

# Cancer Screening for Dermatomyositis: A Survey of Indirect Costs, Burden, and Patient Willingness to Pay

Katherine I. Jicha, MD; Christopher G. Bazewicz, MD; Matthew F. Helm, MD; Melissa Butt, PhD; Cassidy Shumaker, MPH; Galen T. Foulke, MD

## PRACTICE POINTS

- Dermatomyositis (DM) is associated with an increased risk for malignancy. Patient perspective needs to be considered in developing cancer screening guidelines for patients with DM, particularly given the similar efficacy of available screening modalities.
- Current modalities for cancer screening in DM include whole-body positron emission tomography/computed tomography (PET/CT) and a conventional cancer screening panel (CSP), which includes a battery of tests typically requiring multiple visits. Patients may find the simplicity of PET/CT more preferable than the more complex CSP.
- Indirect costs of cancer screening include missed work, travel and childcare expenses, and lost wages. Conventional cancer screening has greater indirect costs than PET/CT.

Dermatomyositis (DM) is a rare idiopathic inflammatory myopathy (IIM) associated with an increased risk for malignancy. Although cancer screening is recommended, no consensus guidelines

currently exist. Whole-body positron emission tomography/computed tomography (PET/CT) has similar cost and efficacy to a more traditional conventional cancer screening panel (CSP). Our study sought to characterize patients' perspective of cancer screening and the indirect costs to patients. We conducted a survey of patients recently diagnosed with DM who were undergoing or had recently undergone a CSP. Patient values and indirect costs need to be considered in choosing a screening modality. This study contributes to a greater understanding of patients' experience of cancer screening in DM, which should be taken into consideration when developing consensus guidelines for cancer screening.

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**D**ermatomyositis (DM) is an uncommon idiopathic inflammatory myopathy (IIM) characterized by muscle inflammation; proximal muscle weakness; and dermatologic findings, such as the heliotrope eruption and Gottron papules.<sup>1-3</sup> Dermatomyositis is associated with an increased malignancy risk compared to other IIMs, with a 13% to 42% lifetime risk for malignancy development.<sup>4,5</sup> The incidence for malignancy peaks during the first year following diagnosis and falls

Dr. Jicha is from the Department of Dermatology, UNC School of Medicine, Chapel Hill, North Carolina. Drs. Bazewicz, Helm, Butt, and Foulke, as well as Cassidy Shumaker, are from the Department of Dermatology, Penn State Health Milton S. Hershey Medical Center, Hershey, Pennsylvania. This work was supported by the James and Joyce Marks Educational Endowment. They had no role in the design of the study or collection, analysis, and interpretation of data or in writing the manuscript. The Penn State Clinical & Translational Research Institute, Pennsylvania State University CTSA, provided funding for the use of REDCap. National Institutes of Health/National Center for Advancing Translational Sciences grant number UL1 TR002014.

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Supplemental information—the Demographics Questionnaire and Independent Questionnaire—is available online at [www.mdedge.com/dermatology](http://www.mdedge.com/dermatology). This material has been provided by the authors to give readers additional information about their work.

Correspondence: Katherine I. Jicha, MD, UNC School of Medicine, 321 S Columbia St, Chapel Hill, NC 27516 (Katherine.jicha@gmail.com).

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gradually over 5 years but remains increased compared to the general population.<sup>6-11</sup> Adenocarcinoma represents the majority of cancers associated with DM, particularly of the ovaries, lungs, breasts, gastrointestinal tract, pancreas, bladder, and prostate. The lymphatic system (non-Hodgkin lymphoma) also is overrepresented among cancers in DM.<sup>12</sup>

