Review Article

Maternal contributors to intergenerational nutrition, health, and well-being: revisiting the Tanjungsari Cohort Study for effective policy and action in Indonesia

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Perinatal and maternal mortalities in Java became of concern in the 1980s. Since some 90% of births took place at home, the Tanjungsari (TS) district of West Java was identfied as a locality where community-based risk management strategy might reduce this health burden. In 1987, traditional birth attendants (TBA) were trained to identify risk factors for unfavourable birth outcomes. From January 1st 1988 to December 1989, some 4,000 pregnant women in TS were followed and assigned either a trained or untrained TBA. In the first year, early neonatal, and maternal mortality rates (MMR) (32.9 per 1000 and 170 per 100,000 deliveries respectively) were reduced, but not sustained in the second year. Nationally, MMR was 446 in 2009 and 126 in 2015. Although possible to improve health worker performance, and community engagement, the most likely explanation for benefit attrition is that people and material resources 'downstream' of the TBA services were inadequate. Three decades later, Indonesian neonatal and maternal mortality rates of 14 per 1000 and 126 per 100,000 live births in 2015 (globally 16.2 in 2009 and 216 in 2015) according to UNICEF, still demanded improvement, despite more hospital-based births. The original 1988 cohort of women, their children and grandchildren, can now be interrogated for medium to long term health outcomes of nutritional, such as birth weight and growth, and other risk factors. The evolving TS cohort health and nutrition intermediates and endpoints are instructive. Maternal and early life factors predict adult energy metabolism and cognitive function.

Key Words: Sundanese, intergenerational, fetal origins of disease, IUGR (intrauterine growth retardation), stature, energy metabolism, cognition

MATERNAL AND CHILD CONTRIBUTORS TO HEALTH IN INDONESIA

The advancement of health, nutrition and well-being among Indonesians, while impressive over the period 1960-2017 for life expectancy and infant mortality (Figure 1),[1,2] remains variable across the nation and problematic overall. By way of example and relevance to the Tanjungsari Cohort (TSC) study, West Java, is reviewed here. West Java is populated principally by those of Sundanese ethnicity, and relatively advantaged according to its vital statistical indices. The factors responsible merit scrutiny (Table 1).[3,4] A key question is whether and how economic development can be accompanied by accessible, equitable and sustainable health advancement. UNDP (United Nations Development Program) criteria would indicate that multisystem integration is required, with dependence on community consensus about governance of the commons as proposed by Ostrom.[5,6]

To achieve 'Health for All for Indonesians', an appreciation of its ancient and recent history, its cultural diversity and societal structures is required. Indonesian independence was achieved after some 350 years of Dutch colonisation in 1945, but following a Japanese occupation (1942-1945) and in the face of the Indonesian revolution (1945-1949). In 1950, five years after independence, Indonesia's health systems were in ruins. A public health initiative was set in train after 10 years of independence

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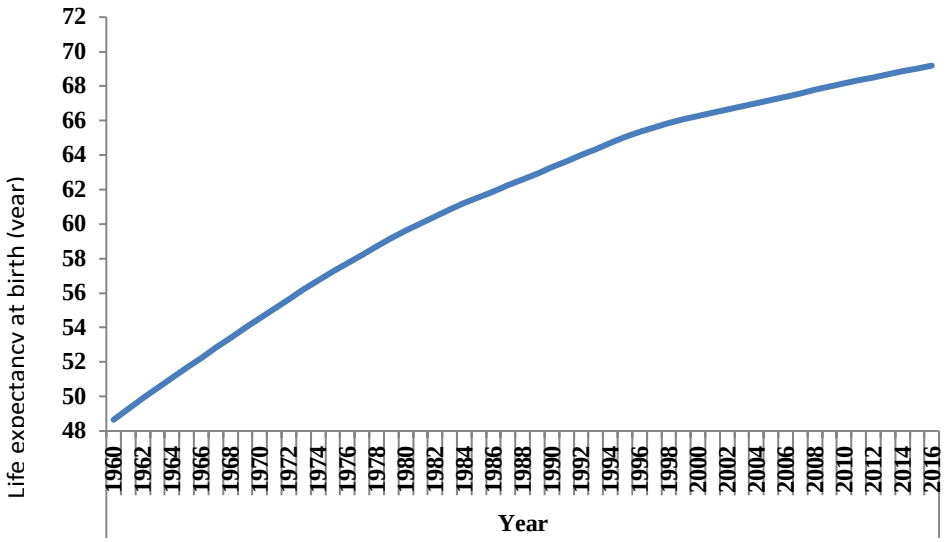
Manuscript received and initial review completed 14 January 2019. Revision accepted 19 January 2019.
doi: 10.6133/apjcn.201901_28(S1).0001

(a) Indonesian life expectancy at birth (years) by year[1]



(b) Indonesian Infant mortality rate (per 1,000 live births) by year

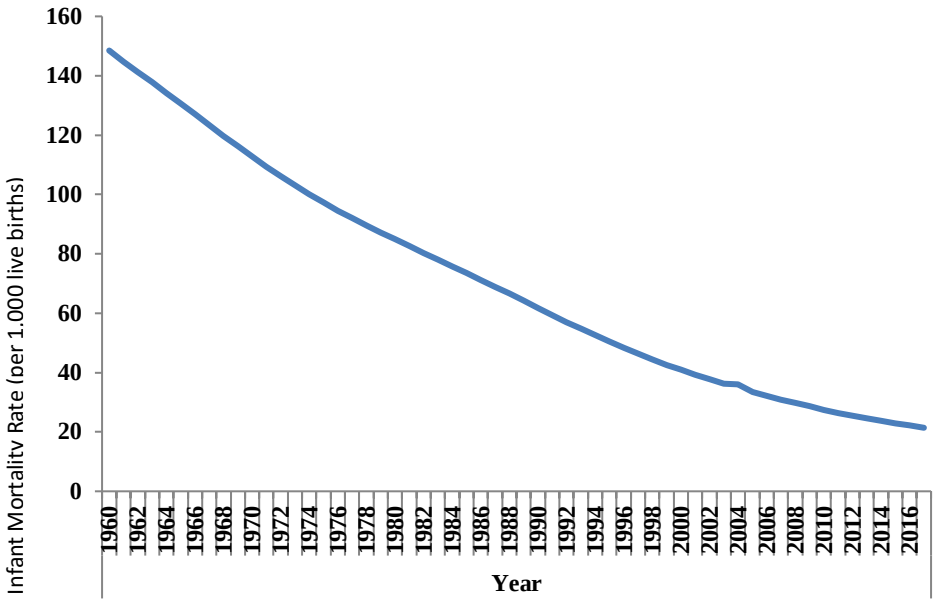


Figure 1. Indonesian Infant mortality rates (per 1,000 live births) and life expectancy at birth by year. (a) Indonesian life expectancy at birth (years) by year[1]; (b) Indonesian Infant mortality rate (per 1,000 live births) by year.[2] Source: World Bank Open Data.

with health regarded as key to nation building. There was a focus on infectious disease control and improvement of nutritional status along with major public health emergency programs.

