



## Epilepsy-related stigma and cost in two onchocerciasis-endemic areas in South Sudan: A pilot descriptive study



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### ABSTRACT

**Introduction:** Epilepsy is a major public health concern in sub-Saharan Africa, particularly in resource-limited rural villages where persons with epilepsy (PWE) are often confronted with a wide treatment gap, frequent stigma and high cost of epilepsy care. We investigated stigma and economic cost related to epilepsy in the states of Maridi and Amadi in South Sudan, two onchocerciasis endemic areas with high epilepsy prevalence.

**Methods:** Between November 2019 and February 2020, community-based surveys were conducted in eight villages of Maridi and Amadi States. Consenting PWE were identified via a door-to-door approach, and perceived stigma was assessed using the validated Kilifi stigma scale. Additional data about household income, as well as epilepsy-related direct and indirect costs were collected.

**Results:** 239 PWE were recruited (95 from Maridi, 144 from Amadi). Stigma scores were higher in Maridi compared to Amadi (mean scores: 13.9 vs 6.5,  $p < 0.001$ ). Mean weighted epilepsy costs per month in Maridi (38.4 USD) were double those observed in Amadi (17.6 USD). The main epilepsy-related expenditure was the purchase of anti-epileptic drugs (AED). Stigma scores correlated with epilepsy cost (Spearman-rho = 0.24,  $p < 0.001$ ) and were positively associated with traditional medicine use (regression estimate = 1.9;  $p = 0.027$ ).

**Conclusion:** In rural South Sudan, PWE and their families often experienced stigma from the community. Higher perceived stigma was associated with traditional medicine use, which increased the overall cost of epilepsy management. Demystifying epilepsy and making AED more accessible would improve the quality of life of PWE and their families, and reduce the economic burden of epilepsy.

### 1. Introduction

An estimated 50 million persons are currently affected by epilepsy around the globe, 80% of whom live in low- and middle-income countries [1]. Besides experiencing seizures and related complications (traumatic injuries, burns, cognitive impairment), persons with epilepsy (PWE) and their families are often confronted with stigma. In sub-Saharan Africa (SSA), epilepsy stigma is often fuelled by deeply rooted misconceptions and compounded by sociocultural realities [2]. Understandably, one of the main aims of the Global Campaign Against Epilepsy is to reduce misconceptions and increase the acceptability of PWE [3]. Seizure control also appears to determine the level of stigma,

as persons who experienced seizures every month reported higher perceived stigma in a study in Ethiopia [4].

In low-income settings as found in SSA, about 75% of PWE do not receive adequate treatment and are therefore prone to more frequent seizures [1]. Such a situation is likely to create a vicious cycle of more seizures leading to higher perceived stigma. Previous studies have highlighted the fact that high cost of epilepsy care, beyond the reach of most PWE and their families, is a major driving force behind this wide treatment gap [5–7]. We investigated epilepsy-related stigma and cost of care among PWE in Maridi and Amadi States in South Sudan; these are onchocerciasis-endemic areas with a high burden of epilepsy (including nodding syndrome) [8,9].

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## 2. Methods

### 2.1. Study setting and design

We conducted a cross-sectional descriptive, community-based study in five villages in Maridi State in South Sudan (Kazana I, Kazana II, Matara, Gabat, Tarawa) and three additional sites in Amadi State (Amadi, Mundri, and Lui). These are remote rural settings where the predominant activity is farming. Previous surveys in these sites found a prevalence of epilepsy/nodding syndrome (NS) ranging from 2.3% to 6.7%, with the majority of diagnosed PWE meeting the criteria for onchocerciasis-associated epilepsy (OAE) [8–10].

### 2.2. Study procedures and tools

Before initiating this community-based survey, we first contacted the community leaders (chiefs, religious authorities, teachers) in the study sites to explain the aims and procedures of the study and obtain their collaboration. They were instrumental in sensitising the village residents and preparing them for the study. The study was carried out in two phases: November 2019 in Maridi State, and February 2020 in Amadi State. The research team visited each study village using a door-to-door approach. We asked each household whether one or more PWE were residing there (defined as any person who experienced two or more unprovoked seizure attacks during the past year [11]), in which case we obtained verbal consent for an interview from the household head. The 2-part study questionnaire was administered in the local language (Arabic) to PWE and/or their caregivers, and the collected information noted on paper forms. Interviews were conducted in a quiet space in the compound of the investigated household. In case an empty house was encountered, we enquired from neighbours if any person from that household had epilepsy, in which case we revisited the house once.

