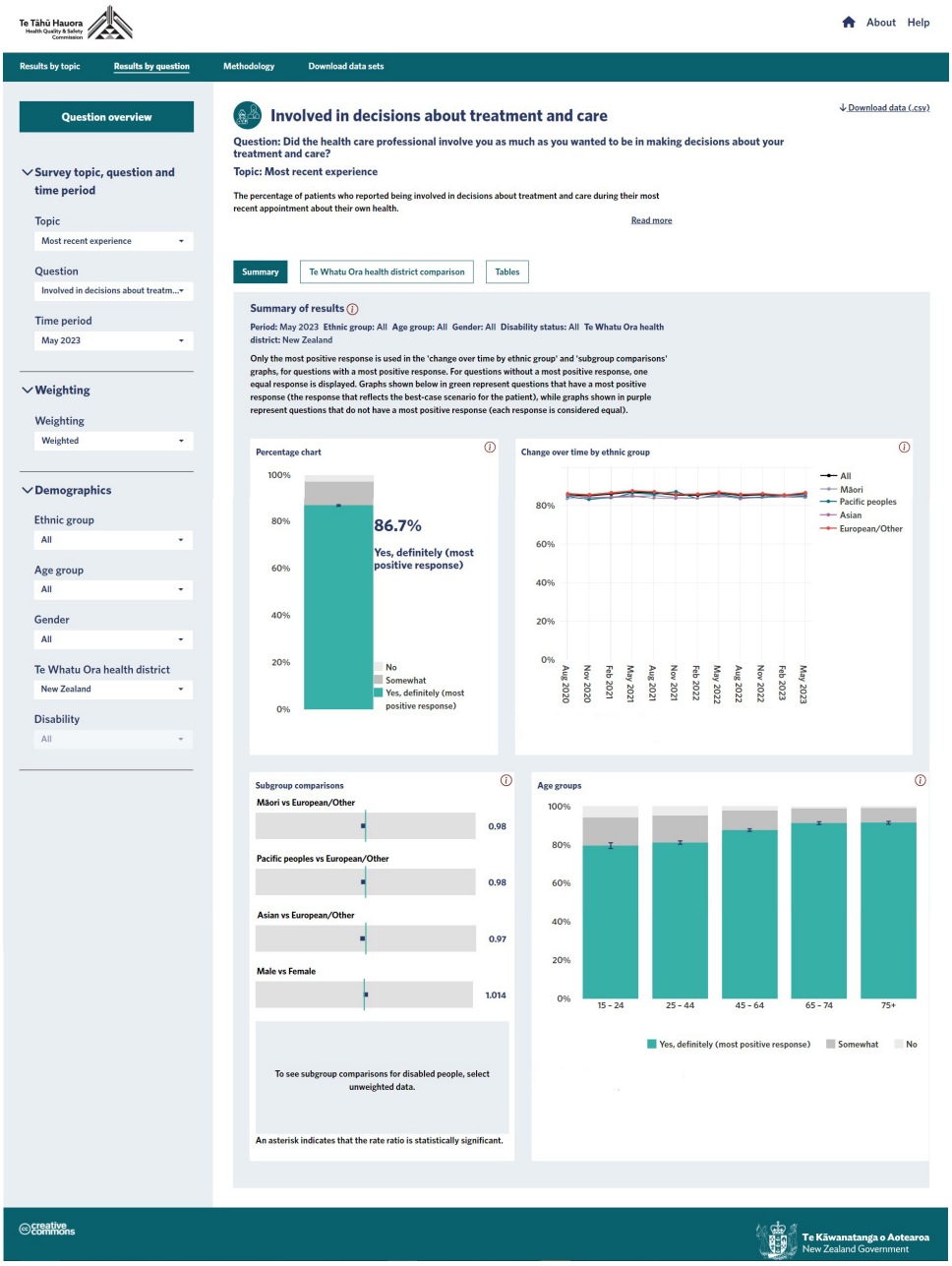
At local and regional levels, providers use survey data to monitor what is working well and what could be improved. For instance, during COVID-19 lockdowns, some hospitals were reporting their survey data to staff to show that, despite all the challenges associated with providing care in personal protective equipment and lockdown, patients were continuing to report positive experiences and were grateful for the care provided. Examples of specific quality improvement work drawing on survey findings are those which have been pursued at four district health boards (DHBs)—Northland, Waikato, Bay of Plenty, and Nelson Marlborough, addressing patient awareness of medication side effects and condition

management post-discharge.[44,45] Survey data for quality improvement work has also been embedded in the annual System Level Measures Improvement Plans of alliances in every district from 2016 to 2022. Examples are available from Auckland, Waitematā and Counties Manukau alliances for the 2020–2021 and 2021–2022 financial years.[46,47]

Health providers participating in the survey can access their own local results through a secure online portal. These dashboards enable people to access, cut and use the data in different ways, by local regions, demographic variables, question and domain, according to their needs and interests. Providers and researchers interested in investigating the data are able to and encouraged to get in touch (contact, in the first instance, the corresponding author Catherine Gerard).

Both the adult primary care patient and adult hospital inpatient surveys now also have publicly available data explorers[48,49] to make access to and investigation of experience data public, easy

Figure 2: Adult primary care patient experience explorer screenshot.



and intuitive. The explorers also allow data to be filtered by survey topics and questions, locations, periods and demographic elements of responders such as age, gender, ethnicity and disability status.

See Figure 2 for an example screenshot of the adult primary care patient experience data explorer, showing most-positive responses to the question: "Did the healthcare professional involve you as much as you wanted to be in making decisions about your treatment and care?"