The health programs paid attention to social factors as framed by Seno Sastroamidjojo, a leading social medicine exponent during the Soekarno era in Indonesia. For nation building, the socio-economic development and public health approach emphasized an equitable distribution and adequate intake of foods. This embraced all phases of an individual's life including the prenatal, postnatal, childhood, adolescent, adulthood and older age groups. Conceptually, it had prescience for the Developmental Origins of Health and Disease (DOHaD) enunciated by David Barker in 1986.[7] Sastroamidjojo was concerned about malnutrition. The high prevalence of oedema among Indonesian infants in the 1950s, according to Sastroamidjojo, was due to a shortage of food; a malnourished child mirrored the poor health of the economy. Sastroamidjojo referred to this as *pembangunan* or developmentalism.[8]

Notwithstanding these early post-Independence policy directions, major public health problems continue to face Indonesians, despite increasing annual national health budgets and enduring efforts to promote health and prevent disease. Communicable disease like tuberculosis is still rampant, and Indonesia is now listed as the country with the third-largest burden of tuberculosis after India and China;[9] and, at the same time, malnutrition, in any of its forms – underweight, wasting and shortness or stunting (pathological shortness), and in any of the recognised at-risk populations – pregnant women, the newborn and underfive children. These infectious disease and dys-nutritional problems are now compounded by an escalation of so-called non-communicable diseases (NCDs). Progress with nutritionally-related disease (NRD)

Table 1. Life expectancy (years) and infant mortality rates (per 1,000 live births) at West Java Province[3,4]

Vital statistics	Years															
	1971	1980	1990	1994	1997	2000	2002	2007	2010	2011	2012	2013	2014	2015	2016	2017
Life expectancy, male (years)									69.4	69.68	69.95	70.22	70.35	70.54	70.57	70.58
life expectancy, female (years)									73.27	73.53	73.79	74.05	74.18	74.36	74.39	74.42
Infant mortality rates (per 1,000 live births)	167	134	90	89	61	57	44	39	26	30	27	27	26	26		
Human Development Index									66.15	66.67	67.32	68.25	68.8	69.5	70.05	70.69

Source: Indonesian Central Bureau of Statistics Data.

Table 2. Gross Domestic Product (GDP), Life expectancy at birth, Infant Mortality Rate (IMR), low birth weight (LBW), and nutritional status of underfive children in 1995, 2007, 2013, and 2018 in Indonesia.[10-14]

	GDP (in constant 2010 USD)			
	1995	2007	2013	2018
	2,219.81	2,750.62	3,560.11	4,130.66
Life expectancy at birth (years)	65.03	67.58	68.68	69.19 (2016)
IMR per 1000 live births	50.4	30.9	24.5	21.4 (2017)
LBW (%)	10.3 (1997)	11.5	10.2	6.2
Underweight (%)	30.3	18.4	19.6	17.7
Wasting (%)	14.9 (1995)	13.6	12.1	10.2
Stunting (%)	48.1 (1995)	36.8	37.2	30.8

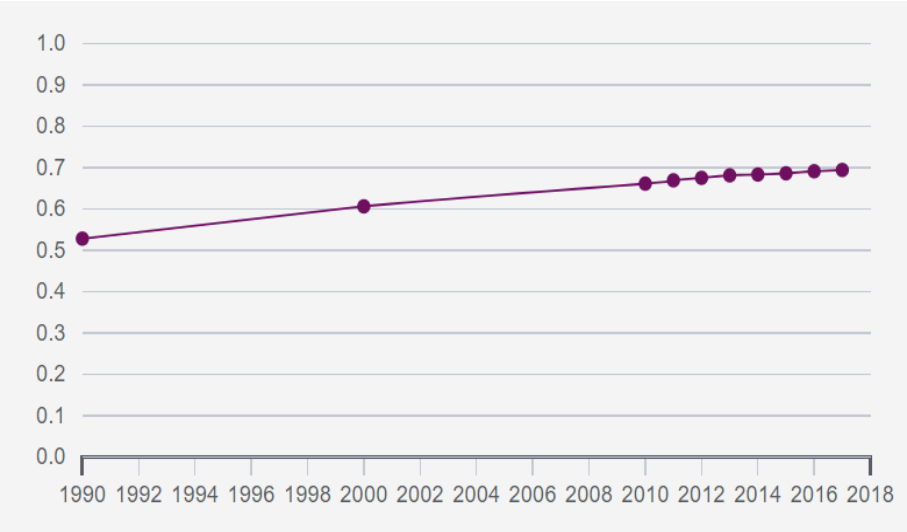


Figure 2. Indonesian Human Development Index (HDI) ranked 116 out of 189 countries.[15] Source: Human Development Reports, UNDP.

has been claimed in the 2018 Baseline Health Research report, with a further slight decline in infant mortality rate, prevalence of low birth weight and underfive malnutrition,[10-11] but not meeting the aspirations of earlier commentators.[12] They may have overstated the role of simplistic situational analysis like that with GDP rather than the broader scope of measures like the UNDI (United Nations Development Index) (Table 2 and Figure 2).[10-15]

Health, wellbeing and their nutritional dependence are usefully targeted as a package - across various life stages,[16] with an ecological perspective,[17,18] and community participation at the village level or its urban counterpart.[5] On this basis, the Tanjungsari cohort study[19] was devised, in its first iteration in 1988, with a focus on the improvement of maternal and child health, especially in pregnancy, childbirth and infancy. The Tanjungsari cohort study now merits revisitation for at least 3 reasons: observations of 3 generations since 1988 have been made, re-analysis for potential links between ecological factors and nutritionally-related health (NRD) outcomes has been possible, and valuable insights into public health and nutritional policy across the lifespan may be provided, not only for West Javanese, but for Indonesians in general. .

