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- Prevalence of teenage pregnancy in 2015–2016 and its obstetric outcomes compared to non-teenage pregnancy at Hospital Tuanku Ja'afar Seremban (HTJS), Negeri Sembilan, Malaysia: A retrospective case-control study based on the national obstetric registry
- Validity and reliability of the Patient Assessment on Chronic Illness Care (PACIC) questionnaire among patients with type 2 diabetes mellitus in Malaysia: English version
- Pay-for-performance challenges in family physician program



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The Malaysian Family Physician Journal

Academy of Family Physicians of Malaysia,

Unit 1-5, Level 1 Enterprise 3B Technology Park Malaysia (TPM)

Jalan Innovasi 1 Lebuhraya Puchong-Sungai Besi

Bukit Jalil, 57000 Kuala Lumpur.

Tel: +603 8993 9176

Email: journal.mfp@gmail.com

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- Standard journal article: Corporate Author International Committee of Medical Journal Editors. Uniform requirements for manuscripts submitted to biomedical journals. N Engl J Med. 1997 Jan 23; 336(4):309-16.
- Books and other monographs: Personal Author(s) Stewart M, Brown JB, Weston WW, et al. Patient-Centered Medicine: Transforming the Clinical Method. Thousand Oaks, California: Sage Publications; 1995.
- Books and other monographs: Corporate Author WONCA International Classification Committee. International Classification of Primary Care, ICPC-2. 2nd ed. Oxford: Oxford University Press; 1998.

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We, the frontliners

Su May Liew

Chief Editor

In the past few months, our lives have been altered drastically by the COVID-19 pandemic. After more than two months under the Movement Control Order, Malaysians are warily stepping out and trying to get back to their regular routines. This is challenging because the situation still seems so uncertain and we are almost afraid to make plans that may have to be unceremoniously abandoned.

In my practice as an academic family physician in a teaching hospital, I have been so proud to see our trainees rising to the challenges. I watched them struggle with fears and anxieties of being infected and worse, infecting their families. Yet, they plodded on in their hot personal protective equipment and learned to take swabs and talk to patients through phone and mask. I have also observed colleagues at my department and in clinics all over the country deal with new operational procedures and coming up to fore. We have always known ourselves as frontliners but never has the label been used so widely or regarded as highly as it is now.

Doctors are being called heroes, wira in Malay.¹ We are seen as soldiers who fight for their country. But no one signed up to medicine thinking that we might have to risk our lives and those that we love. Thankfully we have not seen the lack of PPE here as reported in countries elsewhere. However, it was with great sorrow that I read about the 45 general practitioners who were infected with COVID.² To all my colleagues, trainees and doctors in the country, I ask that you protect yourself with evidence based knowledge, skills and practice during this time. We at the Malaysian Family Physician salute you for your efforts.

References

1. Sakini MS. Fikirlah: Terima kasih wira negara! Bernama. 2020 March 31. Accessed on 27 May 2020 at https://bernama.com/bm/am/news_covid-19.php?id=1826892.
2. Loheswar R. After 45 GPs down with Covid-19, Health D-G urges health workers to take every precaution. The Malay Mail. 2020 April 22. Accessed on 27 May 2020 at <https://www.malaymail.com/news/malaysia/2020/04/22/after-45-gps-down-with-covid-19-health-d-g-urges-health-workers-to-take-eve/1859265>.

Prevalence of teenage pregnancy in 2015–2016 and its obstetric outcomes compared to non-teenage pregnancy at Hospital Tuanku Ja'afar Seremban (HTJS), Negeri Sembilan, Malaysia: A retrospective case-control study based on the national obstetric registry

Nagandla K, Kumar K

Nagandla K, Kumar K. Prevalence of teenage pregnancy in 2015–2016 and its obstetric outcomes compared to non-teenage pregnancy at Hospital Tuanku Ja'afar Seremban (HTJS), Negeri Sembilan, Malaysia: A retrospective case-control study based on the national obstetric registry. *Malays Fam Physician*. 2020;15(2);2–9.

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Authors:

Kavitha Nagandla

(Corresponding author)
Department of Obstetrics & Gynecology, International Medical University, Seremban

Krishna Kumar

Department of Obstetrics & Gynecology, Hospital Tuanku Ja'afar Seremban, Seremban

Abstract

Objective: To determine the prevalence of teenage pregnancy and compare its obstetric and perinatal outcomes with those of non-teenage pregnancy.