The first part of the questionnaire assessed stigma perceived by the PWE using the Kilifi stigma scale, which had been validated in Kenya and showed high internal consistency, excellent test-retest reliability, and yielded similar results upon interviewing PWE or their caregivers [12]. This scale consists of 15 questions, each scored on a Likert scale ranging from 0 to 2 (maximum score of 30). The scoring is done based on the responses to the questions, as follows: 0 for “not at all”, 1 for “sometimes”, and 2 for “always”. PWE with scores  $\geq 20$  (that is, the 66th percentile of 30) were considered as experiencing high perceived stigma. In addition to the 15 standard questions, two additional questions were asked to complete the stigma assessment: (i) whether the PWE had been sexually harassed and (ii) whether the PWE had ever been physically abused. The answers to these additional questions also followed the 0–2 Likert score format.

In the second part of the questionnaire, we asked questions aimed at estimating epilepsy-related costs. The tool we employed had previously been used in the Democratic Republic of the Congo and provides estimates for epilepsy-related direct costs (cost of anti-epileptic drugs [AED], medical consultations, hospitalisations, transport and traditional medicine), indirect costs (days of work lost by the PWE or caregiver), as well as estimated household income per month [13]. We asked the family to convert all payments in kind (usually to traditional healers) to an estimated monetary value. Furthermore, we considered one day of work lost to be worth the Gross Domestic Product (GDP) per capita, per day; this involved dividing the 2019 GDP per person of South Sudan (275.18 USD) [14] by 365 days, to obtain 0.75 USD per day. All costs provided in local currency (South Sudanese Pounds or SSP) were converted to USD at the rate of 1 SSP = 0.0077 USD, which was applicable in December 2019.

### 2.3. Sample size

We calculated our sample size using the Raosoft online calculator

([www.raosoft.com/samplesize.html](http://www.raosoft.com/samplesize.html)). Assuming a 10% margin of error, 95% confidence level, 50% response distribution, and a total PWE population of 1,000 in each state (based on previous findings in Maridi where 774 PWE were reported [8], and making allowance for unreported cases), our study required at least 88 participants from each state.

### 2.4. Data preparation

In case missing data were encountered among the responses to the 15 questions on the stigma scale, the partial stigma score was adjusted to reflect the 30-point scale using the following formula:

$$\text{Adjusted Score (on 30)} = \frac{\text{Total score for } n \text{ questions}}{2n} \times 30$$

In addition, when evaluating the proportion of household income that is represented by the epilepsy cost per household, we attributed an arbitrary income of 1 USD for the 30 households whose income was recorded as 0. This implied that the epilepsy cost ( $C$ ) for these households was equivalent to  $C$  times their income.

### 2.5. Data analysis

The collected data were transferred from paper forms into an electronic database, codified and cleaned. Data analysis was mainly descriptive and was done using the software R version 3.6.2. Continuous variables were summarised as means and standard deviations (SD); the Shapiro-Wilk normality test was done for each continuous outcome, and parametric or non-parametric statistical tests were used as appropriate. Categorical variables were expressed as frequencies and compared using the Chi-squared test (or Fisher's exact test for counts below five). P-values less than 0.05 were considered as statistically significant.

We performed a multivariable linear regression analysis to assess factors associated with perceived epilepsy stigma. The independent variable of interest was the household income (selected based on previous research [15]), adjusted for purposively selected socio-demographic parameters, history of sexual harassment or physical abuse, and epilepsy treatment options (AED, traditional medicine).

### 2.6. Ethical considerations

The study protocol was approved by the Ministry of Health of South Sudan. Informed consent was obtained from all participants, and the collected data were codified to ensure anonymity. The outcomes of this study will be shared with the health authorities in each state, with whom the research team is currently working to contribute to an epilepsy management strategy for the affected villages.

