

Estimated Incremental Direct All-Cause Healthcare Costs Associated With Multimorbidity Amongst Patients With Sickle Cell Disease (SCD) in England

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Aim: To estimate the all-cause direct incremental healthcare costs associated with having SCD-related complications amongst patients with SCD in England.

Background

- SCD can cause acute and chronic complications due to the 'sickling' of red blood cells and the end-organ damage that results from long-term reduced blood flow and ongoing haemolysis.¹ These complications can affect multiple organ systems.¹
- Patients with SCD have much higher rates of certain complications (e.g., stroke, kidney disease) than people in the general population^{1,2} and experience a significant complication burden from even early life stages.^{1,2}
- As such, it is important to have accurate estimates of all-cause healthcare costs for patients with SCD-related complications and accurate estimates of how healthcare costs increase with each additional complication.
- Cost estimates among patients in England can inform SCD treatment and management and guide resource allocation decisions by the English National Health Service (NHS).

Methods

We conducted a retrospective study that estimated healthcare costs amongst patients with SCD who had no complications, 1 complication, or ≥ 2 complications. We also estimated incremental healthcare costs per complication.

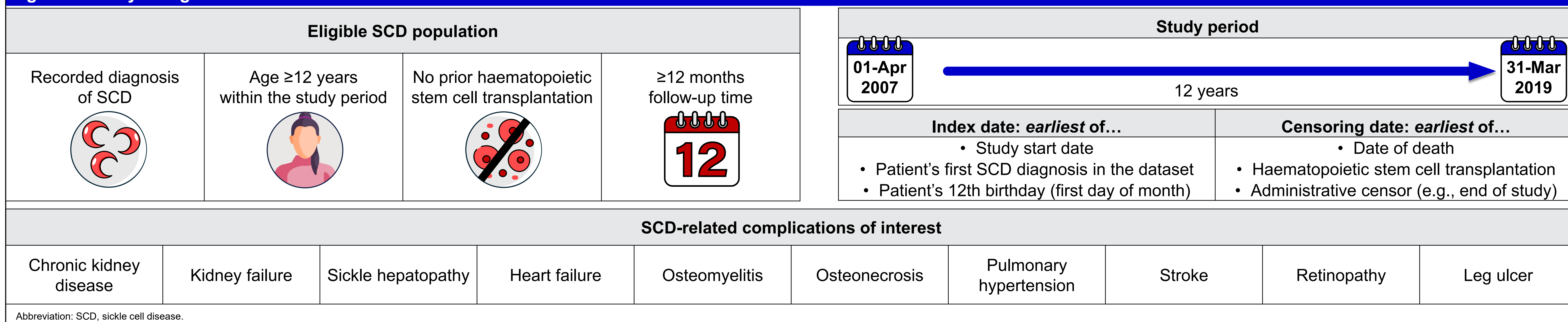
Study design

- We identified patients with SCD and estimated their healthcare resource using primary care data from the Clinical Practice Research Datalink Aurum dataset³ linked with secondary care data from the Hospital Episode Statistics reimbursement dataset (Figure 1).⁴
- We calculated a patient's maximum total number of SCD-related complications of interest across the study period (Figure 1).
- Complications of interest were chronic conditions or associated with persistent detriments (Figure 1); selection of complications was informed by discussion with clinicians experienced in treating SCD.
- All-cause direct healthcare costs were estimated between patients' index and censor dates (Figure 1).
- Costs were estimated based on the eligible patients with complete cases (i.e., data for all model covariates).
- Costs for primary care staff time were obtained from the Unit Costs of Health and Social Care 2018/19.⁵
- Costs for secondary care were obtained from the NHS Payment Grouper 2018/19.^{6,7}
- All costs were inflated to 2020/21 pounds sterling using the NHS Cost Inflation Index.⁸

Model procedures

- We estimated costs using a multiple generalised linear model with gamma variance and identity link functions.
- Costs associated with having a maximum total number of 0, 1, or ≥ 2 complications during baseline and follow up were analysed using the maximum number of complications as a categorical variable (0, 1, ≥ 2).
- Incremental costs per complication were estimated using the maximum total number of complications as a continuous variable.
- Models were adjusted for age at index, gender, ethnicity, deprivation quintile, and follow-up time, which was centered on the median follow-up time.

Figure 1. Study Design



Results

Patient population

- 7,265 patients were included in the eligible SCD population (Figure 2; Table 1).
- Eligible patients had a mean (standard deviation) age of 30 (16) years at baseline. Most were female (61.3%) and reported black ethnicity (75.8%) (Table 1).
- Median follow-up time was 9.75 years.
- 7,112 (97.9%) of eligible patients had data for all covariates and thus were included in regression analyses.

Patients with 0, 1, or ≥ 2 complications

- 16.3% of patients had a maximum total of 1 complication. 9.0% of patients had in total ≥ 2 complications.