TANJUNGSARI COHORT STUDY: HISTORICAL PERSPECTIVE

The Tanjungsari Cohort Study described by Alisjahbana et al[19] was preceded by a prospective study designed by Dr Hellen Wallace (a WHO Consultant) and supported by the WHO Regional Office for Southeast Asia (WHO-SEARO). This prospective study commenced in 1978 in view of the high infant mortality rates (IMR) and low birth weights (LBW) in Indonesia at the time. The objectives twere to study the natural history of pregnancy and its outcome in one or more rural and in one or more urban areas of the province of West Java; and to study in-depth the problems of perinatal mortality and morbidity and of LBW in one or more teaching hospital precincts, connected with a medical school.[20] The Medical School of Universitas Padjadjaran was the institution engaged.

The study observational parameters were expanded by Dr Anna Alisjahbana, a neonatologist and Head of the Neonatal Ward and Intensive Care Unit at Hasan Sadikin General Hospital that linked to the Department of Child Health - Medical School of Universitas Padjadjaran in Bandung, West Java. Information on perinatal mortality and morbidity, stillbirth and late fetal death in communities were gathered. Home deliveries by semiliterate Traditional Birth Attendants (TBAs) were dominant (about 90%), especially in rural areas and, therefore, assessed for quality of after-care of livebirths.

Support from the National Research and Development Agency, Ministry of Health of the Republic of Indonesia, allowed rural coverage and expansion to the Ujung Berung subdistrict in September 1978, a training precinct of the Medical School at Universitas Padjadjaran. Ujung Berung subdistrict was purposively selected to ensure the involvement of interdisciplinary trained personnel/experts and research capacity within the Medical School at Universitas Padjadjaran and Indonesia at large.

Table 3. Maternal mortality rate, its pregnancy outcomes, and direct causes in rural study areas of Ujung Berung subdistrict.[20]

Indicator	Statistics (per 1,000 deliveries)	Direct causes (practically preventable)
Maternal Mortality Rate (MMR)	1.7	Toxaemia or eclampsia, haemorrhage, infection
Stillbirth Rate (SR)	13.7	anoxia/asphyxia, congenital abnormalities, other
Perinatal Mortality Rate (PMR)	45.0	Prematurity/LBW, anoxia/asphyxia, infections, congenital abnormalities, other respiratory problems
Early Neonatal Mortality Rate (ENMR)	32.9	Prematurity/LBW, anoxia/asphyxia, infections, congenital abnormalities, other respiratory problems, others

A total of 2,342 and 2,888 pregnant women were observed in the three remote villages of Ujung Berung subdistrict and at Hasan Sadikin Hospital, respectively. The natural history of pregnancy, birth outcomes, and major risks factors in rural and urban settings were recorded. Inter alia, socio-economic factors were considered as putative determinants of pregnancy outcomes. Thus the first comprehensive possible indicators of pregnancy outcomes, especially for rural areas in Indonesia, were obtained (Table 3)[20] and early evaluation of the strategy for training semiliterate TBAs was possible.

An earlier study (December 1984 – February 1985) at Sumedang Selatan subdistrict (located 60 km outside Bandung city) served as a feasibility test for the Risk Approach Strategy (RAS) that later would be applied in the TSC study. This used simplified aids like coloured spiral weighing scales, pictorial mother-child cards, and a Problem-Action-Guidelines (PAG) booklet.[21-23] The training strategy constructed for the Ujung Berung study was tested for its effectiveness in helping the illiterate TBAs perform their duties.

A conceptual diagram is provided to illustrate how the TSC study was envisaged to address principle questions about the progressive optimisation of maternal health, pregnancy, birth, infancy, childhood, adolescence and ultimately adulthood and subsequent intergenerational well-being in West Java, Indonesia (Figure 3).

The findings from these early studies which established the TCS framework have been published[20,24-27] and represent the Tanjungsari subdistrict activities of 1988-1990 to assess whether the RAS of MCH care had utility for often aging and semiliterate TBAs (Figure 4a, 4b) through adaptive training (Figure 5a, 5b). As indicated above, simple technology (eg. coloured spiral weighing scale - Figure 6) and tools (e.g. PAC – Figure 7a-7c., MCC – Figure 7d, referral card) were its premise.

The Tanjungsari sub-district is located about 15-20 km from Bandung and consisted of 27 villages when the study began. It had a total population of about 87,000 (based on the 1990 census) and a mountainous terrain. Only one health centre operated in the area, with one GP and one midwife. At the village level, the informal health services were delivered through an integrated health service post, a so-called *Posyandu*, which was run by volunteer women health workers, known as cadres.

During the two-year observation period, almost 90% of the women delivered at home and were attended by TBAs. There were 4,108 singletons recorded and 38 (pairs) twin births. Study extension until March 1990, allowed further recruitment and increased cohort size to 4,698 singletons. Figure 8 shows the sample sizes for the different cohorts reported by Alisjahbana et al,[28] Sofiatin et al,[29] Sasongko et al,[30] and Nugraha et al.[31]

The TSC study has been conducted in accordance with the WMA Declaration of Helsinki – 'Ethical Principles for Medical Research Involving Human Subjects' from 1983 onwards.

TANJUNGSARI: THEN AND NOW

Compared to the situation in 1988 when the study began, there have been major administrative, demographic, socioeconomic and geographic changes in the Tanjungsari subdistrict. With increased population density, the administrative area of Tanjungsari sub-district has been further divided from 27 into 32 villages in 2001 (Figure 9a, 9b). The wide and extensive terraced rice fields documented in 1988 are now much narrower and often replaced with rows of houses (Figure 10a, 10b). The previous rural setting has been transformed into a peri-urban landscape with better road access (Figure 11a–11d). Food-oriented agriculture has partly given way to more lucrative tobacco crops, with their own socioeconomic and health implications.

After some three decades, the TSC now comprises intergenerational groups of cohorts (Figure 12) and cadres (Figure 13) allowing various studies in a changing Tanjungsari. Among previously recruited staff and study personnel, field supervisors and TBA are no longer actively involved with the TSC study. On account of government regulation and a changing health system, TBA are slowly disappearing in the community and being replaced by village midwifes. In Tanjungsari, only one TBA remains, with no successor (Figure 14).

