RESEARCH PAPER

Types and prevalence of birth defects and the geospatial mapping of cases presenting to Rabaul Provincial Hospital, East New Britain Province, Papua New Guinea: A hospital based, mixed methods observational study

Anna Toti

MMED 2

A research paper presented as a requirement for:

Master of Medicine in Child Health Part 2

University of Papua New Guinea

School of Medicine and Health Sciences

Discipline of Child Health: Post Graduate Programme

Completed under the guidance and supervision of the following paediatricians:

Trevor Duke, Royal Childrens' Hospital, Melbourne, Victoria, Australia

Beryl Vetuna, Deputy Chief of Paediatrics, New Guinea Islands Region

Veronica Kalit, Paediatrician, Rabaul Provincial Hospital

Patrick Kiromat, Director Clinical Services, East New Britain Provincial Health Authority

Special Acknowledgement

East New Britain Provincial Health Authority

Rabaul Provincial Hospital Rural Outreach Team

Medical officers:

Danny Toti, Coordinator of Rural Outreach Programme

Sonny Kambual, Deputy Coordinator

Support staff:

Thomas Jubilee

For providing East New Britain Province population, Census and geospatial data

“Birth defects are structural or functional abnormalities, including metabolic disorders present from birth.”

- World Health Organisation 2014[2]

[Associated terms: congenital anomalies, congenital malformations, congenital abnormalities]

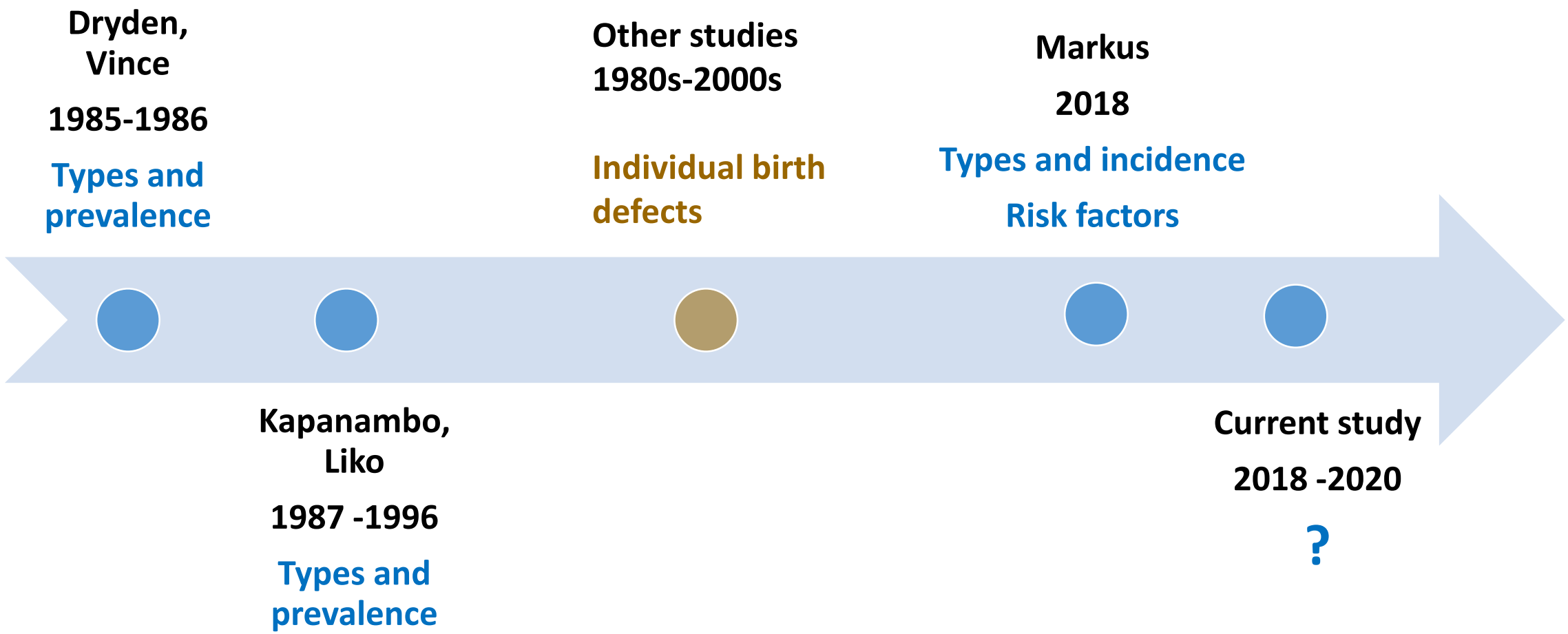
Global burden of birth defects

- 7.9 million (6%) children born with a birth defect per year
- Under 5 deaths from birth defects: 3.3 million per year
- 3.2 million children live with a disability due to a birth defect
- 94% birth defects and 95% of deaths occur in low and middle income countries (LMICs)
- The care and prevention of birth defects in LMICs has been shown to be cost effective and feasible and includes birth defects surveillance

March of Dimes: Global Report on Birth Defects: The hidden toll of dying and disabled children. 2006[1]

Birth defects research in Papua New Guinea

What is the contribution of the Rabaul birth defects study?



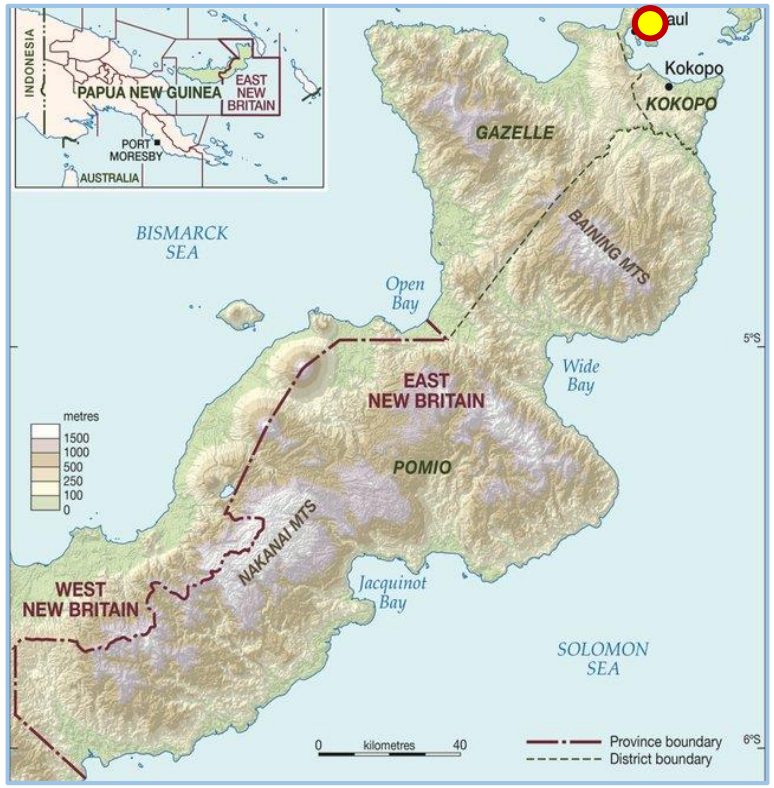


Image 1

Study location

Facility	Rabaul Provincial Hospital formerly Nonga General Hospital
Geography	Rabaul District, East New Britain Province, Papua New Guinea
	Northern coast less than 100m above sea level along the Pacific Ring of Fire
Population	328, 369 people in the province Pop. Density 21/km^2
	12, 073 births reported in 2019
Health service coverage	32 registered functioning health facilities 1 specialist medical officer per 16, 419 people

