Title:

Cost evaluation of out-of-country care for patients with eating disorders in Ontario: a populationbased study

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None declared.

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intended or should be inferred. Parts of this material are based on data and information compiled and provided by the Canadian Institute for Health Information (CIHI). However, the analyses, conclusions, opinions, and statements expressed herein are those of the authors, and not necessarily those of CIHI.

Abstract

Background: Eating disorders, specifically anorexia nervosa, bulimia nervosa and eating disorder not otherwise specified, represent a substantial burden to the health care system. To address this, the Ontario government made an investment in eating disorders treatment in 2007. We soughtOur goal was to estimate the economic burden of patients who received specialised out-of-country eating disorder care.

Method: We conducted a cost-of-illness study evaluating-all health care costs among individuals in Ontario who received specialised out-of-country eating disorder care from 2003 to 2011, from the public third-party payer perspective. Using linked administrative health care-databases, we estimated net costs of eating disorders for two patient groups – patients who received specialised out-of-country and in-province inpatient care (n=160), and patients who received specialised out-of-country inpatient care only (n=126).

Results: Patients approved for specialised out-of-country eating disorder care were mostly young females from high-income, urban neighbourhoods. Total annual net costs varied by yearanually, and were higher for patients treated out-of-country and in province (\$11.0 million before 2007, \$6.5 million after) compared to those treated out-of-country alone (\$5.0 million before 2007, \$2.0 million after). The main cost drivers were out-of-country care and physician services.

Interpretation: Costs associated with eating disorder care are large and represent a substantial economic burden to the Ontario health care system. Given the high costs of out-of-country care, there may be opportunity to redirect these funds to increase capacity and expertise for eating disorder treatment within Ontario; this may potentially reduce patient and systems costs, improve patient access and continuity of care, and create new jobs in the province<u>Ontario</u>.

Background

Approximately 3% of women are affected by an eating disorder (ED) in their lifetime (1). These disorders are associated with high levels of comorbid psychopathology, such as mood and anxiety disorders, post-traumatic stress disorder, and substance use disorders (2). In addition, anorexia nervosa has the highest mortality rate of any psychiatric disorder (3). These consequences are reflected in high costs of care (2). Prior research suggests that age-adjusted treatment costs for anorexia nervosa and bulimia nervosa are comparable to those for schizophrenia (4). Other work has shown that eating disorders are associated with high direct and indirect health care costs (5). Yet, precise cost estimates are currently lacking.

Eating disorders represent a substantial economic burden to the Ontario health care system; yet, precise cost estimates are currently lacking. To address this burden, the Ontario government made an investment in eating disorders treatment within the province in 2007. In addition to funding treatment within the province, the Ontario Ministry of Health and Long-term Care spends funds to send patients to the United States to receive specialised eating disorder treatment for complex cases, which cannot be treated in-province due to lack of resources, or when wait times for in-province treatment are deemed unacceptably long. This care is specifically for patients with serious, treatment-interfering post-traumatic stress disorder or substance use disorders concurrent with an ED. Furthermore, some patients may receive additional specialised care in-province; this is typically based on the care protocol defined by the specialist.

To be eligible for out-of-country care, patients must be diagnosed with an eating disorder by a specialist at one of Ontario's funded inpatient programs for eating disorders. The specialist submits the application on behalf of the patient to the Ontario Out-of-Country Health Coverage Program, which makes decisions regarding coverage for medical and hospital services performed outside of Canada. This program reviews these applications, undertaking research and/or consults with experts as required, and decides whether applicants are approved; these decisions are largely based on disease severity.

It may be possible to reinvest funds spent on out-of-country eating disorder care to develop and augment programs within Ontario. This could potentially reduce overall costs, including out-of-pocket expenses, improve patients' experiences and create additional jobs in Ontario (6). Currently there is no protocol for transitional or follow-up care upon a patient's return to Ontario. Instead, follow-up care is provided by the patient's current care provider. Given the high risk of relapse during this transition period, disjointed care in different jurisdictions may contribute to poorer patient outcomes and additional treatment costs, although these data have not been collected.

The purpose of this study was to estimate the total costs of patients with an eating disorder who received *specialised out-of-country care*, from the perspective of the public third-party payer . A careful evaluation of these costs may be useful to determine whether developing further specialised in-province services is financially justified. These findings will have significant policy implications for the Ontario government and other jurisdictions with similar out-of-country programs, such as British Columbia.

Method

Study design and setting

We conducted a cost of illness study. Our patient cohort included all individuals with an eating disorder covered under the Ontario Health Insurance Plan (OHIP) who applied and received specialised out-of-country care from April 1 2000 to March 31 2012. This cohort was divided into two groups, according to whether patients received additional specialized in-province inpatient care or not. We matched each patient to a set of control subjects with no history of an eating disorder to estimate net costs.

During our analysis period, namely in 2007, the Ontario government made a significant investment in eating disorders treatment within the province, which led to a decrease in the number of out-of-country applications.

Data Sources

Data on all applicants who received out-of-country care for eating disorders were obtained from the Ministry of Health and Long-term Care's out-of-country program database, which includes all records on out-of-country care. These data included information on the status of the application (approved, denied, cancelled, awaiting information, pending), name of the out-ofcountry provider, total amount billed and paid by the province, service start and end dates, and the condition for which care was provided (eating disorder, substance abuse, other psychiatric disorders). Further information on the out-of-country care program can be found elsewhere (7).

