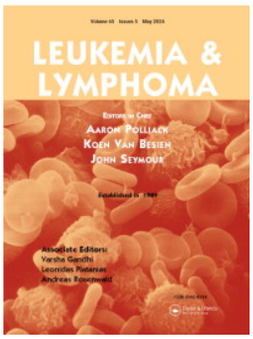

Leukemia & Lymphoma

ISSN: (Print) (Online) Journal homepage: informahealthcare.com/journals/ilal20

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To cite this article: Jingbo Yu, James Nelson, Taylor Marlin, Evan Braunstein & Michelle Jerry (27 Apr 2024): Direct and indirect costs for patients with myeloproliferative neoplasms, Leukemia & Lymphoma, DOI: 10.1080/10428194.2024.2338849

To link to this article: https://doi.org/10.1080/10428194.2024.2338849

Published online: 27 Apr 2024.

LEUKEMIA & LYMPHOMA
https://doi.org/10.1080/10428194.2024.2338849

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ORIGINAL ARTICLE

Direct and indirect costs for patients with myeloproliferative neoplasms

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ABSTRACT

Myeloproliferative neoplasms (MPNs) are associated with substantial healthcare resource use and productivity loss. This retrospective cohort analysis used disability leave and medical claims data to measure direct and indirect healthcare costs associated with MPNs. The analysis included 173 patients with myelofibrosis (MF), 4477 with polycythemia vera (PV), 6061 with essential thrombocythemia (ET), and matched controls (n = 519, n = 13,431, and n = 18,183, respectively). Total healthcare costs were significantly higher for cases versus controls in each cohort (mean cost difference: MF, $67,456; PV, $10,970; ET, $22,279). Cases were more likely than controls to take disability leave and incurred higher disability-related costs. Among subgroups with thrombotic events, direct and indirect costs were higher for cases versus controls. Thrombotic events substantially increased direct costs and disability leave for patients with PV or ET compared with the full PV or ET cohorts. These findings demonstrate increased economic burden for patients with MPNs.

ARTICLE HISTORY
Received 23 January 2024
Revised 28 March 2024
Accepted 31 March 2024

KEYWORDS
Myeloproliferative neoplasm; thrombocythemia; healthcare utilization; healthcare costs; disability leave; indirect costs

Introduction

The myeloproliferative neoplasms (MPNs) are classified into three subtypes of generally increasing severity: essential thrombocythemia (ET), polycythemia vera (PV), and myelofibrosis (MF). Although signs and symptoms vary, MPNs are generally associated with elevated blood counts, splenomegaly, constitutional symptoms, and an increased risk of thrombotic events [1–4]. The disease phenotype is burdensome for most patients, with a survey of patients with MPNs in the USA indicating that 71–80% of patients experience fatigue, 33–40% pruritus, and 31–53% abdominal discomfort, among other constitutional symptoms [2].

Over the course of illness, MPNs impart substantial direct healthcare costs (e.g. inpatient and outpatient visits, emergency room services, and pharmacy costs) and indirect costs (e.g. lost work productivity and medical leave) [2,5–7]. In an insurance claims analysis, annual direct costs for patients with MPNs in the USA were estimated to be 5-fold higher for MF, 2-fold higher for PV, and 4-fold higher for ET compared with age- and sex-matched control populations without MPNs [5]. Danish and Canadian matched cohort studies similarly showed significantly increased healthcare resource utilization and costs for patients with an MPN versus matched controls without MPNs, with the highest costs associated with MF [8,9]. Beyond the direct healthcare costs, a patient survey in the USA revealed that 50.5% of patients who were employed at diagnosis made at least one change in employment status due to their MPN, including terminating or changing a job (30.2%), reducing hours of employment (21.8%), and requiring medical disability leave because of their disease (24.8%) [6]. Similarly, a separate US patient survey found that 37–53% believed having an MPN interfered with daily activities, 30–59% of patients reduced work hours, and 11–31% voluntarily terminated their job as a result of their MPN diagnosis, depending on MPN subtype [2].

This study was conducted to evaluate healthcare resource utilization, healthcare costs, short- and long-term disability leave, and lost productivity-related costs for employed adult patients with MPNs in the USA. Patients with ET, PV, and MF, including a subset of patients who experienced thrombotic events, were evaluated separately to provide data on MPN subtypes and the effect of debilitating disease complications.

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Materials and methods

Study design and patients

This retrospective cohort study used data from the Merative™ MarketScan® Commercial and Health and Productivity Management databases. The Commercial database contains the inpatient, outpatient, and outpatient prescription drug claims of employees and dependents covered under fee-for-service and managed care health plans, including exclusive provider organizations, preferred provider organizations, point-of-service plans, indemnity plans, and health maintenance organizations. For a subset of employees in the Commercial database, the Health and Productivity Management database contains data related to workplace absence, short-term disability, long-term disability, and workers' compensation. Because this was a retrospective study that used de-identified administrative claims data, ethics approval and written informed consent from patients were not obtained.

