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ORIGINAL ARTICLE

Diagnosing and characterizing inflammatory myopathies at an Australian tertiary public hospital: Resource utilization and direct healthcare costs

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Abstract

Aim: To determine the direct health service costs and resource utilization associated with diagnosing and characterizing idiopathic inflammatory myopathies (IIMs), and to assess for limitations and diagnostic delay in current practice.

Methods: A retrospective, single-center cohort analysis of all patients diagnosed with IIMs between January 2012 and December 2021 in a large tertiary public hospital was conducted. Demographics, resource utilization and costs associated with diagnosing IIM and characterizing disease manifestations were identified using the hospital's electronic medical record and Health Intelligence Unit, and the Medicare Benefits Schedule.

Results: Thirty-eight IIM patients were identified. IIM subtypes included dermatomyositis (34.2%), inclusion body myositis (18.4%), immune-mediated necrotizing myopathy (18.4%), polymyositis (15.8%), and anti-synthetase syndrome (13.2%). The median time from symptom onset to diagnosis was 212 days (IQR: 118–722), while the median time from hospital presentation to diagnosis was 30 days (8–120). Seventy-six percent of patients required emergent hospitalization during their diagnosis, with a median length of stay of 8 days (4–15).

The average total cost of diagnosing IIM was $15 618 AUD (STD: 11331) per patient. Fifty percent of patients underwent both MRI and EMG to identify affected muscles, 10% underwent both pan-CT and PET-CT for malignancy detection, and 5% underwent both open surgical and percutaneous muscle biopsies. Autoimmune serology was unnecessarily repeated in 37% of patients.

Conclusion: The diagnosis of IIMs requires substantial and costly resource use; however, our study has identified potential limitations in current practice and highlighted the need for streamlined diagnostic algorithms to improve patient outcomes and reduce healthcare-related economic burden.

Shereen Oon and Jessica Day contributed equally.

KEYWORDS
direct cost, healthcare resource, inflammatory myopathy, myositis, retrospective

1 | INTRODUCTION

Idiopathic inflammatory myopathies (IIM), commonly known as myositis, are a heterogenous group of chronic autoimmune diseases characterized primarily by the potential for skeletal muscle inflammation. Subtypes include dermatomyositis (DM), inclusion body myositis (IBM), immune-mediated necrotizing myopathy (IMNM), anti-synthetase syndrome (ASyS), polymyositis (PM) and overlap myositis (OM), which occurs in the context of other connective tissue diseases. While uncommon, with an annual incidence in Australia of approximately 8 per million person-years, IIMs confer substantial morbidity and disability, and may be life-limiting,[1,2] with the potential for life-threatening bulbar, cardiac, respiratory, and gastrointestinal muscle involvement. Extra-muscular manifestations may include rash, interstitial lung disease, arthritis, and constitutional symptoms. IIMs are also associated with malignancy, with a significantly increased risk, particularly in patients with DM.[3]

While there have been significant advances in serological, radiographic and histological characterization of IIM in the past decades, there remains no standardized diagnostic approach or commonly accepted diagnostic criteria. Patients commonly undergo a series of investigations including electromyography (EMG), musculoskeletal magnetic resonance imaging (MRI), muscle biopsy, respiratory function tests, and malignancy screening tests, many of which are difficult to obtain in busy public hospitals and lead to diagnostic delays and prolonged inpatient admissions.

Several North American and European studies have shown that IIMs impose a substantial economic burden on healthcare systems.[4–8] Between 2002 and 2012, there were 9687 admissions in the United States with a primary diagnosis of dermatomyositis, which cost the healthcare system over $168 million USD.[9] To our knowledge, an analysis of direct healthcare costs associated specifically with a *diagnosis* of IIM in a healthcare system is yet to be published.

Our study aimed to (i) determine the direct healthcare costs and resource utilization associated with diagnosing and characterizing IIMs and (ii) assess diagnostic delay in our current practice.

2 | METHODS

2.1 | Study population

A retrospective single-center cohort analysis was conducted on patients diagnosed with IIM between 1 January 2012 and 31 December 2021 at The Royal Melbourne Hospital (RMH), Victoria, Australia; a large tertiary metropolitan hospital that provides specialized inpatient and outpatient rheumatology care.

To be included in the study, individuals needed to: (i) be ≥18 years of age; (ii) have either ≥1 inpatient admission associated with an IIM diagnosis, or ≥2 outpatient visits associated with an IIM diagnosis; (iii) present with symptoms suggestive of an IIM between 2012 and 2021 (including proximal girdle and/or truncal weakness, dysphagia, dyspnoea, rash, arthralgias, chest pain and/or constitutional symptoms); and (iv) to have completed their diagnostic work up at the RMH.

