

The cost of paediatric and perianal Crohn's disease in Canterbury, New Zealand

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Abstract

Aims The aim of this study was to determine the direct and indirect costs of Crohn's disease (CD) in paediatric and perianal patients in Canterbury in one year.

Methods A retrospective cross-sectional analysis was performed. Paediatric CD patients and adult patients with perianal CD were recruited over a three month period. Interviews were conducted to obtain information regarding demographic, socioeconomic factors, and indirect costs. Hospital clinical notes were reviewed to determine direct health care utilisation and costs.

Results Forty-nine patients (24 paediatric and 25 perianal CD) were enrolled. In one year the total costs per patient for paediatric CD were \$14,375 with direct and indirect costs comprising \$12,583 and \$1,792, respectively. The total costs per patient for perianal CD were \$20,366 with direct and indirect costs comprising \$18,261 and \$2,105, respectively. Extrapolating these data across New Zealand, the total cost of paediatric and perianal CD in one year is approximately \$25.9 million and \$36.7 million, respectively.

Conclusions Paediatric and perianal CD are high-cost diseases with significant costs borne by patients and their families. Expensive pharmaceuticals comprise a significant proportion of the costs: increased access to these drugs might decrease hospital admissions and prevent work absenteeism and loss of carer productivity.

Crohn's disease (CD) is a chronic inflammatory bowel disease characterised by transmural segmental inflammation of the gastrointestinal tract. The incidence of CD is increasing worldwide¹ and the peak age of onset is between 15 and 35 years.² CD remains incurable and a proportion of patients will endure recurrent and prolonged periods of illness requiring extensive medical and surgical interventions during what would otherwise be a highly productive time of life. Hence CD presents an increasingly significant health problem not only in terms of morbidity, but also in cost to the individual patient and society.

Previous studies have indicated that the majority of the total cost associated with this disease relates to "extensive interventions required by a small proportion of severely affected individuals".³ Several clinical markers of disease severity have been documented including diagnosis at a young age and the presence of perianal disease.⁴ In addition to being independent markers of a more severe disease course, paediatric patients have a longer temporal exposure to CD-related complications and perianal disease patients develop local perianal complications requiring frequent intervention⁵ hence, both these patient groups are more likely to use significant health resources and incur the largest costs as a result of the disease.

A number of international studies have considered the cost of inflammatory bowel disease overall,⁶⁻¹⁵ however, to the authors' knowledge, there are no data documenting the average patient cost of IBD in New Zealand or Australia and no previous work specifically investigating the paediatric and perianal CD groups.

Evidence is accumulating that newer therapies, such as anti-tumour necrosis factor alpha (anti-TNF α) antibodies, have significant efficacy in inducing and maintaining remission and have particular roles in complicated CD, such as perianal CD.^{16,17} However, these agents are expensive and predicting which patients will benefit from their early use remains a challenge.⁴ The introduction of these modern biological treatments has created a need for government agencies to consider the economic impact of therapeutic alternatives. To achieve this, the cost of CD and its societal burden requires further study.

This is a cross-sectional retrospective study, which aimed to estimate the direct and indirect costs of paediatric and perianal CD in one year in Canterbury using patient-based data. The costs of CD are borne not only by the taxpayer through government funded healthcare but the patient and their family, therefore the cost perspective is approached from a societal point of view.

Methods

This study was performed in Christchurch Hospital, a tertiary university hospital serving a population of around 500,000. Ethical approval for the study was obtained from the regional ethics committee. All patients with CD according to previously documented diagnostic criteria¹⁸ presenting to the institution during the period November 2009 to February 2010 were eligible for entry into the study.

Paediatric patients were defined as those aged 16 years or less. Perianal disease was defined as any symptomatic perianal lesion included in the American Gastroenterological Association classification.¹⁹ Patients were recruited through Gastroenterology, Colorectal surgical and Paediatric outpatient clinics and hospital admissions.

After giving written informed consent, patients or their parents were submitted to a structured interview to obtain information regarding demographic and socioeconomic factors, work and school absenteeism, alternative health resource use and other related data for the preceding twelve month period.

Participants were also offered the opportunity to nominate other costs that were not mentioned in the interview. Following the structured interview, hospital clinical notes were reviewed to determine direct health care utilization. This included hospital inpatient and outpatient visits and prescription drug use.