Conclusion

Te Tāhū Hauora has administered national patient experience surveys since 2014. Since its inception, the programme has continuously grown and evolved, delivering valid, reliable, relevant, systematic and practical patient experience surveys and resulting data reflecting patient experiences of specific aspects of care. The patient-reported data and trends are translated into relevant metrics and indicators, easily accessible, to give providers a guide to how well their services are being delivered to their populations.

Te Tāhū Hauora patient experience survey data supplies coordinates and a compass to guide more patient-centred care. These surveys place patient feedback at the heart of quality improvement and health system responsiveness. The myths debunked above, we suspect, derive simply from suspicion premised on unfamiliarity.

To this we would add: thanks to the survey, we have heard from a representative sample of New Zealanders about the experience of healthcare they have had. We have this data, and we have a statutory imperative. We are now legally and ethically[50,51] bound to listen and to use the results for improvement.

COMPETING INTERESTS
Nil.

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REFERENCES
1. Anhang Price R, Elliott MN. Debunking Myths About Patient Experience Surveys [Internet]. Santa Monica, CA (US): RAND Corporation; 2016 [cited 2023 Nov 14]. Available from: https://www.rand.org/pubs/commentary/2016/05/debunking-myths-about-patient-experience-surveys.html.
2. Anhang Price R, Elliott MN, Cleary PD, et al. Should health care providers be accountable for patients' care experiences? J Gen Intern Med. 2015;30(2):253-6. doi: 10.1007/s11606-014-3111-7.
3. Fink JW, Paine SJ, Gander PH, et al. Changing response rates from Māori and non-Māori in national sleep health surveys. N Z Med J. 2011;124(1328):52-63.
4. Reeves R, Seccombe I. Do patient surveys work? The influence of a national survey programme on local quality-improvement initiatives. Qual Saf Health Care. 2008;17(6):437-41. doi: 10.1136/qshc.2007.022749.
5. Gooding P. From deinstitutionalisation to consumer empowerment: Mental health policy, neoliberal restructuring and the closure of the Big bins in Victoria. Health Sociol Rev. 2016;25(1):18-32. doi: 10.1080/14461242.2015.1134261.
6. Crowe M, O'Malley J, Gordon S. Meeting the needs of consumers in the community: a working partnership in mental health in New Zealand. J Adv Nurs. 2001;35(1):88-96. doi: 10.1046/j.1365-2648.2001.01825.x.
7. Winter JA. The development of the disability rights movement as a social problem solver. DSQ. 2003;23(1):33-61. doi: 10.18061/dsq.v23i1.399.
8. Scotch RK. Politics and policy in the history of the disability rights movement. Milbank Q. 1989;67 Suppl 2 Pt 2:380-400.
9. Coney S. Effective consumer voice and participation for New Zealand: a systematic review of the evidence. Wellington (NZ): New Zealand Guidelines Group; 2004 [cited 2023 Aug 22]. Available from: https://www.moh.govt.nz/.
10. Roseman D, Osborne-Stafsnes J, Amy CH, et al. Early lessons from four 'aligning forces for quality' communities bolster the case for patient-centered care. Health Aff (Millwood) 2013;32(2):232-41. doi: 10.1377/hlthaff.2012.1085.
11. Hamilton Lopez M, Holve E, Rein A, Winkler J. Involving patients and consumers in research: new opportunities for meaningful engagement in research and quality improvement [Internet]. Washington DC (US): EDM Forum Community, AcademyHealth; 2012 [cited 2023 Aug 22]. Available from: https://academyhealth.org/sites/default/files/Involving%20Patients%20and%20Consumers%20in%20Research_%20New%20Opportunities%20f.pdf.
12. Holden RJ, Valdez RS, Anders S, Ewart C. The Patient Factor: Involving Patient and Family Stakeholders as Advisors, Co-Designers, Citizen Scientists, and Peers. Proc Hum Factors Ergon Soc Annu Meet. 2020;64(1):622-26. doi: 10.1177/1071181320641141.
13. Gandhi TK, Kaplan GS, Leape L, et al. Transforming concepts in patient safety: a progress report. BMJ Qual Saf. 2018;27(12):1019-1026. doi: 10.1136/bmjqs-2017-007756.
14. Doyle C, Lennox L, Bell D. A systematic review of evidence on the links between patient experience and clinical safety and effectiveness. BMJ Open. 2013;3(1):e001570. doi: 10.1136/bmjopen-2012-001570.
15. Anhang Price R, Elliott MN, Zaslavsky AM, et al. Examining the role of patient experience surveys in measuring health care quality. Med Care Res Rev. 2014;71(5):522-54. doi: 10.1177/1077558714541480.

