

# South Sudan Nodding Syndrome Study

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# Introduction

## What is known about NS

- ***Age-groups effected?***
  - 5-18 years
  - 2-22 years
- ***First symptoms***
  - Head nodding/seizures, triggered by food or cold wheather
- ***Disease progression and outcome***
  - Mental retardation, delayed growth and puberty
  - Psychiatric symptoms
  - School dropout
  - Psychosocial and economic complications

## NS and OAE (Onchicerciasis Associated Epilepsy )

- OAE include 3 conditions: NS, Epilepsy and Nakalanga Syndrome

# Introduction

## **Geographical locations of NS cases**

- Tanzania (southern), 1934
- Liberia, 1983
- South Sudan (Western Equatoria), 1991
- Uganda (western & northern), 1994 and 2003 respectively
- Democratic Republic of Congo, 2016
- Cameroon, 2018
- Central African Republic, 2019

## **Outstanding questions?**

- Size of the NS problem in Mundri, South Sudan
- Distribution of NS cases in Mundri, South Sudan
- Is there clustering of cases?
- Does the epidemiology study helps us find the cause of the disease?

# Epidemiology study – methods

## **Prevalence household survey**

- Villages in Mundri West and East counties

## **Case-control studies**

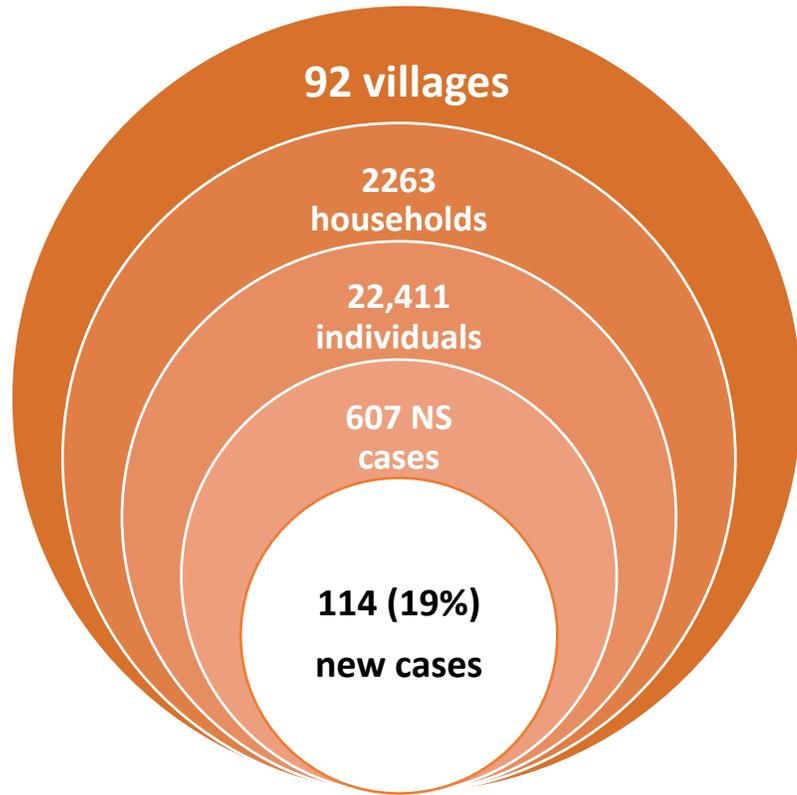
## **Questions asked in each household**

- Identify NS cases in households (affected and number)
- Asked questions on behaviour and living conditions
- Asked questions on household resources and socioeconomic status
- Exposure to animals, source of water, internal displacement, meals, ivermectin

## **Analysis**

- Geographical distribution and clustering of NS cases, spatial & binomial regression
- Epidemiological risk factors, univariate and multivariate models