For the purposes of this analysis the costs were classified as direct or indirect. Direct costs included hospital (Emergency Department visits, laboratory tests, radiological investigations, endoscopy, pharmaceuticals, inpatient care and operating theatre costs) and outpatient (General Practice visits, specialist clinic visits, alternative health professional visits, non-prescription medications, pharmaceuticals, laboratory tests, District Nurse and Social Work services) associated costs. Indirect costs included; lost productivity, travel, carers, tutors and additional phone or internet requirements.

The costs of hospital resources were determined through the Costing Department of the Canterbury District Health Board (CDHB) and the Ministry of Health. Hospital costs were calculated using DRG codes assigned to the patient each time they visited the hospital. A different code is given for each service required during each visit. Based on the quantity used during the visit the cost is calculated for each service used in a given visit.

The cost department for the CDHB supplied the authors with all the codes and costs accrued by the patients during the study period. For primary care cost calculation, it was assumed that all patients were enrolled in a primary health organisation (PHO). The New Zealand Government provides subsidies to lower the cost of general practitioner (GP) visits for eligible people enrolled in a PHO. The cost of GP services was estimated using the average cost of an appointment by age as obtained from Pegasus Health PHO and the 2010 yearly capitation rates provided by the Government. The capitation rates took into account whether or not the patient had a high user health card (HUHC).

Pharmaceutical costs were calculated from the cost to the Pharmaceutical Management Agency in New Zealand (PHARMAC) provided by their pharmaceutical schedule accessed 1 December 2009. Additionally a 4% mark-up was added to pharmaceutical costs plus a \$5.80 dispensing fee attributable to pharmacists. The co-payments paid by the patient are a transfer not a cost therefore these were not included.

The human capital method as described by Drummond et al²⁰ was employed in calculating indirect costs. Patients were asked the number of days they had off work as either unpaid or annual leave related to CD. This was transferred into hours off work and was multiplied by their gross hourly wage. For those patients not in work their indirect costs are discussed descriptively as monetary values were not able to be estimated. Government welfare payments received by patients not in work were not included as they represent a transfer rather than a cost.

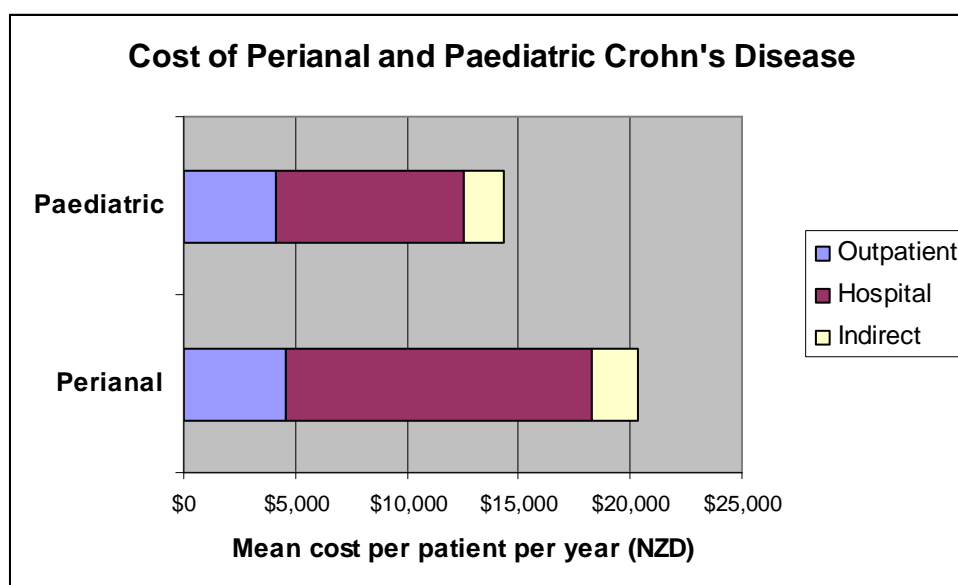
It is conservatively estimated that there are approximately 9000 individuals with IBD in NZ, with perianal and paediatric patients consisting of about 20% each. This estimation was used to extrapolate the data to obtain values for the cost to society in New Zealand of perianal and paediatric CD in one year.

Results

In total 49 patients were entered into the study; 24 paediatric CD patients (mean age 12 years, range 4 to 15 years) and 25 adult patients with perianal CD (mean age 33 years, range 17 to 73 years). The paediatric group contained 16 males and 21 of the patients were of New Zealand European ethnicity.