PREDICTORS OF AND BY INTRA-UTERINE GROWTH RETARDATION: THE SURROGACY FOR HEALTH AND NUTRITION STATUS OF LOW BIRTH WEIGHT AND SHORTNESS IN TANJUNGSARI

Low birth weight (LBW), which is found among preterm infants, is increasingly recognised as a reflection and predictor of the developmental origins of health and disease.[32,33] Although most preterm infants are LBW, term infants may also be LBW as a result of maternal malnutrition and other intrapartum health challenges, which affect intrauterine fetal growth.[34,35]

Likewise, ***shortness*** may be an indicator of compromised growth velocity, with reference to childhood malnutrition or recurrent infection and referred to as ***stunting***.[36,37] However, shortness may be physiological or, if pathological, attributable to intragenerational nutritional

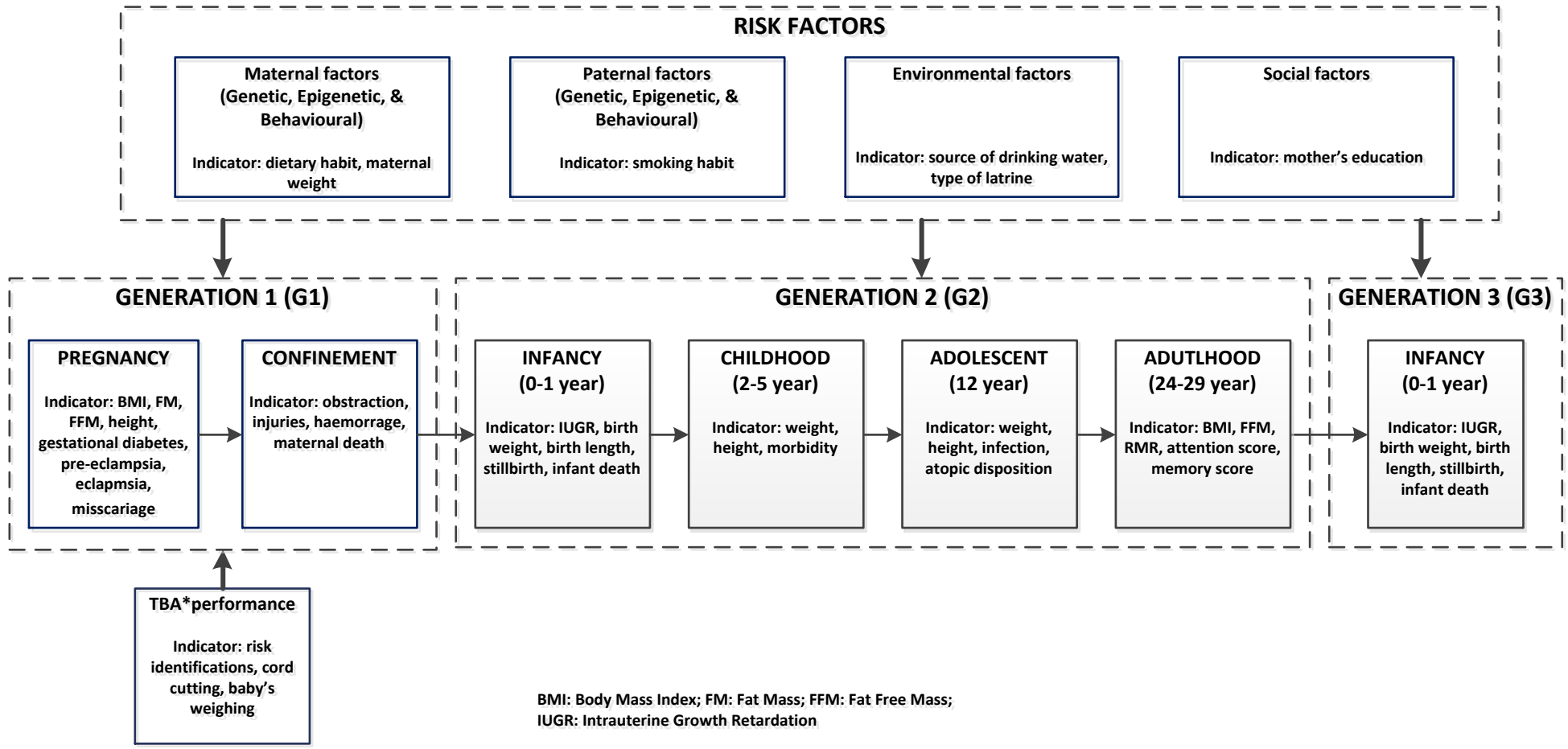


Figure 3. Conceptual Framework for the intra-and inter-generational Tanjungsari Cohort study of maternal and child health with example indicators.

a. A TBA in Tanjungsari subdistrict[19]

b. An untrained TBA in Tanjungsari assisting a delivery19

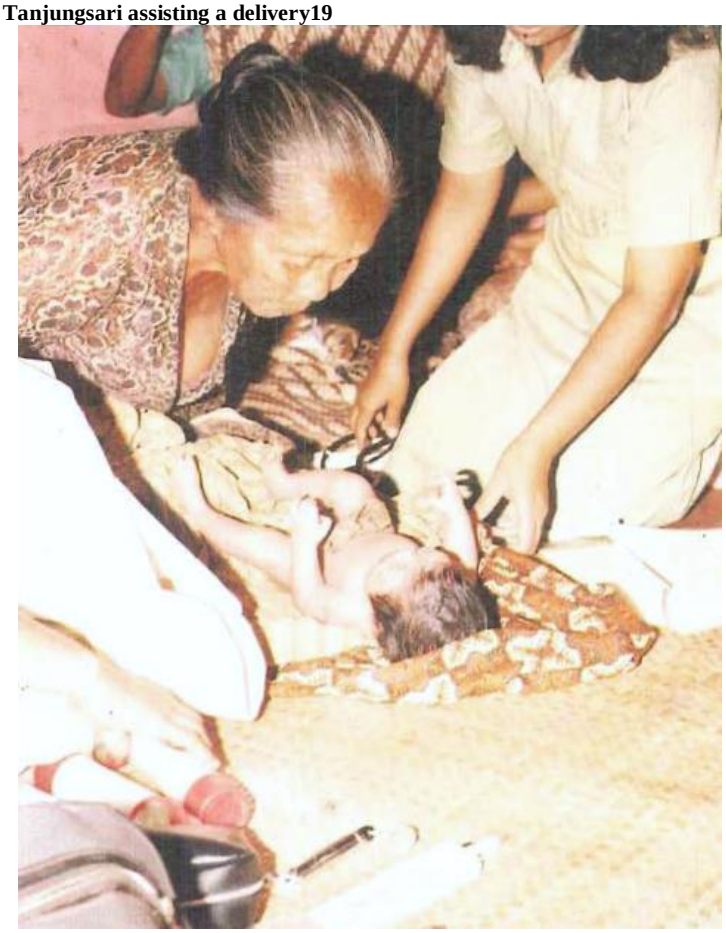


Figure 4. Traditional Birth Attendants (TBA) in Tanjungsari subdistrict.