# Epidemiology study – findings 1

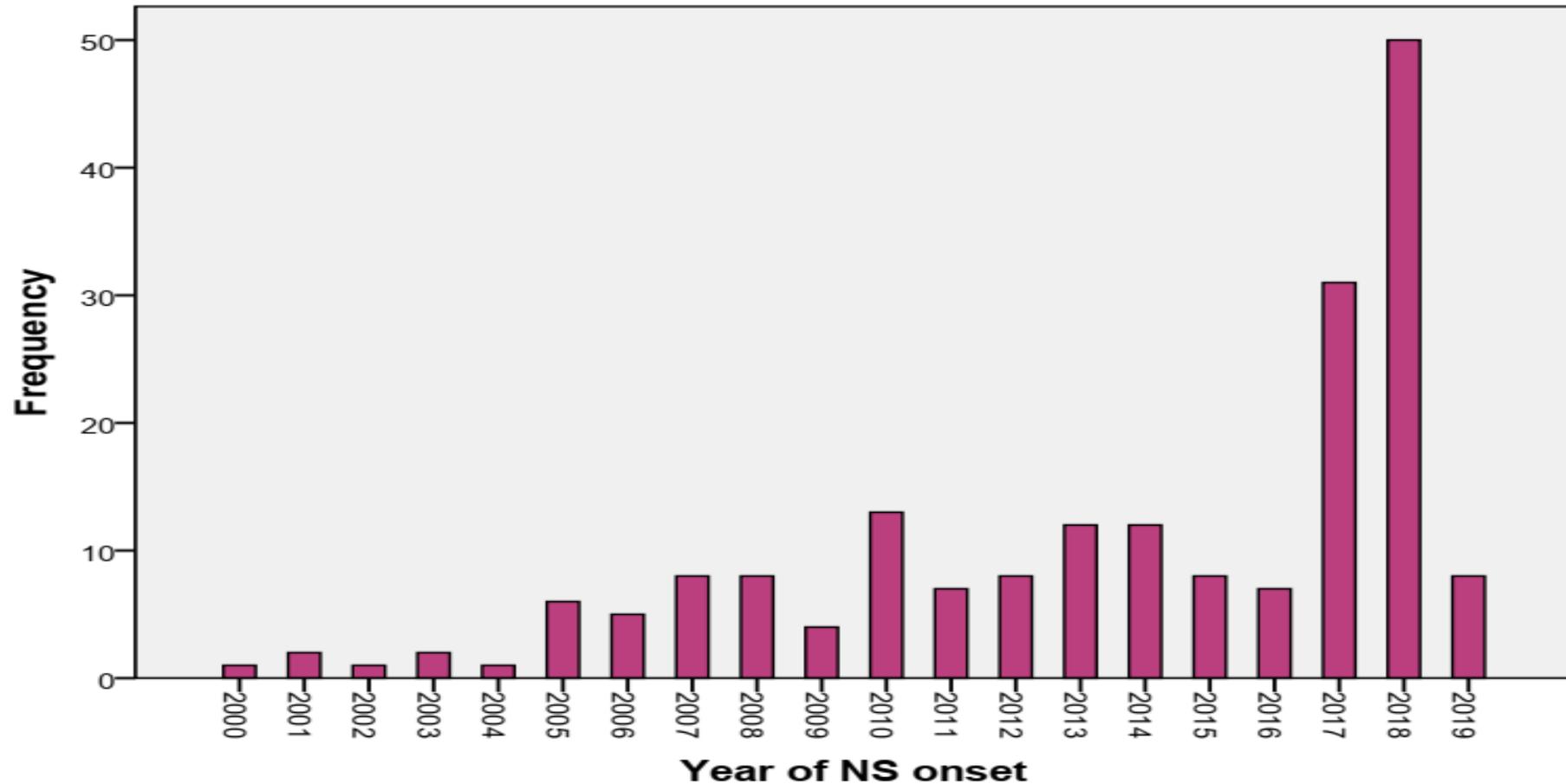


Higher numbers/rates compared to other countries

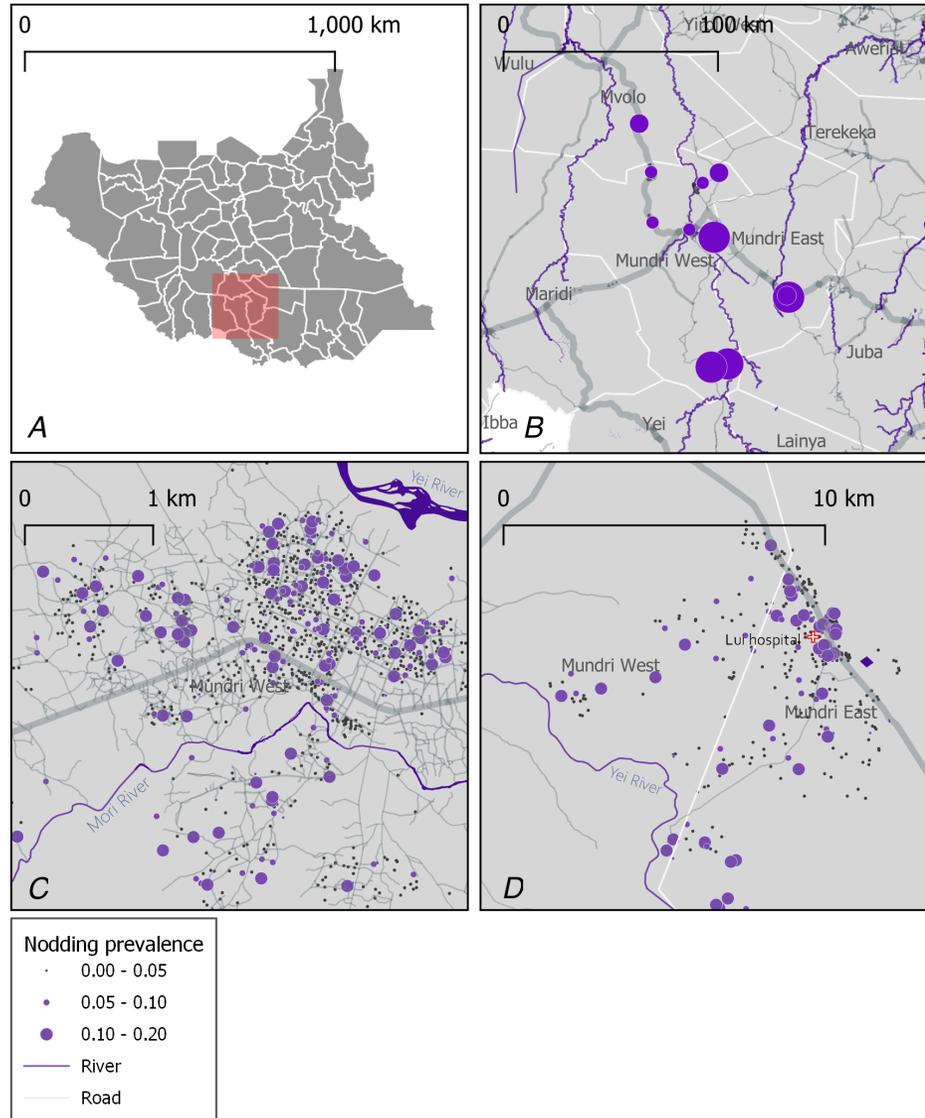
Two or more NS cases in some households

Study area	Prevalence of NS (all age groups)	Prevalence of NS ( $\leq 18$ years)
Amadi	2.4	1.8
Biyokoriwa	2.5	0.2
Delewa	3.5	0.3
Diko	13.7	3.1
Gulu	3.5	0.8
Kulundu	2.2	0.6
Lui	1.2	0.2
Mbara	1.6	0.6
Mideh	5.6	1.9
Moba	2.7	3.8
Mundri	2.2	0.4
Witto	4.0	4.5
Yeri	4.5	4.2
<b>Total</b>	<b>2.7</b>	<b>0.9</b>