Because of the increased malignancy risk and cancer-related mortality in patients with DM, cancer screening generally is recommended following diagnosis.<sup>13,14</sup> However, consensus guidelines for screening modalities and frequency currently do not exist, resulting in widely varying practice patterns.<sup>15</sup> Some experts advocate for a conventional cancer screening panel (CSP), as summarized in Table 1.<sup>15-18</sup> These tests may be repeated annually for 3 to 5 years following the diagnosis of DM. Although the use of myositis-specific antibodies (MSAs) recently has helped to risk-stratify DM patients, up to half of patients are MSA negative,<sup>19</sup> and broad malignancy screening remains essential. Individualized discussions with patients about their risk factors, screening options, and risks and benefits of screening also are strongly encouraged.<sup>19-22</sup> Studies of the direct costs and effectiveness of streamlined screening with positron emission tomography/computed tomography (PET/CT) compared with a CSP have shown similar efficacy and lower out-of-pocket costs for patients receiving PET/CT imaging.<sup>16-18</sup>

The goal of our study was to further characterize patients' perspectives and experience of cancer screening in DM as well as indirect costs, both of which must be taken into consideration when developing consensus guidelines for DM malignancy screening. Inclusion of patient voice is essential given the similar efficacy of both screening methods. We assessed the indirect costs (eg, travel, lost work or wages, childcare) of a CSP in patients with DM. We theorized that the large quantity of tests involved in a CSP, which are performed at various locations on multiple days over the course of several years, may have substantial costs to patients beyond the copay and deductible. We also sought to measure patients' perception of the burden associated with an annual CSP, which we defined to participants as the inconvenience or unpleasantness experienced by the patient, compared with an annual whole-body PET/CT. Finally, we examined the relative value of these screening methods to patients using a willingness-to-pay (WTP) analysis.

**Materials and Methods**

*Patient Eligibility*—Our study included Penn State Health (Hershey, Pennsylvania) patients 18 years or older with a recent diagnosis of DM—*International Classification of Diseases, Ninth Revision* code 710.3 or *International Classification of Diseases, Tenth Revision* codes M33.10 or M33.90—who were undergoing or had recently completed a CSP. Patients were excluded from the study if they had a concurrent or

preceding diagnosis of malignancy (excluding nonmelanoma skin cancers) or had another IIM. The institutional review board at Penn State Health College of Medicine approved the study. Data for all patients were prospectively obtained.

*Survey Design*—A survey was generated to assess the burden and indirect costs associated with a CSP, which was modified from work done by Tchuente et al<sup>23</sup> and Teni et al.<sup>24</sup> Focus groups were held in 2018 and 2019 with patients who met our inclusion criteria with the purpose of refining the survey instrument based on patient input. A summary explanation of research was provided to all participants, and informed consent was obtained. Patients were compensated for their time for focus groups. Audio of each focus group was then transcribed and analyzed for common themes. Following focus group feedback, a finalized survey was generated for assessing burden and indirect costs (survey instrument provided in the Supplementary Information). REDCap (Vanderbilt University), a secure web application, was used to construct the finalized survey and to collect and manage data.<sup>25</sup>

Patients who fit our inclusion criteria were identified and recruited in multiple ways. Patients with appointments at the Penn State Milton S. Hershey Medical Center Department of Dermatology were presented with the opportunity to participate, Penn State Health records with the appropriate billing codes were collected and patients were contacted, and an advertisement for the study was posted on StudyFinder. Surveys constructed on REDCap were then sent electronically to patients who agreed to participate in the study. A second summary explanation of research was included on the first page of the survey to describe the process.

**TABLE 1. Conventional Cancer Screening Panel for Dermatomyositis<sup>15-18</sup>**

History and physical examination
Laboratory tests (CBC, serum chemistry panel)
CT of the chest, abdomen, and pelvis without contrast
Tumor markers: <ul style="list-style-type: none"> <li>• CA19-9</li> <li>• Carcinoembryonic antigen</li> <li>• CA125 (women)</li> </ul>
Prostate-specific antigen (men)
Additional tests for women: <ul style="list-style-type: none"> <li>• Transvaginal ultrasound</li> <li>• Cervical cytopathology</li> <li>• Mammography</li> </ul>
Age-appropriate cancer screenings

Abbreviations: CA, cancer antigen; CBC, complete blood cell count; CT, computed tomography.

