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ORIGINAL RESEARCH

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The clinical and economic burden of illness in the first two years after ostomy creation: a nationwide Danish cohort study

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ABSTRACT

Background: Living with an ostomy is often associated with costly complications. This study examined the burden of illness the first two years after ostomy creation.

Methods: Data from Danish national registries included all adult Danes with an ostomy created between 2002 and 2014.

Results: Four cohorts consisted, respectively, of 11,385 subjects with a colostomy and 4,574 with an ileostomy, of which 1,663 subjects had inflammatory bowel disease (IBD) and 1,270 colorectal cancer as cause of their ileostomy. The healthcare cost was significantly higher for cases versus matched controls for all cohorts. In the first year, the total healthcare cost per person-year was €27,962 versus €4,200 for subjects with colostomy, €29,392 versus €3,308 for subjects with ileostomy, €15,947 versus €2,216 when IBD was the underlying cause, and €32,438 versus €4,196 when it was colorectal cancer. Healthcare costs decreased in the second year but remained significantly higher than controls. Hospitalization and outpatient services were primary cost drivers, with ostomy-related complications comprising 8–16% of hospitalization expenses.

Conclusion: Compared to controls, subjects with an ostomy bear a significant health and financial burden attributable to ostomy-related complications, in addition to the underlying disease, emphasizing the importance of better ostomy care to enhance well-being and reduce economic strain.

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KEYWORDS
Burden of illness; colorectal cancer; colostomy; healthcare cost; ileostomy; inflammatory bowel disease

1. Introduction

Surgical treatment of diseases such as colorectal cancer, bladder cancer, inflammatory bowel disease (IBD), and diverticulitis may require the creation of a temporary or permanent ostomy [1]. An ostomy diverts the flow of feces or urine through an artificial opening in the abdominal wall [1]. It is estimated that, in Denmark alone 14,000 subjects (2.5 out of 1000 Danish citizens) are living with an ostomy, and that 4,000 new ostomies are created each year [2]. The Danish healthcare system is universal and based on the principles of providing free and equal access to healthcare for all citizens. Most healthcare services are financed through general taxes.

The impact on quality of life due to creation of an ostomy can vary widely. For some, it represents a positive change, relieving symptoms, and even saving lives, while a significant number of people encounter challenges and limitations [3,4]. However, having an ostomy created significantly alters life, and the physical and emotional adjustments can be overwhelming [5–7]. Moreover, ostomy-related complications are common. Wound infections, ileus, dehydration, and pain often occur shortly after surgery, whereas parastomal hernias, ostomy prolapse, stenosis, or retraction represent long-term complications [5,6]. In addition, leakage of fecal output under the baseplate touching the skin can lead to costly peristomal skin complications (PSCs) [8–10]. Leakage and the worry about it are major concerns for people with an ostomy [5,11–13]. While the risk of developing ostomy-related complications remains lifelong, the incidence is highest close to discharge from ostomy creation, and readmissions often occur within the first 30 days after discharge [14,15]. Dealing with these complications lead to frequent interactions with the healthcare system, resulting in substantial healthcare costs [7–10,16–18].

Cost-effectiveness is a growing concern in clinical decision making, but there is a notable gap in understanding the complete economic impact of living with an ostomy caused by a severe underlying disease, along with ostomy-related complications. However, earlier research has shown a rise in healthcare costs in the first year after ostomy creation, compared to the year before ostomy creation, and compared with the general population [17,18]. What is more, a recent study has revealed significant increases in annual direct and indirect costs for up to ten years after ostomy creation, compared to matched controls from the general population [7]. Furthermore, living with an ostomy can negatively affect the quality of life to an extent that the ability to work is affected [5,6]. This is supported by evidence of increased sickness pay and early retirement pension [7]. The hypothesis of this study was that the economic burden associated with living with an ostomy comprises not

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only direct ostomy-related expenses but also encompasses a significant portion of costs attributed to managing the underlying condition compared to matched controls from the general population.

Hence, the aim of this study was to estimate the annual societal (direct and indirect) cost in Denmark in the first two years after creation of a colostomy or an ileostomy compared to matched controls from the general population and, furthermore, to examine if subjects with an ileostomy incurred varying societal cost depending on whether their underlying disease was IBD or colorectal cancer.

2. Methods

2.1. Data sources

This was a nationwide case-control study based on retrospectively collected data from Danish national registries and using an incidence approach. Data were obtained by linking several national registries using the civil registration number unique to all Danish citizens. The applied registries were the Civil Registration System [19], the Danish National Patient Register [20], the National Health Service Registry [21], the Danish Prescription Register [22], and Statistics Denmark [23–25].

Diagnoses of diseases and complications were based on ICD-10 codes, the Danish Medical Classification System was used for procedure (SKS) codes [26], and information regarding prescription medications was based on ATC codes (Tables S1-S3).

2.2. Study design and study population

The selection process of the incident cases is illustrated in Figure 1. Subjects with a procedure code for the creation of a colostomy or an ileostomy between 2002 and 2014 were included in the study. Subjects who were below the age of 18 years, had more than one ostomy, had the ostomy reversed during follow-up, and died during index admission or within 30 days after were excluded. The included subjects were followed for each of the first two years after the index date (the date of ostomy creation) or until death. The study continued until the end of 2016 to allow two years' follow-up of all subjects. One cohort consisted of subjects which had a colostomy created and one which had an ileostomy created. Furthermore, the ileostomy cohort was stratified into two subcohorts based on the underlying disease; one subcohort with colorectal cancer and one with IBD. A pool of controls from the general population consisting of subjects who, at any time during 2000–2014, were living without an ostomy, was created. Each case subject was matched at the index date with four subjects randomly selected from the control pool based on age, sex, marital status, residential area, and education.