# Epidemiology study – findings 2



# Epidemiology study – findings 3



NS is significantly clustered between adjacent households (OR 6.9, 95% CI 4.2-11.5)

Multiple clusters of high NS prevalence exist within the communities surveyed and concentrated within Mundri town and along the main branch of the Yei River

# Epidemiology study – findings 4



All behaviors around river and using river water increases the risk of having NS in households:

- Drinking (OR 1.9, 95% CI 1.1-3.2)
- Cooking (OR 1.8,  $p = 0.024$ )
- Hand washing (OR 1.8, 95% CI 1.1-3.1)
- Bathing (OR 1.8, 95% CI 1.1-3.0)

Living near a river significantly increases the risk of having NS in households (OR 0.9, 95% CI 0.9-1.0)

- ??? *Onchocerca volvulus* (OV)
- ??? Blackflies
- ??? Something else

# Epidemiology study – findings 5



Exposure to poultry increase the risk of having NS case in households (OR 1.4, 95% CI 1.1-1.8)

No association with displacement, nutritional status and ivermectin

# Epidemiology study – findings 6

Clinical presentation of NS  
in Mundri

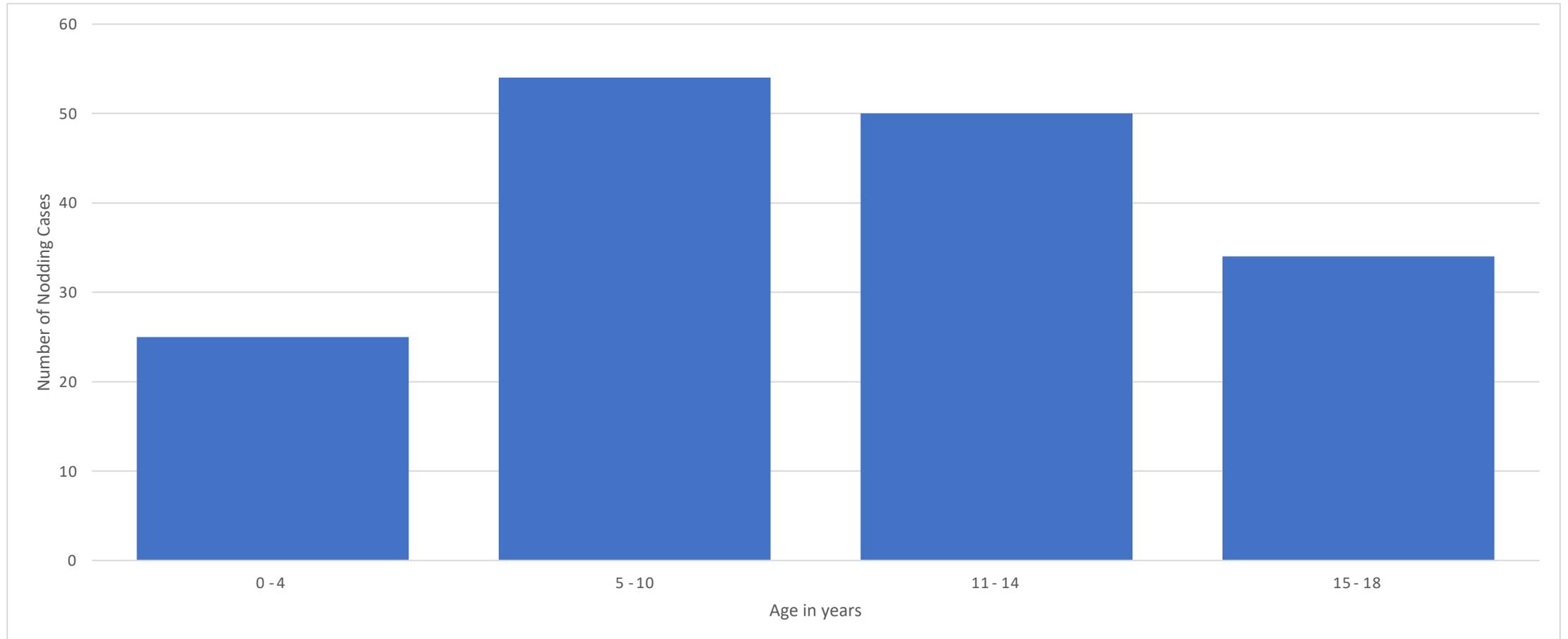
Individual risk factors

**FURTHER ANALYSIS OF DATA**

Behaviour

Others

# NS onset by age group



## Clinical features of NS in Mundri, South Sudan

<b>Features</b>	<b>Number (%)</b>
Head nodding seizure only	50 (22.32%)
Head nodding plus other seizures	174 (77.68%)
Generalized convulsions	160 (71.4)
Triggered by cold temperature	116 (59.8)
Triggered by the sight of food	108 (55.7)
Wasting	17 (23.6)
Stunted growth	16 (22.2)
Speech disorder	14 (19.4)
Absences	10 (13.9)
Delayed sexual development	9 (17.3)
Behavioral abnormalities	4 (5.6)
<b>Total cases</b>	<b>224</b>

	Cases		Controls		OR (95% CI)	P value
	#	%	#	%		
MALE GENDER	126	56.2			1.28 (0.97 - 1.69)	0.07
CONSUMPTION OF BUSH MEAT						
Monkey	41	46.1	29	43.3	1.12 (0.56 - 2.23)	0.73
Baboon	34	38.2	22	32.8	1.26 (0.62 - 2.61)	0.49