16. Ocloo J, Matthews R. From tokenism to empowerment: progressing patient and public involvement in healthcare improvement. BMJ Qual Saf. 2016;25(8):626-32. doi: 10.1136/bmjqs-2015-004839.
17. Te Tāhū Hauora Health Quality & Safety Commission. Code of expectations for health entities' engagement with consumers and whānau [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/consumer-hub/engaging-consumers-and-whanau/code-of-expectations-for-health-entities-engagement-with-consumers-and-whanau/.
18. *Pae Ora (Healthy Futures) Act 2022*.
19. Te Tāhū Hauora Health Quality & Safety Commission. Code of expectations for health entities' engagement with consumers and whānau. Part 2.2 [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/consumer-hub/engaging-consumers-and-whanau/code-of-expectations-for-health-entities-engagement-with-consumers-and-whanau/.
20. Te Tāhū Hauora Health Quality & Safety Commission. Te aratohu tikanga: Code of expectations implementation guide [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/consumer-hub/engaging-consumers-and-whanau/implementing-the-code/.
21. Dunsch F, Evans DK, Macis M, Wang Q. Bias in patient satisfaction surveys: a threat to measuring healthcare quality. BMJ Global Health. 2018;3(2):e000694. doi: 10.1136/bmjgh-2017-000694.
22. Institute for Healthcare Improvement (IHI). Triple Aim and Population Health [Internet]. Boston, MA (US): Institute for Healthcare Improvement; 2023 [cited 2023 Aug 22]. Available from: https://www.ihi.org/Engage/Initiatives/TripleAim/Pages/default.aspx.
23. Sikka R, Morath JM, Leape L. The Quadruple Aim: care, health, cost and meaning in work. BMJ Qual Saf. 2015;24(10):608-10. doi: 10.1136/bmjqs-2015-004160.
24. Institute of Medicine (IOM). Crossing the Quality Chasm: A New Health System for the 21st Century. Washington DC (US): National Academy Press; 2001.
25. Te Tāhū Hauora Health Quality & Safety Commission. Patient-reported measures [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/our-data/patient-experience/.
26. Te Tāhū Hauora Health Quality & Safety Commission. New Zealand patient experience survey programme refresh 2019/20 [Internet]. Wellington (NZ): Te Tāhū Hauora; 2021 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/resources/resource-library/new-zealand-patient-experience-survey-programme-refresh-201920/.
27. Te Tāhū Hauora Health Quality & Safety Commission. New Zealand COVID-19 patient experience survey questionnaire [Internet]. Wellington (NZ): Te Tāhū Hauora; 2020 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/our-data/patient-reported-measures/patient-experience/survey-results/#COVID.
28. Booker QS, Austin JD, Balasubramanian BA. Survey strategies to increase participant response rates in primary care research studies. Fam Pract. 2021;38(5):699-702. doi: 10.1093/fampra/cmab070.
29. Creavin ST, Creavin AL, Mallen CD. Do GPs respond to postal questionnaire surveys? A comprehensive review of primary care literature. Fam Pract. 2011;28(4):461-7. doi: 10.1093/fampra/cmr001.
30. Ricci-Cabello I, Avery AJ, Reeves D, et al. Measuring patient safety in primary care: the development and validation of the "Patient Reported Experiences and Outcomes of Safety in Primary Care" (PREOS-PC). Ann Fam Med. 2016;14(3):253-61. doi: 10.1370/afm.1935.
31. Medina-Lara A, Grigore B, Lewis R, et al. Cancer diagnostic tools to aid decision-making in primary care: mixed-methods systematic reviews and cost-effectiveness analysis. Health Technol Assess. 2020;24(66):1-332. doi: 10.3310/hta24660.
32. Thomson M, Pledger M, Hamblin R, et al. Comparing initial and follow-up responders to a New Zealand patient experience survey. N Z Med J. 2018;131(1482):46-58.
33. Johnson TP, Wislar JS. Response rates and nonresponse errors in surveys. JAMA. 2012;307(17):1805-6. doi: 10.1001/jama.2012.3532.
34. Ahmed F, Burt J, Roland M. Measuring patient experience: concepts and methods. Patient. 2014;7(3):235-41. doi: 10.1007/s40271-014-0060-5.
35. Groves RM, Peytcheva E. The impact of nonresponse rates on nonresponse bias: a meta-analysis. Public Opin Q. 2008;72(2):167-89. doi: 10.1093/poq/nfn011.
36. Stats NZ Tatauranga Aotearoa. A guide to good survey design: Fifth edition [Internet]. Wellington (NZ): Stats NZ Tatauranga Aotearoa; 2019 [cited 2023 Nov 14]. Available from: https://www.stats.govt.nz/assets/Uploads/Methods/A-guide-to-good-survey-design-fifth-edition/a-guide-to-good-survey-