In addition, we used administrative health care databases available through the Institute for Clinical Evaluative Sciences in Toronto, Ontario, to obtain data on all health services provided within province and covered by the Ministry of Health and Long-term Care (only). These databases have been validated and described in the literature (8), and have been used previously for costing analyses in Ontario (9-11). A full description of each database can be found in the Appendix. These databases were linked using unique encoded identifiers and analyzed at the Institute for Clinical Evaluative Sciences. This study was approved by the Research Ethics Board at Sunnybrook Health Sciences Centre, Toronto, Canada.

Patient Cohort

We included patients diagnosed with anorexia nervosa, bulimia nervosa and eating disorder not otherwise specified (International Classification of Diseases (ICD)-9 codes 307.1, 307.50, 307.51; ICD-10 codes F50.0-F50.3, F50.8, F50.9; and Diagnostic and Statistical Manual of Mental Disorders, 4th edition codes 307.1, 307.50, 307.51). We excluded individuals diagnosed with Prader-Willi syndrome (ICD-9 759.81 code; no specific ICD-10 code was available), a rare genetic disorder associated with excessive eating and obesity; individuals who did not reside in Ontario at time of diagnosis; those who did not have a valid OHIP number during the analysis period; and patients with missing information on age and/or sex. We also excluded patients who first applied for out-of-country care outside the study period, those who were denied or cancelled out-of-country care, and patients with missing information on the out-of-country application status. Our final cohort included all patients who received specialised out-of-country eating disorder treatment (and thus similar disease severity). This cohort was further divided into two groups, differing only in terms of whether they received specialised in-province care or not.

Analysis

Socio-demographic and clinical characteristics

We compared both patient groups on socio-demographic characteristics, such as age, sex, neighbourhood income quintile (at the Census tract level) and urban/rural residence, and comorbidity, such as diabetes, asthma and hypertension, which were defined using validated algorithms (12-14).

Estimation of costs of care

Costs for out-of-country care were available in the OHIP claims database (fee codes H700, H820, H830 and H850). Costs, in American dollars, were converted to Canadian dollars using monthly exchange rates, and adjusted to 2012 values.

To estimate direct costs for in-province care, we used a costing algorithm (8) available at the Institute for Clinical Evaluative Sciences. Complete cost data were not available for the entire analysis period, and therefore estimated from 2003 to 2011 only. These included costs of inpatient hospitalizations (psychiatric and non-psychiatric), emergency department visits, other ambulatory care, physician visits, lab tests, outpatient prescription drugs covered under the public provincial drug program, complex continuing care, long-term care, home care, rehabilitation and assistive devices (see Appendix). All costs were adjusted to 2012 Canadian dollars.

Page 11 of 52

Standard costing methodology employed a bottom-up (micro-costing) approach to cost services at the individual level (8). This approach made use of individual episodes of care or utilization in the health system and attached prices (or costs or amounts paid by the Ministry of Health and Long-term Care) to each one. A top-down approach, which allocates corporate aggregate (institutional) costs to individual visits or to cases or episodes of care, was applied when individual unit costs were not available (for example, for institutional care settings) (8). Further details on the costing methodology can be found elsewhere (8).

Many cost-of-illness studies have been criticized for attributing all costs to a single disorder, despite most patients having other health problems that also require care (15). Therefore, we employed the 'net cost' method to obtain an estimate of the cost of an eating disorder. This method consists of matching patients who have an eating disorder to patients who do not (control subjects) on variables associated with resource use, and subtracting the costs incurred by the former from those incurred by the latter (16-18). Patients with an eating disorder were matched to control subjects on age, sex and neighbourhood income quintile on a 1:4 ratio, for a total of 1,144 control subjects. The net cost method was only applied to costs incurred in Ontario; out-of-country costs were over and above net costs.

Results

We started with all out-of-country applicants (n=1,854). We excluded those who did not meet our eating disorder care definition (n=1,295), those who first applied outside our study window (n=62), and those who were denied out-of-country care, whose care was cancelled or had missing information (n=211) (Figure 1). Patients who were approved care were younger than those denied care (median age of 18 versus 23, respectively), but similar on all other patient characteristics (results available upon request). This was also the case for those with cancelled care and missing information. Our final cohort included all patients who first applied and were approved for out-of-country care between fiscal years 2000 and 2012 (n=286), which was further divided into two patient groups – those who did not receive additional specialised in-province inpatient care during the study period (n=126), and those who did (n=160) .

Socio-demographic and clinical characteristics

Patient groups differed mostly in terms of age and sex, but were similar on all other sociodemographic and clinical characteristics (Table 1). There was a slightly higher proportion of female patients among those who received specialised out-of-country and in-province inpatient care (98% vs. 91%). Patients who received both specialised out-of-country and in-province inpatient care were roughly 4 years younger than those who received specialised out-of-country care only. Most patients were from high-income, urban neighbourhoods. Less than 7% had diabetes or hypertension; a quarter of patients had asthma.

Estimation of costs

The number of funded out-of-country applications increased from 2000 to 2006, and decreased thereafter (Figure 2). Total annual net costs were estimated from 2003 to 2011; these varied accordingly with the number of funded applications approved each year. Overall, there was an inverted u-shape curve in costs over time for both patient groups (Figures 3a and b).

Total annual costs per year were approximately \$5 million before 2007 and \$2 million thereafter for patients who received specialised out-of-country inpatient care only (Figure 3a), and \$11 million before 2007 and \$6.5 million after 2007 for patients who received both specialised out-of-country and in-province inpatient care (Figure 3b).