Patients were included in the study if they had at least 1 medical claim for a doctor visit with a diagnosis of MF, PV, or ET between 1 January 2009 and 31 December 2019. Additionally, patients were required to be 18–64 years old as of the diagnosis date (index date) and to have continuous enrollment with medical, pharmacy, and disability insurance for at least 6 months before (pre-index period) and 12 months after (follow-up period) the index date. Patients were excluded if they had evidence of pregnancy during follow-up or a diagnosis of acute myeloid leukemia (any cohort) or MF or myelodysplastic syndrome (PV and ET cohorts only) during the pre-index or follow-up periods. The earliest diagnosis in each calendar year during the study window was considered as a possible index date for each patient, and all other selection criteria were evaluated assuming that index date. For patients who met all eligibility criteria for more than one possible index date, the latest qualifying index date was selected for the study to maximize the number of prevalent patients.

A control cohort was also selected that included patients who met the same medical and disability benefit enrollment criteria between 1 January 2009 and 31 December 2019, but who did not have evidence of MPNs, acute myeloid leukemia, or myelodysplastic syndrome at any point during the study period. For control patients, the index date was randomly assigned according to the distribution of index dates observed in patients with MPNs. MPN cohorts were matched with controls using a hybrid matching approach. Direct matching was used for key variables such as age and sex. Propensity score matching was used for variables such as comorbidity index score category, index year, and employee status. For each disease cohort (MF, PV, and ET), control patients were matched with case patients at a 3:1 ratio.

A subgroup analysis was conducted among patients with a thrombotic event during the 12-month follow-up period. Thrombotic events were identified based on the presence of a nondiagnostic medical claim with an International Classification of Diseases, 9th Revision, Clinical Modification (ICD-9-CM) or 10th Revision (ICD-10-CM) diagnosis code in any position. Among the subset of patients in each disease cohort (MF, PV, and ET) with thrombotic events, control patients with thrombotic events were matched with case patients at a 1:1 ratio.

Outcomes

All study measures were reported for the overall MPN cohorts (MF, PV, and ET) and their respective matched controls, as well as the thrombotic event subcohorts (MF, PV, and ET) and their respective matched controls.

Baseline demographic characteristics were reported as of the index date. The National Cancer Institute-adapted Comorbidity Index was reported during the 6-month pre-index period.

Productivity loss measures were evaluated during the 12-month follow-up period and included the percentage of patients with short-term and/or long-term disability leave, the duration of leave, and the number of periods of leave. Indirect costs associated with disability leave were estimated based on the following equation:

$$\begin{aligned} \textit{Annual indirect costs} = {} & \textit{number of workdays lost} \\ & \times \textit{median daily wage} \\ & \times \textit{0.70 wage adjustment} \end{aligned}$$

The median daily wage was set at US$183.40 based on the 2019 Labor Statistics/Bureau of the Census Current Population Survey Annual Social and Economic Supplement [10]. Disability claims could overlap with the start or end of the 12-month follow-up period, and only days that occurred during the follow-up period were counted, so the number of days of disability leave represents the mean number of days lost to disability in the 12-month follow-up period (not the mean number of days of a short- or long-term disability claim overall). The number of days lost to disability leave accounted for workdays (5 per week) and could total a maximum of 261 d during the 12-month follow-up period.

Healthcare resource utilization and costs were evaluated based on the percentage of patients with inpatient admissions, emergency room visits, outpatient office visits, and other outpatient services; the number of inpatient admissions/visits and (for inpatient admissions) length of stay; and total costs associated with those admissions or visits. Healthcare costs were based on paid amounts of adjudicated claims, including insurer and health plan payments as well as patient cost-sharing in the form of copayment, deductible, and coinsurance. Costs for services provided under capitated arrangements were estimated using payment proxies based on paid claims at the procedure level using the MarketScan Commercial database. The percentage of patients with outpatient pharmacy claims and total outpatient pharmacy costs were also assessed.

Statistical analysis

For comparisons of demographic and clinical characteristics between case and control groups, a standard mean difference of less than 0.1 was considered to be well balanced. All costs were adjusted for inflation using the Consumer Price Index and standardized to 2019 US dollars. Chi-square tests were used to evaluate statistical significance in the differences of categorical variables. Continuous variables were analyzed using *t* tests. Statistical significance was set at $p < 0.05$. Descriptive reporting (including generation of *p* values) was conducted using World Programming System (WPS) version 4.2 (World Programming, London, UK); propensity score matching was conducted using R version 4.3.2.

Results

Patients

In total, 42,844 patients were included in the analysis, including 173 with MF and 519 matched controls, 4477 subjects with PV and 13,431 matched controls, and 6061 subjects with ET and 18,183 matched controls. For each cohort, the standard mean differences in demographic and baseline clinical characteristics between case and control patients were less than 0.1, indicating that case patients were well balanced versus controls on all factors (Table 1).