Figure 2. Study population flow chart

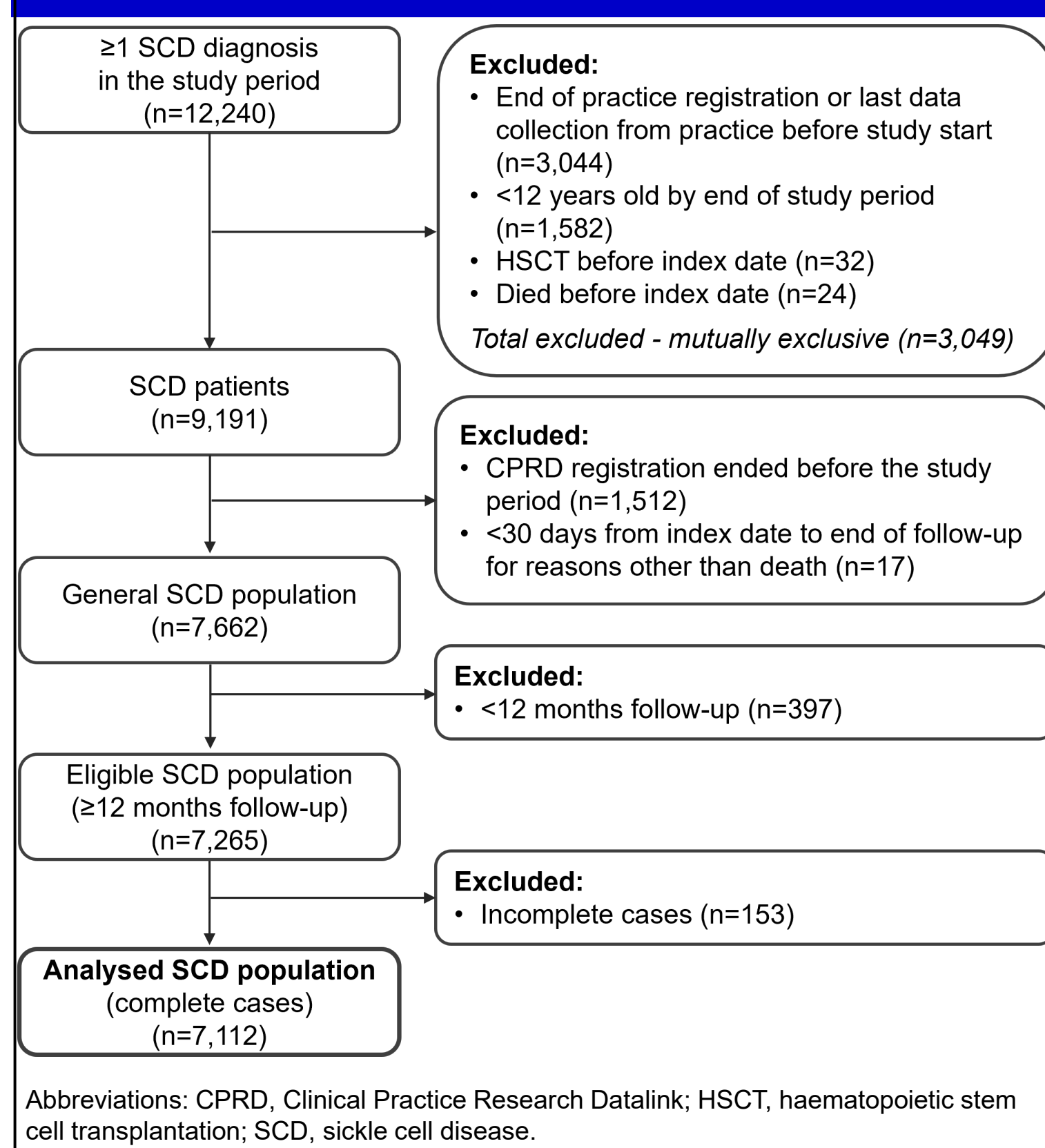


Table 1. Baseline characteristics

Characteristic	Patients (n=7,265) ^a
Age in years, mean (SD)	29.9 (16.3)
Female sex, n (%)	4,453 (61.3)
Ethnicity	
Black	5,508 (75.8)
White	657 (9.0)
South Asian	298 (4.1)
Mixed or "Other"	656 (9.0)
Unknown	146 (2.0)
Deprivation, IMD quintile, n (%)	
1 – least deprived	470 (6.5)
2	649 (8.9)
3	1,093 (15.0)
4	2,240 (30.8)
5 – most deprived	2,809 (38.7)
Unknown	4 (0.0)

Abbreviations: IMD, Index of Multiple Deprivation; SD, standard deviation.

^a Eligible patient population.

Limitations

- We treated all complications as 'chronic' in the regression model, but this may not accurately reflect that costs are not accumulated at the same rate over time in certain complications (e.g., leg ulcers).
- Costs were spread out over the whole period that the patient was in the study, rather than the specific period when the patient had complications. This may have led us to underestimate the true cost of having SCD-related complications.