and non-nutritional socio-environmental factors commencing at conception, act during intrauterine life, and continue through growth and development into adulthood.[38-40] In turn, there may be an association between LBW and shortness, which would most likely be of pathological significance although potentially amenable to recovery or 'catch-up'.[41]

The TSC Study has provided an opportunity to review these considerations in a relatively socio-economically disadvantaged population, and with the passage of time and generation, in West Java, Indonesia. Associations operative when the study began around 1988 may not be as evident some 3 decades later, but the investigators have examined whether such associations are predictive into later life and relevant for progeny, notwithstanding societal change. Table 4 summarises the associations between IUGR and indices of growth and development at 5 years, 12 years and in adulthood where the detail is available elsewhere.[28-31] An intergenerational study has also been published to do with the maternal and offspring associa-

a. Training of TBAs[19]

b. Risk assessment in a meeting with TBAs and pregnant mothers using the Mother Child Card[19]

Figure 5. Activities of Traditional Birth Attendant (TBA) in Tanjungsari subdistrict.

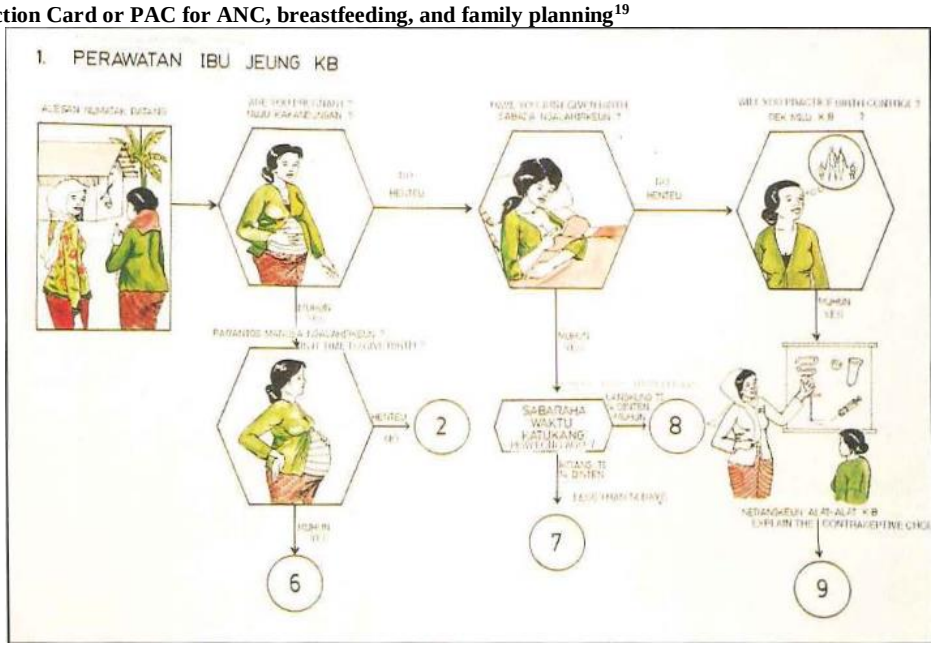
tions of LBW.[42] A composite prediction by IUGR, early life growth and development, maternal education, and environmental sanitation of adult energy metabolism and cognitive function is evident.

SUMMARY

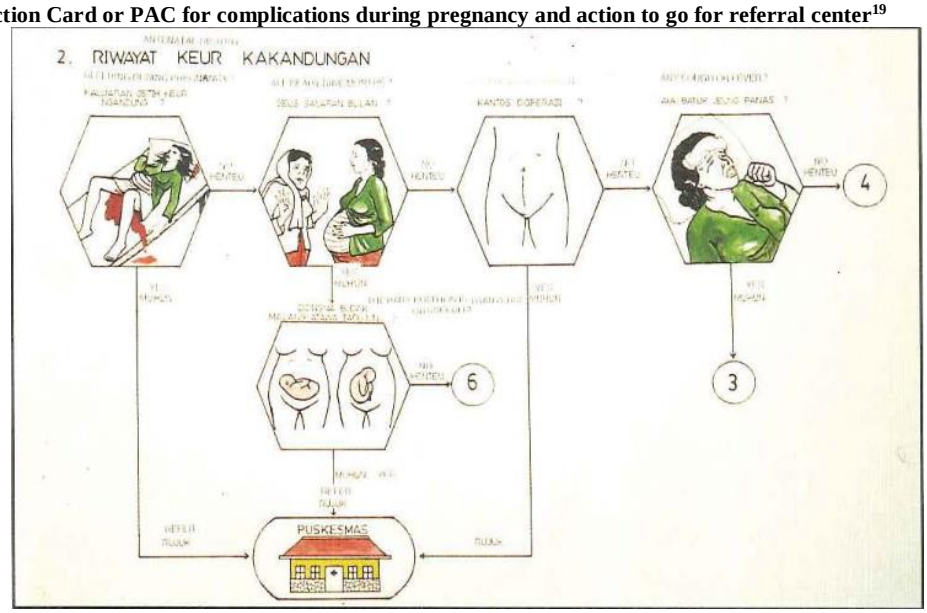
The TSC study began as a community-oriented effort to train and better equip traditional birth attendants (TBA) to decrease the burden of neonatal mortality, maternal mortality and low birth weight in the late 1980s in Tanjungsari, West Java.It was a time when these health outcomes in Tanjungsari were better by comparison with t he national and international experience. The early success of the training intervention was not sustained over the reference non-intervention community,for complex reasons, which include 'leakage' from the trained to the untrained personnel. Birthing centres known as 'Polindes (*Pondok Bersalin Desa*) facilities' for pregnant women have now been incorporated into the wider health system. The Tanjungsari study participants's children and grand-