2.3. Resource utilization and costs

To estimate the cost of inpatient and outpatient healthcare, the Diagnose-Related Group (DRG) system was employed. DRG is a case-mix system that organizes patients with similar diseases and expenses into groups, each with its own tariff, and which is updated annually. The cost of healthcare resource utilization

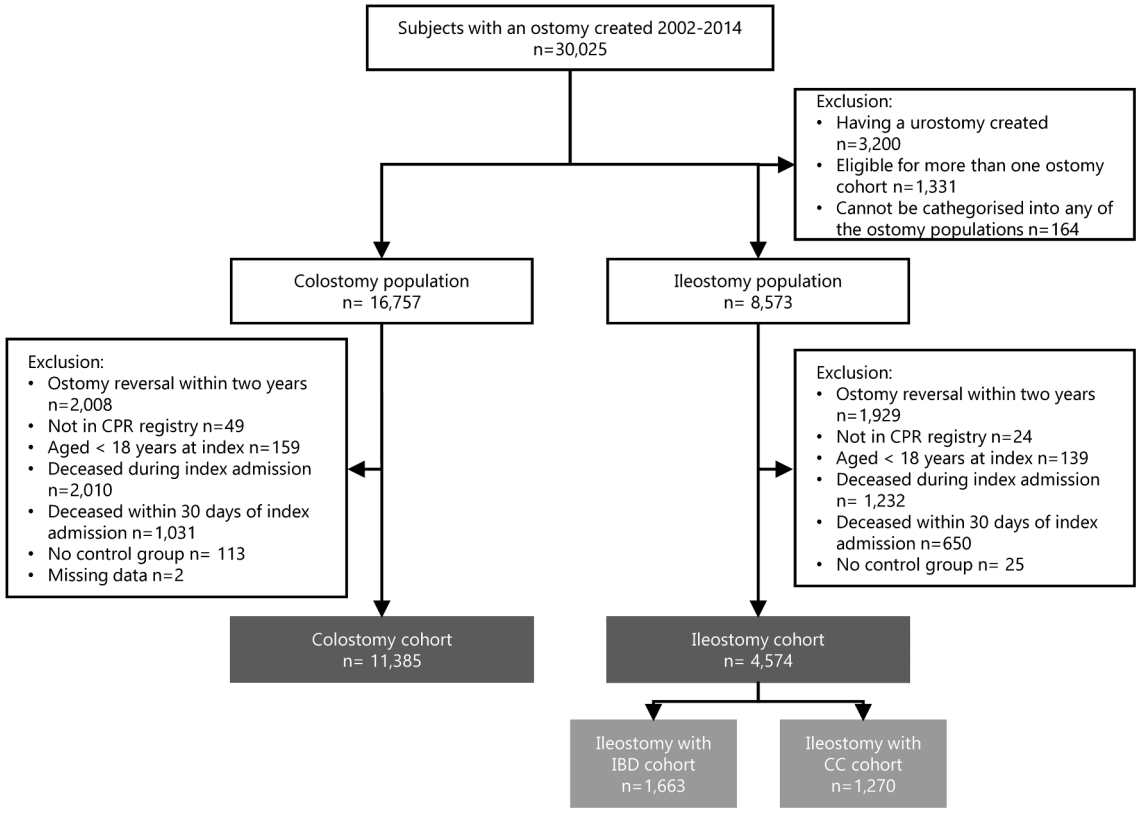


Figure 1. CC: colorectal cancer; IBD: inflammatory bowel disease.

(HCRU) in this study was based on these DRG weights and tariffs. Unit cost of services provided by the general practitioner was derived from the prevailing National Health Insurance fee schedules. The cost of prescription medication was calculated by multiplying the retail price of each drug (including dispensing costs) by the number of transactions. Predicted HCRU and the associated costs were estimated by dividing the aggregated number of incidents and costs with the aggregated number of patient-years. The annual estimates were presented for years 1 and 2 after ostomy creation, and the cost of the index admission was not included in year 1. The cost was adjusted to 2016 prices and presented in euros (€) (€1 = 7.45 Danish kroner (DKK)).

The total HCRU and associated costs consisted of all interactions within the healthcare system during each of the two years' follow-up. HCRU encompassed the number of hospital admissions, days in hospital, outpatient visits (including emergency room services), 30-day readmissions, and primary care sector services (contacts). One primary care sector contact could consist of multiple services.

The total healthcare cost included cost of hospitalization (excluding psychiatric costs), outpatient services (including emergency room services and excluding psychiatric costs), primary care sector services, and prescription medications (excluding medications given in secondary care).

The inpatient cost of common ostomy-related complications was analyzed separately. The complications included dehydration, sepsis, kidney disease, fecal incontinence and constipation, hernia, post-operative wound infection, and common skin disorders. The 20 most common reasons for hospitalization and 30-day readmission (diagnosis given at readmission) were established based on frequency of ICD-10 codes.

The cost of contacts to the general practitioner and prescription medications were also obtained, and the cost of medication related to treatment of common ostomy-related complications was analyzed separately: medication often used for common skin disorders such as PSCs, pain (opioids), mood disorders, and bowel dysfunction.