The study cohort was identified by searching both the current *Epic* electronic medical record (EMR) and a hospital network drive containing historical Rheumatology outpatient letters that predated EMR implementation in August 2020. As International Classification of Diseases (ICD) codes are often too rigid to reflect clinical subtleties with large interobserver variability,[10] a list of search terms was developed based on all predetermined key words within the *Epic* EMR that could classify a myositis syndrome. These terms were inputted into Epic's SlicerDicer data analytics tool, which searched the obligatory "principle diagnosis" assigned to every patient encounter in the EMR. The same terms were searched for in the free text of outpatient letters prior to the EMR. The search terms were: "myositis, dermatomyositis, polymyositis, anti-synthetase syndrome, inclusion body myositis, immune-mediated necrotizing myopathy, myositis antibodies, muscle biopsy, HMG-CoA reductase antibodies." The diagnosis was then confirmed through a review of physical or electronic hospital records. Patients who did not fulfill the above inclusion criteria or for whom we were unable to locate sufficient diagnostic information were excluded. Patients were not necessarily excluded if they did not meet the 2017 European Alliance of Associations for Rheumatology/American College of Rheumatology (EULAR/ACR) classification criteria for IIM,[11] as this classification system is not intended as diagnostic criteria and does not account for all currently known myositis-specific autoantibodies.

2.2 | Direct healthcare cost calculation

Direct healthcare costs were calculated for all patient episodes from the time of onset of symptoms suggestive of an IIM, to the date of diagnosis (defined as the date a muscle biopsy was performed). If a muscle biopsy was not performed, the earliest date with a documented clinical diagnosis of IIM was used. Calculated costs needed to directly contribute to a diagnosis of IIM, and consisted of outpatient physician visits, presentations to the emergency department related to IIM symptoms, and hospitalizations. Total inpatient costs generated by the hospital's Health Intelligence Unit (HIU) were filtered to include direct ward, unit, and operating theater costs up until the date of diagnosis. Non-diagnostic health service costs such as allied health (e.g., physiotherapy,

occupational therapy, etc.) were excluded. The number and cost of pathology tests, radiology scans, neurophysiology, and other procedures that directly contributed to the diagnosis and/or characterization of IIM manifestations such as interstitial lung disease, arthritis, rash, cardiac involvement, and malignancy screening consistent with recently published guidelines,[12] were also recorded. Tests that were repeated for monitoring purposes (e.g., serial creatine kinase levels) were excluded.

2.3 | Data collection

Data collected included patient demographics, comorbidities, and relevant clinical characteristics. Patient comorbidities were used to calculate the Charlson Comorbidity Index (CCI), a validated clinical research tool, with higher scores indicating more severe comorbid conditions and greater 10-year mortality risk.[13] Patient race was self-reported and documented in hospital registration data using predefined categories.

The number of relevant investigations performed was extracted from the patient's medical record. Cost of investigations, outpatient visits, and inpatient episodes per patient were obtained from billing data spreadsheets generated by the HIU. If costs for a specific investigation or episode were not directly available from the HIU, we estimated them using cost data from the nearest preceding year. If these costs were still not available, we extrapolated costs using the 2022 Australian Medicare Benefits Schedule (MBS), which detailed government-funded medical services and fees.[14]

2.4 | Project approval

The study was approved by the Melbourne Health Office for Research Ethics & Governance (QA2022003).

2.5 | Statistical analysis

Continuous variables were expressed as median with interquartile range (IQR). Costs were expressed as mean with standard deviation (SD). We used one-way ANOVA in Microsoft Excel to determine statistically significant differences between categorical variables.

3 | RESULTS

3.1 | Patient characteristics

The study identified 38 patients (Figure S1) who were diagnosed with an IIM at the Royal Melbourne Hospital between January 2012 and December 2021 (Table 1). The median age of patients at diagnosis was 62 years (IQR 55–70). Most were female ($n=28$, 74%), from metropolitan areas[15] ($n=24$, 63%), with a median CCI of 2.5 (IQR 1–4).

DM was the most common subtype of IIM diagnosed ($n=13$, 34.2%), followed by IBM ($n=7$, 18.4%), IMNM ($n=7$, 18.4%), PM ($n=6$, 15.8%) and ASyS ($n=5$, 13.2%). Fifteen (40%) patients presented with skin rashes characteristic of IIM, 11 (29%) with dysphagia, six (16%) with interstitial lung disease, and five (13%) were diagnosed with a malignancy within 3 years of their IIM diagnosis (Table 1).