Pre-research observations

2 groups of infants born with birth defects are managed at Rabaul Provincial Hospital:

i. Newborns delivered at the hospital
ii. Newborns referred in from peripheral health facilities

Research background

Problem statement

- The appearance of birth defects in the population are significant health occurrences in the East New Britain Province

- No population based or hospital based studies of birth defects have been conducted in East New Britain, therefore their prevalence remains unknown

Research focus

Aim

1. Describe the types of birth defects seen in infants delivered at or referred to Rabaul Provincial Hospital

2. Determine the prevalence of birth defects in a sample population of live born infants delivered at Rabaul Provincial Hospital

Research focus

Objectives

1. Perform a literature review to determine the current practice of conducting birth defects research and surveillance and update information throughout the lifespan of the study

2. Design a hospital based prevalence study of birth defects occurring in a consecutive series of 2000 live births

3. Utilise existing geographic information system (GIS) geodata for East New Britain Province to map cases in the live birth series as well as referral cases

4. Critically analyse study findings to determine the contribution of this study to ongoing birth defects research in Papua New Guinea

Literature search of current practices in birth defects research

Diagnosis of birth defects	▪ Birth Defects Surveillance: Quick reference handbook of selected congenital anomalies and infections[3] ▪ Birth defects in Papua New Guinea[4] ▪ Comprehensive New Born Screening: Handbook for Screening Visible Birth Defects at All Delivery Points[5]
Case ascertainment	▪ **Birth Defects Surveillance: A manual for programme managers 2nd Ed[2]**
Case coding ICD-10-CM: Q00-Q99	▪ **Birth Defects Surveillance: Quick reference handbook of selected congenital anomalies and infections[3]** ▪ International classification of diseases Tenth Revision, Clinical Modification: ICD-10-CM
Data collection methods	▪ Birth Defects Surveillance: A manual for programme managers 2nd Ed[2]
Data analysis and interpretation	▪ Birth Defects Surveillance: A manual for programme managers 2nd Ed[2]
Application of spatial epidemiology to birth defects study	▪ Birth Defects Surveillance: A manual for programme managers 2nd Ed[2] ▪ Public Health Informatics and Information systems

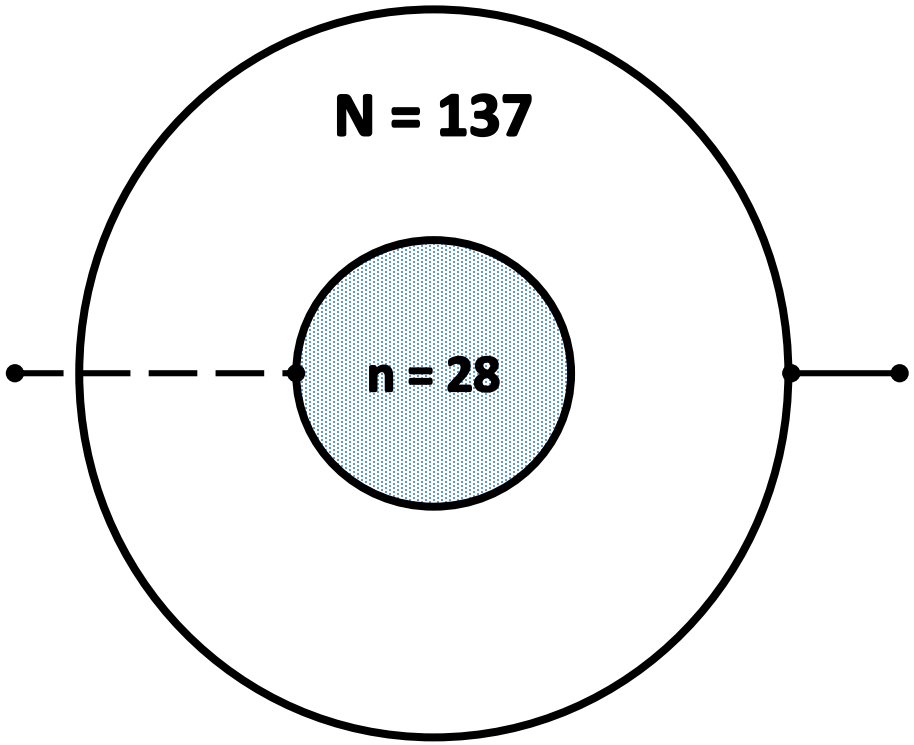
Study design

1. Observational Study: 2018 - 2020

Live birth study

- Cross section: Hospital deliveries
- Timeline: January 1st 2019 to December 19th 2019
- Prospective
- Subpopulation: All live born babies with a recognisable birth defect (numerator) **observed in a series of 2000 consecutive live births** (denominator)

Primary Outcome: Types and prevalence of birth defects



Mapped cases

- Case series: All cases - Referrals and hospital deliveries
- Timeline: January 1st 2018 to November 31st 2020
- Retrospective - prospective
- Population: All live born babies with a recognisable birth defect **observed at Rabaul Provincial Hospital including those in the live birth series**