design-fifth-edition.pdf.
37. Leeuw ED, Hox JJ, Dillman DA, editors. International handbook of survey methodology. New York (US): Lawrence Erlbaum Associates; 2009.
38. Te Tāhū Hauora Health Quality & Safety Commission. Survey information and methodology [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Nov 14]. Available from: https://www.hqsc.govt.nz/our-data/patient-reported-measures/patient-experience/survey-information-and-methodology/.
39. Rolstad S, Adler J, Rydén A. Response burden and questionnaire length: is shorter better? A review and meta-analysis. Value Health. 2011;14(8):1101-8. doi: 10.1016/j.jval.2011.06.003.
40. Te Whatu Ora – Health New Zealand. Zero Data [Internet]. Wellington (NZ): Te Whatu Ora – Health New Zealand; 2023 [cited 2023 Aug 22]. Available from: https://www.tewhatuora.govt.nz/our-health-system/digital-health/zero-data/.
41. Te Tāhū Hauora Health Quality & Safety Commission. From PES to PDSA – Workbook: Using adult primary care patient experience survey data for quality improvement [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Nov 7]. Available from: https://www.hqsc.govt.nz/resources/resource-library/from-pes-to-pdsa-workbook-using-adult-primary-care-patient-experience-survey-data-for-quality-improvement/.
42. Te Tāhū Hauora Health Quality & Safety Commission. A window on quality 2022 (Part 2): COVID-19 and impacts on our broader health system (Part 2) | He tirohanga kounga 2022: Me ngā pānga ki te pūnaha hauora whānui (Wāhanga 2) [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://www.hqsc.govt.nz/resources/resource-library/a-window-on-quality-2022-part-2-whakarapopototanga-matua-he-tirohanga-kounga-2021-wahanga-2/.
43. Manatū Hauora – Ministry of Health. Health and Disability System Review – Interim Report [Internet]. Wellington (NZ): Manatū Hauora – Ministry of Health; 2019 [cited 2023 Aug 22]. Available from: https://www.health.govt.nz/publication/health-and-disability-system-review-interim-report.
44. Jaine R, Giness D, Hohaia J, et al. Raising the bar on the National Patient Experience Survey [Internet]. Wellington (NZ): Health Quality & Safety Commission New Zealand; 2017 [cited 2023 Nov 7]. Available from: https://www.hqsc.govt.nz/assets/Consumer-hub/Partners-in-Care/Publications-resources/Raising-the-bar-on-the-National-Patient-Experience-Survey-Final-REVIEWED-Report-19-05-2017.pdf.
45. George V, Jaine R, Opferkuch C, et al. Raising the Bar on the National Adult Inpatient Experience Survey Phase 2: Co-Designing Nudges [Internet]. Wellington (NZ): Health Quality & Safety Commission New Zealand; 2018 [cited 2023 Nov 7]. Available from: https://www.hqsc.govt.nz/assets/Our-data/Publications-resources/Co-designing-nudges-raising-the-bar-on-the-national-adult-inpatient-experience-survey-Nov-2018.pdf.
46. Auckland, Waitemata & Counties Manukau Health Alliances. 2020/2021 System Level Measures Improvement Plan [Internet]. Auckland (NZ): Waitemata DHB; 2021 [cited 2023 Nov 7]. Available from: https://www.waitematadhb.govt.nz/assets/Documents/health-plans/Metro-Auckland-SLM-Improvement-Plan-2020-21-Final.pdf.
47. Auckland, Waitemata & Counties Manukau Health Alliances. 2021/2022 System Level Measures Improvement Plan [Internet]. Auckland (NZ): Auckland DHB; 2022 [cited 2023 Nov 7]. Available from: https://www.adhb.health.nz/assets/Documents/About-Us/Planning-documents/Metro-Auckland-SLM-Improvement-Plan-2021-2022.pdf.
48. Te Tāhū Hauora Health Quality & Safety Commission. Adult primary care patient experience explorer [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://reports.hqsc.govt.nz/APC-explorer/.
49. Te Tāhū Hauora Health Quality & Safety Commission. Adult hospital inpatient experience explorer [Internet]. Wellington (NZ): Te Tāhū Hauora; 2023 [cited 2023 Aug 22]. Available from: https://reports.hqsc.govt.nz/AHI-explorer/.
50. Locock L, Montgomery C, Parkin S, et al. How do frontline staff use patient experience data for service improvement? Findings from an ethnographic case study evaluation. J Health Serv Res Policy. 2020;25(3):151-161. doi: 10.1177/1355819619888675.
51. Coulter A, Locock L, Ziebland S, Calabrese J. Collecting data on patient experience is not enough: they must be used to improve care. BMJ. 2014;348:g2225. doi: 10.1136/bmj.g2225.

Discordant thyroid function tests—beware of albumin variants

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Familial dysalbuminaemic hyperthyroxinaemia (FDH) is a benign euthyroid condition caused by albumin variants with increased binding affinity for thyroxine (T4). FDH falsely elevates T4 by common indirect/analogue methods confounding patient diagnosis.[1,2] Uncommonly, FDH may be present in a patient with Graves' disease, as seen in the case below, making interpretation of tests used for monitoring difficult.

Case report

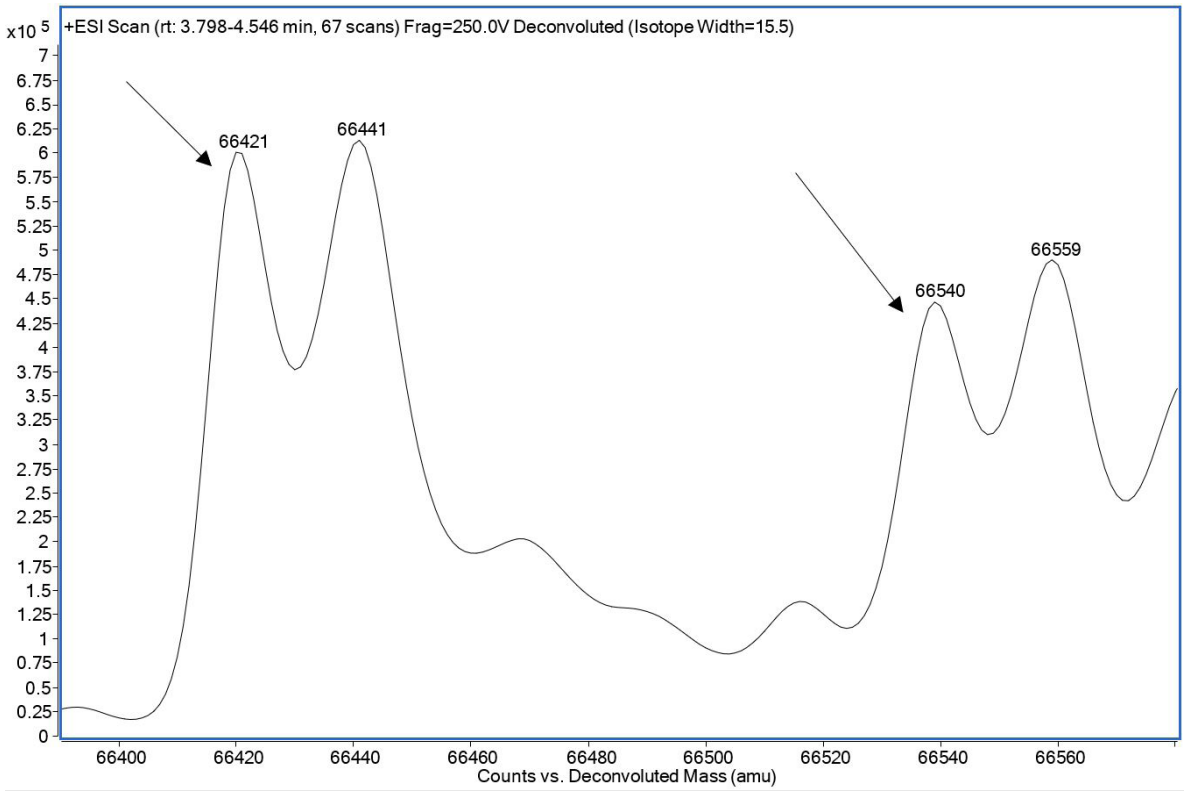
A 24-year-old Afghan woman, G1P1, was referred with hyperthyroidism in April 2022 following sinus tachycardia during labour, which persisted following delivery despite fluids and labetalol. She had three presentations with hyperemesis gravidarum requiring anti-emetics and intravenous fluids in the first 14 weeks of pregnancy. No thyroid function tests (TFTs) were performed during pregnancy. Community bloods 3 years earlier (October 2019) demonstrated free thyroxine (FT4) >77pmol/L (reference interval [RI] 7–16), free triiodothyronine (FT3) 31.6pmol/L (RI 3.6–6.5) and thyroid stimulating hormone (TSH) <0.01mU/L (RI 0.3–5.0) (Table 1). She denied symptoms of thyrotoxicosis and was unaware of her previous abnormal result. It was unclear why these results were not followed up at the time. Clinically, she was mildly thyrotoxic, with a small diffuse goitre and no thyroid eye disease. Initial TFTs were thought somewhat atypical for Graves' disease with discordance between FT4 and FT3 levels. TSH receptor antibodies were positive at 3.1U/L (RI <2U/L) consistent with a diagnosis of Graves' disease. Carbimazole was commenced, increasing to 15mg per day. Thyroid hormone levels increased in May 2022, requiring a further