Figure 6. Coloured infant weighing scale, developed for the Tanjungsari study.[19]

a. Problem Action Card or PAC for ANC, breastfeeding, and family planning[19]



b. Problem Action Card or PAC for complications during pregnancy and action to go for referral center[19]



c. Problem Action Card or PAC for complications during delivery and action at the referral center[19]

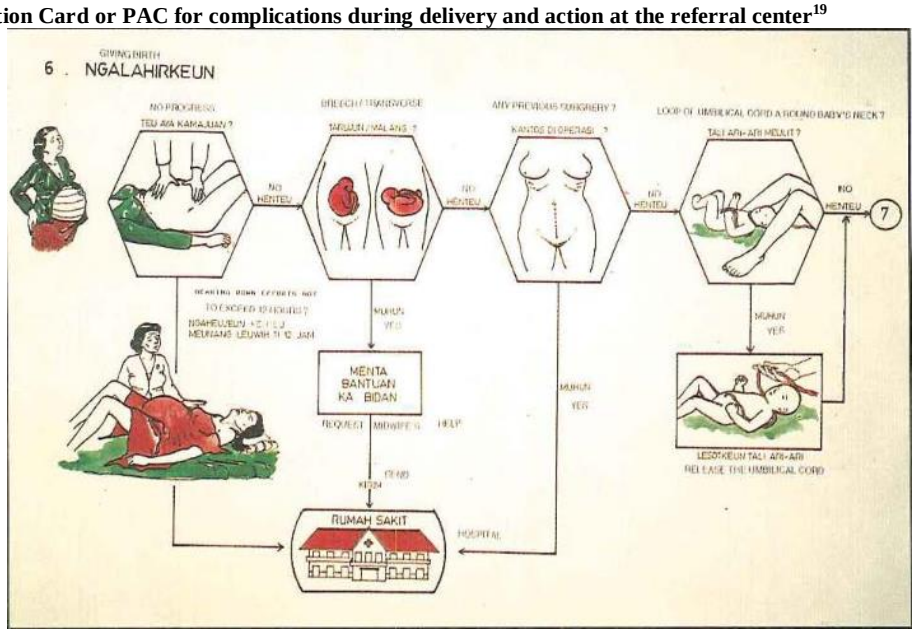


Figure 7. Problem action and mother-child cards.

d. Mother-Child Card or MCC[19]

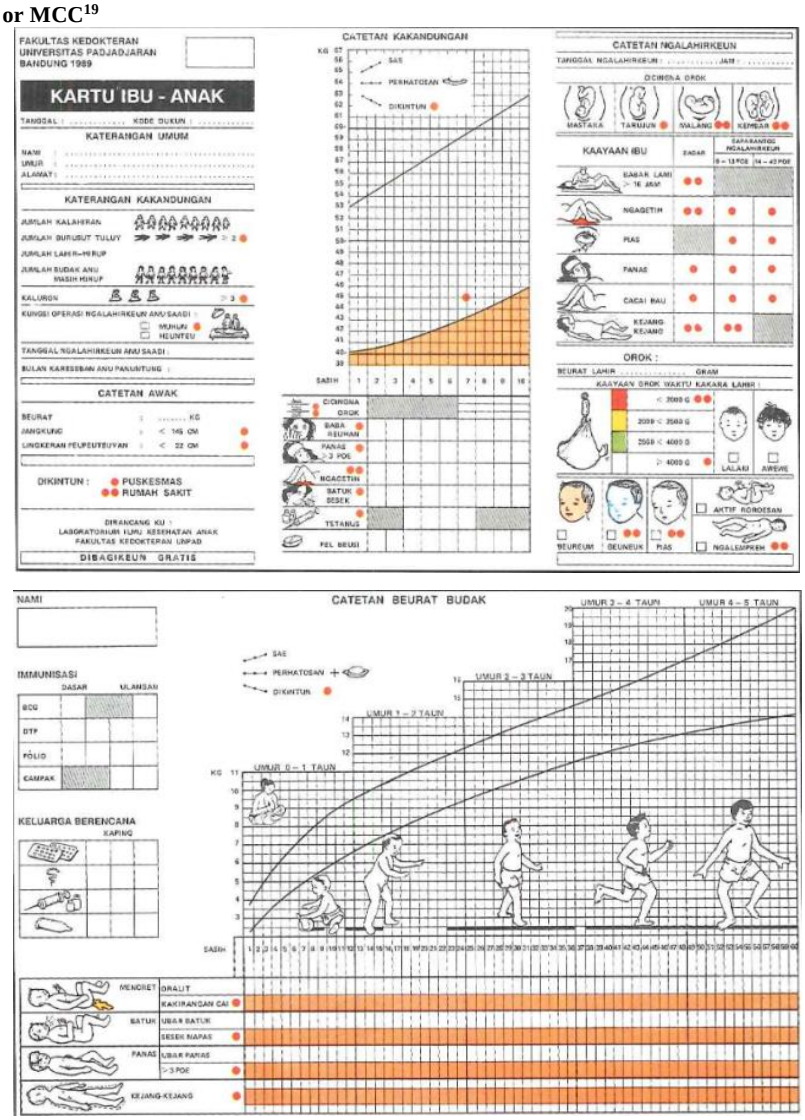


Figure 7. Problem action card and mother child card (cont.).