The indirect cost included earned income and income transfer payments, which included disability pension, early retirement pension, unemployment benefit, sick pay (public funded), and age pension, as well as home care (available from 2009).

2.4. Statistical analysis

The SAS SURVEYSELECT procedure was used for selecting probability-based random control cohorts. A chi-squared test was employed to compare cases with matched controls regarding baseline characteristics which were not part of the matching process. A Poisson model and a generalized linear model (GLM) were applied to predict the number of incidents, as well as cost per patient-year, and to test if differences between cases and controls were significant. A Poisson model was applied to predict (HCRU), as it is a count variable and observations with a value of 0 is included. A 2-step one model GEE (generalized estimating equation) regression model with gamma distribution and log link was applied to predict cost, since it is a continuous variable and includes 0 in the response variable. A GEE model ensures robust standard errors. Weighting for exposure time was used to adjust for subjects who died during the follow-up period. Cost and resource utilization were reported as mean and 95% confidence interval (CI). A p-value of < 0.05 was considered statistically significant. Analyses were performed using SAS version 9.4 (SAS Institute, Cary, NC, U.S.A.).

2.5. Ethical considerations

Since the study applied retrospective anonymized registry data, no approval from the Danish Ethics Committee was required by Danish law.

3. Results

3.1. Demographic and clinical characteristics

A total of 11,385 (71.3%) subjects with a colostomy and 4,574 (28.7%) with an ileostomy created between 2002 and 2014 were included in the study (Figure 1). The share of male and female subjects was comparable (46.4% males and 53.6% females in both cohorts), and the mean (± SD) age was 69.9 ± 13.2 years in the colostomy cohort and 60.2 ± 17.2 years in the ileostomy cohort (Table 1). In the colostomy cohort, 24.7% died in year 1 and 13.1% in year 2, and in the ileostomy cohort these numbers were 19.3% and 7.4%. The cohort with IBD as the cause of the ileostomy consisted of 1,663 subjects (36.4% of the ileostomy cohort), and the cohort with colorectal cancer as the cause consisted of 1,270 subjects of which 56, in addition, suffered from IBD (27.8% of the ileostomy cohort). The age at ileostomy creation was more than 20 years lower in the ileostomy cohort with IBD (48.3 ± 17.1 years) than in the cohort with colorectal cancer (69.3 ± 11.6 years).

3.2. Resource utilization and cost of healthcare

The HCRU (Table S4) and the associated total healthcare cost per patient-year (Figure 2) was significantly higher in both years compared with controls for all case cohorts. In year 1, the absolute total healthcare cost per person-year was comparable for subjects living with a colostomy or an ileostomy, whereas, compared to controls, the excess cost was higher when living with an ileostomy (€29,392 vs. €3,308) than with a colostomy (€27,962 vs. €4,200). The absolute total healthcare cost per patient-year, as well as the excess cost compared to controls was higher when the reason for ileostomy creation was colorectal cancer (€32,438 vs. €4,196) than when it was IBD (€15,947 vs. €2,216). In year 2, the absolute and excess total healthcare cost was comparable in the ileostomy cohort (€14,956 vs. €3,466) and the colostomy cohort (€16,194 vs. €4,299) and it remained higher in the ileostomy cohort with colorectal cancer (€17,112 vs. €4,122) than in the ileostomy cohort with IBD (€10,394 vs. €2,421).

The primary driver of the total healthcare cost was inpatient admissions, which was significantly higher for case cohorts compared to matched controls (Figure 2). In year 1, the absolute and excess cost per patient-year of inpatient admissions was higher for subjects with an ileostomy (€20,052 vs. €1,499) than for those with a colostomy (€15,336 vs. €2,093) and higher for subjects with an ileostomy and colorectal cancer

Table 1. Baseline characteristics of case and control cohorts at index (unless otherwise stated) stratified by ostomy type and underlying disease.