A total of 689 patients who experienced thrombotic events during the follow-up period were included in the subanalysis, including 19 (11%) cases in the MF cohort, 288 (6%) in the PV cohort, and 382 (6%) in the ET cohort, with equal numbers of matched controls for each condition, for a total of 1378 patients in the analysis. As in the overall cohorts, the subgroups of patients with thrombotic events were well balanced between case patients and controls (data not shown).

Healthcare costs and resource utilization

Total direct healthcare costs and each cost component were significantly higher for each MPN disease type compared with controls (Figure 1). The mean difference between MPN and control groups in total direct annual cost was \$67,456 ($p < 0.001$) for MF, \$10,970 ($p < 0.001$) for PV, and \$22,279 ($p < 0.001$) for ET. In all 3 MPN cohorts, statistically significant differences ($p < 0.01$) were also observed between case and control patients for each cost component (i.e. inpatient, emergency room, outpatient office visits, other outpatient services, and outpatient pharmacy). For the PV and ET cohorts, inpatient costs and other outpatient services costs (excluding office visits and pharmacy costs) were the primary contributors to the direct healthcare costs, whereas for the MF cohort, inpatient costs, other outpatient services, and outpatient pharmacy costs were the primary contributors to the direct healthcare costs.

Among patients with a thrombotic event during follow-up, direct healthcare costs were higher for case compared with control patients for each MPN cohort. For subgroups with thrombotic events in the ET and PV cohorts, total direct costs and component costs were statistically significantly higher for cases versus controls and were numerically higher (statistical significance was not assessed) for cases in the thrombotic event subgroups versus cases in the overall groups (Figure 1). Total direct healthcare costs for ET patients with thrombotic events increased 2.1-fold over controls with thrombotic events and 3.4-fold over ET patients overall. For PV patients with thrombotic events, total direct healthcare costs increased 1.6-fold over the controls with thrombotic events and 3.2-fold over PV patients overall. Direct healthcare costs among patients with ET or PV and thrombotic events were primarily driven by inpatient costs. For MF, total direct costs and individual cost components were numerically higher for cases versus controls, with pharmacy costs the primary driver (statistical significance not assessed due to small sample size; Figure 1). Patients with MF and a thrombotic event also had numerically higher (1.3-fold) direct healthcare costs relative to the full MF group, as well as higher costs for all individual components except other outpatient services.

Table 1. Patient demographics and clinical characteristics at index date.

Patient characteristic	Myelofibrosis		Polycythemia vera		Essential thrombocythemia	
	Control $n=519$	MF $n=173$	Control $n=13,431$	PV $n=4477$	Control $n=18,183$	ET $n=6061$
Age, median (IQR), years	54.0 (47.0–58.0)	54.0 (47.0–58.0)	51.0 (43.0–57.0)	51.0 (43.0–57.0)	48.0 (40.0–55.0)	48.0 (40.0–55.0)
Male, *n* (%)	303 (58.4)	101 (58.4)	11,070 (82.4)	3690 (82.4)	6552 (36.0)	2184 (36.0)
Index year, *n* (%)						
2009–2014	174 (33.5)	58 (33.5)	6603 (49.2)	2210 (49.4)	5261 (28.9)	1766 (29.1)
2015–2019	345 (66.5)	115 (66.5)	6828 (50.8)	2267 (50.6)	12,922 (71.1)	4295 (70.9)
Prevalence/incidence status, *n* (%)*						
Incident	N/A	83 (48.0)	N/A	2667 (59.6)	N/A	3954 (65.2)
Prevalent	N/A	79 (45.6)	N/A	1585 (35.4)	N/A	1745 (28.8)
Unknown	N/A	11 (6.4)	N/A	225 (5.0)	N/A	362 (6.0)
Geographic region, *n* (%)						
Northeast	93 (17.9)	32 (18.5)	1696 (12.6)	569 (12.7)	2713 (14.9)	945 (15.6)
North Central	132 (25.4)	41 (23.7)	3170 (23.6)	1030 (23.0)	3533 (19.4)	1182 (19.5)
South	169 (32.6)	57 (32.9)	5949 (44.3)	2001 (44.7)	8389 (46.1)	2717 (44.8)
West	125 (24.1)	43 (24.9)	2585 (19.2)	872 (19.5)	3514 (19.3)	1208 (19.9)
Unknown	0	0	31 (0.2)	5 (0.1)	34 (0.2)	9 (0.1)
Employment status, *n* (%)						
Full time	494 (95.2)	166 (96.0)	13,148 (97.9)	4375 (97.7)	17,389 (95.6)	5785 (95.4)
Part time	6 (1.2)	2 (1.2)	113 (0.8)	38 (0.8)	239 (1.3)	86 (1.4)
Other	19 (3.7)	5 (2.9)	170 (1.3)	64 (1.4)	555 (3.1)	190 (3.1)
NCI-adapted Comorbidity Index†, mean (SD)	0.1 (0.3)	0.1 (0.3)	0.1 (0.3)	0.1 (0.3)	0.1 (0.3)	0.1 (0.3)

ET: essential thrombocythemia; MF: myelofibrosis; MPN: myeloproliferative neoplasm; N/A: not applicable; NCI: National Cancer Institute; PV: polycythemia vera

*Only for MPN cases. Prevalent patients are those with another diagnosis for the given MPN condition prior to the index date (using full database history back to 2006). Incident patients are those who have at least 12 months of enrollment prior to the index date and no diagnosis for the given MPN condition prior to the index date. Unknown patients are those who do not meet the criteria for prevalent or incident patients (those with no prior diagnosis for the given MPN condition and without 12 months of enrollment prior to the index date).