Secondary Outcome: Mapped data

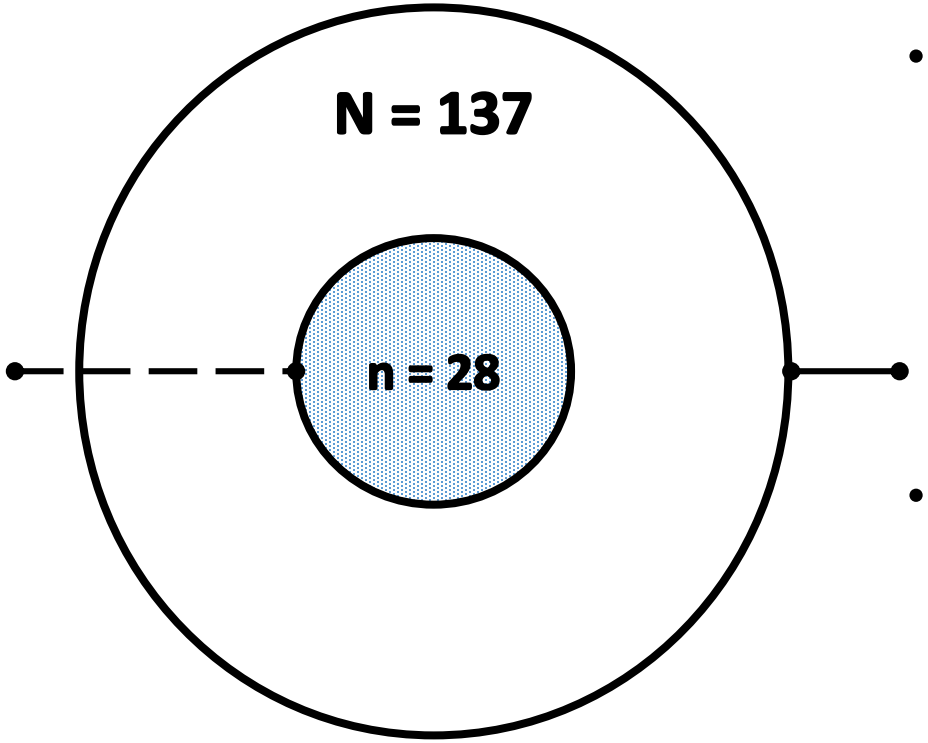
Study design

2. Study population: Sampling and selection

Live birth study

- Purposive sampling
- Inclusion criteria
  - Live born
  - Age at diagnosis: ≤28 days
  - Recognisable birth defect
  - Both major and minor
- Exclusion criteria
  - Still born or miscarriage
  - Gestational age less than 28 weeks
  - Weight < 1000 grams

No comparison group



Mapped cases

- Convenience sampling (referrals)
- Inclusion criteria
  - Live born
  - Age at diagnosis: ≤28 days
  - Recognisable birth defect
  - Both major and minor
  - Maternal residence known
- Exclusion criteria
  - Gestational age less than 28 weeks
  - Weight < 1000 grams

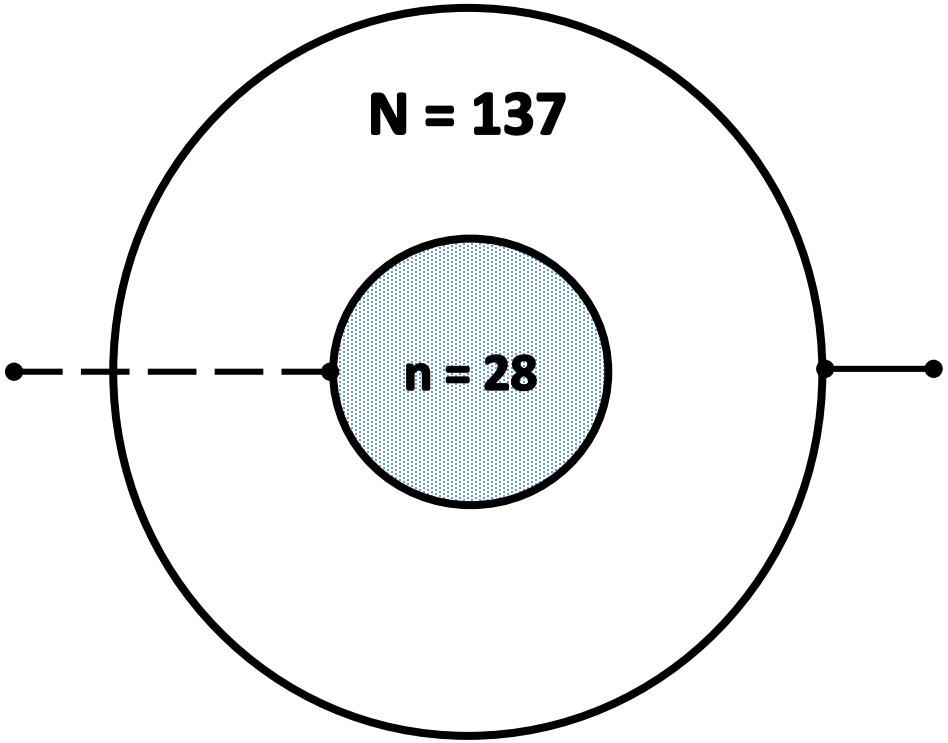
No comparison group

Methodology

1. Data collection and management

Live birth study

- Electronic Proforma PDF form
- Designed using Adobe LiveCycle ES 8.2 software for use with Android operating system
- Printable version filled manually
- Data organised into database spreadsheet in Microsoft Office Excel 2016 software
- Descriptive data analysis using IBM-SPSS v23



Mapped cases

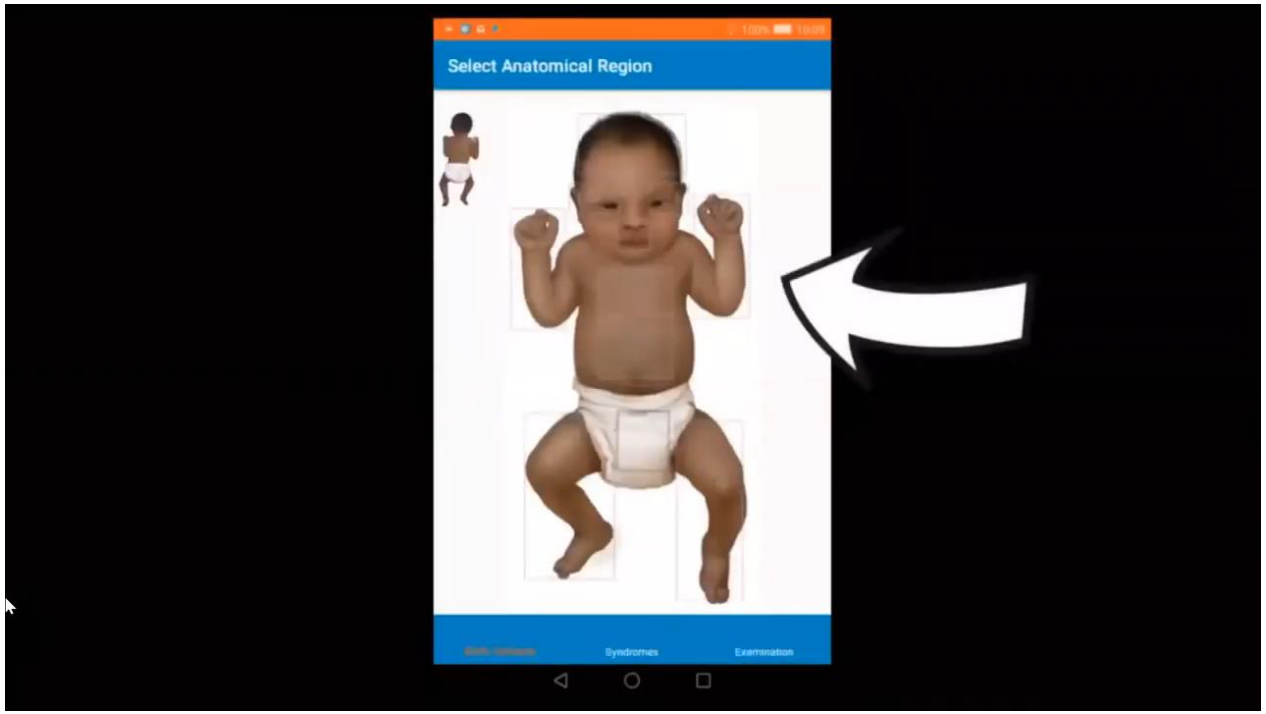
- Electronic Abstractor PDF form
- Designed using Adobe LiveCycle ES 8.2 software for use with Android operating system
- Printable version filled manually
- Data organised into database spreadsheet in Microsoft Office Excel 2016 software
- Residential data exported from Excel to open source Quantum GIS software for mapping