## 3. Results

A total of 239 participants were recruited (95 from Maridi, 144 from Amadi), from whom stigma and economic cost data were collected. Although there were several missing values for age (60 missing data) and sex (73 missing data), from the available data a mean age of 21.8 years (SD = 7.4, range 4–50) and 52.4% of males were determined in our study population. PWE in Amadi State were older than those in Maridi State (mean ages: 23.3 vs 16.2,  $p < 0.001$ ). Additional information was collected in Amadi State only. This included: seizure type (93% PWE with generalized tonic-clonic seizures, 25.3% reporting nodding seizures, and 7.7% absences); monthly seizure frequency (median: 3; IQR: 2–11); duration of epilepsy (median: 8 years; IQR: 4–14); education status (13.2% unschooled, 82.6% having primary education and 4.2% with secondary education); and person who responded to the investigator's questions (caretakers in 84.7% of cases, and PWE themselves in 15.3%).

**Table 1**  
Epilepsy stigma scores in the study sites.

	Mean score (SD)	P-value <sup>a</sup>	Score ≥ 20: n (%)	P-value <sup>b</sup>
<b>Maridi State</b>				
Kazana 1 (n = 28)	14.4 (6.5)	0.272	4 (14.3%)	0.616
Kazana 2 (n = 21)	14.3 (5.8)		3 (14.3%)	
Matara (n = 21)	14.5 (3.0)		2 (9.5%)	
Tarawa (n = 14)	13.8 (6.0)		3 (21.4%)	
Gabat (n = 11)	10.6 (4.6)		0 (0%)	
<b>Amadi State</b>				
Amadi (n = 23)	5.0 (5.9)	< 0.001	0 (0%)	0.030
Lui (n = 23)	13.8 (5.3)		3 (13.0%)	
Mundri (n = 98)	5.1 (5.7)		1 (1.0%)	

<sup>a</sup> Kruskal Wallis test.

<sup>b</sup> Fisher's Exact test.

### 3.1. Epilepsy stigma scores

Kilifi scale stigma scores in our study population ranged from 0 to 25 and differed significantly between states (mean scores:  $13.9 \pm 5.4$  in Maridi vs  $6.5 \pm 6.5$  in Amadi;  $p < 0.001$ ). Furthermore, villages in Amadi state showed significant inter-village variability in stigma scores (Table 1).

Among the responses to the Kilifi scale questions, there were 33 missing data from 26 participants whose final scores were adjusted: 21 PWE had missing data for one question, three PWE had missing data for two questions, and two PWE had three missing answers. Stigma scores did not correlate with participant age ( $p = 0.264$ ) and did not vary by sex ( $p = 0.652$ ). In contrast, higher mean stigma scores were observed with increasing frequency of past physical abuse ( $p < 0.001$ ) and sexual harassment ( $p = 0.018$ ). There were no differences in the prevalence of both sexual harassment ( $p = 0.923$ ) and physical abuse ( $p = 0.644$ ) among male and female PWE. Table 2 reports the stigma scores of our participants by summarising each question based on the theme it addresses as suggested by Mbuba et al [12].

Considering the data from Amadi State ( $n = 144$ ), stigma scores did not correlate with monthly seizure frequency ( $p = 0.366$ ) nor with education level ( $p = 0.963$ ). However, PWE with nodding seizures had higher stigma scores compared to those who did not report nodding seizures (mean scores:  $8.6$  vs  $5.8$ ;  $p = 0.025$ ). Furthermore, perceived stigma increased with increasing duration of epilepsy (Spearmanrho =  $0.28$ ,  $p < 0.001$ ). We also observed that caretakers reported higher stigma scores compared to when PWE themselves responded to the questions ( $p = 0.006$ ).