Table 1: TFT result comparison.

TFT	October 2019	April 2022	May 2022	October 2022	Reference interval
Roche					
TSH (mU/L)		<0.02		0.45	0.27–4.2
FT4 (pmol/L)		37		32	11–22
FT3 (pmol/L)		8.5		6.3	3.1–6.8
Beckman Coulter					
TSH (mU/L)	<0.01		<0.01	0.42	0.3–5.0
FT4 (pmol/L)	>77		67	31	7–16
FT3 (pmol/L)	31.6		9.1	6.1	3.6–6.5
TT4 (nmol/L)				281	78–157
FTI (nmol/L)				98	75–170
TT3 (nmol/L)				1.70	1.30–2.70

TFT = thyroid function test; TSH = thyroid stimulating hormone; FT4 = free thyroxine; FT3 = free T3; TT4 = total thyroxine; FTI = free thyroxine index; TT3 = total T3

Figure 1: Electrospray ionisation time-of-flight mass spectrometry (ESI-TOF-MS) deconvoluted albumin mass spectrum showing the expected major albumin isoform (66,441 Da) and cysteinylated derivative (66,559 Da), as well as variant masses for each component (66,421 and 66,540 Da; annotated by arrows).



dose increase to 25mg per day. Subsequently, in October 2022, TFTs measured on Roche e801 showed TSH had normalised (0.45mU/L [RI 0.27–4.2]). However, FT4 remained elevated (32pmol/L [RI 12–22]), while FT3 was normal (6.3pmol/L [RI 3.1–6.8]), raising the possibility of assay interference. Total T4 (TT4) was elevated on Beckman Coulter Access 2 (281nmol/L [RI 78–157]) but free T4 index (FTI) was normal (98 [RI 75–170]), as was total T3 (TT3) (1.70nmol/L [RI 1.30–2.70]).

Electrospray ionisation time-of-flight mass spectrometry (ESI-TOF-MS) identified the expected albumin isoform (66,441 Da) and cysteinylated derivative (66,559 Da) alongside variant albumin masses, approximately 19 Da fewer (Figure 1). Genetic sequencing confirmed heterozygosity for FDH variant NM_000477.5(*ALB*):c.725G>A, p.(Arg242His), also known as Arg218His by protein nomenclature.

Neonatal bloods for her daughter at day 5 showed FT4 >100pmol/L (RI 11–35), FT3 13.2pmol/L (RI 1.7–11) and TSH <0.02mU/L (RI 0.5–11), consistent with neonatal Graves' disease, despite relatively low maternal TSH receptor antibodies following delivery. It is likely that antibody levels may have been higher earlier in the third trimester. Treatment with carbimazole was commenced for 5 weeks, resulting in normalisation of FT3 and TSH but continued mild elevation of FT4. By 13 months of age, FT4 remained elevated (27pmol/L [RI 7–16]), with normal FT3 (7pmol/L [RI 3–10) and TSH (1.48mU/L [RI 0.3–5]).

Discussion

In patients with discordant TFTs showing elevated T4 or T3 but non-suppressed TSH, analytical artefacts should be considered. Discordant results on multiple platforms may represent FDH, transthyretin variants, thyroid hormone resistance, thyroid hormone autoantibodies or, rarely, TSH-secreting pituitary adenomas.[3,4] An ESI-TOF-MS method was developed locally to screen for FDH and transthyretin variants.[5] Arg218His is the most widespread FDH variant and has previously been detected

in Caucasian, Hispanic, Chinese, Korean and Anatolian Turkish patients.[3,5–10] A decrease in albumin mass of 19 Da is highly suggestive of Arg218His in combination with discordant TFTs. Genetic testing was undertaken for confirmation in our case, as this FDH variant has not previously been reported in Afghan patients.