†Measured during the 6-month pre-index period.

Disability leave and costs

Patients with MPNs were more likely than matched controls to take at least one disability leave during the 12-month follow-up period (Figure 2). The percentage of patients who took short-term or long-term disability leave was statistically significantly greater ($p < 0.001$) for MPN versus control patients in each cohort, except for long-term disability in the MF cohort ($p = 0.103$). Patients with MPNs also took more days of disability leave compared with controls; these differences reached statistical significance ($p < 0.05$) for both short-term and long-term disability in all cohorts (Figure 3).

For the subgroups of patients with thrombotic events in the PV and ET cohorts, significantly higher percentages of cases versus controls took short-term disability leave, and a significantly higher percentage of PV cases versus controls took long-term disability leave (Figure 2). In addition, a substantially larger proportion of patients with PV or ET with thrombotic events took short-term or long-term disability leave relative to the overall cohorts. Similar results were observed for the duration of disability leave, with statistically significantly more days of short-term and long-term disability for cases versus controls in the PV and ET cohorts (Figure 3). In the MF cohort, the subgroup of patients with thrombotic events was too small to assess statistical significance.

In each MPN cohort, indirect costs related to short-term and long-term disability leave were significantly higher for case versus control patients (Table 2). Among subgroups of patients with thrombotic events, patients with PV and ET had significantly higher indirect costs related to short-term disability leave compared with controls. Patients with PV with thrombotic events also had significantly higher indirect costs for long-term disability leave compared with controls.

Discussion

This real-world analysis identified a high economic burden among employed patients with MPNs. Compared with matched controls, patients with MF, PV, or ET had significantly higher direct medical costs versus matched controls, with the highest direct costs associated with MF, followed by ET then PV. For the PV and ET cohorts, inpatient costs and other outpatient services costs (excluding emergency room visits, office visits, and pharmacy costs) were the primary contributors to direct healthcare costs, whereas for the MF cohort, outpatient pharmacy costs were an additional driver of high costs. Furthermore, the presence of thrombotic events increased direct costs by 3.2- and 3.4-fold for patients with PV or ET, respectively, and by 75% for patients with MF. These increases were primarily due to