Ethical approval: Hospital administration; informed consent for participants of live birth study
Patient confidentiality: Unique study identifier assigned to each case. Electronic files encrypted

Methodology

2. Additional tool for diagnosis in case ascertainment

Global Birth Defects Description and Coding (GBDDC) Application



Video 1

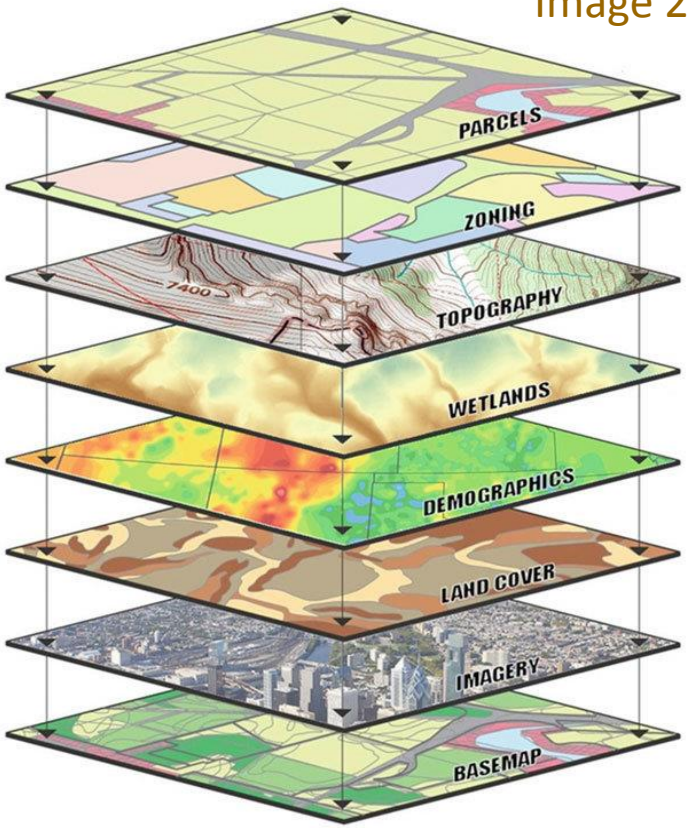
Methodology

3. Geographic information system data mapping

- Enabling technology for spatial epidemiology
- Links together geographically referenced information
- Basic function – cartography (mapping)
- Complex functions – spatial analysis and disease modelling, use of Bayesian statistics

Current birth defects study: Mapping function only

Image 2



RESULTS:

I. Live birth study

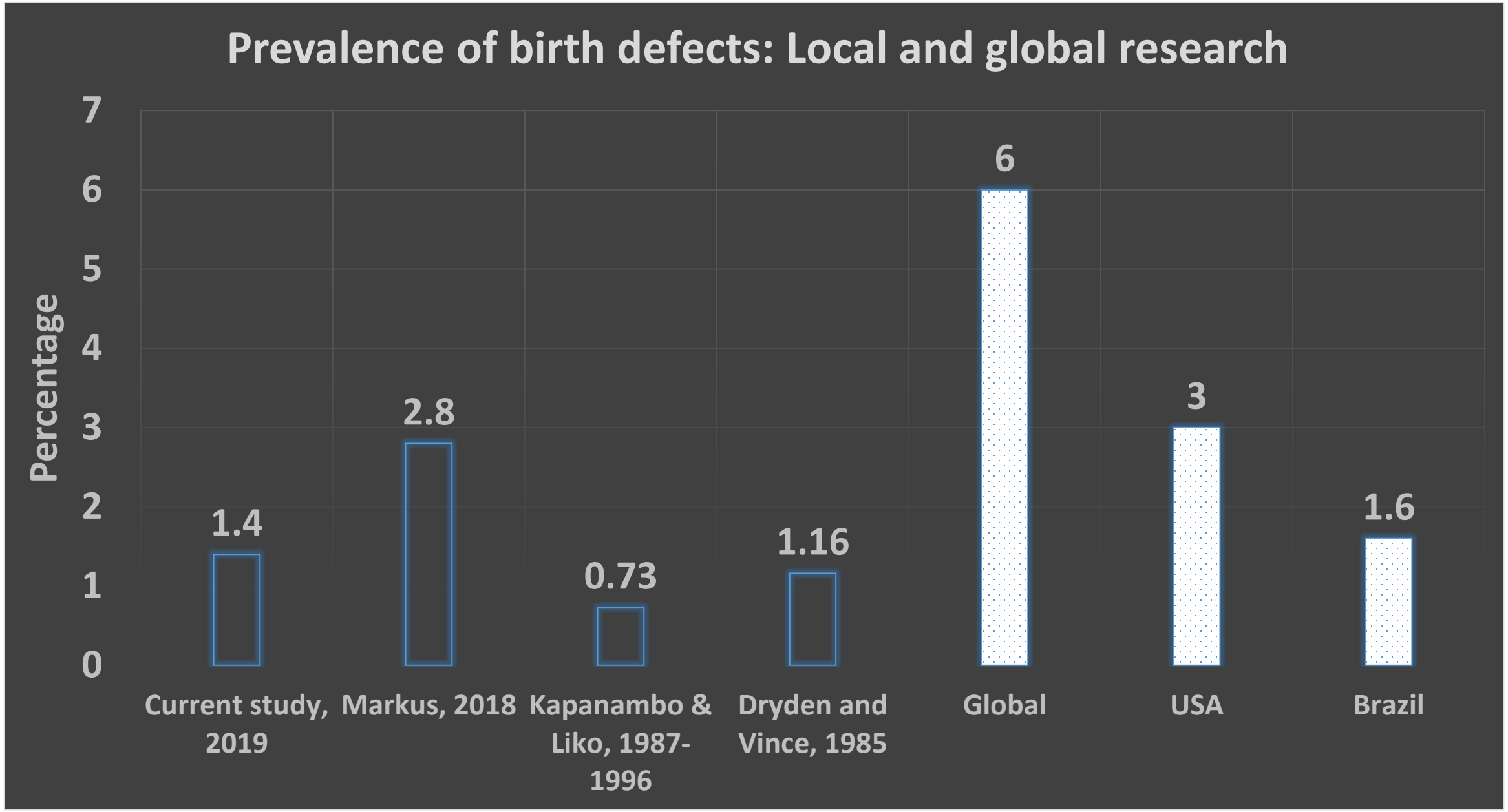
Types and prevalence of birth defects

Table 1. DEMOGRAPHIC PROFILE OF PATIENTS [n= 28]