Patients with FDH usually have normal TSH and high T4 and/or T3 depending on the variant, and no clinical symptoms of thyrotoxicosis. However, FDH may present alongside thyroid disease, as seen in our patient with Graves' disease. Identification of FDH in patients with Graves' disease requires high clinical suspicion. In the first instance, given clear evidence of Graves' disease, it was appropriate to treat for Graves' disease. However, with normalisation of TSH and FT3, the discordant pattern became clear, raising clinical suspicion for FDH.

FDH is inherited in an autosomal dominant manner. There was no known family history of hyperthyroidism for our case and the patient's mother and sister both had normal TSH and FT4. Her father was deceased. As such, it is likely she inherited FDH from her father. Given the high FT4 measured in the setting of normal TSH and FT3 for the daughter, it is likely she has also inherited the FDH variant. FDH can coexist with thyroid disease, complicating patient management when not diagnosed. Unfortunately, the daughter's elevated FT4 initially led her paediatrician to restart carbimazole unaware of her mother's FDH diagnosis. As such, it is recommended to identify FDH in first-degree relatives in order to prevent future misdiagnosis and inappropriate management from discordant TFTs.

Equilibrium dialysis mass spectrometry for FT4 or FTI calculated from TT4 and thyroid uptake by immunoassay appear unaffected by Arg218His, but these methods are not widely available in Australasia.[1] Although FTI was previously available at Canterbury Health Laboratories in Christchurch, the discontinuation of thyroid uptake reagent by Beckman Coulter has led to discontinuation of FTI reporting in New Zealand, though TT4 and TT3 remain available for specialist requestors for interference investigations. FT3 assays are usually unaffected or only slightly affected by Arg218His and were helpful in monitoring treatment for Graves' disease in our patient.[10] Other rarer forms of FDH may present with higher FT3 on routine immunoassays. When TFTs are discordant and unexpected, liaison with the laboratory is recommended to investigate potential interferences and avoid unnecessary investigations for thyroid hormone resistance or TSH-oma, or misattribution to hyperthyroidism.

COMPETING INTERESTS
Nil.

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URL
www.nzmj.org.nz/journal/vol-137-no-1588/discordant-thyroid-function-tests-beware-of-albumin-variants

REFERENCES
1. Cartwright D, O'Shea P, Rajanayagam O, et al. Familial dysalbuminemic hyperthyroxinemia: a persistent diagnostic challenge. Clin Chem. 2009 May;55(5):1044-6. doi: 10.1373/clinchem.2008.120303.
2. Koulouri O, Moran C, Halsall D, et al. Pitfalls in the measurement and interpretation of thyroid function tests. Best Pract Res Clin Endocrinol Metab. 2013 Dec;27(6):745-62. doi: 10.1016/j.beem.2013.10.003.
3. Li B, Moore J, Potter H, et al. Two cases of discordant thyroid function tests: familial dysalbuminaemic hyperthyroxinaemia and dystransthyretinaemic hyperthyroxinaemia. Intern Med J. 2023 Jan;53(1):152-154. doi: 10.1111/imj.15981.
4. Glendenning P, Siriwardhana D, Hoad K, Musk A. Thyroxine autoantibody interference is an uncommon cause of inappropriate TSH secretion using the Immulite 2000 assay. Clin Chim Acta. 2009 May;403(1-2):136-8. doi: 10.1016/j.cca.2009.02.002.
5. Ryan JB, Brennan SO, Potter H, et al. Familial dysalbuminaemic hyperthyroxinaemia: a rapid and novel mass spectrometry approach to diagnosis. Ann Clin Biochem. 2016 Jul;53(Pt 4):504-7. doi: 10.1177/0004563215598168.
6. Sunthornthepvarakul T, Angkeow P, Weiss RE, et al. An identical missense mutation in the albumin gene results in familial dysalbuminemic hyperthyroxinemia in 8 unrelated families. Biochem Biophys Res Commun. 1994 Jul 29;202(2):781-7. doi: 10.1006/bbrc.1994.1998.
7. Pappa T, Ferrara AM, Refetoff S. Inherited defects of thyroxine-binding proteins. Best Pract Res Clin Endocrinol Metab. 2015 Oct;29(5):735-47. doi: 10.1016/j.beem.2015.09.002.
8. Mimoto MS, Karaca A, Scherberg N, et al. Homozygous Mutation in Human Serum Albumin and Its Implication on Thyroid Tests. Thyroid. 2018 Jun;28(6):811-814. doi: 10.1089/thy.2017.0564.
9. Brown A, Hofman P, Li B, et al. A Positive Newborn Screen for Congenital Hypothyroidism in a Clinically Euthyroid Neonate-Avoiding Unnecessary Treatment. Int J Neonatal Screen. 2023 Mar 23;9(2):16. doi: 10.3390/ijns9020016.
10. Mahendhar R, Shahbaz A, Riaz M, et al. Effect of Albumin Polymorphism on Thyroid Hormones: A Case Report and Literature Review. Cureus. 2018 Jul 1;10(7):e2903. doi: 10.7759/cureus.2903.

Cancer care in New Zealand: thoughts from afar

Murray F Brennan

I am the most fortunate of people. I received a remarkable education at the University of Otago in the early 1960s. It was meant to be a medical education; however, I learnt very little about medicine but a great deal about life. I was no academic star as I stumbled through somewhere in the midst of the pack. During that time, however, I was able to grow from an immature youth to some semblance of an adult. I was fortunate to be involved in high-level rugby, be active in student politics, complete a degree in math, and had an opportunity to travel internationally while completing medical school. Once graduated, I realised how little I knew, and how I would have to give up most, if not all, of these extracurricular activities to learn some medicine. This began in Dunedin, and it was matured when I moved to Boston to the Peter Bent Brigham Hospital, where I completed a surgical residency. Following that, I spent 6 years at the National Cancer Institute, developing my own clinical and basic research interest in cancer.