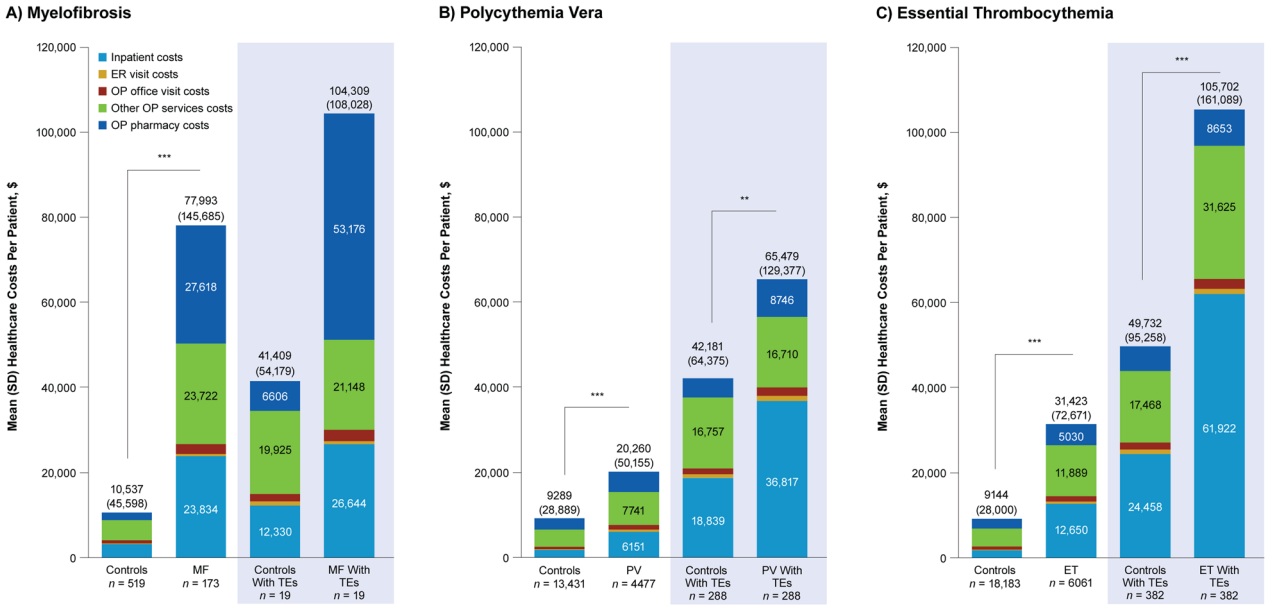

Figure 1. Mean direct healthcare costs per patient. Mean total direct healthcare costs per patient, measured over 12 months of follow-up, are shown for each MPN population ([A] MF, [B] PV, and [C] ET): total case, total control, subgroup case that experienced a TE, and subgroup control that experienced a TE. Total direct healthcare costs are divided into mutually exclusive categories (i.e. inpatient, ER visit, OP office visit, other OP services, OP pharmacy). $^{**}p < 0.01$; $^{***}p < 0.001$. ER: emergency room; ET: essential thrombocythemia; MF: myelofibrosis; OP: outpatient; PV: polycythemia vera; TE: thrombotic event.

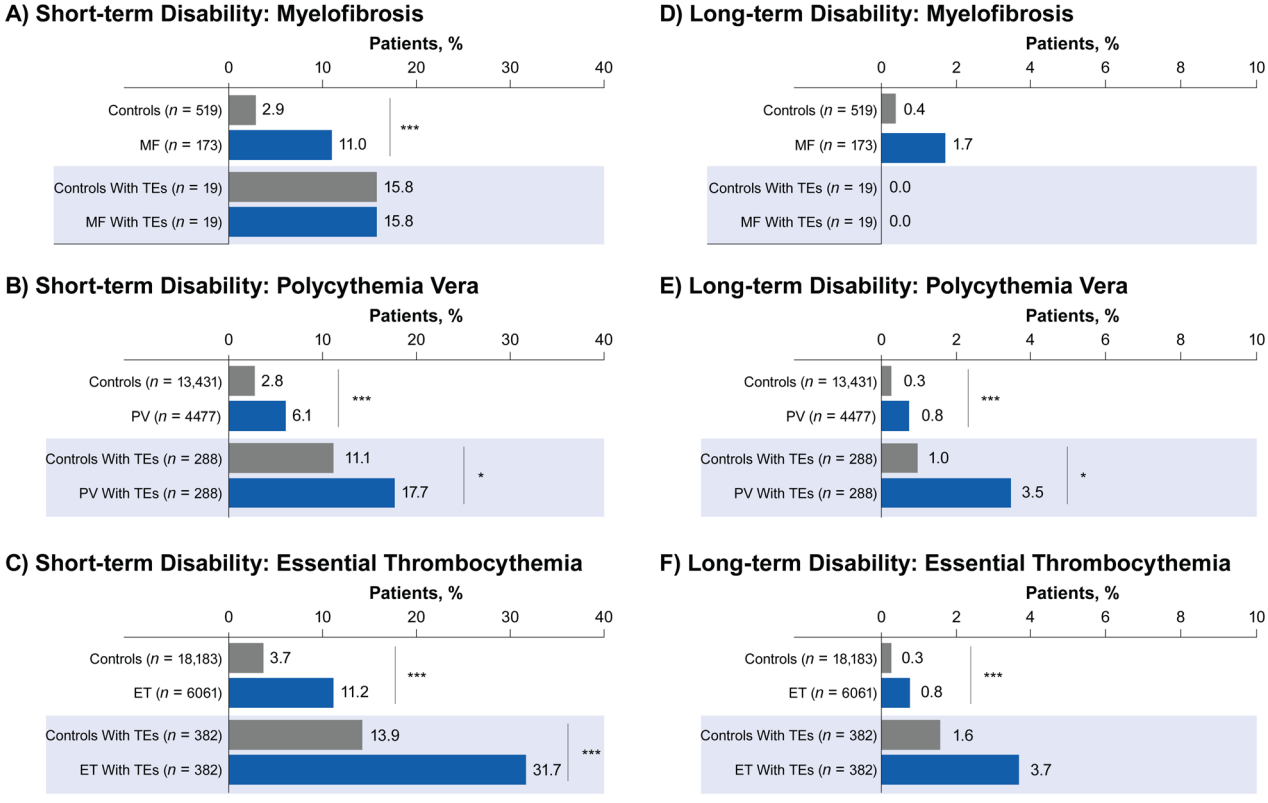

Figure 2. Prevalence of short-term and long-term disability leave per MPN subtype. Mean prevalence of short-term and long-term disability leave is shown for each MPN population ([A, D] MF, [B, E] PV, and [C, F] ET): total case, total control, subgroup case that experienced a TE, and subgroup control that experienced a TE. $^{*}p < 0.05$; $^{***}p < 0.001$. ET: essential thrombocythemia; MF: myelofibrosis; MPN: myeloproliferative neoplasm; PV: polycythemia vera; TE: thrombotic event.