The survey had 3 main sections. The first section collected demographic information. In the second section, we surveyed patients regarding the various aspects of a CSP that focus groups identified as burdensome. In addition, patients were asked to compare their feelings regarding an annual CSP vs whole-body PET/CT for a 3-year period utilizing a rating scale of strongly disagree, somewhat disagree, somewhat agree, and strongly agree. This section also included a willingness-to-pay (WTP) analysis for each modality. We defined WTP as the maximum out-of-pocket cost that the patient would be willing to pay to receive testing, which was measured in a hypothetical scenario where neither whole-body PET/CT nor CSP was covered by insurance.<sup>26</sup> Although WTP may be influenced by external factors such as patient income, it can serve as a numerical measure of how much the patient values each service. Furthermore, these external factors become less relevant when comparing the relative value of 2 separate tests, as such factors apply equally in both scenarios. In the third section of the survey, patients were queried regarding various indirect costs associated with a CSP. Descriptions for a CSP and whole-body PET/CT, including risks and benefits, were provided to allow patients to make informed decisions.

**Statistical Analysis**—Because of the rarity of DM and the subsequently limited sample size, summary and descriptive statistics were utilized to characterize the sample and identify patterns in the results. Continuous variables are presented with means and standard deviations, and proportions are presented with frequencies and percentages. All analyses were done using SAS Version 9.4 (SAS Institute Inc).

## Results

**Patient Demographics**—Fifty-four patients were identified using StudyFinder, physician referral, and search of the electronic health record. Nine patients agreed to take part in the focus groups, and 27 offered email addresses to be contacted for the survey. Of those 27 patients, 16 (59.3%) fit our inclusion criteria and completed the survey. Patient demographics are detailed in Table 2. The mean age was 55 years, and most patients were White (88% [14/16]), female (81% [13/16]), and had at least a bachelor's degree (69% [11/16]). Most patients (69% [11/16]) had an annual income of less than \$50,000, and half (50% [8/16]) were employed. All patients had been diagnosed with DM in or after 2013. Two patients were diagnosed with basal cell carcinoma during or after cancer screening.

**Patient Preference for Screening and WTP**—A majority (81% [13/16]) of patients desired some form of screening for occult malignancy following the diagnosis of DM, even in the hypothetical situation in which screening did not provide survival benefit (Figure 1). Twenty-five percent (4/16) of patients expressed that a CSP was burdensome, and 12.5% of patients (2/16) missed a CSP appointment; all of these patients rescheduled or were planning to reschedule. Assuming that both

**TABLE 2. Characteristics of Sample Population (N=16)<sup>a</sup>**

Variable	Patients with DM
Mean age (SD), y	55.3 (16.1)
Sex, n (%)	
Female	13 (81.3)
Male	3 (18.8)
Race, n (%)	
White/Caucasian	14 (87.5)
Black or African American	1 (6.3)
Other	1 (6.3)
Education, n (%)	
High school diploma or GED	3 (18.8)
Some college/no degree	2 (12.5)
Bachelor's degree	6 (37.5)
Master's degree	3 (18.8)
Doctoral degree	2 (12.5)
Annual income, n (%)	
<\$20,000	4 (25.0)
\$20,000–\$34,999	3 (18.8)
\$35,000–\$49,999	4 (25.0)
\$50,000–\$74,999	3 (18.8)
\$150,000–\$199,999	1 (6.3)
≥\$200,000	1 (6.3)
Employment status, n (%)	
Full-time student	1 (6.3)
Full-time employed	5 (31.3)
Part-time employed	3 (18.8)
On disability/social security income	2 (12.5)
On pension/retired	4 (25.0)
Unemployed	1 (6.3)
Year diagnosed with DM	
2013	4 (25.0)
2016	6 (37.5)
2017	3 (18.8)
2018	3 (18.8)
Cancer diagnosis, n (%)	
No	14 (87.5)
Yes—basal cell carcinoma	2 (12.5)

Abbreviations: DM, dermatomyositis; GED, General Educational Development test.