In-province costs for patients who receive specialised out-of-country inpatient care only were just under \$3 million per year until 2007, and decreased to around \$1.5 million per year thereafter (Figure 3a). Out-of-country costs for this group were slightly lower than in-province costs and followed a similar temporal pattern (\$2.3 million before 2007, \$0.5 million after) (Figure 3a).

In-province costs were substantially higher for patients who received both specialised out-ofcountry and in-province care – just under \$7 million annually until 2007 (except in 2003), and about \$5 million per year afterwards (3) (Figure 3b). Out-of-country costs were slightly lower, but followed a similar pattern (around \$4 million in the first years and about \$1.5 million in the later years) (Figure 3b).

On average, out-of-country and physician costs were the largest contributors to the overall cost for both patient groups. Costs of inpatient hospitalizations were also important cost drivers, mainly for patients who received specialised in-province care. Costs with other health services represented a small percentage ($\leq 5\%$) of the overall cost for all patients (not shown).

Interpretation

Patients with eating disorders have high health care utilization and costs (5). The management of health care costs and avoidance of out-of-country care are a high priority for Ontario; however, little is known about the costs of treating patients with eating disorders. We estimated total *net* costs for two patient groups that were approved for and received specialised out-of-country care. Most patients were young, female and from high-income, urban neighbourhoods. Total net costs were higher for patients who received both specialised out-of-country and in-province inpatient care, suggesting these patients are complex cases. The major contributors to the overall cost were out-of-country care and physician services, and to a lesser degree, in-province inpatient hospitalizations. Although we observed a decrease in costs in 2007, due to significant changes in the out-of-country care program (19), the costs of this program remained a relatively large component of the economic burden of eating disorders to the province. A comprehensive literature review identified only six studies worldwide on health services use

A comprehensive interature review identified only six studies worldwide on health services use and costs among individuals with an eating disorder. Most studies were dated and results varied greatly (5). In the UK, the direct health care system cost of anorexia nervosa was estimated at £4.2 million in 1990 (20); in Germany, the cost was €65 million for anorexia nervosa and €10 million for bulimia nervosa in 1998 (21). In Australia health care costs of EDs were Aus\$22 million in 1993/1994 (22). Other cost studies that focused mostly on inpatient care reported highly variable estimates. More recent work from the UK has shown that total treatment costs to the National Health Service may vary between £3.9 billion and £ 4.6 billion (23). Despite high health care utilization among these individuals, results in most studies were likely gross underestimates of the full magnitude of the economic burden as only inpatient costs were captured (21), or results were based on limited cost data (4). Furthermore, only one study

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reported specific information on service use of eating disorder not otherwise specified (4), the most common diagnosis encountered in clinical practice (24). We did not find any studies that estimated net costs or that examined an out-of-country program; thus, our estimates are not directly comparable to previous work. Nonetheless, we found that our estimates were quite substantial even for a very small group of patients (n=286), with out-of-country costs representing a small proportion of the Ontario health care budget (0.005% in 2012) (25). Evidence suggests that average daily treatment costs in-province are lower than those out-of-country (\$918 versus \$1,285, respectively) (26).

Complete cost estimates for patients with eating disorders are required to determine costeffective treatment options (5); yet, these are generally lacking in the literature (27). Few studies have examined the cost-effectiveness of eating disorder-related interventions (28. Thus, our cost estimates may be useful inputs for researchers undertaking economic evaluations. Furthermore, they may help inform policy discussions in Ontario and other jurisdictions with similar funded out-of-country care programs, such as British Columbia.

The ongoing need for out-of-country care in Ontario likely reflects inability to access care and/or to adequately manage complexity within eating disorder cases. Overall, there is a need for greater variability and availability of comprehensive care across the province, ideally aligned with patients' need; in other words, regionalised eating disorder care with higher levels of coordination, perhaps like the Ontario cancer care system. An ideal model of care might provide availability of basic services across the province, but have expertise for more complex cases centralised in large urban centres, like in the Netherlands (29). This alignment could potentially

improve care transitions and follow-up care, which are crucial given the high rates of relapse among these patients (30,31).

Strengths and limitations

Our study examined an important policy issue within the Ontario health care context, and addressed a gap in the literature. Thus far, even internationally, there have been little data and research on the costs of eating disorders (4,32). Our work estimated *total* net costs for a specific group of patients with an eating disorder; most studies have examined only inpatient and outpatient costs for subgroups of patients. Furthermore, our study is the only one we are aware of that has estimated *net* costs. Contrary to other studies, our cost estimates represents the direct cost of eating disorders themselves.

We were not able to account for addiction-related health care costs from community-based agencies, where a large part of addiction treatment is provided (33). In addition, we were not able to capture OHIP-funded outpatient eating disorder treatment, other than that provided directly by psychiatrists. We were only able to include costs of outpatient prescription drugs for patients covered by the public provincial drug program (for this cohort, individuals receiving social assistance). Thus, our cost values are likely an underestimate. We also had limited ability to examine subpopulations of interest, including sex, type of eating disorder, and psychiatric comorbidity, and only examined patients who were approved for out-of-country care. Finally, we estimated direct costs incurred by the health care system and did not estimate other relevant costs, such as those covered under private health care plans, out-of-pocket costs, costs related to

inability to work (lost productivity) for patients and family members, and costs with death, which are also lacking in the literature. Future research should examine this.