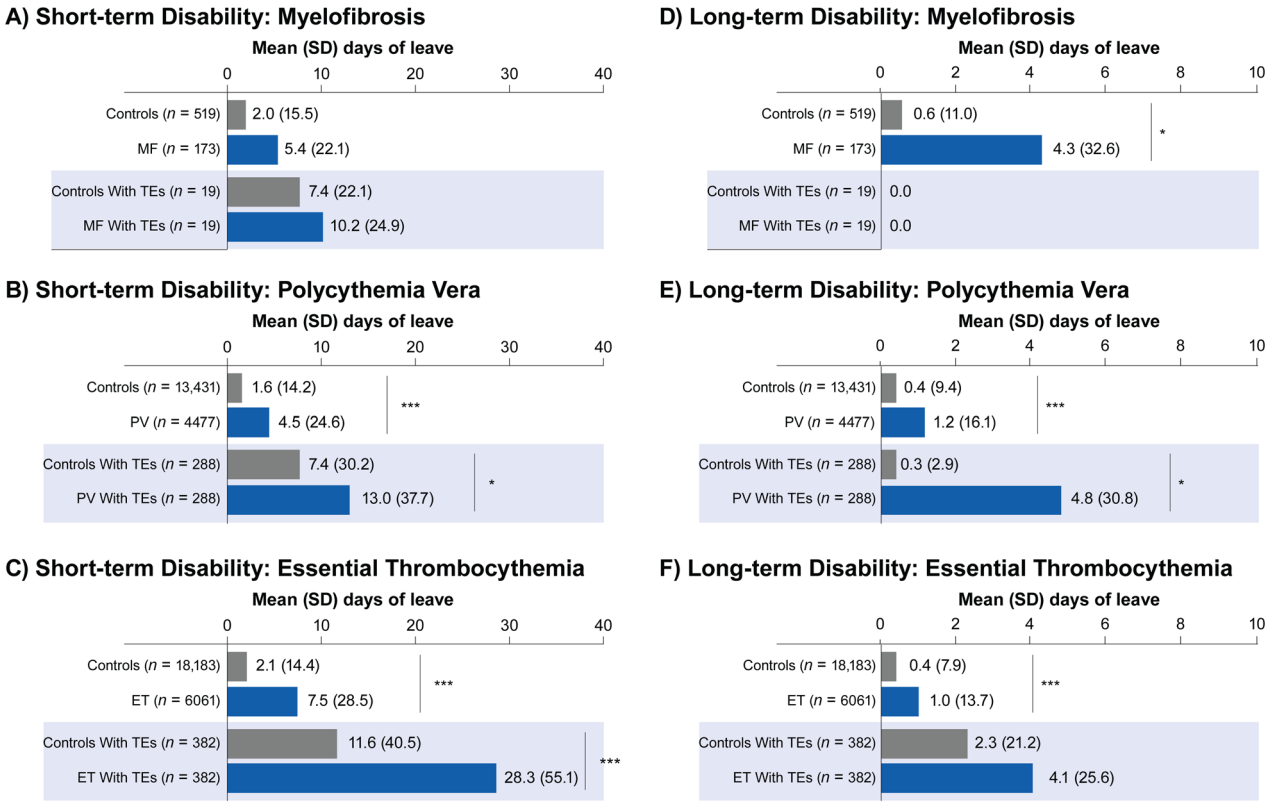

Figure 3. Cumulative duration of short-term and long-term disability leave per MPN subtype. Mean cumulative duration of short-term and long-term disability leave is shown for each MPN population ([A, D] MF, [B, E] PV, and [C, F] ET): total case, total control, subgroup case that experienced a TE, and subgroup control that experienced a TE. *$p < 0.05$; ***$p < 0.001$. ET: essential thrombocythemia; MF: myelofibrosis; MPN: myeloproliferative neoplasm; PV: polycythemia vera; TE: thrombotic event.

Table 2. Indirect costs (2019 US$) due to disability leave.

	Myelofibrosis				Polycythemia vera				Essential thrombocythemia			
Mean (SD) cost	Control n = 519	MF n = 173	Controls with TE n = 19	MF with TE n = 19	Control n = 13,431	PV n = 4477	Controls with TE n = 288	PV with TE n = 288	Control n = 18,183	ET n = 6061	Controls with TE n = 382	ET with TE n = 382
Short-term disability	257 (1995)	697 (2841)*	953 (2843)	1304 (3198)	210 (1826)	573 (3159)†	946 (3871)	1663 (4837)*	263 (1849)	962 (3661)†	1491 (5196)	3629 (7076)†
Long-term disability	72 (1415)	553 (4185)*	N/A	N/A	56 (1205)	153 (2061)†	37 (369)	621 (3950)*	47 (1008)	133 (1758)†	293 (2716)	526 (3289)