Thirty-two (84%) patients presented with elevated muscle enzymes, with a median peak serum creatine kinase level of 1062 U/L (IQR 274–3392). Twenty-three (61%) patients were seropositive for either a myositis-specific or myositis-associated antibody. Of note, the EUROLINE Inflammatory Myopathies 16 Ag line blot immunoassay was not available at the RMH until 2019. As such, truly seropositive patients are likely under-represented, with many "seronegative" cases prior to 2019 potentially possessing undetected myositis antibodies.

3.2 | IIM diagnosis

The median time from symptom onset to diagnosis was 212 days (IQR 118–722) (Table 1). Single factor ANOVA showed significant variation in time to diagnosis among IIM subtypes ($p=.0009$). Time to diagnosis for patients with IBM was substantially longer than other IIM subtypes at 1233 days (IQR 1012–1281). The median time to diagnosis for patients with non-IBM subtypes of IIM was 191 days (IQR 103–288). The median time between presentation to the RMH and diagnosis across all subtypes was 30 days (IQR 8–120). Even after presentation to the RMH, there was a delay in diagnosis of the IBM subtype, with the longest median time of 138 days (IQR 113–217).

3.3 | Resource utilization

3.3.1 | Hospital encounters

Most (76%) patients required emergent hospitalization during their diagnostic workup, with a median length of stay of 8 days (IQR 4–15) (Table 2). Five (13%) patients obtained an IIM diagnosis through muscle biopsy as an elective day admission, while few (10%) patients did not require hospitalization at all. Seven (18%) patients required emergent hospitalization and an additional elective day admission to complete their diagnostic workup. Four (11%) people presented to the emergency department while being investigated for an IIM. The number of outpatient specialist clinic visits associated with securing an IIM diagnosis ranged between 0 and 21 per patient, the latter being a case of IBM.

3.3.2 | Investigations

The median number of investigations (including pathology, imaging, and procedural tests) to diagnose and characterize the

TABLE 1 Baseline demographic, comorbidity, and clinical characteristics.

	All ($n=38$)	DM ($n=13$, 34.2%)	IBM ($n=7$, 18.4%)	IMNM ($n=7$, 18.4%)	PM ($n=6$, 15.8%)	ASyS ($n=5$, 13.2%)
Sex						
Female	28 (73.7%)	10 (76.9%)	5 (71.4%)	4 (57.1%)	5 (83.3%)	4 (80.0%)
Age (years)						
Median (IQR)	62 (55.5–70)	57 (49–65)	68 (66–73.5)	70 (68.5–74)	58 (52–60.5)	60 (55–61)
Residence						
Metropolitan	24 (63.2%)	8 (61.5%)	3 (42.9%)	4 (57.1%)	5 (83.3%)	4 (80.0%)
Regional	14 (36.8%)	5 (38.5%)	4 (57.1%)	3 (42.9%)	1 (16.7%)	1 (20.0%)
Race						
White	34 (89.5%)	10 (76.9%)	7 (100.0%)	7 (100.0%)	6 (100.0%)	4 (80.0%)
Asian	3 (7.9%)	3 (23.1%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)
First Nations	1 (2.6%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	0 (0.0%)	1 (20.0%)
Employment						
Unemployed	25 (65.8%)	8 (61.5%)	6 (85.7%)	6 (85.7%)	4 (66.7%)	1 (20.0%)
Median CCI (IQR)	2.5 (1–4)	1 (1–3)	4 (3–5)	4 (2.5–5)	1.5 (1–2)	2 (1–3)
Muscle involvement	33 (86.8%)	10 (76.9%)	7 (100%)	7 (100%)	5 (83.3%)	4 (80.0%)
Skin involvement	15 (39.5%)	12 (92.3%)	0 (0.0%)	0 (0.0%)	1 (16.7%)	2 (40.0%)
Dysphagia	11 (28.9%)	3 (23.1%)	2 (28.6%)	1 (14.3%)	4 (66.7%)	1 (20.0%)
Interstitial lung disease	6 (15.8%)	1 (7.7%)	0 (0.0%)	0 (0.0%)	2 (33.3%)	3 (60.0%)
Joint involvement	10 (26.3%)	4 (30.8%)	1 (14.3%)	0 (0.0%)	1 (16.7%)	4 (80.0%)
Malignancy[a]	5 (13.2%)	2 (15.4%)	2 (28.6%)	1 (14.3%)	0 (0.0%)	0 (0.0%)
Median peak CK (IQR)	1062 (274–3392)	658 (141–1554)	705 (328–823)	5919 (3900–7129)	2359 (825–4489)	680 (277–3445)
Antibody positive						
MSA	20 (52.6%)	7 (53.8%)	0 (0.0%)	7 (100%)	1 (16.7%)	5 (100%)
MAA	7 (18.4%)	2 (15.4%)	1 (14.3%)	0 (0.0%)	2 (33.3%)	2 (40.0%)
Neither	15 (39.5%)	6 (46.2%)	6 (85.7%)	0 (0.0%	3 (50.0%)	0 (0.0%)
EULAR/ACR IIM 2017 criteria						
Definite	19 (50.0%)	9 (69.2%)	2 (28.6%)	3 (42.9%)	2 (33.3%)	3 (60.0%)
Probable	13 (34.2%)	2 (15.4%)	3 (42.9%)	4 (57.1%)	3 (50.0%)	1 (20.0%)
Neither	6 (15.8%)	2 (15.4%)	2 (28.6%)	0 (0.0%)	1 (16.7%)	1 (20.0%)
Median time to diagnosis (days) (IQR)						
From symptoms	212 (118–722)	164 (99–282)	1233 (1012–1281)	107 (68–182)	259 (207–285)	212 (183–668)
From presentation to a tertiary center	30 (8–120)	50 (9–76)	138 (113–217)	15 (10–58)	14 (8–25)	19 (8–30)