I moved to the Memorial Sloan-Kettering Cancer Centre (MSK) in 1981, where I have stayed ever since. MSK is a unique place and premiere cancer hospital, present for over 150 years. Working in this focussed environment has been a privilege and a constant challenge. Having similarly focussed colleagues around me has provided constant stimulus. While I have been invited to look elsewhere I cannot imagine an environment more conducive to challenge, innovation and progress in cancer care.

People used to ask why I did not return to New Zealand. It was not that I did not wish to return or that I did not have enormous affection for the country, particularly for central Otago where I have two sons. Both were raised in New York but have embraced New Zealand culture, as if native-born. I did not return because there was nothing to return to that would allow me to fill my professional aspirations. What do I mean by that and why do I bother even writing this brief tome? When I began more than four decades ago, the only potentially curative therapy for solid tumours was a surgical operation, often a major operation with significant morbidity. Remember John Hunter, who in the eighteenth century said, "*Surgery is like an armed savage trying to render by force what a civilized man would render by stratagem*".[1] Chemotherapy for haematological and paediatric malignancies began in the 1950s. Radiation therapy, while discovered early in the century, was relatively ineffective and viewed predominantly as palliation, which in itself was accompanied by significant side effects. For me there was a great opportunity for understanding the cancer patient and the response in man to the presence of a cancer.

We now know that where you get your initial cancer diagnosis, investigation and initiation of a treatment plan determines your outcome. We can clearly show that survival outcome and minimisation of complications of cancer treatment are improved when delivered in a cancer centre. What is most exciting is that new innovations, delivered precisely and to minimise morbidity, can provide superior outcome. Cancer care in the 2020s is a team sport. There is no one player that dominates the delivery of care. Leadership yes, research yes, sensitivity and insight yes, but it is all done within the team.

New Zealand is a relatively small country in terms of population, but diverse in culture. In my opinion, New Zealand deserves two focussed cancer centres, one in each main island. New Zealand has unique challenges in cancer care; some relate to the diversity of the population, and, as in the United States, the limitation of access for certain sections of the populace. However, with the advent of video consultation and remote evaluation it is no longer necessary that every patient with cancer be referred to a centre of excellence. It is, again in my opinion, essential that there be centres of excellence to provide consultation and access for the more complicated cancers, and those requiring more sophisticated or innovative evaluation and treatment. An innovative centre providing a clinical and research environment will attract clinicians and information scientists back into New Zealand.

Why bother with this preamble? I have been

involved in providing some advice and support for the proposed cancer centre in Christchurch. This is not to pre-empt other parts of the nation, but is in a community with strong clinical, financial and intellectual strengths with a supportive private sector that would allow a model that could be applied elsewhere. I believe citizens of New Zealand, given the strong historical record of excellent clinical care, deserve and need a focussed national cancer centre network with hubs in Auckland and Christchurch. Such dedicated cancer centres would contain a strong research component in all areas of cancer and would provide an environment that would attract and retain clinicians, scientists and healthcare professionals who want to live in one of the most attractive countries in the world, but need the intellectual, physical and societal ambiance that allows them to grow and contribute to the better care of the cancer patient.

COMPETING INTERESTS
Nil.

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www.nzmj.org.nz/journal/vol-137-no-1588/cancer-care-in-new-zealand-thoughts-from-afar

REFERENCES

1. Hunter J. Lectures on the Principles of Surgery. Philadelphia: Haswell, Barrington, and Haswell; 1839. Chapter 1, Introduction, p. 9.

Asthma

NZMJ, 1924

By E. H. Williams, M.B.

In the first place I must express my appreciation of the honour you have done me in asking me to open this discussion on asthma. Except that I have interested myself in this subject in its more recent conception, I can lay no claim to fitness for so important a position. If the date of this discussion were 1913 instead of 1923, our remarks would be in terms of *Curschman's* spirals, *Leyden's* crystals, or the perles of *Lannec*, and we would be repeating what we had learned as students. During the last ten years, however, much has been explained that was hardly guessed at before that time, and a most interesting or even fascinating study has centred round the symptom called asthma and some allied conditions. While my contribution to this discussion is almost entirely the result of my own personal experience in practice, and that chiefly among children, it will be necessary for me to mention a few scientific facts just so far as they have been helpful to me in explaining certain phenomena.

Of the more technical aspect *Dr. Carmalt Jones* will speak more fully and give us the results of special study in immunity and its relation to anaphylaxis. I wish it understood that in my remarks I refer to spasmodic asthma, excluding renal and cardiac dyspnoea, obstructive dyspnoea from tracheo-bronchial gland enlargement, abnormalities or infections of the nasopharynx, reflex causes, bacterial infection generally, primary lung condition such as bronchitis and emphysema preceding the asthmatic complication.

In *Progressive Medicine*, 1922, it says: "In the study of ex-service men the greatest difficulty has been experienced in distinguishing between chronic bronchitis with exacerbations and with the varied types of dyspnoea incident to this condition and true bronchial asthma with added bronchitis. Neither the clinical history and observation nor a complete physical examination will differentiate between them. Unless such conditions as I have just mentioned are first eliminated, treatment by desensitisation will be disappointing; and even when the asthma is primary to bronchitis and emphysema, in cases older than 40, though there may be great relief from treatment, disappointment follows.