Conclusions

- About 25% of patients with SCD had ≥ 1 complication during the observed period.
- Over a 9.75-year period, costs can be up to 10-fold higher for patients with ≥ 2 complications than for patients with no complications.
- Costs increase substantially with each additional complication.
- These data can help decision-makers and clinicians understand the healthcare burden of multimorbidity amongst patients with SCD in England and highlight the need for treatments that can prevent the development of complications.

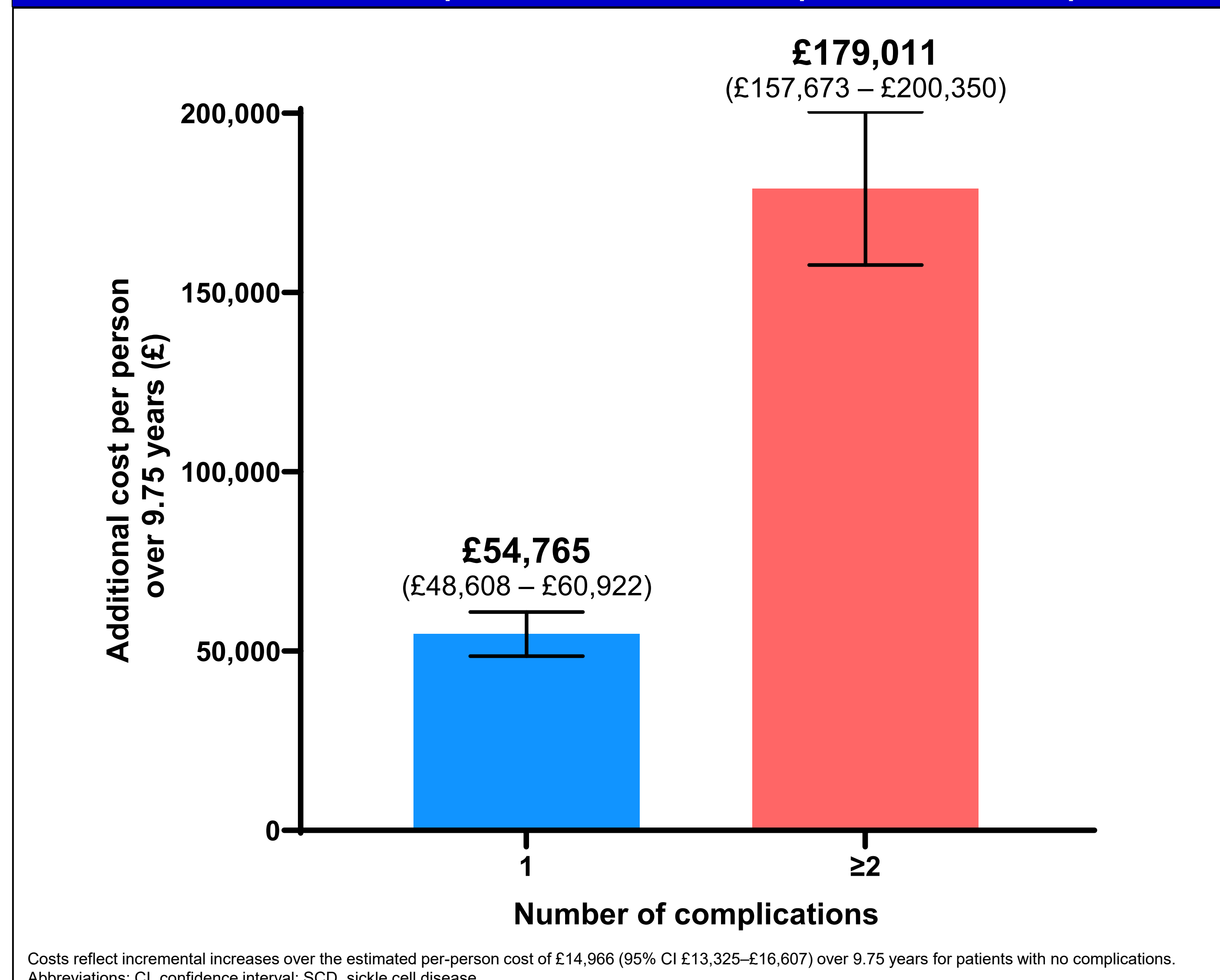
Estimated costs

- Estimated per person costs over the median follow-up time of 9.75 years were significantly greater for patients with ≥ 2 complications ($p < 0.001$) or 1 complication ($p < 0.001$) compared to patients with 0 complications (Figure 3).

- Each additional complication was associated with an estimated incremental cost of £49,572 (95% CI £45,608–£53,536) per person over 9.75 years.



Figure 3. Estimated additional per person costs (95% CI) over 9.75 years for patients with 1 and ≥ 2 SCD-related complications above costs of patients with 0 complications.



Costs reflect incremental increases over the estimated per-person cost of £14,966 (95% CI £13,325–£16,607) over 9.75 years for patients with no complications. Abbreviations: CI, confidence interval; SCD, sickle cell disease.

References

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Disclosures

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