**Table 2**  
Participant responses to the Kilifi stigma scale in Maridi and Amadi States, South Sudan.

Stigma scale questions	Not at all n (%)	Sometimes n (%)	Always n (%)	Number of responses
Different	131 (54.8)	68 (28.5)	40 (16.7)	239
Lonely*	137 (57.8)	59 (24.9)	41 (17.3)	237
Embarrassed*	116 (48.9)	83 (35.0)	38 (16.0)	237
Disappointed*	98 (41.9)	95 (40.6)	41 (17.5)	234
Rewarding life*	119 (50.6)	70 (29.8)	46 (19.6)	235
Society*	133 (55.9)	68 (28.6)	37 (15.5)	238
Public places*	131 (55.3)	74 (31.2)	32 (13.5)	237
Uncomfortable*	113 (48.1)	79 (33.6)	43 (18.3)	235
Occasions	109 (45.6)	65 (27.2)	65 (27.2)	239
Inferior*	112 (47.9)	78 (33.3)	44 (18.8)	234
Avoid*	119 (50.4)	78 (33.1)	39 (16.5)	236
Greetings*	154 (65.0)	63 (26.6)	20 (8.4)	237
Mistreated	134 (56.1)	81 (33.9)	24 (10.0)	239
Discriminate	146 (61.1)	76 (31.8)	17 (7.1)	239
Outcast*	133 (56.4)	73 (30.9)	30 (12.7)	236
<b>Additional questions</b>				
Physically abused	189 (79.1)	43 (18.0)	7 (2.9)	239
Sexually harassed*	213 (90.3)	20 (8.5)	3 (1.2)	236

\* Missing values (not all participants had answers for this item).

### 3.2. Economic cost of epilepsy

#### 3.2.1. Epilepsy cost in Maridi state

The weighted mean total cost of epilepsy in Maridi was estimated at 38.4 USD per PWE per month (85.4% direct cost, 14.6% indirect cost). Given the mean monthly income per household of 30.3 USD (SD: 43.3), this implies that on average, epilepsy care amounts to 1.3 times the household budget in Maridi. The bulk of the direct cost was spent on AED purchase (36.6%), followed by traditional medicine expenses representing 27.1% of the direct cost (Table 3).

#### 3.2.2. Epilepsy cost in Amadi State

In Amadi, the weighted mean total cost of epilepsy was 17.6 USD per PWE per month (67% direct cost, 33% indirect cost), representing 33.5% of the average household income of 52.6 USD (SD: 57.4). Similarly as was observed in Maridi, the largest amount was spent on AED (48.3% of direct cost) (Table 4). Epilepsy cost in Amadi was neither correlated with seizure frequency ( $p = 0.913$ ), nor associated with education level ( $p = 0.148$ ), duration of epilepsy ( $p = 0.486$ ), or history of nodding seizures ( $p = 0.798$ ).

Considering all the study sites, epilepsy cost did not correlate with age ( $p = 0.938$ ), sex ( $p = 0.236$ ), history of physical abuse ( $p = 0.522$ ), or sexual harassment ( $p = 0.985$ ). Of the 219 participants who reported spending on AED, 124 (56.6%) purchased them in a clinic, 32 (14.6%) in a hospital, while 61 (27.9%) bought the drugs at a pharmacy; in two PWE, the source of AED was not documented. The AED cost was significantly different depending on the place of purchase (Kruskal Wallis  $p$ -value  $< 0.001$ ). Post-hoc analysis using Dunn's multiple comparisons test showed that compared to untreated PWE (AED cost = 0), AED cost was highest for those who obtained the drugs from private clinics ( $z = 8.5$ ), followed by hospitals ( $z = 5.6$ ) and pharmacies ( $z = 4.6$ ).

### 3.3. Epilepsy stigma and economic cost

We investigated possible associations between epilepsy cost and stigma using data from all 239 participants. Although no significant correlation was found between the total cost of epilepsy and stigma scores ( $p = 0.938$ ), we noted that the level of perceived stigma tended to be directly proportional to the amount of money spent on traditional medicine (Spearman rho =  $0.13$ ,  $p = 0.050$ ). Moreover, a significant positive correlation was observed between the stigma scores and the total epilepsy cost expressed as multiples of household income (Spearman rho =  $0.24$ ;  $p < 0.001$ ) (Fig. 1). There was a negative correlation, albeit weaker, between household income and stigma