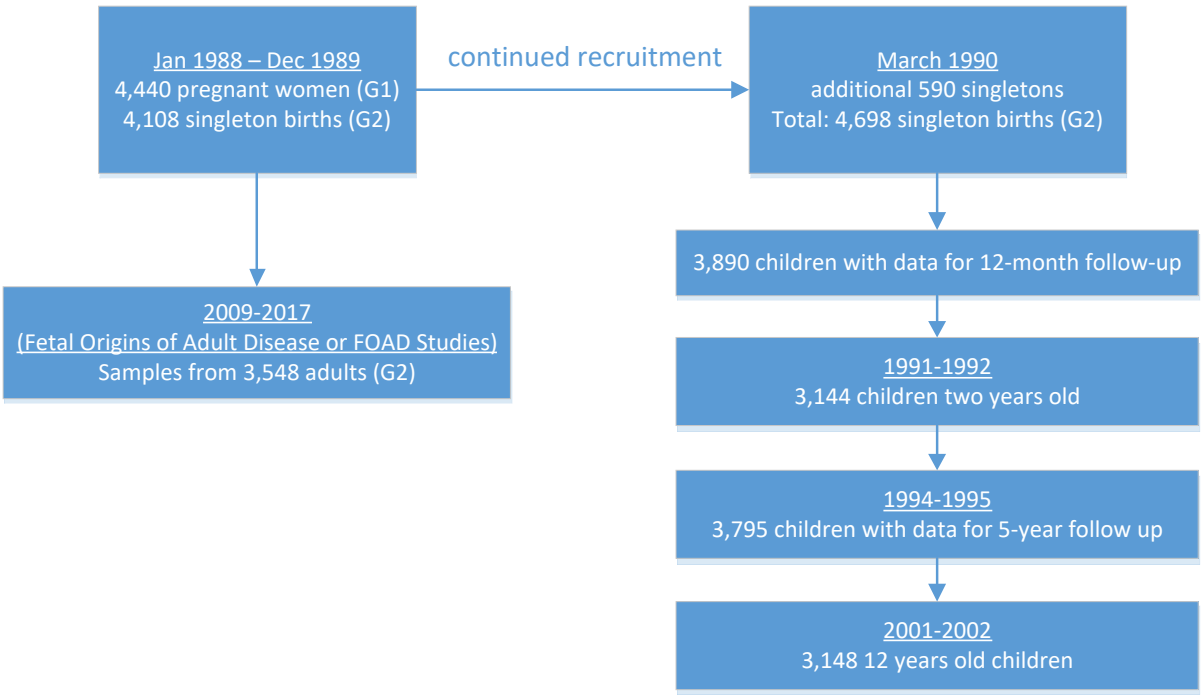
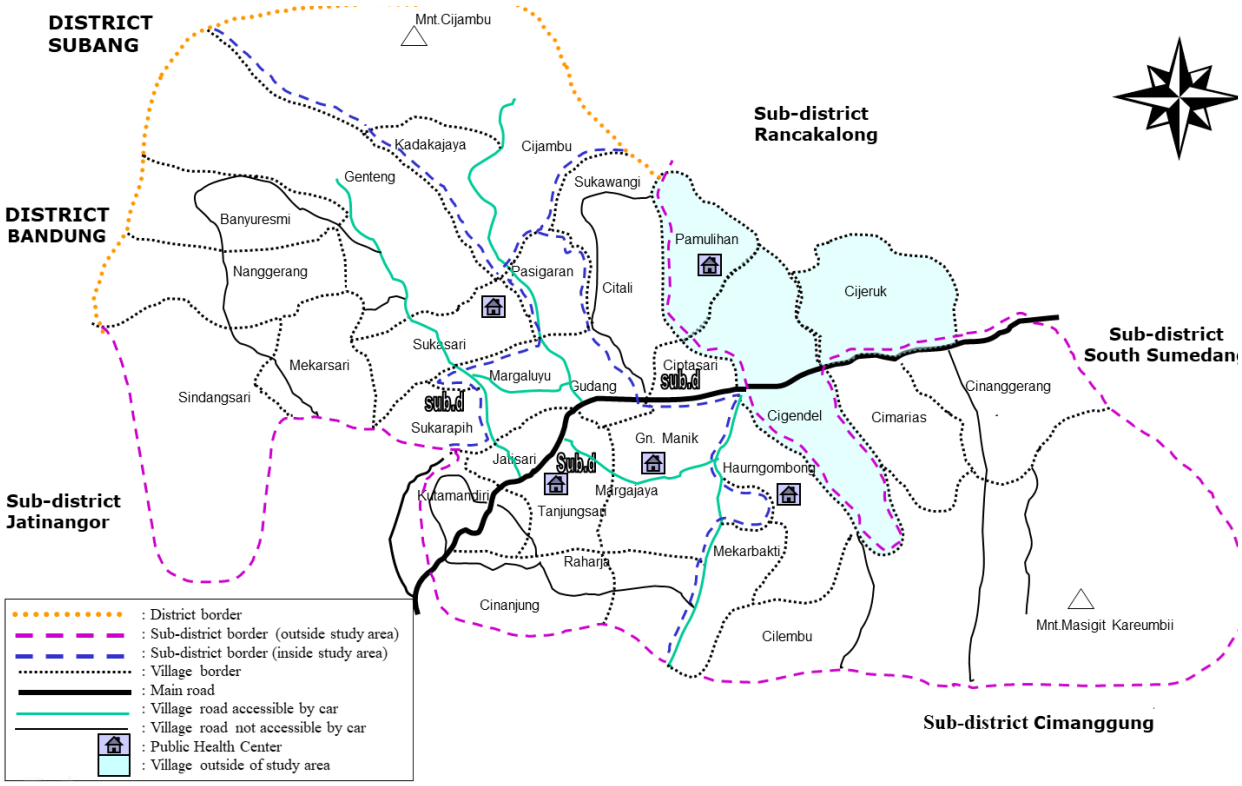


Figure 8. Sample size of Tanjungsari cohorts (G1 and G2) from 1988 to 2018.

a. Before area expansion in 2001.



b. After area expansion in 2001.