Characteristic	n	%
Gender (%)		
Male	14	50
Female	14	50
Mean postnatal age at diagnosis (days)	1	-
Maturity at birth		
Term	18	64
Preterm	10	36
Twinning	2	7
Weight at birth (kg)		
Low <2.5	13	46
Normal 2.5-3.5kg	14	50
High >3.5	1	4
Maternal		
Mean age	28	-
Median parity	3	-
Primary residence		
Rural	22	79
Urban	6	21

Table 2. TYPES & PREVALENCE OF BIRTH DEFECTS [n=28]

System or body region	Cases (n)	Prevalence (%)
Thorax	**10/2000**	**0.5**
Congenital heart disease	9	0.45
Tracheo-oesophalgeal fistula	1	0.05
Genetic faults	**8/2000**	**0.4**
Down syndrome	4	0.2
Treacher-Collin syndrome	1	0.05
Undiagnosed syndrome	2	0.15
Central nervous system, Head and neck	**7/2000**	**0.35**
Microcephaly	2	0.1
Isolated soft cleft palate	2	0.1
Cleft lip and palate	1	0.05
Isolated cleft palate	1	0.05
Pierre Robin sequence	1	0.05
Gastrointestinal system, Abdomen	**2/2000**	**0.1**
Gastroschisis	1	0.05
Duodenal atresia	1	0.05
Musculoskeletal, Limbs	**1/2000**	**0.05**
Syndactyly	1	0.05


Prevalence of birth defects: Local and global research
Percentage
7
6
5
4
3
2
1
0
1.4
2.8
0.73
1.16
6
3
1.6
Current study, 2019
Markus, 2018
Kapanambo & Liko, 1987-1996
Dryden and Vince, 1985
Global
USA
Brazil

Graph 1

RESULTS:

II. Case series

Mapped birth defects

Map 1: Chloropleth map of distribution of birth defects

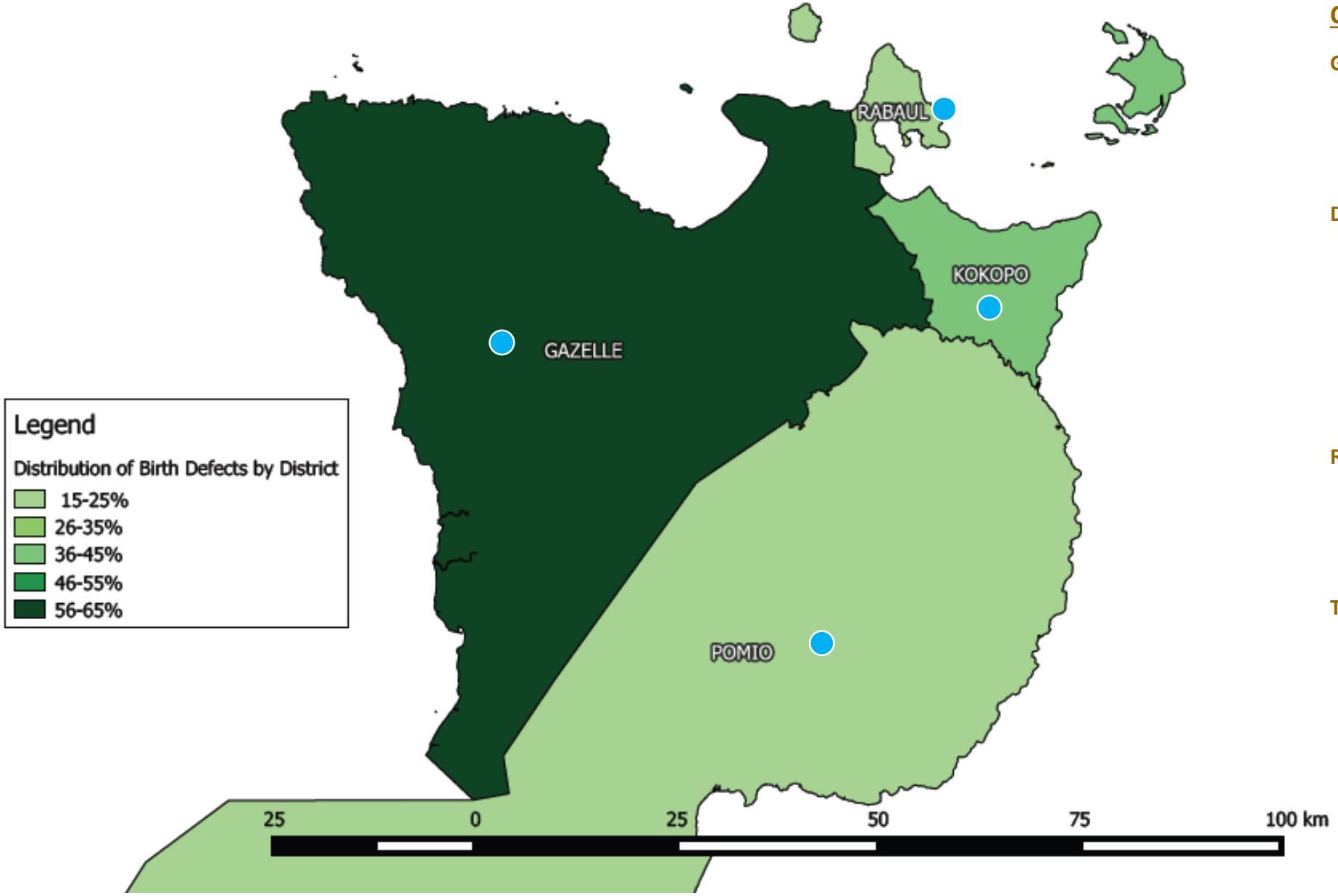
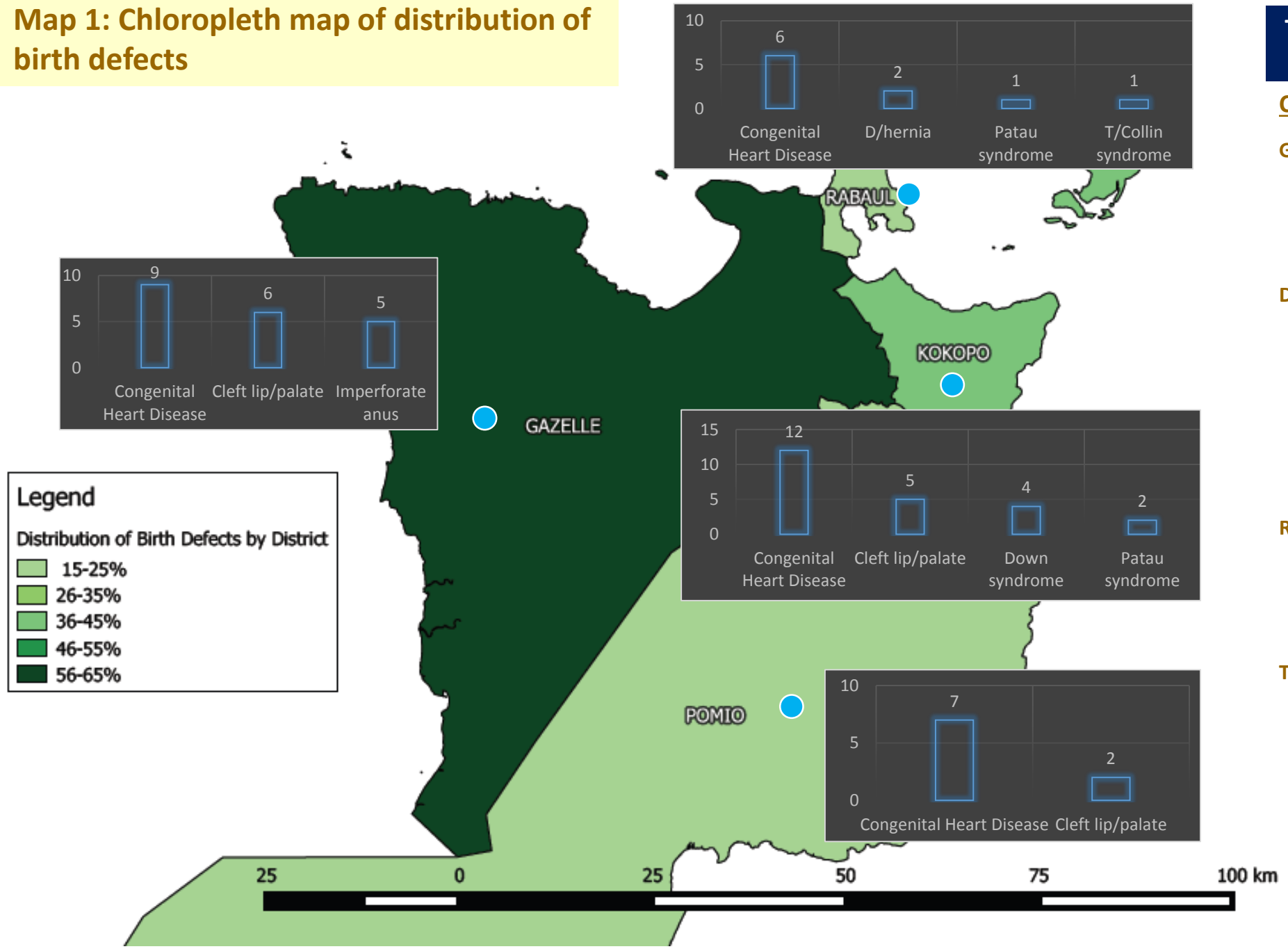