	Colostomy		Ileostomy		Ileostomy with IBD		Ileostomy with CC	
	Case	Control	Case	Control	Case	Control	Case	Control
Number of subjects	11385	43713	4574	17780	1663	6541	1270	4883
Number of patient-years								
Year 1	9894	42952	4081	17629	1632	6520	1099	4818
Year 2	7781	41115	3512	17262	1593	6458	861	4671
Underlying disease/comorbidities 12 months before index, mean (%)								
CC	5557 (48.8%)	98 (0.2%)	1270 (27.8%)	33 (0.2%)	0 (0%)	6 (0.1%)	1270 (100.0%)	14 (0.3%)
IBD	248 (2.2%)	73 (0.2%)	1719 (37.6%)	31 (0.2%)	1663 (100.0%)	10 (0.2%)	56 (4.4%)	9 (0.2%)
Sex, mean (%)								
Male	5283 (46.4%)	20124 (46.0%)	2124 (46.4%)	8201 (46.1%)	764 (45.9%)	3005 (45.9%)	663 (52.2%)	2527 (51.8%)
Female	6102 (53.6%)	23589 (54.0%)	2450 (53.6%)	9579 (53.9%)	899 (54.1%)	3536 (54.1%)	607 (47.8%)	2356 (48.2%)
Age (years), mean (%)								
18–20	11 (0.1%)	44 (0.1%)	67 (1.5%)	268 (1.5%)	60 (3.6%)	240 (3.7%)	0 (0%)	0 (0%)
21–30	94 (0.8%)	368 (0.8%)	286 (6.3%)	1127 (6.3%)	249 (15.0%)	979 (15.0%)	6 (0.5%)	24 (0.5%)
31–40	217 (1.9%)	860 (2.0%)	386 (8.4%)	1526 (8.6%)	317 (19.1%)	1252 (19.1%)	21 (1.7%)	83 (1.7%)
41–50	641 (5.6%)	2528 (5.8%)	498 (10.9%)	1975 (11.1%)	279 (16.8%)	1106 (16.9%)	65 (5.1%)	257 (5.3%)
51–60	1546 (13.6%)	6084 (13.9%)	764 (16.7%)	3013 (16.9%)	294 (17.7%)	1159 17.7%)	162 (12.8%)	639 (13.1%)
61–70	2796 (24.6%)	10971 (25.1%)	1095 (23.9%)	4276 (24.0%)	272 (16.4%)	1071 (16.4%)	371 (29.2%)	1453 (29.8%)
71–80	3499 (30.7%)	13422 (30.7%)	1047 (22.9%)	4010 (22.6%)	158 (9.5%)	605 (9.2%)	450 (35.4%)	1708 (35.0%)
>80	2581 (22.7%)	9436 (21.6%)	431 (9.4%)	1585 (8.9%)	34 (2.0%)	129 (2.0%)	195 (15.4%)	719 (14.7%)
Age, mean ± SD	69.9 ± 13.2	69.6 ± 13.1	60.2 ± 17.2	59.9 ± 17.2	48.3 ± 17.1	48.2 + 17.0	69.3 + 11.6	69.0 ± 11.6
Marital status, n (%)								
Living alone	5153 (45.3%)	19501 (44.6%)	1826 (39.9%)	6998 (39.4%)	572 (34.4%)	2225 (34.0%)	513 (40.4%)	1933 (39.6%)
Married or cohabiting	6232 (54.7%)	24212 (55.4%)	2748 (60.1%)	10782 (60.6%)	1091 (65.6%)	4316 (66.0%)	757 (59.6%)	2950 (60.4%)
Education, n (%)								
Primary	5117 (44.9%)	19943 (45.6%)	1754 (38.3%)	6888 (38.7%)	519 (31.2%)	2057 (31.4%)	539 (42.4%)	2099 (43.0%)
Secondary	181 (1.6%)	622 (1.4%)	175 (3.8%)	670 (3.8%)	121 (7.3%)	476 (7.3%)	22 (1.7%)	84 (1.7%)
Vocational	3506 (30.8%)	13773 (31.5%)	1621 (35.4%)	6412 (36.1%)	609 (36.6%)	2423 (37.0%)	463 (36.5%)	1817 (37.2%)
Short college	236 (2.1%)	785 (1.8%)	146 (3.2%)	521 (2.9%)	67 (4.0%)	246 (3.8%)	34 (2.7%)	114 (2.3%)
Bachelor	1020 (9.0%)	3898 (8.9%)	528 (11.5%)	2044 (11.5%)	233 (14.0%)	924 (14.1%)	113 (8.9%)	426 (8.7%)
Master/PHD	305 (2.7%)	1097 (2.5%)	192 (4.2%)	715 (4.0%)	81 (4.9%)	314 (4.8%)	43 (3.4%)	150 (3.1%)
Unknown	1020 (9.0%)	3595 (8.2%)	158 (3.5%)	530 (3.0%)	33 (2.0%)	101 (1.5%)	56 (4.4%)	193 (4.0%)
Residential area, n (%)								
Northern region	1406 (12.3%)	5415 (12.4%)	440 (9.6%)	1714 (9.6%)	180 (10.8%)	715 (10.9%)	137 (10.8%)	523 (10.7%)
Central region	2060 (18.1%)	7953 (18.2%)	802 (17.5%)	3131 (17.6%)	413 (24.8%)	1627 (24.9%)	174 (13.7%)	670 (13.7%)
Southern region	2612 (22.9%)	10021 (22.9%)	971 (21.2%)	3768 (21.2%)	319 (19.2%)	1250 (19.1%)	285 (22.4%)	1102 (22.6%)
Capital region of Denmark	3269 (28.7%)	12479 (28.5%)	1458 (31.9%)	5665 (31.9%)	438 (26.3%)	1722 (26.3%)	434 (34.2%)	1666 (34.1%)
Region Zealand	2038 (17.9%)	7845 (17.9%)	903 (19.7%)	3502 (19.7%)	313 (18.8%)	1227 (18.8%)	240 (18.9%)	922 (18.9%)
Dead during follow-up, n (%)								
Year 1	2808* (24.7%)	1686 (3.9%)	884* (19.3%)	327 (1.8%)	57* (3.4%)	49 (0.7%)	326* (25.7%)	143 (2.9%)
Year 2	1495* (13.1%)	1802 (4.1%)	339* (7.4%)	374 (2.1%)	24 (1.4%)	72 (1.1%)	154* (12.1%)	139 (2.8%)

CC: colorectal cancer; IBD: inflammatory bowel disease; SD: standard deviation; *: $p < 0.000$.

(€18,677 vs. €2,027) in contrast to those with ileostomy and IBD (€10,703 vs. €877). In year 2, the absolute and excess cost per patient-year of inpatient admissions was comparable between ostomy types (ileostomy: €8,624 vs. €1,614 and colostomy: €8,016 vs. €2,144). In contrast, it continued to be higher when ileostomy was created due to colorectal cancer (€8,573 vs. €1,994) than IBD (€6,280 vs. €1,039).