ET: essential thrombocythemia; MF: myelofibrosis; N/A: not applicable; PV: polycythemia vera; TE: thrombotic event
Costs associated with absenteeism and disability leave used a wage constant (US$183.40/day) equivalent to the median hourly wage for employed full-time wage and salary workers from the US Bureau of Labor Statistics (2019). Benefits typically pay 70% of wages; therefore, we reported 70% of the wage rate per day as the indirect costs incurred due to disability leave.
*$p < 0.05$ versus controls; †$p < 0.001$ versus controls.

higher outpatient pharmacy costs for patients with MF and increases in inpatient and other outpatient services costs for the PV and ET cohorts. Regarding indirect costs, short-term disability prevalence was significantly higher versus controls for all MPN subtypes, with the highest prevalence of short-term disability leave in the ET cohort. The prevalence of long-term disability leave was also significantly higher versus controls for all MPN subtypes. Furthermore, the presence of thrombotic events nearly doubled the cumulative duration of short-term disability for MF, nearly tripled for PV, and more than tripled for ET; for long-term disability, the presence of thrombotic events quadrupled the cumulative duration for PV and ET.

The highest direct medical costs overall were incurred by ET patients with thrombotic events, with direct medical costs 3.4-fold higher than the full ET group and 2.1-fold higher than controls with thrombotic events. The increase in medical costs in ET patients with a thrombotic event was mostly driven by inpatient costs, which were 2.5-fold higher than controls with a thrombotic event. Because ET patients and

their respective controls were matched for comorbidities, it can be inferred that this increase in inpatient costs was due to ET disease-related complications.

This analysis builds on previous studies by providing additional focus on disability leave usage and the effects of thrombotic events, for which information in the literature is limited. One online survey of patients who had been diagnosed with an MPN for a mean of 6.1 years at the time of the survey found that 38%, 22%, and 15% of those with MF, ET, and PV, respectively, went on medical disability leave [6,7], whereas in this study, 11%, 6%, and 11% of patients with MF, ET, and PV, respectively, went on short-term disability leave. The lower frequencies of disability leave in this analysis may be underestimated due to insufficient follow-up duration; 62.6% of patients in this study were incident patients (with only 12 months of follow-up post-diagnosis), and average time since diagnosis of the prevalent patients was only ~2.5 years. Additionally, patients who changed jobs, retired early, or were on unemployment were included in the survey, whereas such patients may have been excluded in this study due to inclusion criteria that limited patients to those who remained with the same employer for at least 1 year of the follow-up.

Increased short-term and long-term disability leave usage is perhaps most surprising for patients with ET and PV, which are considered to have a less severe phenotype than MF. Although direct explanations based on patient surveys for why patients with ET or PV go on disability leave are not available, we expect several drivers contribute. First, based on this analysis, patients with ET or PV have substantial outpatient and inpatient usage, which can result in large time constraints. Second, the MPN Landmark survey reported that bothersome symptoms are common in patients with ET or PV and are often cause for calling in sick to work when severe [2]. Third, data from the MPN Landmark survey also showed a substantial strain of frequent phlebotomy in patients with PV, including fatigue, negative impact on quality of life, reduced productivity, and increased amount of sick days [11]. Finally, the same survey found that a sizeable proportion of patients across MPN subtypes spent multiple days in bed in the past 30 d (25% patients with ET and 23% with PV spent ≥1 d in bed), and 45% with ET and 52% with PV had activities limited due to pain and discomfort [2], indicating limits on patient availability and productivity.

This analysis also builds on previous economic studies in MPN by providing information on the effect of thrombotic events on healthcare costs, for which there is limited information in the literature. Specifically, the Canadian cohort study found that patients with PV and ET with a history of venous thrombosis had higher costs [8]. Furthermore, the MPN Landmark survey showed that patients with a history of thrombotic events were more likely to take disability leave (36%) compared with those without a thrombotic event (22%) [2]. These findings are in alignment with this study's results showing the significant impact of thrombotic events on increasing the direct and indirect healthcare costs across MPN subtypes. It should be underlined that the costs reported in our study are total direct and indirect costs and thus reflect the combined high costs and healthcare resource utilization associated with thrombotic events and the underlying MPNs, which are chronic diseases with burdensome symptoms that often require long-term management [2,5–9]. Because the higher costs and need for disability leave of MPNs versus controls are observed with or without thrombotic events, at least a portion of the cost difference can be attributed to expenses related to managing MPNs in general.

Limitations of this study are based on its retrospective observational design. Data in the MarketScan databases were used as entered by healthcare providers for billing purposes and may be subject to miscoding or inaccurate/incomplete records. In addition, diagnostic criteria were not standardized for this analysis, which could result in variability among patient cohorts. However, these limitations would likely affect all study groups. This analysis only included patients who worked for employers who provided disability insurance and thus did not include patients who were working without disability benefits or who were self-employed. Also, patients who died early in the course of MPN diseases were not included in the analysis due to the criteria of continuous benefit enrollment. Based on the previous evidence that most disability leave by MPN patients did not start until about 2 years after disease diagnosis, the frequency and length of disability leave, as well as the estimated indirect costs, are likely underestimated. Additionally, no corrections for multiple testing were conducted. Finally, we do not have data regarding the etiology of MF in our population (i.e. primary, post-PV, or post-ET), which could have an impact on the observed results. In addition, the relatively high rate of thrombosis in the MF cohort was unexpected and may have contributed to the high costs observed in our analysis.