Table 3. DISTRIBUTION OF BIRTH DEFECTS: N=137

Characteristic	n	%
Gender		
Male	75	55
Female	62	45
Distribution across districts		
Gazelle	58	42
Kokopo	44	32
Pomio	19	14
Rabaul	16	12
Rural – Urban distribution		
Rural	112	**82**
Urban	25	18
Top 6 defects		
Congenital heart disease	34	25
Genetic faults	30	22
Oral maxillofacial defects	19	14
Perineal defects	14	10
Head and neck defects	12	8
Neural tube defects	9	7

Map 1: Chloropleth map of distribution of birth defects



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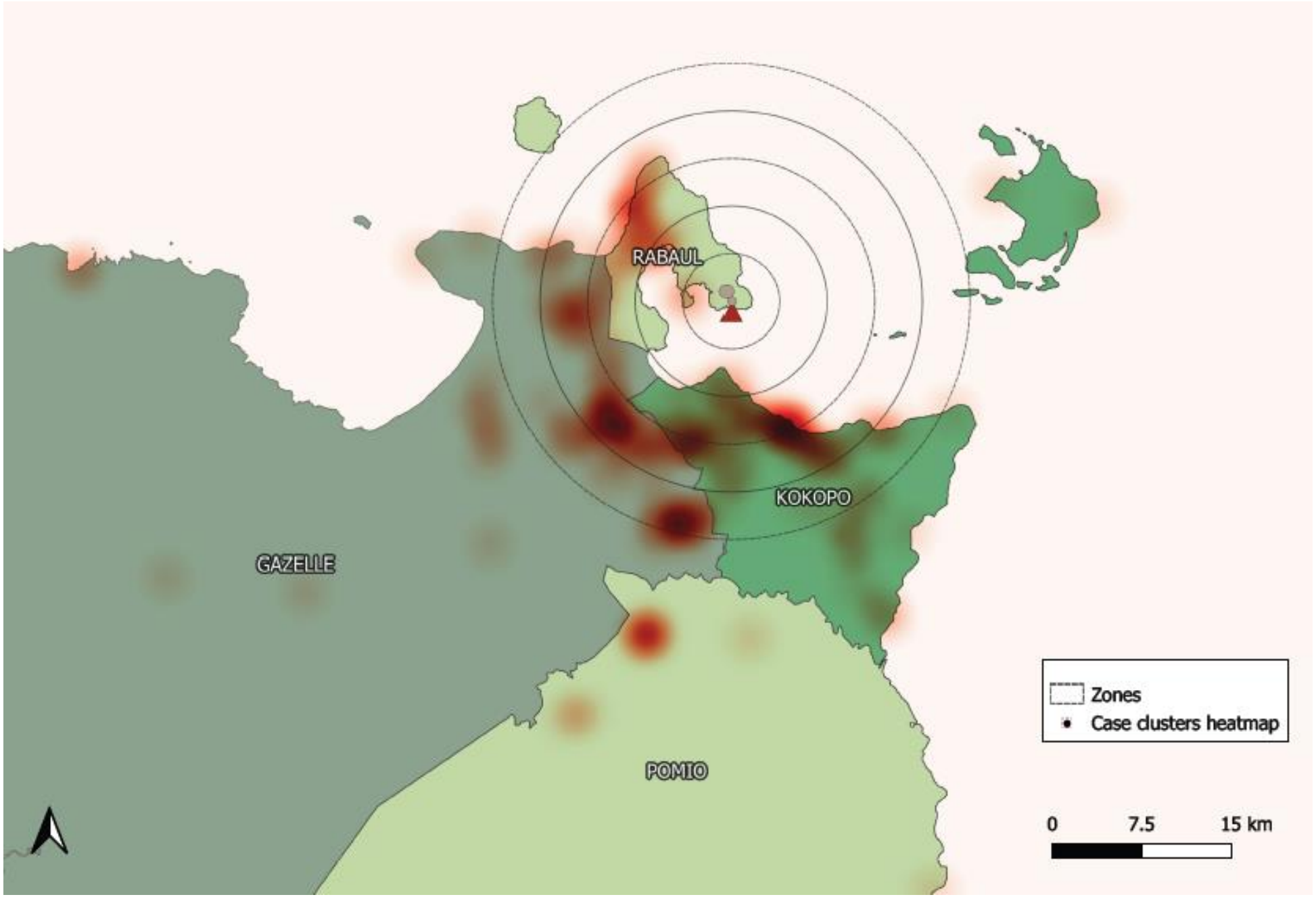
Map 2: Heat map of birth defect cluster intensity