**Table 3**  
Monthly costs of epilepsy in Maridi State, South Sudan.

	Persons concerned by this item: n (%) <sup>a</sup>	Cost in USD: mean (SD)	Weighted mean cost <sup>b</sup> in USD	Proportion
<b>Direct cost items</b>				
Medical consultations	43 (45.3)	2.5 (18.6)	1.1	3.4%
Hospitalisations	32 (33.7)	22.4 (79.5)	7.5	22.8%
Anti-epileptic drugs	81 (85.3)	14.1 (41.6)	12.0	36.6%
Transport	30 (31.6)	10.5 (42.4)	3.3	10.1%
Traditional medicine	30 (31.6)	28.3 (135.1)	8.9	27.1%
<i>Total weighted direct cost</i>			32.8	100%
<b>Indirect cost items</b>				
Incurred by PWE	58 (61.1)	2.6 (3.7)	1.6	28.6%
Incurred by caregiver	77 (81.1)	4.9 (9.7)	4.0	71.4%
<i>Total weighted indirect cost</i>			5.6	100%
<b>Total cost items</b>				
Direct cost			32.8	85.4%
Indirect cost			5.6	14.6%
<i>Total weighted epilepsy cost</i>			38.4	100%

PWE: Persons with epilepsy; SD: Standard deviation; USD: American dollars.

<sup>a</sup> Proportion of participants who reported a non-zero economic cost for the investigated item.

<sup>b</sup> Weighted mean cost = Proportion of persons concerned by this item × Mean cost.

**Table 4**  
Monthly costs of epilepsy in Amadi State, South Sudan.

	Persons concerned by this item: n (%) <sup>a</sup>	Cost in USD: mean (SD)	Weighted mean cost <sup>b</sup> in USD	Proportion
<b>Direct cost items</b>				
Medical consultations	48 (33.3)	1.1 (3.3)	0.4	3.4%
Hospitalisations	37 (25.7)	3.7 (16.2)	1.0	8.5%
Anti-epileptic drugs	138 (95.8)	5.9 (3.8)	5.7	48.3%
Transport	85 (59.0)	5.7 (20.0)	3.4	28.8%
Traditional medicine	41 (28.5)	4.4 (9.3)	1.3	11.0%
<i>Total weighted direct cost</i>			11.8	100%
<b>Indirect cost items</b>				
Incurred by PWE	94 (65.3)	2.8 (3.2)	1.8	31.0%
Incurred by caregiver	130 (90.3)	4.4 (3.3)	4.0	69.0%
<i>Total weighted indirect cost</i>			5.8	100%
<b>Total cost items</b>				
Direct cost			11.8	67.0%
Indirect cost			5.8	33.0%
<i>Total weighted epilepsy cost</i>			17.6	100%

PWE: Persons with epilepsy; SD: Standard deviation; USD: American dollars.

<sup>a</sup> Proportion of participants who reported a non-zero economic cost for the investigated item.

<sup>b</sup> Weighted mean cost = Proportion of persons concerned by this item × Mean cost.

scores (Spearman rho = -0.17,  $p = 0.007$ ).

The linear regression model found that residing in Maridi, traditional medicine use, and a history of physical abuse in PWE were all associated with increased stigma scores (Table 5). The inclusion of age and sex in the model excluded many participants due to several missing values, thereby reducing the performance of the model ( $R^2 = 0.26$ ). Upon excluding age and sex, the performance of the final model was improved ( $R^2 = 0.32$ ). Of note, both models yielded similar outcomes in terms of the covariates which were significantly associated with epilepsy stigma.

#### 4. Discussion

Our study reveals that PWE in South Sudan are confronted with low to moderate perceived stigma, but high economic cost because of epilepsy. Mean stigma scores ranging from 6.5 in Amadi State to 13.9 in the Maridi State place most participants in the lower half of the stigma scale, although 16 (6.7%) of them experienced high perceived stigma. A

positive correlation was noted between stigma scores and epilepsy cost relative to household income, suggesting that increased perceived stigma causes the affected families to spend more on epilepsy care, sometimes beyond their income. Given that epilepsy stigma is often fuelled by misconceptions about the cause and treatment of seizures [2], it is expected that addressing the latter will go a long way to discourage traditional medicine use, thereby reducing the financial burden of epilepsy. The negative correlation between stigma scores and household income, as depicted by our two study sites (Maridi: low income and high stigma, vs Amadi: high income and low stigma) also suggests that as the financial situation of the household improves, less stigma is perceived by the PWE. These findings align with observations in Turkey where PWE with higher incomes had the lowest perceived stigma scores [15]. A possible explanation is that with the higher incomes, PWE can afford optimal epilepsy care and improve their quality of life. However, we could not demonstrate a direct association between stigma scores and household income in the multivariable model; this warrants further research using different approaches for stigma assessment in PWE and their families.