<sup>a</sup>Variables with no responses are not listed.

screening methods had similar predictive value in detecting malignancy, all 16 patients felt annual whole-body PET/CT for a 3-year period would be less burdensome than a CSP, and most (73% [11/15]) felt that it would decrease the likelihood of missed appointments. Overall, 93% (13/14) of patients preferred whole-body PET/CT over a CSP when given the choice between the 2 options (Figure 2). This preference was consistent with the patients' WTP for these tests; patients reliably reported that they would pay more for annual whole-body PET/CT than for a CSP (Figure 3). Specifically, 75% (12/16) and 38% (6/16) of patients were willing to spend \$250 or more and \$1000 or more for annual whole-body PET/CT, respectively, compared with 56% (9/16) and 19% (3/16), respectively, for an annual CSP. Many patients (38% [6/16]) reported that they would not be willing to pay any out-of-pocket cost for a CSP compared with 13% (2/16) for PET/CT.

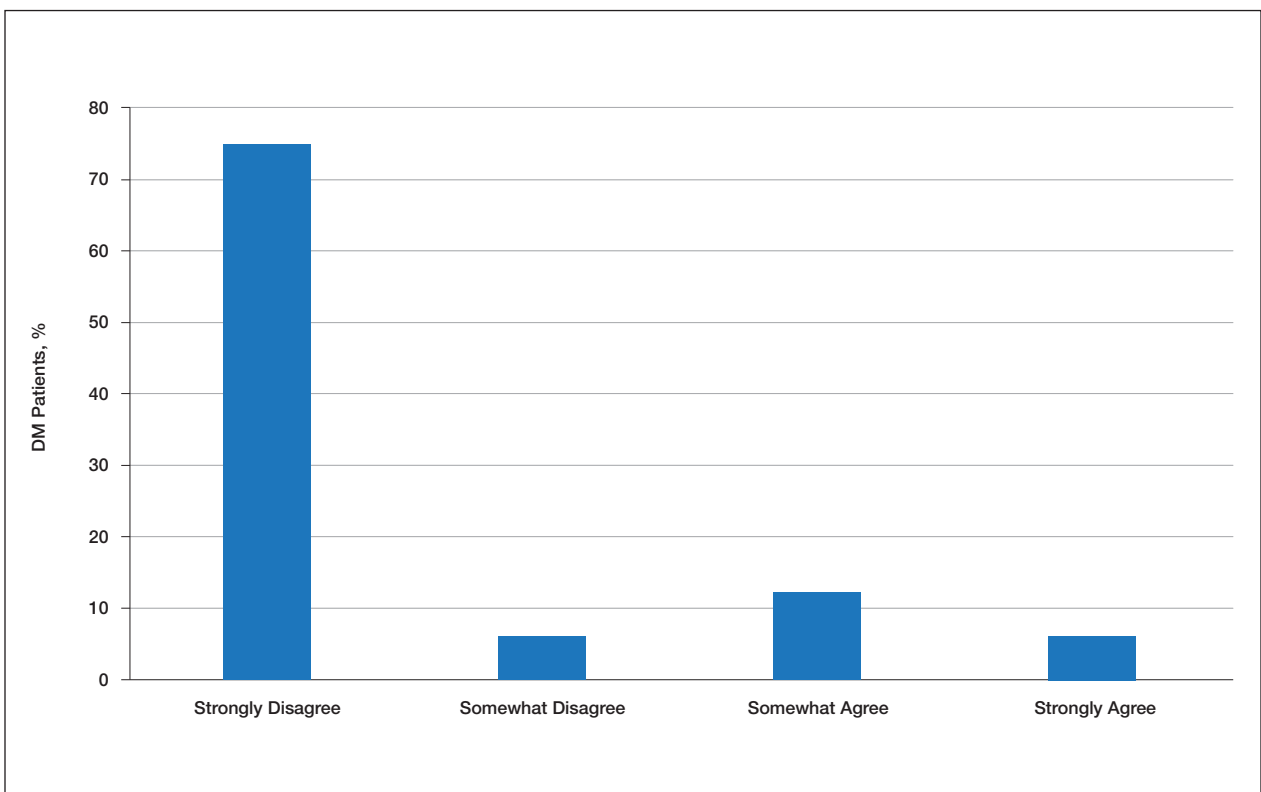
*Indirect Costs of Screening for Patients*—Indirect costs incurred by patients undergoing a CSP are summarized in Table 3. Specifically, a large percentage of employed patients missed work (63% [5/8]) or had family miss work (38% [3/8]), necessitating the use of vacation and/or sick days to attend CSP appointments. A subset (25% [2/8]) lost income (average, \$1500), and 1 patient reported that a family member lost income due to attending a CSP