In conclusion, this retrospective analysis of claims data in the USA demonstrated significantly higher healthcare costs, higher incidence of medical disability leave, and longer duration of disability leave for patients with MPNs compared with control patients.

Direct costs and the frequency and duration of disability leave were also higher for patients with PV and ET who experienced a thrombotic event compared with those who did not experience an event and compared with control patients who did experience an event. These findings support prior studies that demonstrated an increased economic burden for patients with MPNs relative to control patients and add to this growing knowledge base by providing information about indirect costs associated with disability leave and the effects of thrombotic events. Furthermore, the data across cohorts showed that in addition to MF, ET and PV are also associated with significant direct and indirect healthcare costs, which has important implications given the broader patient population affected. As the treatment landscape evolves for MPNs, additional analyses will be warranted to understand the effect of treatments on resource utilization and work productivity and the associated costs, as well as any relevant differences among MPN subtypes.

Acknowledgments

Writing assistance was provided by ICON (Blue Bell, PA) and was funded by Incyte Corporation (Wilmington, DE).

Disclosure statement

JY and EB are employees and stockholders of Incyte. JN and TM were employees of Merative at the time the study was conducted. MJ is an employee of Merative, a paid consultant of Incyte.

Funding

Incyte Corporation sponsored this study. Authors employed by Incyte participated in study design development, analysis, and interpretation of the data, and writing the manuscript. The authors prepared the manuscript with sponsor-funded writing assistance. All authors had full access to all study data and had the final responsibility to submit the manuscript for publication.

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Data availability statement

The data that support the findings of this study are available from Merative. Restrictions apply to the availability of these data, which were used under license for this study.

References

[1] Arber DA, Orazi A, Hasserjian R, et al. The 2016 revision to the World Health Organization classification of myeloid neoplasms and acute leukemia. Blood. 2016; 127(20):2391–2405. doi:10.1182/blood-2016-03-643544
[2] Mesa R, Miller CB, Thyne M, et al. Myeloproliferative neoplasms (MPNs) have a significant impact on patients' overall health and productivity: the MPN Landmark survey. BMC Cancer. 2016;16(1):167. doi:10.1186/s12885-016-2208-2
[3] Price GL, Davis KL, Karve S, et al. Survival patterns in United States (US) Medicare enrollees with non-CML myeloproliferative neoplasms (MPN). PLoS One. 2014; 9(3):e90299. doi:10.1371/journal.pone.0090299
[4] Rungjirajittranon T, Owattanapanich W, Ungprasert P, et al. A systematic review and meta-analysis of the prevalence of thrombosis and bleeding at diagnosis of Philadelphia-negative myeloproliferative neoplasms. BMC Cancer. 2019;19(1):184. doi:10.1186/s12885-019-5387-9
[5] Mehta J, Wang H, Fryzek JP, et al. Health resource utilization and cost associated with myeloproliferative neoplasms in a large United States health plan. Leuk Lymphoma. 2014;55(10):2368–2374. doi:10.3109/10428194.2013.879127
[6] Yu J, Parasuraman S, Paranagama D, et al. Impact of myeloproliferative neoplasms on patients' employment status and work productivity in the United States: results from the Living With MPNs survey. BMC Cancer. 2018;18(1):420. doi:10.1186/s12885-018-4322-9
[7] Yu J, Paranagama D, Geyer HL, et al. Relationship between symptom burden and disability leave among patients with myeloproliferative neoplasms (MPNs): findings from the Living With MPN patient survey. Ann Hematol. 2019;98(5):1119–1125. doi:10.1007/s00277-019-03610-4
[8] Bankar A, Zhao H, Iqbal J, et al. Healthcare resource utilization in myeloproliferative neoplasms: a population-based study from Ontario, Canada. Leuk Lymphoma. 2020; 61(8):1908–1919. doi:10.1080/10428194.2020.1749607
[9] Christensen SF, Svingel LS, Kjaersgaard A, et al. Healthcare resource utilization in patients with myeloproliferative neoplasms: a Danish nationwide matched cohort study. Eur J Haematol. 2022;109(5):526–541. doi:10.1111/ejh.13841
[10] US Census Bureau. March 2019 Annual Social and Economic Supplement (ASEC). Washington, DC: Bureau of the Census for the Bureau of Labor Statistics; 2019.
[11] Fazal S, Miller CB, Mascarenhas JO, et al. Phlebotomy frequency and quality of life and productivity in patients with polycythemia vera: results from the MPN Landmark survey in the United States. Blood. 2015; 126(23):5184–5184. doi:10.1182/blood.V126.23.5184.5184