Conclusion

Costs of care associated with eating disorders are large and represent a substantial financial burden to the health care system. We found that patients treated out-of-country and in Ontario had higher costs than those treated out-of-country alone. Furthermore, we found that the main cost drivers were out-of-country care and physician services. Given the high costs associated with out-of-country care, there may be opportunity to redirect these funds to increase capacity and expertise for eating disorder treatment within Ontario; this could reduce overall costs of care, improve patients' experiences and create new jobs in the province. This would require additional analyses around the estimation of cost-savings to the system in the long-run and the implications for patient outcomes. Moreover, there may be scope to reorganise the eating disorder care system in the province to achieve these goals.

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Competing interests:

None declared.

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Abstract

Background: Eating disorders, specifically anorexia nervosa, bulimia nervosa and eating disorder not otherwise specified, represent a substantial burden to the health care system. To address this, the Ontario government made an investment in eating disorders treatment in 2007. Our goal was to estimate the economic burden of patients who received specialised out-of-country eating disorder care.

Method: We conducted a cost-of-illness study evaluating health care costs among individuals in Ontario who received specialised out-of-country eating disorder care from 2003 to 2011, from the public third-party payer perspective. Using linked administrative databases, we estimated net costs of eating disorders for two patient groups – patients who received specialised out-of-country and in-province inpatient care (n=160), and patients who received specialised out-of-country inpatient care only (n=126).

Results: Patients approved for specialised out-of-country eating disorder care were mostly young females from high-income, urban neighbourhoods. Total annual net costs varied anually, and were higher for patients treated out-of-country and in province (\$11.0 million before 2007, \$6.5 million after) compared to those treated out-of-country alone (\$5.0 million before 2007, \$2.0 million after). The main cost drivers were out-of-country care and physician services. **Interpretation:** Costs associated with eating disorder care represent a substantial economic burden to the Ontario health care system. Given the high costs of out-of-country care, there may be opportunity to redirect these funds to increase capacity and expertise for eating disorder treatment within Ontario; this may potentially reduce patient and systems costs, improve patient access and continuity of care, and create new jobs in Ontario.

Background

Approximately 3% of women are affected by an eating disorder (ED) in their lifetime (1). These disorders are associated with high levels of comorbid psychopathology, such as mood and anxiety disorders, post-traumatic stress disorder, and substance use disorders (2). In addition, anorexia nervosa has the highest mortality rate of any psychiatric disorder (3). These consequences are reflected in high costs of care (2). Prior research suggests that age-adjusted treatment costs for anorexia nervosa and bulimia nervosa are comparable to those for schizophrenia (4). Other work has shown that eating disorders are associated with high direct and indirect health care costs (5). Yet, precise cost estimates are currently lacking.

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To be eligible for out-of-country care, patients must be diagnosed with an eating disorder by a specialist at one of Ontario's funded inpatient programs for eating disorders. The specialist

submits the application on behalf of the patient to the Ontario Out-of-Country Health Coverage Program, which makes decisions regarding coverage for medical and hospital services performed outside of Canada. This program reviews these applications, undertaking research and/or consults with experts as required, and decides whether applicants are approved; these decisions are largely based on disease severity.

It may be possible to reinvest funds spent on out-of-country eating disorder care to develop and augment programs within Ontario. This could potentially reduce overall costs, including out-of-pocket expenses, improve patients' experiences and create additional jobs in Ontario (6). Currently there is no protocol for transitional or follow-up care upon a patient's return to Ontario. Instead, follow-up care is provided by the patient's current care provider. Given the high risk of relapse during this transition period, disjointed care in different jurisdictions may contribute to poorer patient outcomes and additional treatment costs, although these data have not been collected.

The purpose of this study was to estimate the total costs of patients with an eating disorder who received *specialised out-of-country care*, from the perspective of the public third-party payer . A careful evaluation of these costs may be useful to determine whether developing further specialised in-province services is financially justified. These findings will have significant policy implications for the Ontario government and other jurisdictions with similar out-of-country programs, such as British Columbia.

Method

Study design and setting

We conducted a cost of illness study. Our patient cohort included all individuals with an eating disorder covered under the Ontario Health Insurance Plan (OHIP) who applied and received specialised out-of-country care from April 1 2000 to March 31 2012. This cohort was divided into two groups, according to whether patients received additional specialized in-province inpatient care or not. We matched each patient to a set of control subjects with no history of an eating disorder to estimate net costs.

During our analysis period, namely in 2007, the Ontario government made a significant investment in eating disorders treatment within the province, which led to a decrease in the number of out-of-country applications.

Data Sources

Data on all applicants who received out-of-country care for eating disorders were obtained from the Ministry of Health and Long-term Care's out-of-country program database, which includes all records on out-of-country care. These data included information on the status of the application (approved, denied, cancelled, awaiting information, pending), name of the out-ofcountry provider, total amount billed and paid by the province, service start and end dates, and the condition for which care was provided (eating disorder, substance abuse, other psychiatric disorders). Further information on the out-of-country care program can be found elsewhere (7).

In addition, we used administrative health care databases available through the Institute for Clinical Evaluative Sciences in Toronto, Ontario, to obtain data on all health services

provided within province and covered by the Ministry of Health and Long-term Care (only). These databases have been validated and described in the literature (8), and have been used previously for costing analyses in Ontario (9-11). A full description of each database can be found in the Appendix. These databases were linked using unique encoded identifiers and analyzed at the Institute for Clinical Evaluative Sciences. This study was approved by the Research Ethics Board at Sunnybrook Health Sciences Centre, Toronto, Canada.