appointment. Most (75% [12/16]) patients also incurred substantial transportation costs (average, \$243), with 1 patient spending \$1000. No patients incurred child or elder care costs. One patient paid a small sum for lodging/meals while traveling to attend a CSP appointment.

**Comment**

Patients with DM have an increased incidence of malignancy, thus cancer screening serves a crucial role in the detection of occult disease.<sup>13</sup> Up to half of DM patients are MSA negative, and most cancers in these patients are found with blind screening. Whole-body PET/CT has emerged as an alternative to a CSP. Evidence suggests that it has similar efficacy in detecting malignancy and may be particularly useful for identifying malignancies not routinely screened for in a CSP. In a prospective study of patients diagnosed with DM and polymyositis (N=55), whole-body PET/CT had a positive predictive value of 85.7% and negative predictive value for detecting occult malignancy of 93.8% compared with 77.8% and 95.7%, respectively, for a CSP.<sup>17</sup>

The results of our study showed that cancer screening is important to patients diagnosed with DM and that most of these patients desire some form of cancer screening. This finding held true even when patients were presented with a hypothetical situation in which screening was proven to have no survival benefit.



**FIGURE 1.** Patient preference regarding cancer screening in general following the diagnosis of dermatomyositis (“Would you rather have no cancer screenings at all to look for cancer?”)(N=16).

Based on focus group data, this desire was likely driven by the fear generated by not knowing whether cancer is present, as reported by the following DM patients:

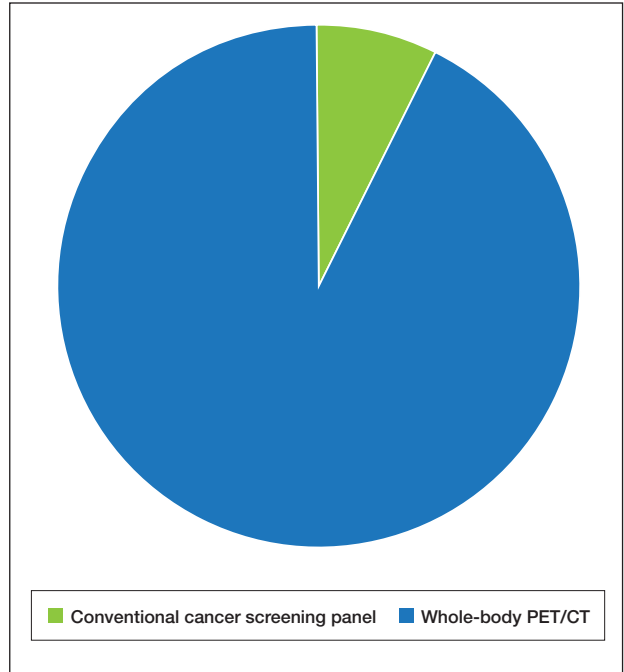
“I mean [cancer screening] is peace of mind. It is ultimately worth it. You know, better than . . . not doing the screenings and finding 3 years down the road that you have, you know, a serious problem . . . you had the cancer, and you didn’t have the screenings.” (DM patient 1)