The perianal group contained 15 males; 21 declared New Zealand European ethnicity. All of the paediatric patients were attending school except for one who was at kindergarten part-time. In the perianal sample, 18 patients were in some form of employment, three were in the education system, three were not participating in any work or education activities and one was retired. In one year, the average total cost per patient for paediatric CD was \$14,375 with direct and indirect costs comprising \$12,583 and \$1792, respectively (Figure 1).

Figure 1. Overall cost of perianal and paediatric CD divided into outpatient, hospital and indirect costs



The most significant direct costs were inpatient costs (Figure 2) followed by pharmaceutical costs (Figure 3 and 4). Foregone productivity as a result of parental absenteeism from work was the greatest indirect cost (Figure 5). The children had an average of 21 days off school during the year.

Figure 2. Outpatient cost of perianal and paediatric CD showing the components that made up the total cost

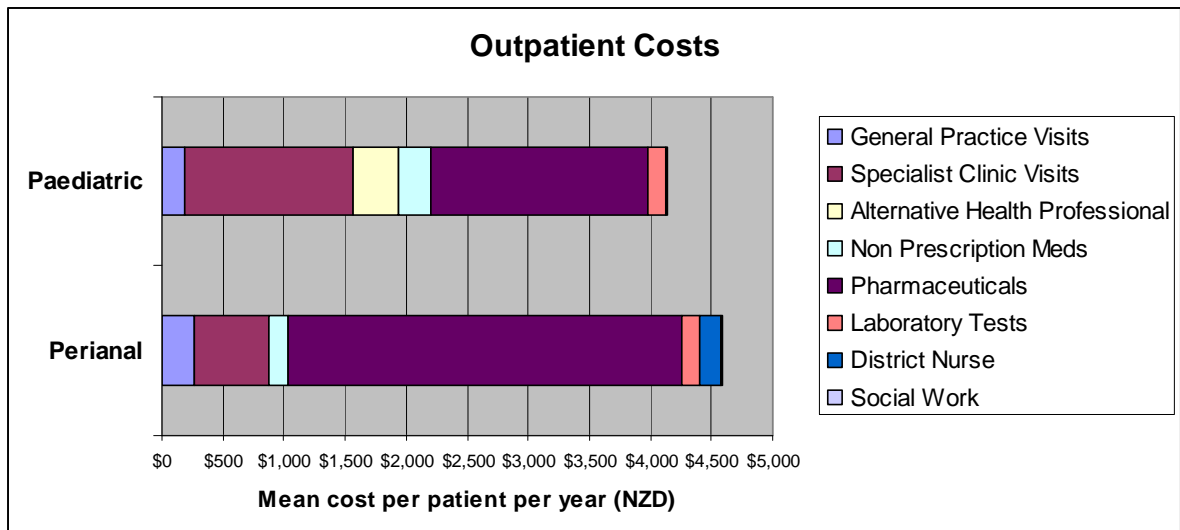


Figure 3. Hospital cost of perianal and paediatric CD showing the components that made up the total cost