Patient Cohort

We included patients diagnosed with anorexia nervosa, bulimia nervosa and eating disorder not otherwise specified (International Classification of Diseases (ICD)-9 codes 307.1, 307.50, 307.51; ICD-10 codes F50.0-F50.3, F50.8, F50.9; and Diagnostic and Statistical Manual of Mental Disorders, 4th edition codes 307.1, 307.50, 307.51). We excluded individuals diagnosed with Prader-Willi syndrome (ICD-9 759.81 code; no specific ICD-10 code was available), a rare genetic disorder associated with excessive eating and obesity; individuals who did not reside in Ontario at time of diagnosis; those who did not have a valid OHIP number during the analysis period; and patients with missing information on age and/or sex. We also excluded patients who first applied for out-of-country care outside the study period, those who were denied or cancelled out-of-country care, and patients with missing information on the out-of-country application status. Our final cohort included all patients who received specialised out-of-country eating disorder treatment (and thus similar disease severity). This cohort was further divided into two groups, differing only in terms of whether they received specialised in-province care or not.

Analysis

Socio-demographic and clinical characteristics

We compared both patient groups on socio-demographic characteristics, such as age, sex, neighbourhood income quintile (at the Census tract level) and urban/rural residence, and comorbidity, such as diabetes, asthma and hypertension, which were defined using validated algorithms (12-14).

Estimation of costs of care

Costs for out-of-country care were available in the OHIP claims database (fee codes H700, H820, H830 and H850). Costs, in American dollars, were converted to Canadian dollars using monthly exchange rates, and adjusted to 2012 values.

To estimate direct costs for in-province care, we used a costing algorithm (8) available at the Institute for Clinical Evaluative Sciences. Complete cost data were not available for the entire analysis period, and therefore estimated from 2003 to 2011 only. These included costs of inpatient hospitalizations (psychiatric and non-psychiatric), emergency department visits, other ambulatory care, physician visits, lab tests, outpatient prescription drugs covered under the public provincial drug program, complex continuing care, long-term care, home care, rehabilitation and assistive devices (see Appendix). All costs were adjusted to 2012 Canadian dollars.

Standard costing methodology employed a bottom-up (micro-costing) approach to cost services at the individual level (8). This approach made use of individual episodes of care or utilization in the health system and attached prices (or costs or amounts paid by the Ministry of Health and

 Long-term Care) to each one. A top-down approach, which allocates corporate aggregate (institutional) costs to individual visits or to cases or episodes of care, was applied when individual unit costs were not available (for example, for institutional care settings) (8). Further details on the costing methodology can be found elsewhere (8).

Many cost-of-illness studies have been criticized for attributing all costs to a single disorder, despite most patients having other health problems that also require care (15). Therefore, we employed the 'net cost' method to obtain an estimate of the cost of an eating disorder. This method consists of matching patients who have an eating disorder to patients who do not (control subjects) on variables associated with resource use, and subtracting the costs incurred by the former from those incurred by the latter (16-18). Patients with an eating disorder were matched to control subjects on age, sex and neighbourhood income quintile on a 1:4 ratio, for a total of 1,144 control subjects. The net cost method was only applied to costs incurred in Ontario; out-of-country costs were over and above net costs.

Results

We started with all out-of-country applicants (n=1,854). We excluded those who did not meet our eating disorder care definition (n=1,295), those who first applied outside our study window (n=62), and those who were denied out-of-country care, whose care was cancelled or had missing information (n=211) (Figure 1). Patients who were approved care were younger than those denied care (median age of 18 versus 23, respectively), but similar on all other patient characteristics (results available upon request). This was also the case for those with cancelled care and missing information. Our final cohort included all patients who first applied and were approved for out-of-country care between fiscal years 2000 and 2012 (n=286), which was further divided into two patient groups – those who did not receive additional specialised in-province inpatient care during the study period (n=126), and those who did (n=160) .

Socio-demographic and clinical characteristics

Patient groups differed mostly in terms of age and sex, but were similar on all other sociodemographic and clinical characteristics (Table 1). There was a slightly higher proportion of female patients among those who received specialised out-of-country and in-province inpatient care (98% vs. 91%). Patients who received both specialised out-of-country and in-province inpatient care were roughly 4 years younger than those who received specialised out-of-country care only. Most patients were from high-income, urban neighbourhoods. Less than 7% had diabetes or hypertension; a quarter of patients had asthma.

Estimation of costs

The number of funded out-of-country applications increased from 2000 to 2006, and decreased thereafter (Figure 2). Total annual net costs were estimated from 2003 to 2011; these varied accordingly with the number of funded applications approved each year. Overall, there was an inverted u-shape curve in costs over time for both patient groups (Figures 3a and b).

Total annual costs per year were approximately \$5 million before 2007 and \$2 million thereafter for patients who received specialised out-of-country inpatient care only (Figure 3a), and \$11 million before 2007 and \$6.5 million after 2007 for patients who received both specialised out-of-country and in-province inpatient care (Figure 3b).

In-province costs for patients who receive specialised out-of-country inpatient care only were just under \$3 million per year until 2007, and decreased to around \$1.5 million per year thereafter (Figure 3a). Out-of-country costs for this group were slightly lower than in-province costs and followed a similar temporal pattern (\$2.3 million before 2007, \$0.5 million after) (Figure 3a).

In-province costs were substantially higher for patients who received both specialised out-ofcountry and in-province care – just under \$7 million annually until 2007 (except in 2003), and about \$5 million per year afterwards (3) (Figure 3b). Out-of-country costs were slightly lower, but followed a similar pattern (around \$4 million in the first years and about \$1.5 million in the later years) (Figure 3b).