Mean stigma scores were higher in Lui compared to the other two villages of Amadi State. This is most likely because in Lui there is a reference hospital which provides epilepsy care and receives PWE from neighbouring villages; the fact that Lui serves as a hub where many PWE come for treatment may have increased the stigma perceived by participants who reside there permanently. Different socio-anthropological contexts may also explain the differences in the level of stigma perceived by PWE in different sites. Therefore, qualitative studies are underway to better explore local epilepsy beliefs in the study villages. When compared with other studies which used the Kilifi scale to assess perceived stigma in PWE, the mean stigma score of 13.9 in Maridi was close to the 15.9 obtained in Ugandan patients [16], but much higher than that of PWE in Ethiopia [17]. These differences can be explained by the fact that participants in both the Ugandan study and the Maridi villages were relatively young (mean ages 14.9 and 16.2 respectively), and younger age has been associated with greater perception of stigma [12,18]. Meanwhile, the higher age of participants in the Ethiopian study (mean age 29.3) and in Amadi State (mean age 23.3) could account for lower stigma scores [17]. We also found that stigma increased with longer duration of epilepsy as well as with increasing frequency of physical abuse and sexual harassment, similar to findings in other SSA settings [12,19]. Other factors that have previously been associated with increased epilepsy stigma in SSA settings include: epilepsy-related injury, other chronic illnesses, longer duration of AED use, and high seizure frequency [4,16,17]. We were however unable to explore all these aspects during this pilot study.

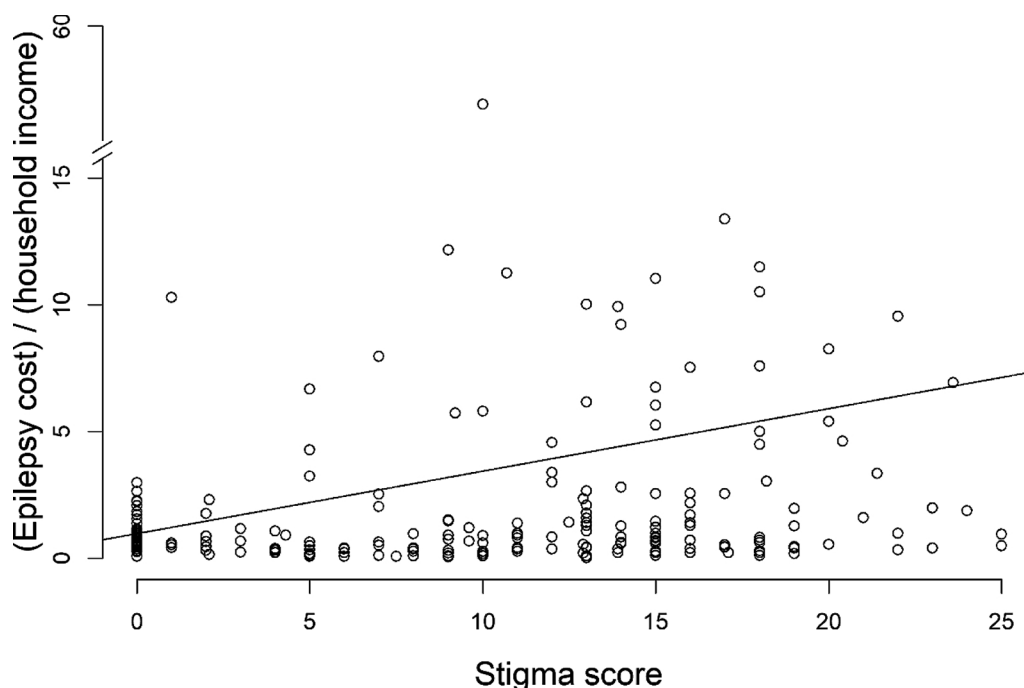


Fig. 1. Correlation plot of epilepsy cost (relative to household income) and stigma score.