Abbreviations: ACR, American College of Rheumatology; CCI, Charlson Comorbidity Index; CK, creatine kinase (U/L); EULAR, European Alliance of Associations for Rheumatology; IIM, idiopathic inflammatory myopathy; MAA, myositis-associated antibody (SSA/Ro, Ku, PM/Scl-75, PM/Scl-100, AMA, U1RNP); MSA, myositis-specific antibody (t-RNA synthetases – Jo-1, PL-7, PL-12, EJ, OJ; Mi-2, SRP, TIF-1gamma, NXP-2, MDA5, SAE, HMG-CoA reductase).

[a]Within 3 years of IIM diagnosis.

muscular and extra-muscular manifestations of an IIM was 21 per patient (IQR 15–26) (Table 2). Individuals with ASyS had the highest number of investigations (median 26, IQR 18–34), while those with IBM had the fewest (median 14, IQR 13–19). Thirteen (34%) patients had more than one ANA assay test—with one patient undergoing four repeat tests—and 10 (26%) had more than one ENA test performed. 2 (6%) patients were repeatedly tested for myositis-specific antibodies. Nineteen (50%) patients underwent both skeletal muscle MRI and EMG to identify abnormal muscle involvement, while four (10%) patients underwent both CT chest-abdomen-pelvis and PET-CT to screen for malignancy. Additional malignancy screening included fecal occult blood testing (16%), colonoscopy (13%), mammography (38% of female patients), breast ultrasound (8% of female patients), and pelvic ultrasound

TABLE 2 Resource utilization and costs.

	All ($n=38$)	DM ($n=13$, 34.2%)	IBM ($n=7$, 18.4%)	IMNM ($n=7$, 18.4%)	PM ($n=6$, 15.8%)	ASyS ($n=5$, 13.2%)
Hospitalizations						
Total	51	20	7	11	9	4
Emergent	38	14	4	10	6	4
Elective[a]	13	6	3	1	3	0
Median number	1	1	1	2	1.5	1
Median LOS (days) (IQR)	8 (4–14.8)	7 (4–12)	5 (1–7.5)	19 (9.5–29)	8 (5.8–14)	11 (0–13.8)
Investigations[b]						
Median number	21 (15–26)	24 (20–28)	14 (13–19)	17 (14–24)	21.5 (19–24)	26 (18–34)
Total number						
ANA	50	18	7	8	8	9
ENA	47	17	7	9	8	6
Myositis blot	36	13	4	7	7	5
Skeletal muscle MRI	28	10	4	6	4	4
EMG	28	8	6	7	4	3
CT-CAP	20	9	3	4	2	2
PET	4	1	1	0	1	1
Muscle biopsy						
Open	23	5	7	4	5	2
Percutaneous	7	4	1	2	0	0
Skin biopsy	7	7	0	0	0	0
Mean cost: AUD (SD)						
Outpatient encounters[c]	$806.12 (1229.05)	$845.75 (1058.59)	$1033.40 (1146.45	$379.23 (1003.35)	$874.95 (1793.39)	$899.96 (1636.01)
Inpatient encounters[d]	$11 726.68 (11 221.82)	$8548.63 (6360.22)	$11 178.95 (12 340.68)	$18 886.49 (13 678.70)	$8497.25 (7425.82)	$14 608.02 (17 787.09)
Investigations	$3084.77 (1258.30)	$3294.61 (743.07)	$2810.61 (1055.86)	$2501.49 (1235.15)	$3121.30 (1300.84)	$3695.82 (2361.07)
Total	$15 617.58 (11 331.17)	$12 688.99 (5713.03)	$15 022.96 (14 055.63)	$21 767.20 (12 521.61)	$12 493.50 (7498.09)	$19 203.79 (19 040.69)

Abbreviations: CT-CAP, CT chest, abdomen, pelvis; LOS, length of stay.