I mentioned without any specific purpose the year 1913—merely because a decade has elapsed since then—but in *Osler's System of Medicine* of that date, besides the terms referred to earlier—*Leyden's* crystals, etc.—one finds certain observations set down which indicate that attention was being drawn in the right direction without the true explanation being forthcoming. *Osler* speaks of swelling of bronchial mucous membrane, spasms of peribronchial muscles, a viscid exudation in bronchioles as observed by *Curschman*, the association of angio-neurotic oedema with asthma, of asthma following upon an attack of whooping cough. He refers, as do other writers, to asthma arising from flowers and grasses and animal emanations: to the effects of certain diets or articles of food. He mentions the phenomenon of anaphylaxis in connection with fatal results following the injection of antitoxin in asthmatic subjects; he refers to the presence of eosinophilia in the blood of those recovering from an attack of asthma. Yet he concludes as follows: "Briefly stated, then, bronchial asthma is a neurotic affection characterised by hyperæmia and turgescence of the mucosa of smaller bronchial tubes and a peculiar exudate of mucin.

Most of *Osler's* observations are explained in the light of our present knowledge, and, while it must be admitted that a medical hobby-horse is often ridden till it drops, the explanation of spasmodic asthma and hay asthma by anaphylaxis or allergy is almost entirely convincing.

In much more recent publications than *Osler* of 1913 one finds similar observations without what appears to be the correct explanation of asthmatic attacks. One must remember, however, that many writers compiled their articles before the war—that is, before recent experimental work helped to explain this subject—and have only recently succeeded in getting their articles published.

In the *Synopsis of Medicine*, 1922, *H. Lethaby Tidy* refers to (1) spasm of muscular coat of smaller bronchi as being the generally accepted

factor. Stimulation of vagus causes constriction of bronchi, distension of lungs with air resulting. Drugs controlling this vagotonia are recommended, belladonna or atropine giving striking results in this direction. (2) Swelling of bronchial mucous membrane, the rapidity of onset being parallel to urticaria, the paroxysm being an urticaria of the mucous membrane. These observations are not in advance of *Osler's*, and though asthma and urticaria are mentioned together, the importance of their relationship does not seem to be appreciated.

Dale and *Auld*, in the *British Medical Journal* during the last two years, have contributed several articles to the subject of specific sensitiveness and anaphylaxis, and without encroaching upon the technical aspect of their work, to which *Dr. Carmalt Jones* may wish to refer, I should like to pick out a few experimental and clinical facts mentioned in their articles which may have some significance:

1. "When a guinea-pig dies suddenly from the intravenous introduction of a foreign protein to which it is sensitive, as a result of anaphylaxis there is a tense contraction of plain muscle surrounding the bronchioles, with the lungs in full inspiration and with right-side heart failure. Recovery, if it occurs, is associated with eosinophilia."—This is strictly comparable to one variety of anaphylactic attack in man or to severe spasmodic asthma, as pointed out by *Tidy*, from whom I have just quoted.
2. In speaking of passive anaphylaxis it is stated "that if a guinea-pig be sensitised to a foreign protein, the serum of this animal injected into another of the same species will cause a similar tendency."—Somewhat analogous to this is the reported case of blood from a donor conveying horse asthma to the recipient, hitherto a stranger to it.
3. The usual interval of eight to ten days that elapses before the appearance of a serum rash in man is the same interval of time noticed in a guinea-pig after the first injection, before the anaphylactic condition develops in that animal.
4. Anaphylaxis produced in rabbits is more often associated with gastro-intestinal symptoms.—In children suffering from anaphylactic manifestations the attack may resemble the pulmonary condition seen in the guinea-pig or the intestinal symptoms found in the rabbit.
5. Anti-anaphylaxis is taken to mean a period of freedom from sensitiveness following an attack. During this period the antigen will not act if injected, and such a person may eat egg, for instance, with impunity.—Though some writers are not in agreement with this statement, it is certainly inconclusive to depend upon the results of skin tests immediately following upon an attack of asthma.

Auld's work is based upon the non-specific treatment by peptone, and his conclusions are supported by *Dale*, who says a dose of peptone not in itself large enough to produce any pronounced reaction will weaken the response of a sensitised guinea-pig to the sensitising antigen, so that after such a dose it is possible to desensitise without at any time producing any pronounced reaction. *Auld* believes that the character of anaphylaxis and peptone poisoning or shock are identical.

The work of *Chandler Walker* is well known to all of us. He is the American representative of the specific protein theory school. To arrive at the particular antigen concerned in asthmatic attacks in a patient, skin tests are performed and the local reaction noted. These tests may be few or very numerous, embracing in some cases most of the proteins known in the animal and vegetable kingdoms. He prefers the dermal to the intra dermal method, the latter being too sensitive. That valuable information can be gained from skin tests is indisputable, but the procedure is apt to be cumbersome and irksome, so that the non-specific treatment with peptone offers considerable advantages.

Herbert French in the *Medical Annual*, 1922, in supporting non-specific protein therapy, says: "It might seem illogical, after thirty years of satisfactory effort to perfect specific therapy, to study non-specific therapy, but if we analyse the subject we will see it is by no means true. Instead of trying to alter or modify the agent causing the disease, whether microbic or toxic, by specific treatment, we may alter the reaction of the body—that is, the inflammatory reaction to the offending agent." During the war it was noted that gonococcal infections were benefitted by typhoid vaccines; in 1918, that numerous cases of arthritis and fibrositis answered to blunderbus injections or influenza and catarrhal vaccine, the effect being probably due to the protein content of those vaccines and serums.