On average, out-of-country and physician costs were the largest contributors to the overall cost for both patient groups. Costs of inpatient hospitalizations were also important cost drivers, mainly for patients who received specialised in-province care. Costs with other health services represented a small percentage ($\leq 5\%$) of the overall cost for all patients (not shown).

Interpretation

Patients with eating disorders have high health care utilization and costs (5). The management of health care costs and avoidance of out-of-country care are a high priority for Ontario; however, little is known about the costs of treating patients with eating disorders. We estimated total *net* costs for two patient groups that were approved for and received specialised out-of-

country care. Most patients were young, female and from high-income, urban neighbourhoods. Total net costs were higher for patients who received both specialised out-of-country and inprovince inpatient care, suggesting these patients are complex cases. The major contributors to the overall cost were out-of-country care and physician services, and to a lesser degree, inprovince inpatient hospitalizations. Although we observed a decrease in costs in 2007, due to significant changes in the out-of-country care program (19), the costs of this program remained a relatively large component of the economic burden of eating disorders to the province.

A comprehensive literature review identified only six studies worldwide on health services use and costs among individuals with an eating disorder. Most studies were dated and results varied greatly (5). In the UK, the direct health care system cost of anorexia nervosa was estimated at £4.2 million in 1990 (20); in Germany, the cost was $\notin 65$ million for anorexia nervosa and $\notin 10$ million for bulimia nervosa in 1998 (21). In Australia health care costs of EDs were Aus\$22 million in 1993/1994 (22). Other cost studies that focused mostly on inpatient care reported highly variable estimates. More recent work from the UK has shown that total treatment costs to the National Health Service may vary between £3.9 billion and £ 4.6 billion (23). Despite high health care utilization among these individuals, results in most studies were likely gross underestimates of the full magnitude of the economic burden as only inpatient costs were captured (21), or results were based on limited cost data (4). Furthermore, only one study reported specific information on service use of eating disorder not otherwise specified (4), the most common diagnosis encountered in clinical practice (24). We did not find any studies that estimated net costs or that examined an out-of-country program; thus, our estimates are not directly comparable to previous work. Nonetheless, we found that our estimates were quite

substantial even for a very small group of patients (n=286), with out-of-country costs representing a small proportion of the Ontario health care budget (0.005% in 2012) (25). Evidence suggests that average daily treatment costs in-province are lower than those out-ofcountry (\$918 versus \$1,285, respectively) (26).

Complete cost estimates for patients with eating disorders are required to determine costeffective treatment options (5); yet, these are generally lacking in the literature (27). Few studies have examined the cost-effectiveness of eating disorder-related interventions (28. Thus, our cost estimates may be useful inputs for researchers undertaking economic evaluations. Furthermore, they may help inform policy discussions in Ontario and other jurisdictions with similar funded out-of-country care programs, such as British Columbia.

The ongoing need for out-of-country care in Ontario likely reflects inability to access care and/or to adequately manage complexity within eating disorder cases. Overall, there is a need for greater variability and availability of comprehensive care across the province, ideally aligned with patients' need; in other words, regionalised eating disorder care with higher levels of coordination, perhaps like the Ontario cancer care system. An ideal model of care might provide availability of basic services across the province, but have expertise for more complex cases centralised in large urban centres, like in the Netherlands (29). This alignment could potentially improve care transitions and follow-up care, which are crucial given the high rates of relapse among these patients (30,31).

Strengths and limitations

Our study examined an important policy issue within the Ontario health care context, and addressed a gap in the literature. Thus far, even internationally, there have been little data and research on the costs of eating disorders (4,32). Our work estimated *total* net costs for a specific group of patients with an eating disorder; most studies have examined only inpatient and outpatient costs for subgroups of patients. Furthermore, our study is the only one we are aware of that has estimated *net* costs. Contrary to other studies, our cost estimates represents the direct cost of eating disorders themselves.

We were not able to account for addiction-related health care costs from community-based agencies, where a large part of addiction treatment is provided (33). In addition, we were not able to capture OHIP-funded outpatient eating disorder treatment, other than that provided directly by psychiatrists. We were only able to include costs of outpatient prescription drugs for patients covered by the public provincial drug program (for this cohort, individuals receiving social assistance). Thus, our cost values are likely an underestimate. We also had limited ability to examine subpopulations of interest, including sex, type of eating disorder, and psychiatric comorbidity, and only examined patients who were approved for out-of-country care. Finally, we estimated direct costs incurred by the health care system and did not estimate other relevant costs, such as those covered under private health care plans, out-of-pocket costs, costs related to inability to work (lost productivity) for patients and family members, and costs with death, which are also lacking in the literature. Future research should examine this.

Conclusion

Costs of care associated with eating disorders are large and represent a substantial financial burden to the health care system. We found that patients treated out-of-country and in Ontario had higher costs than those treated out-of-country alone. Furthermore, we found that the main cost drivers were out-of-country care and physician services. Given the high costs associated with out-of-country care, there may be opportunity to redirect these funds to increase capacity and expertise for eating disorder treatment within Ontario; this could reduce overall costs of care, improve patients' experiences and create new jobs in the province. This would require additional analyses around the estimation of cost-savings to the system in the long-run and the implications for patient outcomes. Moreover, there may be scope to reorganise the eating disorder care system ve these goals. in the province to achieve these goals.