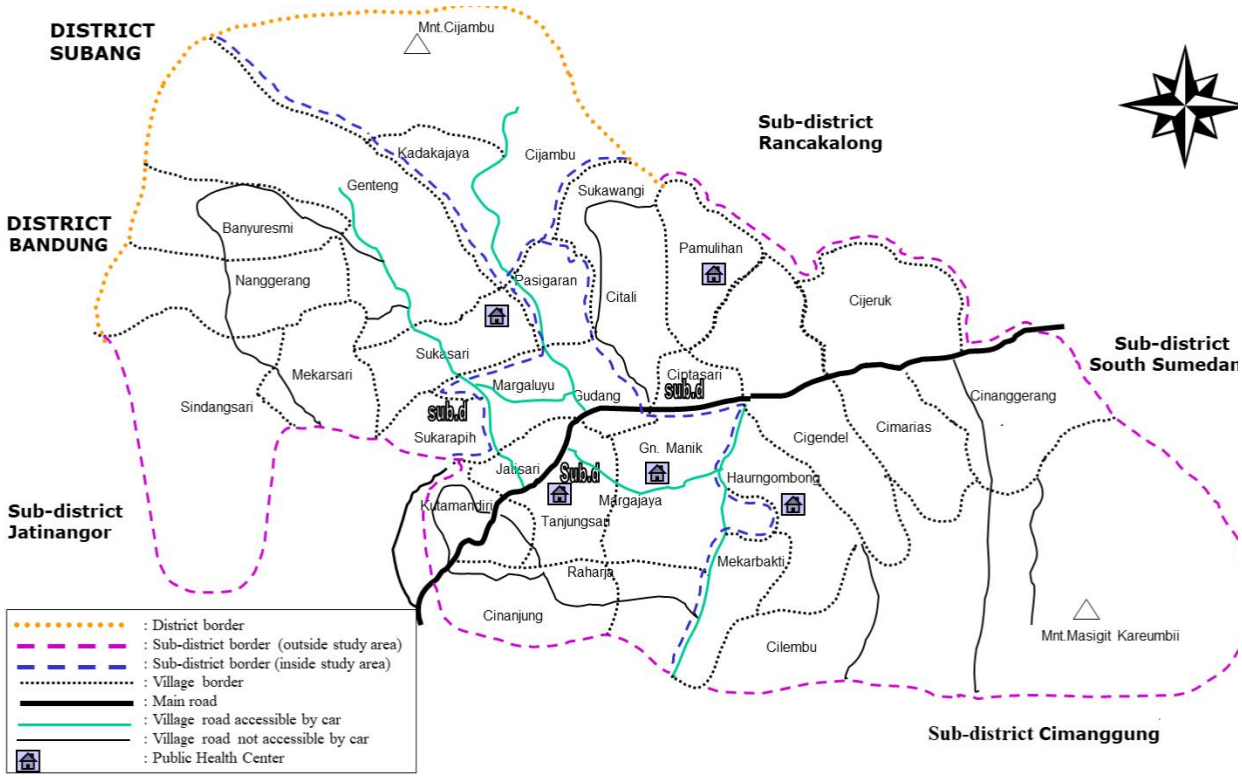


Figure 9. Map of Tanjungsari subdistrict.

chidren, some 3 decades on have become a cohort where, among other indicators, the nutritional observations of birth weight and growth provide opportunities to understand better how nutritionally-related and intergenerational health might be advanced .

A number of examples of these initiatives are provided in the accompanying chart (Figure 3) and in referenced papers .[28-31]

AUTHOR DISCLOSURES

Dr Widjaja Lukito is Director and Ms Lindawati Wibowo a Consultant of the Indonesian Danone Institute Foundation.

a. Terraced rice fields in 1988[19]

b. Rows of houses in 2018 and the piped water system

Figure 10. Eco-agricultural conditions.

Professor ML Wahlqvist has no conflict of interest.

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a. Road conditions in 1988[19]

b. An way to mobilize ill persons in 1988[19]

c. Road conditions in 2018

d. Road conditions in 2018

Figure 11. Transportation system at TS: then and now.

Figure 12. Intergenerational cohort participants in Tanjungsari. From the left to the right: [1] husband of G2; [2] G2; [3] AA - the Principal Investigator of Tanjungsari study; [4] G1 and G3; [5] G2; [6] ML Wahlqvist-advisor

Figure 13. Intergenerational cadres in Tanjungsari.

Figure 14. An informal meeting in front of *Taman Posyandu*. From the left to the right: [1] HS - one of the field supervisors in the Tanjungsari study area, who joined the team in 2003; [2] CW from Sukarapih Village – in the first batch and the oldest health cadre in Tanjungsari study area; [3] AA – the Paediatrician and Principal Investigator of the Tanjungsari study; [4] M – was the youngest TBA trained in the TS study and the last survivor.

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Table 4. Adverse effects of low birth weight and their reversibility from infancy to adulthood with later catch-up growth in the Tanjungsari Cohort (TSC) Study.[28-31]

Study	Publication#1[22]	Publication#2[23]	Publication#3[24]	Publication#4[25]
Cohort	At birth to infancy (0-1 years old)	Childhood` (0-5 years old)	Adolescence (12 years old)	Adulthood (27-29 years old)
Purposes	Birth weight in combination with birth length used to generate a practical classification of IUGR and explore its predictability of growth, morbidity and survival in the TSC during infancy.	Birth weight, genetic polymorphism, maternal- and environmental factors used to predict the risk of growth faltering in the TSC during the first 5 years of life.	Birth weight and shortness at age 2, together with maternal- and environmental factors used to assess any catch-up linear growth in the TSC at the age 12.	Birth weight, catch-up growth in early life, and by adulthood as indicated by anthropometric status, used as predictors of RMR and cognitive function in the TSC in adulthood.
Design	Prospective cohort	Retrospective cohort	Retrospective cohort	Retrospective cohort
Methodology	Comparison of by growth, morbidity and survival between non-IUGR, IUGR, and 'probably preterm' children.	Comparison of the growth of LBW and NBW children as indicated by their WHZ, HAZ, and WAZ. An evaluation of the predictors of child shortness at the age 5.	A determination of the predictors of normal (HAZ ≥-2 SD) and short stature (HAZ <-2 SD) adolescents at the age 12.	Comparison of RMR and cognitive function in ALBW and ANBW. Catch-up growth in early life is indicated by weight and height gain at 2 years and by adulthood anthropometric status as assessed by BMI and FFM.
Findings	The combination of weight and length at birth is a better indicator than birth weight itself to recognise IUGR in field conditions without technological support. This approach sensitively identifies the survival and health risks and survival prospects during infancy.	Birth weight is a determinant of postnatal growth in the first years of life, along with maternal education and the sanitary environment represented by drinking water. Stunting is presumed to be socio-environmentally and nutritionally-related shortness. It is partly an adaptive phenomenon, and may not in itself be invariably pathogenic for shortness-associated health outcomes.	Birth weight (dependent on intrauterine exposures), maternal education, sanitary environment (source of drinking water, type of latrine), and height-for-age at age 2 years are predictors of adolescent stunting. Infectious disease and atopic dispositions are more likely to be found in stunted adolescents.	Birth weight and body weight at 2 years of age are positively associated with RMR, but inversely associated with adult attention scores. BMI and FFM in adult life are positively associated with RMR and memory score. Weight catch-up, at least by 2 years, may be a modulating factor for low birth weight and allow better cognitive function in adulthood

IUGR: Intrauterine Growth Retardation; WHZ: weight for Height Z-score; HAZ: Height for Age Z-score; WAZ: Weight for Age Z-score; RMR: Resting Metabolic Rate; BMI: Body Mass Index; FFM: Fat Free Mass; ALBW: adult with history of Low Birth Weight; ANBW: Adult with history of Normal Birth Weight.

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