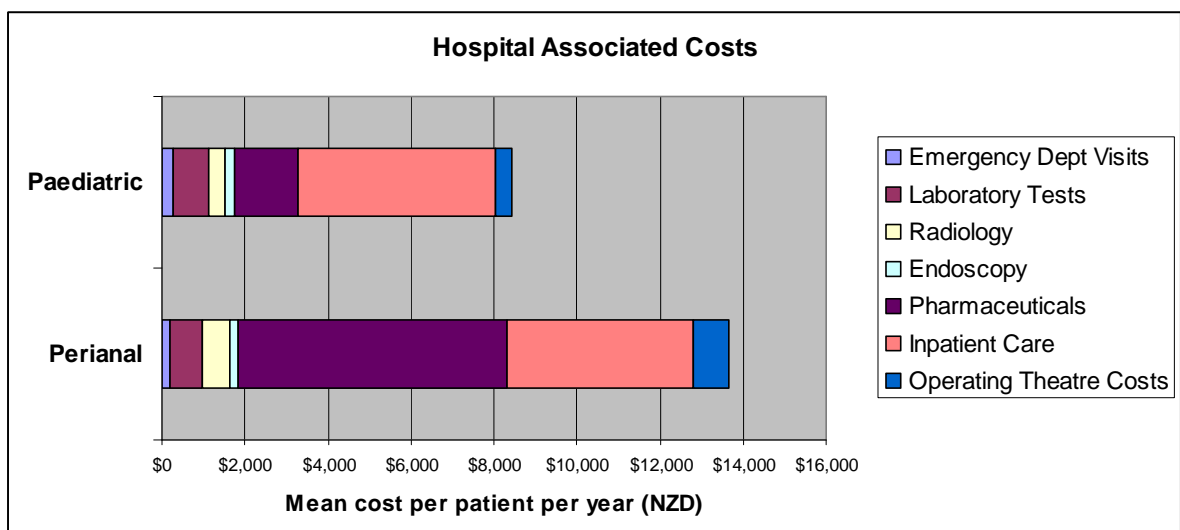


Figure 4. Cost of perianal and paediatric CD with comparison of hospital and pharmaceutical cost

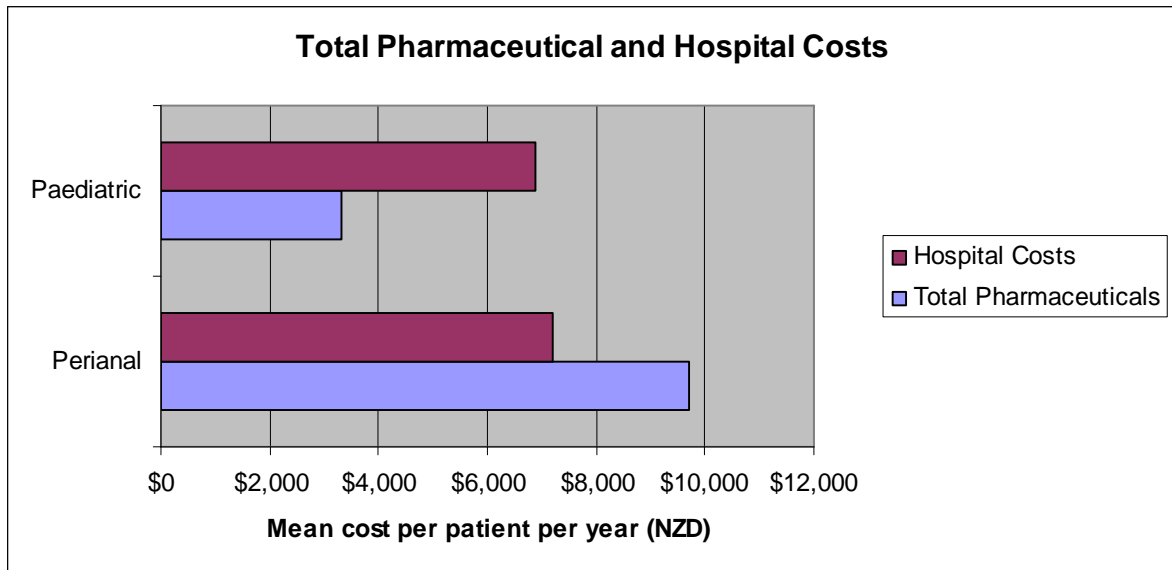
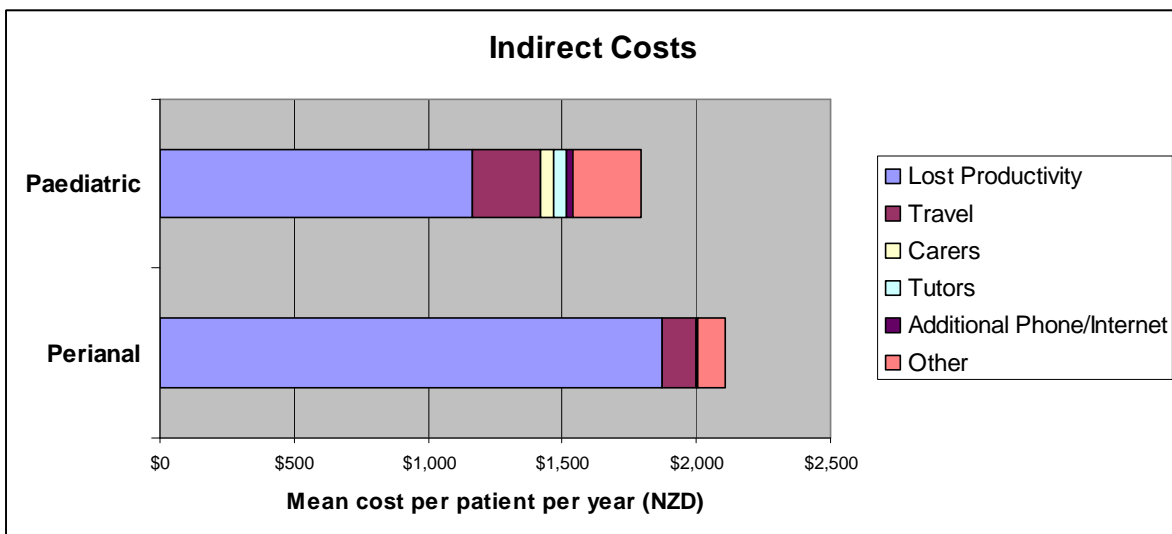