The total inpatient admission cost of common ostomy-related complications was responsible for 8–16% of the total inpatient admission cost (Table 2). Taken together with the 20 most common reasons for hospitalization and 30-day readmission (Tables S7-S14), it became apparent that all case cohorts experienced post-operative wound infection and problems with the artificial opening (colostomy/ileostomy status). Subjects with an ileostomy predominantly suffered from dehydration, volume depletion, and kidney disease, while subjects with a colostomy suffered from fecal incontinence and constipation.

Outpatient services were the second highest driver of the total healthcare cost, and the cost was significantly higher for case cohorts compared to controls in both years (Figure 2). In year 1, the cost of outpatient services per patient-year was higher for subjects with a colostomy (€10,780 vs. €1,019) than an ileostomy (€7,551 vs. €927), and it was higher when the ileostomy was created due to colorectal cancer (€12,041 vs. €1,127) than IBD (€3,854 vs. €676) in both absolute and excess terms. In year 2, the cost of outpatient services was still higher in the colostomy cohort (€6,343 vs. €1,066) than in the ileostomy cohort (€4,672 vs. €966) as well as in the ileostomy cohort with colorectal cancer (€7,004 vs. 1,092) than in the ileostomy cohort with IBD (€2,795 vs. 706).

The cost of total prescription medication constituted small proportions of the total healthcare cost (Figure 2). Notably, the cost of pain medication (opioids) represented ~ 20% of the cost of all prescription medication in all case cohorts and was very high compared to controls (Table 3). Similarly, the cost of mood disorders medication was relatively high, comprising ~ 5% of the total cost of prescription medication.

3.3. Indirect cost

Disability pension and sick pay per patient-year in case cohorts were significantly higher compared to matched controls in