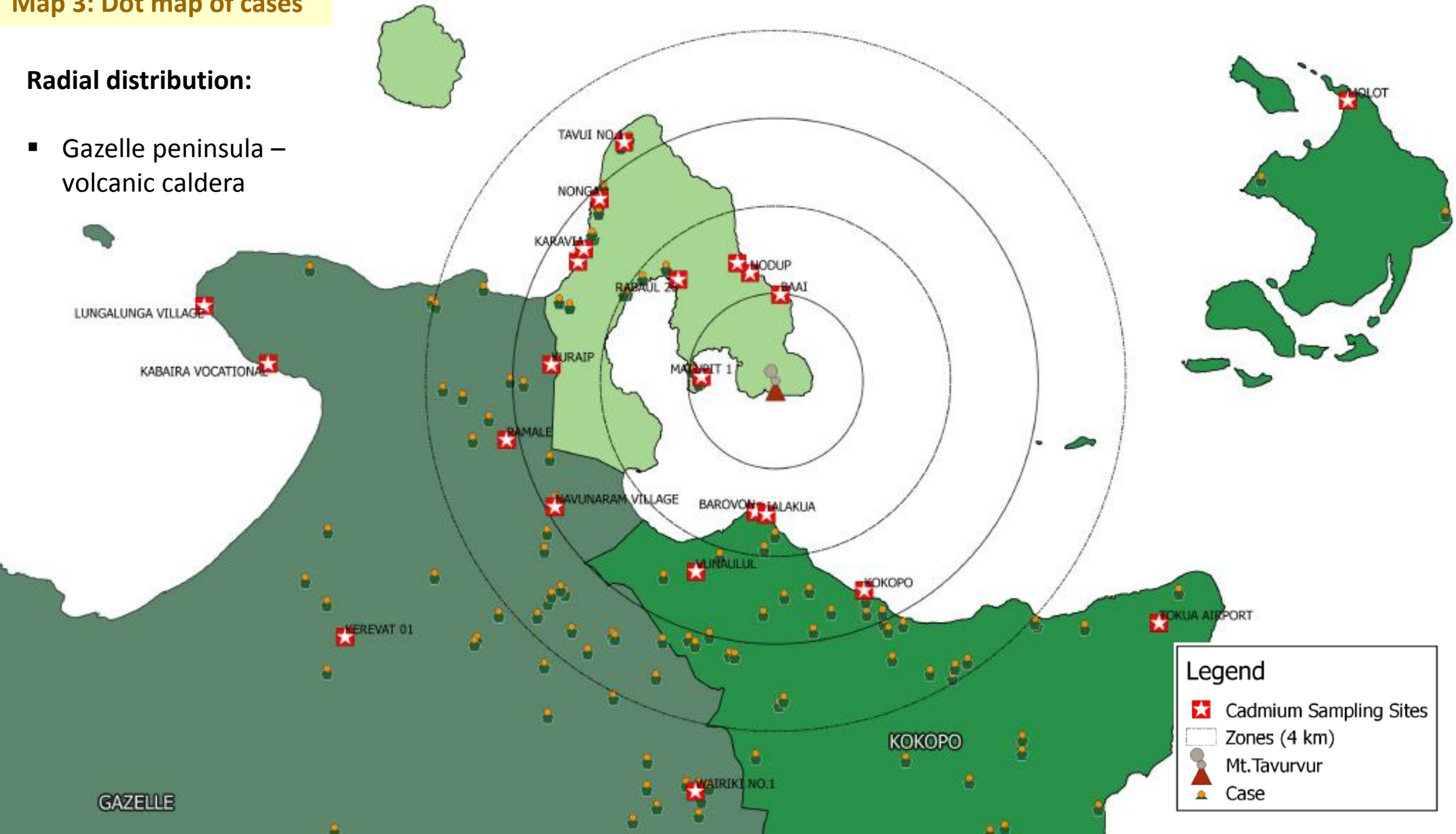
Increased cluster intensity along:

- Gazelle peninsula
- Borders - particularly inter-district borders
- Main mountain ranges

Map 3: Dot map of cases

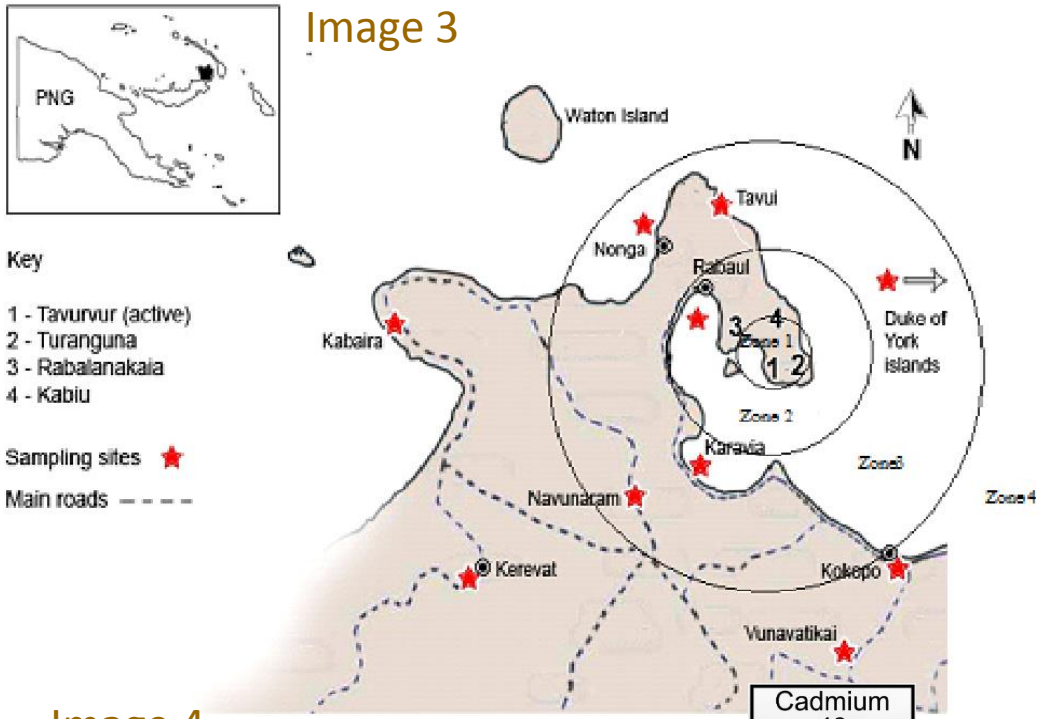
Radial distribution:

- Gazelle peninsula – volcanic caldera



Incidental research observation

Image 3



- Rabaul study dot map of birth defects shares geospatial data points with an agriculture sector study map showing sampling sites for cadmium
- Significant amount of heavy metal cadmium in samples of commonly consumed food in the Gazelle Peninsula
- In excess of WHO recommended provisional tolerable monthly intake (PTMI) > 25µg/kg

Image 4

Cadmium
48
Cd
112.41

Cadmium:

- Natural source in ENBP – volcanic emissions
- Bioaccumulation in major organs: kidneys, liver, pancreas and placenta
- Known human carcinogen
- Some evidence of teratogenicity and embryotoxicity (mainly animal studies)