[a]Elective hospitalizations include: muscle biopsy, bronchoscopy, endoscopy, sleep study.

[b]Investigations recorded include: CRP, ESR, CK, myositis blot, anti-HMG-CoA reductase antibody, anti-mitochondrial antibody, RF, anti-CCP antibody, ANA, ENA, anti-dsDNA antibody, ANCA, complements, thyroid function tests, 25(OH)D_3, anti-acetylcholinesterase antibody, HIV serology, iron studies, histopathology from biopsies, tumor markers, fecal occult blood test, MRI, CT chest +/− abdo/pelvis, PET, chest x-ray, high resolution CT chest, CT pulmonary arteries, ventilation-perfusion scan, CT spine, MRI brain/spine, transthoracic echocardiogram, video fluoroscopy, barium swallow, mammogram, breast US, pelvic US, EMG, respiratory function tests, endoscopy, bronchoscopy, liver US.

[c]Refers to the cost of specialist physician outpatient clinics.

[d]Cost inclusive of ward (room, nursing, medications, meals), unit (specialist clinicians), and operating theater costs from the date of admission to date of diagnosis only, see Methods for further details.

(12% of female patients). Thirty muscle biopsies were performed on 28 patients (74% of the cohort), with two (5%) patients undergoing open surgical muscle biopsy following an inconclusive percutaneous biopsy.

3.3.3 | Costs

The average cost associated with diagnosing IIM was $15 618 AUD per patient (STD: 11 331) (Figure 1A). Diagnosis of IMNM was most costly, with a mean of $21 767 per patient (STD: 12 522), followed by ASyS with a mean of $19 204 per patient (STD: 19 041) (Figure 1B). A substantial proportion of these costs related to longer inpatient hospitalization prior to securing a diagnosis (median 15 and 7 days respectively). Of note, the median number of investigations performed in the IMNM subtype was second lowest at 17 per patient (IQR: 14–24). In recent years, the average overall cost per patient of diagnosing an IIM has trended upwards (Figure 1A), alongside a rise in inpatient costs (Figure 2A), whilst mean outpatient costs and cost of investigations has remained largely static (Figures 3A and

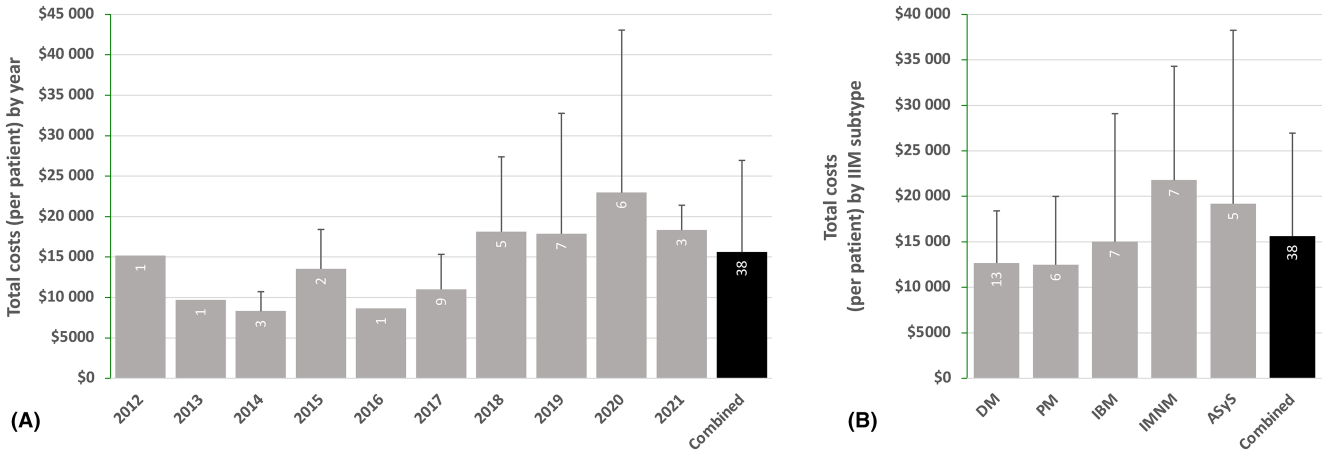

FIGURE 1 Mean *total* cost per patient of diagnosing and characterizing an idiopathic inflammatory myopathy, by year of diagnosis (A) and subtype of IIM (B). Number in column = number of patients in subgroup. Cost in Australian dollars. ASyS, anti-synthetase syndrome; Combined, all subtypes combined; DM, dermatomyositis; IBM, inclusion body myositis; IMNM, immune-mediated necrotizing myopathy; PM, polymyositis.