Elephant	44	49.4	32	47.8	1.07 (0.54 - 2.12)	0.84
Antilope	65	73.0	53	79.1	0.72 (0.31 - 1.61)	0.38
Buffalo	53	59.5	44	65.7	0.77 (0.38 - 1.56)	0.44
Squirrels	48	53.9	39	58.2	0.84 ( 0.42 - 1.67)	0.59
Bats	5	5.6	1	1.5	3.92 (0.42 - 188.55)	0.18
<b>Rat-meat</b>	<b>11</b>	<b>12.4</b>	<b>1</b>	<b>1.5</b>	<b>9.31 (1.27 - 406.51)</b>	<b>0.01</b>
USE OF IVERMECTIN						
<b>Ivermectin intake during the last 5 years</b>	<b>36</b>	<b>64.3</b>	<b>514</b>	<b>42.8</b>	<b>2.40 (1.33 - 4.43)</b>	<b>0.002</b>
Ivermectin doses received: 1	47	83.9	1,018	84.8	0.93 ( 0.44 - 2.21)	0.85
Ivermectin doses received: 2	9	16.1	182	15.2	1.07 (0.45 -2.26)	0.85
EXPOSURE & BEHAVIOR						
<b>Share bedroom with other children currently or in the pass</b>	<b>10</b>	<b>11.2</b>	<b>46</b>	<b>65.7</b>	<b>0.06 (0.02-0.16)</b>	<b>&lt;0.001</b>
<b>Share a bedroom with adults currently or in the past</b>	<b>27</b>	<b>30.3</b>	<b>43</b>	<b>61.4</b>	<b>0.27 (0.13-0.56)</b>	<b>&lt;0.001</b>

# Conclusion and recommendations

- High prevalence of NS in Mundri area
- New NS cases continue to emerge in the Mundri area
- River-related, OV, blackflies, other factors?????
- The novel findings needs to be further evaluated (poultry, bushmeat)

## **Care for current cases and their families**

- Nutritional support
- Economic and psychosocial support

## **Treatment of current cases**

- Does ridged epilepsy control prevent disease progression
- Treat for possible filarial infection

## **Prevention of new cases**

- Filarial treatment studies
- Anticonvulsive studies
- CDTI and blackfly control studies

# Acknowledgment

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- University of Utrecht
- University of Basel
- University of Rotterdam
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# Thank you!

## References

- <https://doi.org/10.3390/tropicalmed6040211>
- <https://doi.org/10.1371/journal.pntd.0010630>