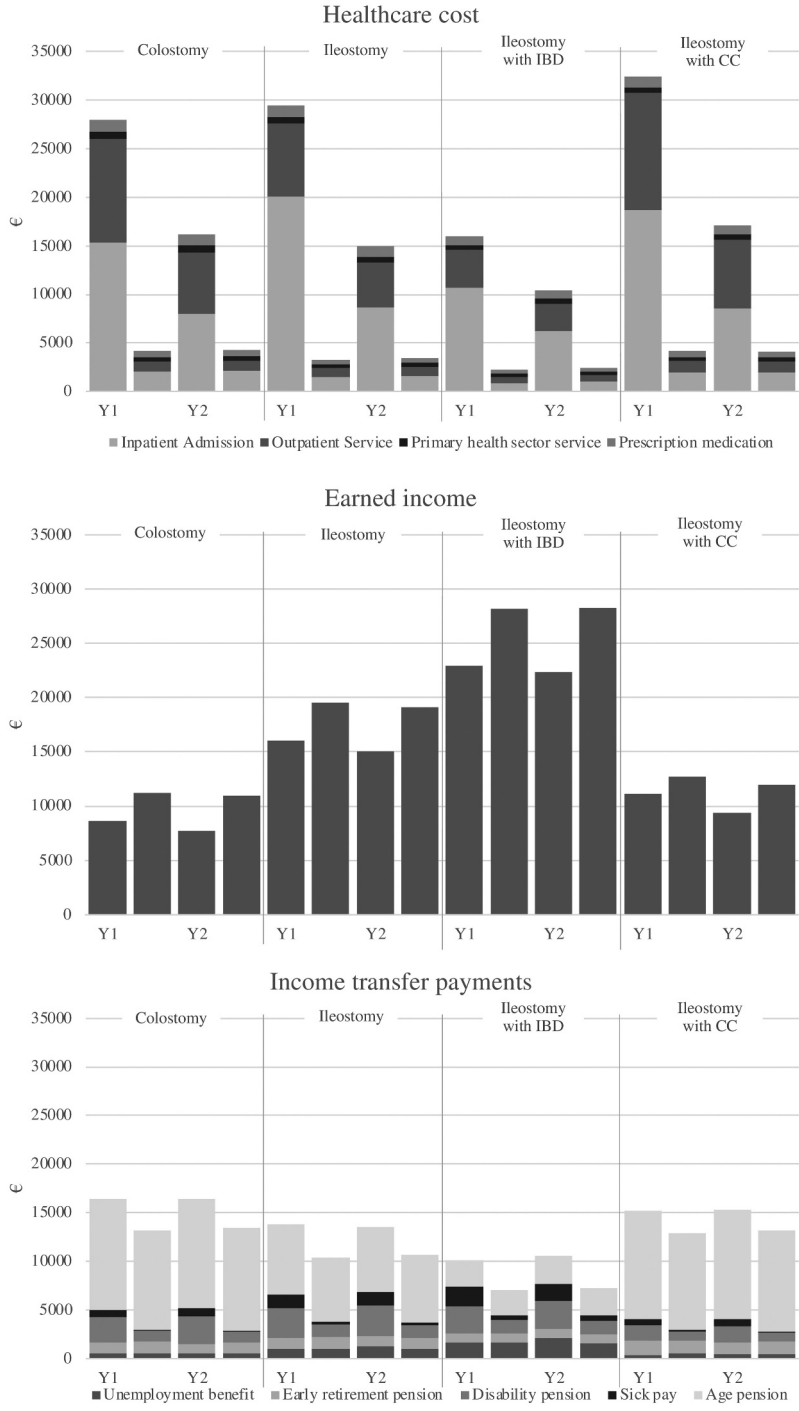


Figure 2. Predicted mean healthcare cost, earned income, and income transfer payments per patient-year (€) of case and control cohorts in year 1 and year 2 stratified by ostomy type and underlying disease. Below the figure: For each year, the cases are represented by the first bar and the controls by the second. IBD: inflammatory bowel disease; CC: colorectal cancer; Y: year. Differences between ostomy and matched control cohorts were significant ($p < 0.05$) except for colostomy: unemployment benefit (p=0.513) in year 2; ileostomy: unemployment benefit (p=0.057) and early retirement pension (p=0.714) in year 1 and age pension (p=0.072) in year 2; ileostomy with IBD: unemployment benefit (p=0.482) in year 1, and early retirement pension (p=0.543) and age pension (0.880) in year 2; ileostomy with CC: unemployment benefit (p=0.418), early retirement pension (p=0.431)), and age pension (p=0.053) in year 2. Please refer to Table S5 for exact values and confidence intervals.

both years, and they were the key drivers of the income transfer payments (Figure 2). In year 1, subjects with an ileostomy had higher disability pension per patient-year (€3,049 vs. €1,339) than those with a colostomy (€2,643 vs. €1,161). Similarly, among subjects with an ileostomy, those with IBD (€2,774 vs. €1,345) had a higher disability pension than those with colorectal cancer (€1,572 vs. €949) in both absolute and excess terms. For sick pay, the same pattern emerged: compared to controls, the ileostomy cohort reached €1,381 vs. €279, and the colostomy cohort €773 vs. €126; the ileostomy cohort with IBD incurred €2,060 vs. €503, and the ileostomy cohort with colorectal cancer €700 vs. €124. In year 2, the sick pay for case and control cohorts were approximately similar to year 1.

Table 2. Predicted mean inpatient costs per patient-year (€) of common ostomy-related complications in case and control cohorts in year 1 and year 2 stratified by ostomy type and underlying disease.

	Colostomy		Ileostomy		Ileostomy with IBD		Ileostomy with CC	
	Case	Control	Case	Control	Case	Control	Case	Control
Common ostomy-related complications, mean [95% CI]								
Dehydration								
Y1	208 [202; 215]*	25 [25; 25]	1000 [952; 1050]*	16 [16; 17]	335 [310; 362]*	5 [5; 5]	1356 [1234; 1489]*	23 [23; 24]
Y2	115 [111; 119]*	29 [29; 29]	261 [248; 275]*	16 [15; 16]	143 [132; 155]*	6 [6; 6]	383 [346; 423]*	15 [15; 15]
Sepsis								
Y1	17 [16; 17]*	5 [5; 5]	74 [71; 78]*	4 [4; 4]	26 [26; 26]*	0 [0; 0]	32 [29; 35]*	8 [8; 9]
Y2	20 [19; 21]*	4 [4; 5]	31 [30; 33]*	3 [3; 3]	7 [7; 8]*	3 [3; 3]	27 [25; 30]*	1 [1; 1]
Kidney disease								
Y1	223 [216; 230]*	24 [24; 25]	606 [577; 636]*	15 [15; 15]	78 [72; 84]*	10 [10; 10]	633 [576; 695]*	24 [23; 25]
Y2	84 [81; 87]*	25 [25; 25]	109 [103; 114]*	18 [18; 18]	34 [31; 37]*	7 [7; 8]	94 [85; 104]*	30 [29; 31]
Fecal incontinence and constipation								
Y1	158 [153; 163]*	8 [8; 8]	72 [69; 76]*	5 [4; 5]	43 [40; 47]*	2 [2; 2]	80 [73; 88]*	1 [1; 1]
Y2	102 [98; 105]*	7 [7; 8]	45 [43; 47]*	7 [7; 7]	39 [36; 42]*	8 [8; 9]	50 [45; 55]*	7 [7; 7]
Hernia								
Y1	71 [68; 73]*	2 [2; 2]	66 [63; 70]*	3 [3; 3]	79 [73; 85]*	1 [1; 1]	64 [58; 70]*	1 [1; 1]
Y2	163 [158; 169]*	3 [3; 3]	157 [149; 165]*	2 [2; 2]	193 [178; 209]*	1 [1; 1]	74 [67; 82]*	4 [4; 5]
Post-operative wound infection								
Y1	436 [422; 450]*	10 [10; 10]	560 [533; 588]*	12 [12; 12]	308 [284; 333]*	5 [5; 5]	740 [673; 812]*	11 [10; 11]
Y2	91 [88; 94]*	9 [9; 9]	68 [64; 71]*	6 [6; 6]	70 [64; 75]*	2 [2; 2]	53 [48; 59]*	9 [9; 9]
Common skin disorders								
Y1	224 [217; 231]*	5 [5; 5]	83 [79; 87]*	5 [5; 5]	33 [27; 28]*	3 [2; 3]	59 [54; 65]*	5 [5; 6]
Y2	130 [126; 135]*	5 [5; 5]	50 [48; 53]*	4 [4; 4]	52 [48; 56]*	2 [2; 2]	53 [47; 58]*	4 [4; 4]

CC: colorectal cancer; CI: confidence interval; IBD: inflammatory bowel disease; Y: year. *$p < 0.000$.