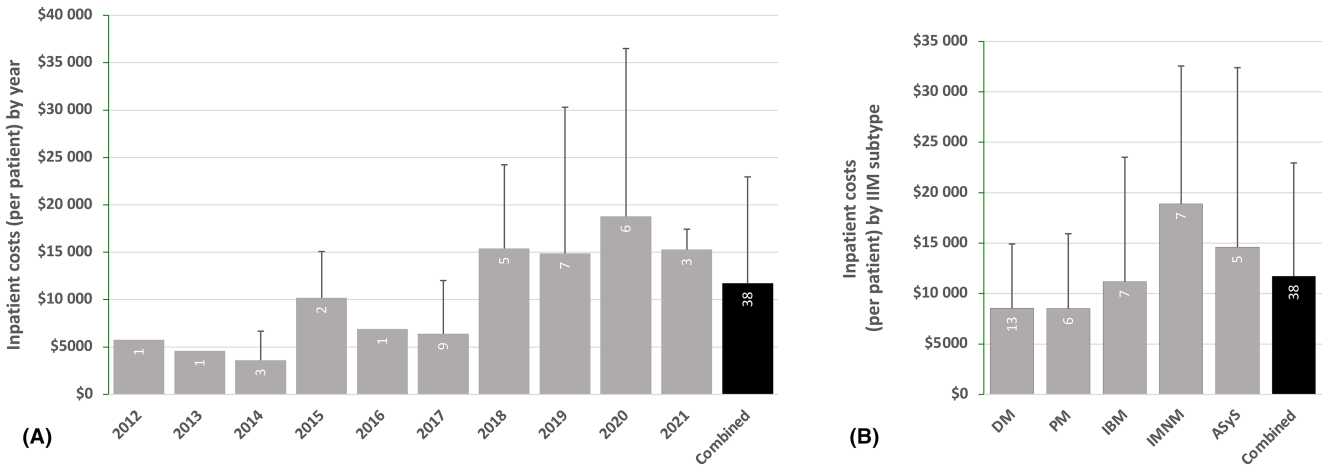

FIGURE 2 Mean *inpatient* costs per patient of diagnosing and characterizing an idiopathic inflammatory myopathy, by year of diagnosis (A) and subtype of IIM (B). Number in column = number of patients in subgroup. Cost in Australian dollars. ASyS, anti-synthetase syndrome; Combined, all subtypes combined; DM, dermatomyositis; IBM, inclusion body myositis; IMNM, immune-mediated necrotizing myopathy; PM, polymyositis.

4A). The cost of commonly performed investigations can be found in Table S1.

4 | DISCUSSION

Our study highlights the significant resource utilization and financial burden of diagnosing inflammatory myopathies in a large Australian tertiary public health service. A substantial component of this cost arises from expenses incurred during hospitalization and the high burden of diagnostic tests. Furthermore, there is evidence of delay in diagnosing an IIM in the current practice at our tertiary center, particularly observed in the IBM subtype, and unnecessary repetitive ordering of serology.

We found that IMNM and ASyS were associated with the highest health service costs per patient (means of AU$21 767 and AU$19 204, respectively). In the case of IMNM, this was due to a longer median duration in hospital prior to reaching a diagnosis (15 days), and thus a higher average cost of hospitalization per patient (AU$18 886), rather than a higher cost of investigations. This is likely secondary to the profound functional impairment associated with active IMNM, prompting early admission to the hospital, and yet time to diagnosis remains prolonged due to clinical uncertainty with often undifferentiated presentations. By comparison, patients with ASyS had a lower median duration in hospital (7 days), but tallied the highest median number of investigations per person ($n = 26$) and hence the highest mean cost of investigations per patient (AU$3696) (Figure 4B). These additional