Figure 5. Indirect costs of perianal and paediatric CD showing the components that made up the total cost



The average total costs per patient for perianal CD were \$20,366 with direct and indirect costs comprising \$18,261 and \$2,105, respectively (Figure 1). Eight of the 25 perianal CD patients (32%) received anti-TNF therapy during the study period, The most significant costs were pharmaceuticals followed by inpatient costs. This was highlighted when the total pharmaceutical bill was compared to other hospital costs (Figure 4).

Anti TNF α medication made up 61% of the pharmaceutical costs for perianal patients. The greatest indirect cost was patient and immediate family absenteeism from work (Figure 5).

Extrapolating these data across New Zealand, the total cost of paediatric and perianal CD in one year is estimated to be at least \$25.9 million and \$36.7 million, respectively.

Discussion

These results demonstrate that both paediatric and perianal Crohn's disease are high-cost disorders. No previous studies in New Zealand or internationally have documented the costs created specifically by these specific groups of patients.

Several studies that considered IBD overall found that CD was associated with greater cost than ulcerative colitis.^{6,13,14} A number of studies have also looked specifically at the cost of CD overall.^{8,9,11,12,15} The heterogeneous nature of these studies in terms of methodology, varying costs in different health care systems and the differential effects of inflation since the period of the study makes direct comparison with the present data difficult.

Juan et al reported the annual cost per patient in a Spanish cohort in 2003 was €6,808 with €2,104 from direct costs and €4,704 from indirect costs.¹² Extrapolating data reported in 2009 by Mesterton et al the average annual cost of CD in Swedish patients was €9,400¹⁵ (approximately 16,240 New Zealand dollars at current exchange rates). Hence this group found comparable figures and also noted that increased cost was predicted by increased severity as measured by the Harvey-Bradshaw index.

While the patients in the present study were not stratified for severity, perianal disease itself is a predictor of severe disease and this may explain the slightly higher direct costs reported here. This is supported by the previous finding that the presence of fistulae doubled the costs of care.¹⁵ Patients recruited from tertiary referral centres, as in the present study, also tend to have more severe disease and this too could have contributed to the higher direct costs reported in this study. This fact and the small sample size in the present study mean some caution should be exercised in interpreting the results of this study when extrapolated to the perianal and paediatric CD population across New Zealand.

In contrast to the present analysis, both the previous European studies determined a higher relative contribution from indirect compared with direct costs. This may reflect the method of estimation of indirect costs. As the patients (or carers) in the present study were asked to estimate the absenteeism over the previous year, an element of recall bias may be present. In addition, no allowance for lost productivity while at work due to the disease (presenteeism) was made in this study.

In some cases parents of CD patients admitted to the use of flexible work or leave arrangements, for example working from home or being able work late to make up time off. There were several cases where parents were only able to work part-time and some not at all that were not included as lost productivity in this analysis as the lost productivity was only partly attributable to CD. Other reasons contributing to the reduced productivity included having more than one child with a sickness and less requirement to work due to their spouse having adequate income.

The patients were not asked if they were working part-time as a result of their disease. This potentially could have increased the value of their lost productivity. There was an economic recession for the majority of 2009 that could have contributed to the reduced work hours; therefore it does not seem appropriate to assume working part-time was a result of their illness. Altogether, these factors likely underestimate the indirect costs associated with CD in the current study.