**Table 5**  
Linear regression model investigating the factors associated with stigma scores.

Model* covariates	Regression coefficient (95% Confidence interval)	P-value
Intercept	11.87 (9.15–14.58)	< 0.001
Household income	0.009 (–0.005 to 0.024)	0.215
Residence in Amadi State	–7.08 (–8.70 to –5.45)	< 0.001
Traditional medicine use	1.91 (0.22–3.59)	0.027
Anti-epileptic drug use	0.16 (–2.63 to 2.96)	0.908
History of physical abuse	3.04 (1.11–4.97)	0.002
History of sexual harassment	1.16 (–1.55 to 3.86)	0.400

\* R-squared = 0.32

Only 6.7% of our participants were identified as experiencing high perceived stigma. This is much lower than what was found in Uganda [16] and Ethiopia [17], where about a third of the recruited PWE were categorised as having high perceived stigma using the Kilifi stigma scale. One possible explanation for this discrepancy could be the fact that the authors of the studies [16] and [17] used the 66<sup>th</sup> percentile of the collected data as cut-off value for high perceived stigma, which inevitably categorized all PWE found between 67<sup>th</sup> to 100<sup>th</sup> percentile as having higher stigma. Our approach to use the 66<sup>th</sup> percentile of the Kilifi scale (i.e. a score of 20) as the cut-off score is more appropriate.

Our study is among the first to provide empirical data on the economic cost of epilepsy in South Sudan. The mean weighted total cost of epilepsy was much lower in Amadi State (17.6 USD) compared to Maridi (38.4 USD); the situation in Amadi State is closer to previous findings in 2017 in the Democratic Republic of the Congo, where a monthly epilepsy cost of 20.1 USD was obtained using a similar approach [13]. The higher costs in Maridi could be explained by more frequent resort to traditional medicine, and also AED costs which are often subsidized at the Lui Hospital in Amadi State. In practice, the real economic impact of epilepsy may be higher if we account for the loss of educational or professional opportunities due to the condition. For instance, a good number of our study participants reported to have stopped school and employment because of epilepsy. Therefore urgent interventions such as provision of affordable or free AED, community-based epilepsy care, and community education about epilepsy should

be implemented to minimise epilepsy-related expenses and facilitate the social reintegration of PWE [20].

Additional data collected during the second phase of the study in Amadi revealed that caretakers more often responded to the investigators’ questions, and frequently reported a higher perceived stigma than PWE themselves. While this finding contrasts with previous observations [12], it brings to light the fact that epilepsy stigma does not only affect the patient, but seems to touch family members to a greater extent. Indeed, research on family stigma due to epilepsy has shown that relatives of PWE are often confronted with shame, and may experience as much stigma as the PWE themselves [21,22]. Moreover in onchocerciasis-endemic areas like our study sites, the occurrence of OAE in clustered homes and communities further compound the stigma experienced by PWE and their families [23].

A limitation of our study is the fact that age and sex information was missing for several participants as some investigators omitted to note these data on the paper questionnaires during the first phase of the study in Maridi. However, the population characteristics obtained from the partial data seem to be a true reflection of the entire study population; indeed, the socio-demographic characteristics of our participants in Maridi showed very close similarities with previous data obtained from over 700 PWE in the Maridi villages in 2018 [10]. In addition, the output of our multivariable analysis were not significantly different with or without the inclusion of age and gender into the model. Another limitation was the fact that more responses were given by caretakers rather than PWE themselves, which may have introduced bias in the stigma evaluation. Lastly, we must note that the performance of the Kilifi stigma scale may be context-dependent, and its outcome must therefore be interpreted with caution when used outside the Kilifi area.

### 5. Conclusion

The findings from this pilot study show that PWE in South Sudan are frequently faced with stigma, which is associated with high economic cost of epilepsy because of traditional medicine use. Demystifying epilepsy and making AED treatment more accessible may significantly reduce epilepsy related stigma and decrease the cost of epilepsy care.

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## Authors' contributions

RC, JNSF and SRJ conceived the study. JNSF, SRJ and MT were involved in performing the survey and data collection. JYC and MYL were involved in facilitating the project. JNSF analyzed the data and wrote the first draft. All authors critically reviewed and approved the final manuscript.

## Declaration of Competing Interest

The authors declare no conflicts of interest.

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