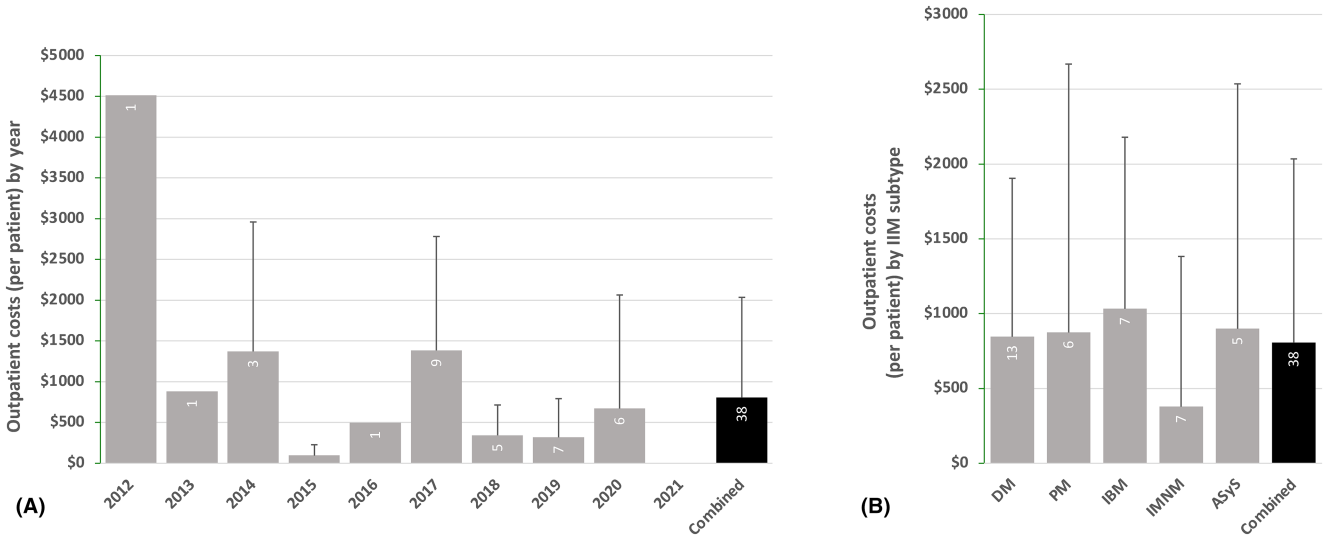

FIGURE 3 Mean *outpatient* costs per patient of diagnosing and characterizing an idiopathic inflammatory myopathy, by year of diagnosis (A) and subtype of IIM (B). Number in column = number of patients in subgroup. Cost in Australian dollars. ASyS, anti-synthetase syndrome; Combined, all subtypes combined; DM, dermatomyositis; IBM, inclusion body myositis; IMNM, immune-mediated necrotizing myopathy; PM, polymyositis.

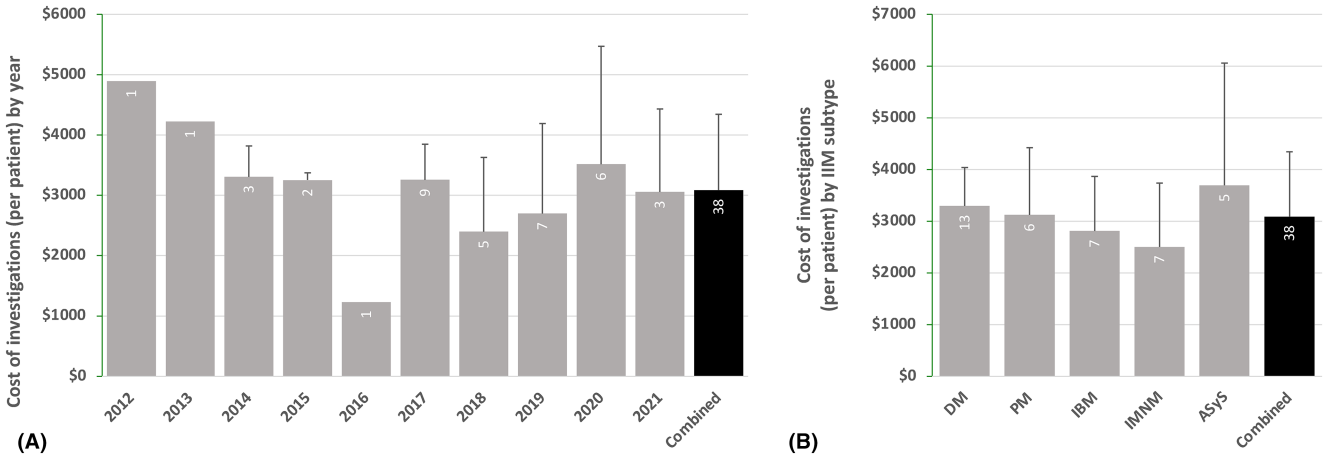

FIGURE 4 Mean cost of *investigations* per patient in diagnosing and characterizing an idiopathic inflammatory myopathy, by year of diagnosis (A) and subtype of IIM (B). Number in column = number of patients in subgroup. Cost in Australian dollars. ASyS, anti-synthetase syndrome; Combined, all subtypes combined; DM, dermatomyositis; IBM, inclusion body myositis; IMNM, immune-mediated necrotizing myopathy; PM, polymyositis.

investigations likely reflect that ASyS is associated with a variety of extra-muscular manifestations that require considerable workup. As such, in calculating these costs, we may have inadvertently captured the effect of influential factors such as disease severity and undifferentiated presentations.