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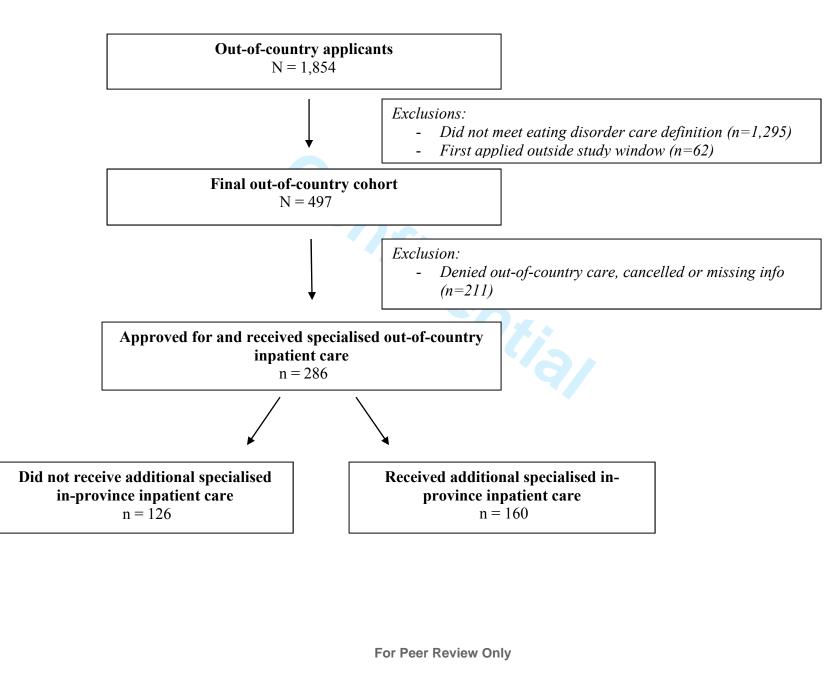
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	Patients who did not receive specialised eating disorder inpatient care in Ontario N=126 (100%)	Patients who received specialised eating disorder inpatient care in Ontario N=160 (100%)	p- value
Sex			
Male	11 (8.7%)	<=5	0.0295
Female	115 (91.3%)	157 (> 98%)	
Age at time of first application			
Mean \pm SD	24.2 ± 10.3	20.3 ± 7.6	0.0003
Median (IQR)	21 (17-28)	17 (16-23)	
Neighbourhood income quintile			
1 (low)	16 (12.7%)	19 (11.9%)	0.9956
2 (medium low)	14 (11.1%)	19 (11.9%)	
3 (medium)	25 (19.8%)	30 (18.8%)	
4 (medium-high)	26 (20.6%)	32 (20.0%)	
5 (high)	45 (35.7%)	60 (37.5%)	
Rural residence	13 (10.3%)	11 (6.9%)	0.2972
Diabetes	6 (4.8%)	<=5	0.3012
Asthma	30 (23.8%)	39 (24.4%)	0.9116
Hypertension	7 (5.6%)	11 (6.9%)	0.3426

Table 1. Socio-demographic and clinical characteristics of patients with eating disorders who received specialised out-of-country inpatient care by patient group

 Figure 1. Patient selection flow chart for out-of-country applicants with an eating disorder (from April 1st, 2000 to March 31st, 2012)



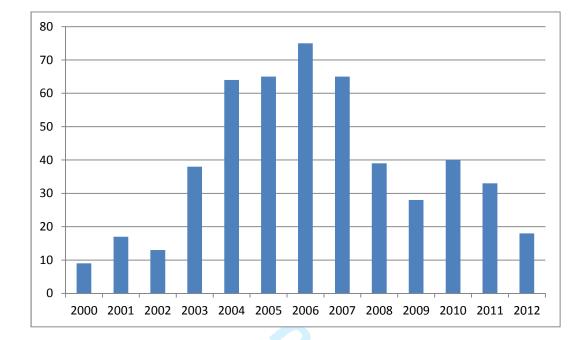


Figure 2. Number of funded out-of-country eating disorder applications by year of application

Note: 2012 does not correspond to the full fiscal year

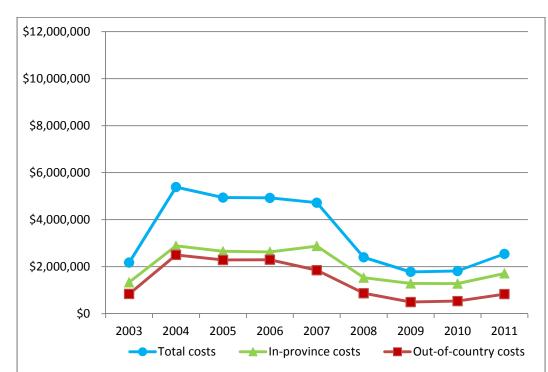
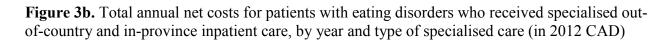
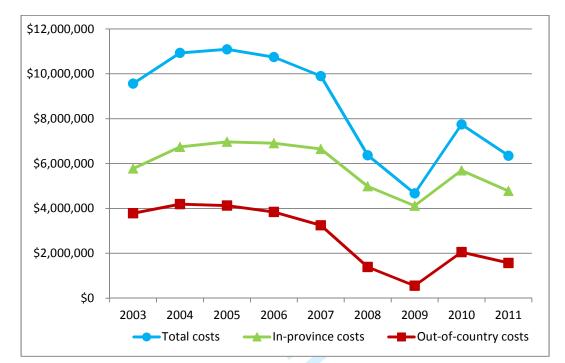


Figure 3a. Total annual net costs for patients with eating disorders who received specialised outof-country inpatient care only, by year and type of specialised care (in 2012 CAD)