These issues highlight the fact that there is some controversy in the literature in regards to the best approach to use when estimating indirect costs. The human capital-cost method is recommended over other approaches by Liljas²¹ and Johannesson²² for cost studies from a societal perspective.

These authors propose it is the most consistent with economic theory, therefore, indirect costs were estimated using the human capital method in this study, consistent with the approach taken in other recent studies.^{14,15} Despite this, the lower proportion of indirect costs found here could indicate the total cost has been underestimated in the present study hence these cost figures should be considered as a minimum.

Studies of this nature do not capture other effects of a chronic disease on productivity because the estimation of indirect costs is calculated based on the patients current gross wage rate. However, this does not take into account the wage rate the patient could have realised had they not been diagnosed with CD. Given that many patients with CD are diagnosed before or during the second or third decades of life, their disease may have potential life-long impact on educational achievement, career prospects and earning potential consequent to disrupted education and work.

Paediatric CD patients in the present study had an average of 21 days absent from school in the previous year consistent with levels of absenteeism documented in two previous case control studies.^{23,24} Of note while the cases in these two studies showed significantly greater absenteeism than controls, decreased ability to present for exams and some degree of discrimination from teachers, there was no difference in level of educational achievement between cases and controls. Despite this, forty seven percent of respondents to a survey conducted in Germany felt that CD had interfered with their career prospects.¹⁴ A separate study revealed 30% of CD patients concealed their diagnosis from their employers.²⁴ Hence more subtle loss of productivity costs are likely to exist which have not been measured in this study.

In addition to both the direct and indirect costs discussed above, this study has not included intangible elements associated with the burden of disease, or loss of wellbeing, associated with CD. A more global assessment of the economic impact of CD would include such costs. Recent economic theory has allowed integration of these concepts into cost calculations.

Using willingness to pay measures of mortality and morbidity associated with disease, economists have developed estimates of the Value of a Statistical Life. This can be used to attach a monetary value to the non-financially derived Disability Adjusted Life Year concept and thereby derive a financial cost associated with the burden of disease.²⁵ This was not attempted in the present study where the aim was to produce estimates for future cost-benefit analysis that will utilise the direct costs associated with CD.

The principal direct costs incurred were for inpatient care and pharmaceuticals. In the adult group with perianal disease, the total pharmaceutical bill was greater than the other hospital associated costs (Figure 4). In several early studies, hospital costs were found to make up a higher proportion of the total cost of care for IBD than pharmaceutical costs.^{7,9,26} However, these three studies were performed prior to the widespread use of anti-TNF α agents.

As the present study was performed in the era of biologic treatments, the proportionally increased pharmaceutical costs are likely to represent a genuine finding and are consistent with the other most recent cost study from Sweden.¹⁵ Anti-TNF α therapy is expensive but it may prove cost-effective if it leads to a reduction in hospitalisation and the high costs associated with this.

This study was not designed to assess this. However, Hay and Hay²⁷ have previously created a model for the cost-effectiveness of expensive drug therapy in CD. Their model demonstrated that if a new drug reduced other costs such as hospitalisation by 20% then, despite a doubling of the pharmaceutical bill, the overall cost of care would reduce by 13%.²⁹

Surgery was another significant direct cost associated with CD in the present study. As patients undergoing surgery tend to more severely affected, direct comparisons of efficacy with medical treatments for similar clinical states are limited. However, Silverstein et al⁸ found in a Markov analysis that despite higher costs for surgery, post surgical remission was longer than for patients treated medically. These authors concluded that surgery may therefore be a more cost-effective option in selected cases.⁸

Laparoscopic surgery is now increasingly used in the treatment of IBD. Short term advantages of laparoscopic surgery include improved pulmonary function, decreased ileus, shorter hospital stay and improved cosmesis.²⁸ In the longer term, there does not appear to be any difference in recurrence rates compared with open surgery.²⁹ While operative time and intraoperative expenses are increased, total hospital costs are reduced with the decreased length of stay.²⁸ Hence, any future cost-benefit analysis will need to allow for the impact of laparoscopic surgery.

This study has confirmed that paediatric and perianal CD patients consume significant health resources. Prior to this, no studies in New Zealand have estimated the cost of IBD and no international studies have estimated the indirect costs of paediatric Crohn's disease. With the advent of increasingly costly and effective medical therapies and evolving surgical treatment, this research will provide valuable information for future cost-effectiveness studies.

Competing interests: None declared.

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