Table 3. Predicted mean costs per patient-year (€) of general practitioner and selected prescription medication according to indication in case and control cohorts in year 1 and year 2 stratified by ostomy type and underlying disease.

	Colostomy		Ileostomy		Ileostomy with IBD		Ileostomy with CC	
	Case	Control	Case	Control	Case	Control	Case	Control
Selected primary care sector services, mean [95% CI]								
General practitioner, contacts								
Y1	400 [388; 413]*	207 [205; 209]	350 [333; 367]*	169 [166; 171]	276 [256; 299]*	137 [133; 140]	389 [355; 428]*	197 [191; 203]
Y2	328 [317; 339]*	213 [211; 215]	272 [258; 286]*	175 [172; 177]	223 [206; 241]*	144 [140; 147]	302 [273; 335]*	201 [195; 207]
Prescription medications according to indication, mean [95% CI]								
Common skin disorders such as PSCs								
Y1	27 [27; 28]*	12 [12; 12]	42 [40; 44]*	11 [11; 11]	23 [21; 24]*	9 [9; 9]	22 [20; 24]*	12 [11; 12]
Y2	25 [25; 26]*	12 [12; 12]	32 [31; 34]*	11 [11; 11]	21 [19; 22]*	9 [8; 9]	22 [20; 25]*	11 [11; 12]
Pain (opioids)								
Y1	271 [262; 279]*	36 [36; 37]	182 [173; 191]*	23 [23; 24]	122 [113; 132]*	13 [13; 13]	180 [164; 197]*	27 [26; 28]
Y2	216 [208; 223]*	36 [35; 36]	179 [170; 189]*	24 [24; 25]	113 [104; 122]*	14 [14; 14]	214 [194; 237]*	28 [28; 29]
Mood disorders								
Y1	68 [66; 70]*	39 [38; 39]	59 [57; 62]*	30 [30; 31]	47 [43; 51]*	24 [24; 25]	49 [45; 54]*	33 [32; 34]
Y2	64 [62; 67]*	35 [35; 35]	55 [53; 58]*	28 [27; 28]	47 [44; 51]*	22 [21; 22]	40 [36; 45]*	30 [29; 31]
Bowel dysfunction								
Y1	34 [33; 35]*	6 [6; 6]	61 [58; 64]*	3 [3; 4]	39 [36; 43]*	2 [2; 2]	58 [53; 64]*	4 [4; 5]
Y2	31 [30; 32]*	6 [6; 6]	67 [64; 71]*	3 [3; 4]	53 [49; 57]*	1 [1; 1]	52 [47; 57]*	5 [5; 5]

CC: colorectal cancer; CI: confidence interval; IBD: inflammatory bowel disease; PSCs: peristomal skin complications; Y: year. *$p < 0.000$.

Earned income per patient-year was significantly lower in all case cohorts in each of the 2 years compared with controls (Figure 2). The difference in earned income between cases and controls was larger among subjects with an ileostomy than with a colostomy. Additionally, the difference was more pronounced when IBD was the cause of ileostomy than when it was colorectal cancer.

4. Discussion

This is the first Danish study to evaluate the annual societal cost in the first two years following ostomy creation and comparing it to matched controls from the general population. The results demonstrated that, in addition to the underlying disease, creation of an ostomy significantly impacted the subjects' health and the societal economic burden. In absolute terms, the total healthcare cost was comparable between subjects with an ileostomy and a colostomy, whereas living with an ileostomy due to colorectal cancer incurred substantially higher healthcare cost than IBD as the underlying disease. When the case cohorts were compared to controls – thereby minimizing a potential effect from the large age variation between cohorts – the excess healthcare cost was higher among subjects with an ileostomy than those with a colostomy in the first year and comparable in the second year. Meanwhile, the excess cost in both years continued to be higher for subjects with an ileostomy and colorectal cancer than for those with an ileostomy and IBD. The key cost drivers were inpatient admissions and outpatient services. Noteworthy, compared to controls, subjects with an ileostomy faced higher cost for inpatient admission than those with a colostomy, and the situation was reversed when it came to

outpatient cost. However, subjects with an ileostomy and colorectal cancer incurred higher cost for both inpatient admission and outpatient services than those with an ileostomy and IBD. These findings suggested that subjects with an ileostomy experienced more severe issues than those with a colostomy during the first year. Furthermore, subjects who were living with an ileostomy created due to colorectal cancer tended to experience greater severity in both the first and second years than those with IBD.

The economic burden of living with an ostomy is scarcely researched and comparability of existing studies is limited because of differences in study designs, study populations, underlying diseases and progression state, treatment modalities, cost calculation methods, data availability, and health systems [7,17,18]. That said, consistent with our study, a recent Swedish registry study established that the societal cost of living with an ostomy significantly increased, even up to ten years after its creation [7]. A German study found that, compared to the year before ostomy creation, the healthcare cost significantly increased in the first year after ostomy creation and remained significantly increased in the second year, albeit on a lower level [18], and a second German study showed that the healthcare cost of living with an ostomy was significantly increased compared to matched controls [17]. Similarly, our study evidenced that, although the healthcare cost declined in year 2, the cost was significantly higher than controls, which likely reflect a sustained high morbidity.