Neural tube defects, congenital heart disease, oral maxillofacial defects, anophthalmia, head and neck anomalies, hypoplastic lungs, undifferentiated limbs, talipes

Determination of Baseline Data on Cadmium Levels for Selected Food Products from Volcanic Areas in ENBP, PNG. IJSBAR 2018

Conclusions, Recommendations & Limitations

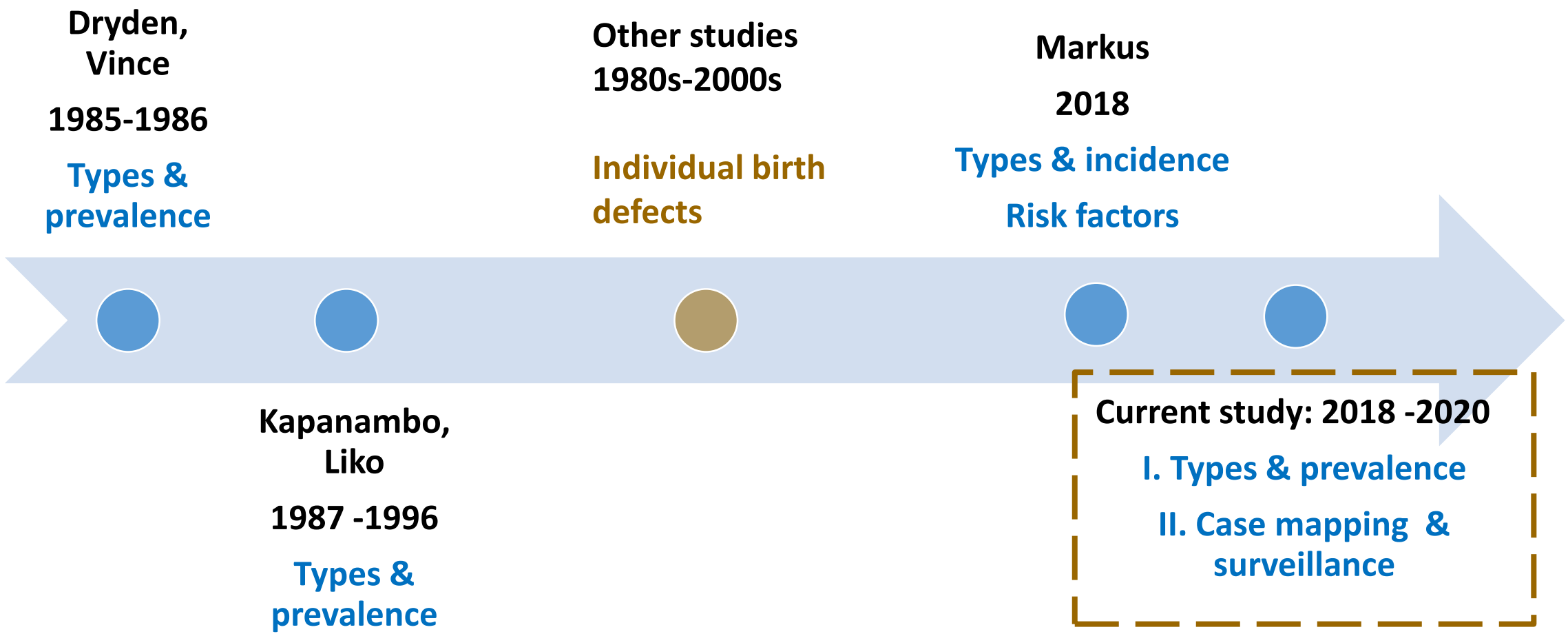
Primary Outcomes	RECOMMENDATIONS
Prevalence of birth defects in a consecutive series of 2000 live births in this study 1.4%	Tools for diagnosis of birth defects in Papua New Guinea to aid diagnosis and ongoing reporting or research
The birth defect with the highest frequency was congenital heart disease	Continue to improve care for children living with congenital heart disease
Secondary Outcomes	
Birth defect highest frequency was congenital heart disease	Continue to improve care for children living with congenital heart disease
Birth defect distribution showed clustering and distinct patterns **Incidental research observation of potential exposure of the sample population to an environmental hazard - cadmium**	Ongoing inter-sector research in ENBP into volcanic natural source of cadmium Might prove useful to informing public health policies to limit anthropogenic sources of cadmium in local agriculture and industry in the province
Surveillance of birth defects	National population based surveillance may not be feasible at this time Sentinel hospital based surveillance via Paediatric Hospital Reporting possible Portfolio of prevention approaches needed to reduce birth defects: Prevention of STIs, legislation controlling management of toxic chemicals, vaccination against rubella and fortification of staple foods with folic acid, iodine and other micronutrients

Limitations:

1. **Hospital rather than population based study**
2. **Stillborn infants not included**
3. **Further analysis of mapped data using GIS limited by time and level of expertise**

Birth defects research in Papua New Guinea

What is the contribution of the Rabaul birth defects study?



“The establishment of appropriate surveillance systems for congenital anomalies is one of the basic components of a national programme for the prevention and care of congenital anomalies.”

- World Health Organisation 2020

Associated terms: Birth defects, congenital malformations or congenital abnormalities

Further acknowledgement

Faith

God for His goodness and blessings

Family and friends

For their love and support

Fellows

Colleague registrars in the Paediatric and Obstetrics & Gynaecology Departments of Rabaul Provincial Hospital

Families

All the families who participated in this study whose children we cared for, even if for a brief moment in time

Reference list and source of citations

Literature:

1. March of dimes Global Report on Birth Defects: The hidden toll of dying and disabled children. March of Dimes Birth Defects Foundation. White Plains New York, USA. 2006
2. Birth Defects Surveillance: A manual for programme managers 2nd Edition. World Health Organisation. 2020.
3. Birth Defects Surveillance: Quick reference handbook of selected congenital anomalies and infections. World Health Organisation. 2020.
4. Dryden R and Vince J. Birth defects in Papua New Guinea. University of Papua New Guinea Press. 1988.
5. Comprehensive New Born Screening. Handbook for screening visible birth defects at all delivery points. Ministry of Health and Welfare. Government of India. September 2016.
6. Mangnuson JA, Dixon BE. Public Health Informatics and Information Systems. 3rd Ed.
7. Preventing disease through healthy environments: Exposure to cadmium: A major public health concern. World Health Organisation. 2010.
8. Calle Toro J et al. Teratogenic effect of cadmium: From the developing embryo to the fetus. Children's Hospital of Philadelphia. Article in: Revisita Colombina de Salud Ocupacianol. June, 2015.

Software:

1. Adobe LiveCycle ES 8.2
2. Microsoft Excel 2016
3. Quantum GIS version 3.10

Images:

1. East New Britain Map. Online: www.researchgate.com
2. Geographic Information Systems. Online: www.admitnetwork.org

Video:

1. Global birth defects description and coding application

Photos: 1-6 and 8-12: Google images, 7 Personal collection

Boina tuna