Appendix

Database	Description
Canadian Institute for Health	The CIHI-DAD is a national database that contains
Information (CIHI)-Discharge	demographic, clinical, and administrative data for acute
Abstract Database (DAD)	inpatient hospital hospitalizations.
CIHI-National Ambulatory Care	The CIHI-NACRS contains data for all ambulatory care
Reporting System (NACRS)	including emergency department visits, and day
	surgery outpatient clinics.
National Rehabilitation Reporting	The NRS contains national data on rehabilitation
System (NRS)	facilities and clients, collected from participating adult
	inpatient rehabilitation facilities and programs.
Ontario Drug Benefit (ODB)	The ODB program includes data on all drugs dispensed
	in community pharmacies and long-term care/nursing
	facilities. The ODB covers all seniors in Ontario (aged
	65 and over) and those on social assistance for all
	prescriptions listed in the provincial formulary.
Ontario Health Insurance Plan 🧹	The OHIP data cover all services and procedures
(OHIP)	provided by health care providers who can claim under
	OHIP (physicians, laboratory services).
Home Care Database (HCD)	The HCD provides data on government-funded services
	coordinated by Ontario's Community Care Access
	Centres (CCAC), for individuals requiring home care.
Continuing Care Reporting	The CCRS contains clinical and demographic
System (CCRS)	information on individuals receiving facility-based
	continuing care. These services include medical long-
	term care, rehabilitation, geriatric assessment, respite
	care, and palliative care, and nursing home care.
Client Profile Database (CPRO)	The CPRO provides the date of admission to LTC
(for 2003-2010)	facilities from 2003 through 2010, while the CCRS
	provides admission and discharge dates from 2010
Continuing Care Reporting	onwards.
System (CCRS) (from 2010	
onwards)	
Ontario Mental Health Reporting	The OMHRS collects, analyzes and reports on
System (OMHRS)	information about individuals admitted to designated
	adult mental health beds in Ontario. Some mental health
	patients are recorded in the CIHI-DAD.
Assistive Devices Program (ADP)	The ADP is a program operated by the Ministry of
	Health that provides consumer support and funding to
	Ontario residents who have long-term physical
	disabilities.

STROBE Statement-checklist of items that should be included in reports of observational studies

	Item No	Recommendation
Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract
		(b) Provide in the abstract an informative and balanced summary of what was done
		and what was found
Introduction		
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported
Objectives	3	State specific objectives, including any prespecified hypotheses
Methods		
Study design	4	Present key elements of study design early in the paper
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment,
0	-	exposure, follow-up, and data collection
Participants	6	(a) Cohort study—Give the eligibility criteria, and the sources and methods of
*		selection of participants. Describe methods of follow-up
		<i>Case-control study</i> —Give the eligibility criteria, and the sources and methods of
		case ascertainment and control selection. Give the rationale for the choice of cases
		and controls
		Cross-sectional study—Give the eligibility criteria, and the sources and methods of
		selection of participants
		(b) Cohort study—For matched studies, give matching criteria and number of
		exposed and unexposed
		<i>Case-control study</i> —For matched studies, give matching criteria and the number of
		controls per case
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect
vulluolos	,	modifiers. Give diagnostic criteria, if applicable
Data sources/	8*	For each variable of interest, give sources of data and details of methods of
measurement	0	assessment (measurement). Describe comparability of assessment methods if there
measurement		is more than one group
Bias	9	Describe any efforts to address potential sources of bias
Study size	10	Explain how the study size was arrived at
Quantitative variables	11	Explain how the study size was unived at Explain how quantitative variables were handled in the analyses. If applicable,
Quantitudive variables	11	describe which groupings were chosen and why
Statistical methods	12	(<i>a</i>) Describe all statistical methods, including those used to control for confounding
Statistical methods	12	(b) Describe any methods used to examine subgroups and interactions
		(c) Explain how missing data were addressed
		(d) Cohort study—If applicable, explain how loss to follow-up was addressed
		<i>Case-control study</i> —If applicable, explain how matching of cases and controls was
		addressed
		<i>Cross-sectional study</i> —If applicable, describe analytical methods taking account of
		sampling strategy
		(\underline{e}) Describe any sensitivity analyses
Continued on next page		

Participants	13*	(a) Report numbers of individuals at each stage of study-eg numbers potentially eligible,
		examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed
		(b) Give reasons for non-participation at each stage
		(c) Consider use of a flow diagram
Descriptive	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and informatio
data		on exposures and potential confounders
		(b) Indicate number of participants with missing data for each variable of interest
		(c) Cohort study—Summarise follow-up time (eg, average and total amount)
Outcome data	15*	Cohort study-Report numbers of outcome events or summary measures over time
		Case-control study-Report numbers in each exposure category, or summary measures of
		exposure
		Cross-sectional study—Report numbers of outcome events or summary measures
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their
		precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and
		why they were included
		(b) Report category boundaries when continuous variables were categorized
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningfu
		time period
Other analyses	17	Report other analyses done-eg analyses of subgroups and interactions, and sensitivity
		analyses
Discussion		
Key results	18	Summarise key results with reference to study objectives
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision.
		Discuss both direction and magnitude of any potential bias
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplici
-		of analyses, results from similar studies, and other relevant evidence
Generalisability	21	Discuss the generalisability (external validity) of the study results
Other informatio	on	
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable,
0		for the original study on which the present article is based

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at http://www.plosmedicine.org/, Annals of Internal Medicine at http://www.annals.org/, and Epidemiology at http://www.epidem.com/). Information on the STROBE Initiative is available at www.strobe-statement.org.