During the SARS-CoV-2 pandemic, our hospital, like many others, experienced delays in the review of new outpatient referrals and prolonged wait times for Telehealth consultations. We observed an increase in the median length of hospital stay as patients presented directly to the emergency department with suspected IIMs, bypassing the less efficient outpatient evaluation process. This shift contributed to an overall increase in the total cost of diagnosis.

Lastly, it is of important to note that our calculated costs do not include patients investigated for a suspected IIM who did not ultimately fulfill the diagnostic criteria. The total economic burden would be far more substantial if these costs were also included.

4.1 | Clinical relevance

This study underscores the limitations of current diagnostic approaches in the evaluation of suspected IIM. We have shown that patients with IIM are subjected to a high burden of investigations and that diagnostic delays are substantial, even within a large

tertiary center. This is of concern, given delay in diagnosis has been shown to be a predictor of poor clinical outcome.[16] Clearly, improved diagnostic algorithms are required. One of our goals should be to prevent inessential hospitalization in order to effectively reduce the financial burden on healthcare systems. In cases where inpatient admission is necessary, awareness of the high associated costs should prompt timely investigation and treatment to maximize the chances of earlier discharge. Furthermore, the unnecessary repetition of various pathology (ANA in 34% of patients, ENA in 26%, and myositis antibody panel in 6%) highlights the need for treating doctors to carefully review previously ordered investigations, in order to minimize additional economic burden, harm to our patients and diagnostic delay.[17,18] We were unable to determine the exact reason for this repetition as this information was not typically documented, but may reflect a lack of clinician awareness and system-level inefficiencies related to accessing external results. We also acknowledge that limited access to investigations that offer a comprehensive evaluation of IIM features such as PET-CT may lead to clinicians request multiple less informative tests, contributing to an increased number of investigations.

4.2 | Strengths and limitations

This study is the first of its kind to quantify the direct costs associated with diagnosing and characterizing IIMs. The accuracy of the results has been enhanced by the inclusion of comprehensive direct cost data, compiled by data analysts from the RMH HIU. Of the 817 investigations on record contributing to a diagnosis of IIM, the exact cost of 52% of tests were available. For the remainder, costs were estimated by extrapolating from data of the adjacent years (28%) or were derived from the 2022 Medicare Benefits Schedule (19%). Only 1% of investigations (e.g., performing a skin biopsy) could not have a specific cost attributed to them.

The study cohort is an accurate representation of patients with IIM. Even though our inclusion criteria did not require a diagnostic muscle biopsy, the majority (84%) of included patients met diagnostic criteria for having a probable or definite IIM as per the 2017 EULAR/ACR classification criteria.[11] While some of our patients did not meet these criteria, it is important to recognize that this classification system is not designed for diagnostic purposes and does not account for all currently known myositis-specific autoantibodies. However, our case identification strategy using predetermined EMR keywords may be limited in the absence of existing literature on searching for IIM in EMR. It is possible that some clinicians use atypical classification terms, for example, myopathy, leading to missed cases which may have influenced our findings.

The study was a retrospective examination of direct costs associated with a diagnosis of IIM at a large public tertiary center over a 10-year period. While data calculated from our center is likely reflective of other similar institutions and populations within the Australian public healthcare system, it was a monocentric study with small subgroup sizes. As such, generalisability and ability to identify predictors of increased cost may be limited. It should also be noted that we did not investigate indirect costs, such as those related to patient work loss or decreased productivity. Previous research has indicated that patients with IIMs experience significantly more work loss than matched controls, primarily due to an increase in medically related absenteeism.[4] Hence the total cost to both patient and healthcare system extends beyond that measured in this study.

5 | CONCLUSION

This study highlights the substantial healthcare resource utilization and economic burden associated with the diagnosis and characterization of IIMs, in part due to significant inpatient care costs. Limitations in current diagnostic practice were also identified, contributing to increased financial burden and longer inpatient admissions. These results should prompt further research aimed at reducing costs and time to diagnosis, with the ultimate goal of improving patient outcomes.

AUTHOR CONTRIBUTIONS

J.D., S.O. and S.C. conceptualised the study with input from M.N. Data was acquired and analysed by V.H. The manuscript was written by V.H. with input from all authors.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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