“I would rather know than not know, even if it is bad news, just tell me. The sooner the better, and give me the whole spiel . . . maybe all the screenings don’t need to be done, done so much, so often afterwards if the initial ones are ok, but I think too, for peace of mind, I would rather know it all up front.” (DM patient 2)

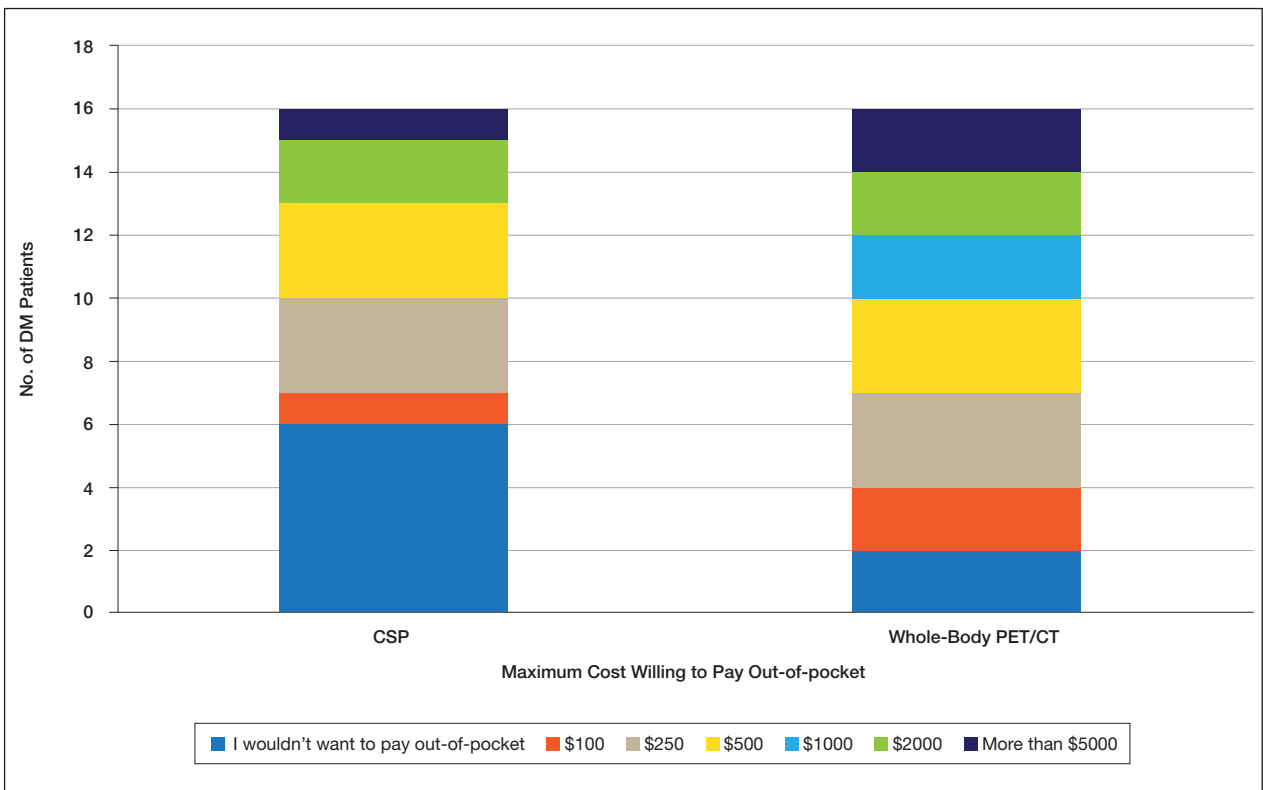
Further, when presented with the hypothetical situation that insurance would not cover screenings, a few patients remarked they would relocate to obtain them:

“I would find a place where the screenings were done. I’d move.” (DM patient 4)

“If it was just sky high and [insurance companies] weren’t willing to negotiate, I would consider moving.” (DM patient 3).



**FIGURE 2.** Patient preference between annual whole-body positron emission tomography/computed tomography (PET/CT) and a conventional cancer screening panel (n=14).