The high total healthcare cost in our study was a result of resource-intensive treatments of the underlying diseases such as colorectal cancer and IBD which have been shown to be costly to treat [27,28], as well as treatment of ostomy-related complications, which have also been shown to increase the use of healthcare resources [8,16,29]. Furthermore, the high mortality rate in our study, together with the requirement of a permanent ostomy, may reflect very advanced disease stages. In our study, particularly subjects with an ileostomy were burdened by readmissions because of dehydration in the first year. This coincides with a previous review demonstrating that dehydration was the leading ostomy-related complication responsible for costly readmissions [14]. Living with a colostomy often results in periods of constipation [30]. This was also evidenced in our study by a common cause of hospitalization and readmission and high medication cost for bowel dysfunction among subjects with a colostomy. The relatively high cost of prescription medication for mood disorders in our study confirm earlier studies pointing out that living with a debilitating – or even life-threatening – disease together with adjusting to a new life with an ostomy has severe mental effects [6,31]. The cost of prescribed pain medication in the form of opioids is high and adds to the evidence of the continuous difficulties these subjects are confronted with. The high intake of opioids could potentially be responsible for the relatively high degree of constipation observed [32]. As the cost of ostomy products are not included in the Danish registries, our cost estimates of total healthcare are conservative. From a Swedish registry study, it became apparent that ostomy products accounted for around 8.5% of the total healthcare costs in the first year after ostomy creation [7].

To our knowledge this is the first Danish study to estimate the earned income and income transfer payment following creation of an ostomy. We found that income transfer payment contributed markedly to the societal cost. Compared to controls, earned income was significantly lower and disability pension and sick pay were significantly higher for all case cohorts in both years. This indicates work disabilities for at least two years after ostomy creation. This finding is in accordance with the Swedish registry study which found that transfer payment to compensate for loss of work was increased in especially the first years [7], and other previous studies highlighted that living with an ostomy diminishes the ability to work [5,8,17,33,34]. Furthermore, the cost of productivity loss due to sickness is seldom fully compensated for by income transfer payments and may thus indicate that living with an ostomy may impact the personal income level, causing societal inequality [35]. The largest decrease in earned income, as well as higher sick pay and disability pension, among subjects with an ileostomy due to IBD can be attributed to their younger age and continued participation in the workforce. As a result, subjects with an ileostomy due to IBD appeared to be impacted the most in terms of their personal income.

To reduce the risk of costly complications, there is a potential for intensifying ostomy education and support. For instance, there is convincing evidence that when qualified stoma care nurses promote and support selfcare among subjects with an ostomy, it can decrease the incidence of complications and thereby reduce the cost [36–40]. Despite that, a recent study showed that 23% of people living with an ostomy in Denmark were unaware of the possibility to consult a stoma care nurse [36].

The most important strength of this study was the use of nationwide registries; they ensured that the number of subjects identified was considered representative for the Danish ostomy population within the study period, allowed us to operate in a real-world setting, and reduced the risk of information and selection bias. The large number of included subjects reduced the risk of random variations. Additionally, the inclusion of four matched controls per case subject increased the power and reduced the bias of the results. The inclusion of earned income and income transfer payments was a significant strength of the methodology, as it illustrates the 'true' cost. The study also has some limitations. The quality of data in registries is dependent on correct coding into the registries. The study period stretched over 15 years and may have impacted data, as treatment modalities have changed over this period affecting the cost (positively or negatively). Despite the study's timeframe (2002–2016), we find its relevance today for various reasons. Firstly, it provides a foundational understanding of the economic impact and challenges faced by individuals with an ostomy. Secondly, even with advancements in surgical techniques, the enduring impact on healthcare expenses and work ability remains significant and the results offer valuable context in a changing healthcare landscape. While we considered supplementing with recent data, the COVID-19 pandemic caused unprecedented disruptions resulting in a substantial shift in available data over several years. The cost of ostomy products, visits at stoma care nurses, patient education, and nursing homes

(which are paid by the municipalities) are not included in the registries. The nature of the study was observational, and causality cannot be inferred. Regardless of these limitations, this study provides new and important information to the current understanding of the burden of living with an ostomy.

5. Conclusions

This study documents that subjects living with an ostomy experience a substantial health burden, leading to increased healthcare expenses and diminished work ability compared to matched controls from the general population. This heightened burden is attributed to complications related to their ostomy per se, in addition to the challenges posed by their underlying disease. Notably, subjects with an ileostomy appear to carry a more substantial health burden than those with a colostomy, especially when their ileostomy is a result of colorectal cancer rather than IBD. These findings underscore the need for optimal management and support for ostomy care, with the goal of enhancing the well-being of those affected and alleviate the economic strain on society.

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Declaration of interest

F Bruun Andersen is an employee of Coloplast Danmark A/S, Denmark. J Kjellberg is employed by VIVE, the Danish Center for Social Science Research, Denmark, an independent research and analysis center. VIVE received funding from Coloplast for the contribution to this study. R Ibsen is an employee of i2Minds, an independent data analysis agency, which received funding from VIVE to this study. C Sternhufvud is an employee of Coloplast AB, Sweden. B Petersen is employed by MedDevHealth, Denmark, an independent consultancy, and received funding from Coloplast for the contribution to the study. The authors have no other relevant affiliation or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript apart from those disclosed.

Author contributions

F Bruun Andersen was involved in conceptualization, methodology, project administration, review and editing. J Kjellberg contributed to methodology, validation, review and editing. R Ibsen provided data curation, formal analysis, investigation, methodology, review and editing. C Sternhufvud was involved in conceptualization, methodology, supervision, validation, review and editing. B Petersen contributed to methodology, review and editing.

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Posters

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