**FIGURE 3.** Patient willingness to pay out-of-pocket for whole-body positron emission tomography/computed tomography (PET/CT) vs a conventional cancer screening panel (CSP) in patients with dermatomyositis (DM)(N=16).

**TABLE 3. Indirect Costs for Patients Associated With a Conventional Cancer Screening Panel**

Item	Patients with DM, n (%)	Mean (SD)	Range
Missed work (n=8)	5 (62.5)	4.2 (3.1)	1–8
Used vacation or sick days (n=8)	5 (62.5)	8.6 (6.1)	2–15
Family missed work (n=8)	3 (37.5)	1 (0.0)	1–1
Family used vacation or sick days (n=8)	3 (37.5)	1.3 (0.6)	1–2
Lost income (n=8)	2 (25.0)	\$1500 (\$707.12)	\$1000–\$2000
Family lost income (n=8)	1 (12.5)	Not reported	Not reported
Transportation cost (n=16)	12 (75.0)	\$242.92 (\$366.30)	\$15–\$1000
Child/elder care costs (n=16)	0 (0.0)	NA	NA
Lodging/meal costs (n=16)	1 (6.3)	\$25 (NA)	NA

Abbreviations: DM, dermatomyositis; NA, not applicable.

Sentiments such as these emphasize the importance and value that DM patients place on being screened for cancer and also may explain why only 25% of patients felt a CSP was burdensome and only 13% reported missing appointments, all of whom planned on making them up at a later time.

When presented with the choice of a CSP or annual whole-body PET/CT for a 3-year period following the diagnosis of DM, all patients expressed that whole-body PET/CT would be less burdensome. Most preferred annual whole-body PET/CT despite the slightly increased radiation exposure associated and thought that it would limit missed appointments. Accordingly, more patients responded that they would pay more money out-of-pocket for annual whole-body PET/CT. Given that WTP can function as a numerical measure of value, our results showed that patients placed a higher value on whole-body PET/CT compared with a CSP. The indirect costs associated with a CSP also were substantial, particularly regarding missed work, use of vacation and/or sick days, and travel expenses, which is particularly important because most patients reported an annual income less than \$50,000.

The direct costs of a CSP and whole-body PET/CT have been studied. Specifically, Kundrick et al<sup>18</sup> found that whole-body PET/CT was less expensive for patients (by approximately \$111) out-of-pocket compared with a CSP, though cost to insurance companies was slightly greater. The present study adds to these findings by better illustrating the burden and indirect costs that patients experience while undergoing a CSP and by characterizing the patient's perception and preference of these 2 screening methods.

Limitations of our study include a small sample size willing to complete the survey. There also was a predominance of White and female participants, partially attributed to the greater number of female patients who develop DM compared to male patients. However, this still may limit applicability of this study to males and patients of other races. Another limitation includes recall bias on survey responses, particularly regarding indirect costs incurred with a CSP. A final limitation was that only patients with a recent diagnosis of DM who were actively undergoing screening or had recently completed malignancy screening were included in the study. Given that these patients were receiving (or had completed) exclusively a CSP, patients were comparing their personal experience with a described experience. In addition, only 2 patients were diagnosed with cancer—both with basal cell carcinoma diagnosed on physical examination—which may have influenced their perception of a CSP, given that nothing was found on an extensive number of tests. However, these patients still greatly valued their screening, as evidenced in the survey.

### Conclusion

Our study contributes to a better understanding of the costs patients face while undergoing malignancy screening for DM and highlights the great value patients assign to undergoing screening regardless of impact on outcome. Our study also shows a preference for streamlined testing, which whole-body PET/CT may represent. Patients incurred substantial indirect costs with a CSP and perceived that a single test, such as whole-body PET/CT, would be less burdensome and result in better compliance with screening. As groups work to establish consensus guidelines for cancer screening in DM, it is



important to include the patient's perspective. Ultimately, prospective trials comparing these modalities are needed, at which time the efficacy, direct and indirect costs, and burden of each modality can be compared.

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