Method: This retrospective hospital-based case-control study was conducted in the Department of Obstetrics and Gynaecology in Hospital Tuanku Ja'afar Seremban. The study made use of the Malaysian National Obstetric Registry (NOR) records of teenage pregnant women aged 11–19 at Hospital Tuanku Ja'afar Seremban over a 12-month period between May 2015 and May 2016 (n=164). Socio-demographic profiles, obstetric outcomes, and perinatal outcomes were detailed for each pregnant woman. The results were compared to a control group of 169 pregnant women aged 20–30 who also delivered in hospital Tuanku Ja'afar Seremban during the same period. The aim of this study was to assess the obstetric outcomes of teenage pregnancy and to compare them with those of the control group. A chi-squared test was used to identify the statistical significance of the relationship between teenage pregnancy rates and obstetric outcomes. Results with $p < 0.05$ was considered statistically significant.

Results: The prevalence of teenage pregnancy was 2.8%. The mean age of the teenage group was 17.9; that of the control group was 26.4. Teenage mothers had a significantly higher risk of anemia ($p < 0.05$), episiotomy ($p < 0.001$), preterm labor ($p < 0.001$), and delivering low-birthweight babies ($p < 0.001$). There were no significant differences between the two groups in mode of delivery, antenatal complications, birth outcomes, APGAR scores at 5th minute, or neonatal complications.

Conclusion: The prevalence of teenage pregnancy in this study is relatively low but is associated with an increased risk of some perinatal complications. The primary care physician's role is pivotal in educating adolescents on sexual health, providing continual care in hospitals, and empowering teenagers in their reproductive health decisions.

Introduction

Teenage pregnancy is defined by the World Health Organization as pregnancy that occurs among adolescent girls aged 10–19. Teenage pregnancy is a global social problem related to a wide range of adverse health and social outcomes impacting teenagers, families, and society. Evidence suggests that approximately 14 in every 1,000 underage Malaysian girls, or 18,000 overall, become pregnant each year.^{1,2}

The Malaysia Welfare Department reported that approximately 111 unmarried young girls

were pregnant and that the recorded rate was 6 births per 1000 women aged 15–19 years.³ In 2012, the Ministry of Health recorded 18,847 pregnant girls aged 10–19 at Malaysian public health facilities, which constituted 3.2% of the estimated 580,536 pregnant mothers that year (32 out of 1,000 pregnancies).⁴ Even though the teenage pregnancy rate in Malaysia is fairly low relative to that in many other countries, it may not reflect the actual figure, as illegal abortion and infant abandonment are on the rise among teenage mothers.

The issue of teenage pregnancy remains stark

throughout the rest of the world; the UK still has one of the highest teenage birth rates in Western Europe at 6.4 live births to every 1,000 women aged 15–17 in 2015.^{5–7} The country with the highest rate of adolescent pregnancy is the United States at 57 pregnancies per 1,000 adolescents in 2010, followed by New Zealand at 51 per 1000.⁷

Sixteen million girls between the ages of 15 and 19 are estimated to give birth each year, representing 11% of all global births. Ninety-five percent of these births occur in developing countries and about 89% occur outside of marriage.^{8,9} Gynecologic age (GA), defined as age in years at conception minus age at menarche, serves as an indicator of physiological maturity. Low gynecologic age is associated with an increased chance of obstetric and perinatal complications.¹⁰ Pregnancy-related complications include anemia, bleeding during pregnancy, premature delivery, placental insufficiency, pre-eclampsia, maternal alcohol use, maternal illicit drug use, gestational diabetes, and maternal infection. Delivery complications include emergency cesarean section, preterm birth, prematurity, protracted labor due to underdeveloped pelvis, vacuum extraction/forceps delivery, breech presentation, asphyxia at birth, and nuchal cord entanglement. The perinatal complications include neonatal jaundice and infections in the baby.¹¹ Babies born to mothers under 20 in low-and middle-income countries face a 50% higher risk of being stillborn or dying in the first few weeks than those born to mothers aged 20–29.¹² Although most research links teenage pregnancy with preterm delivery, fetal growth restriction, low birth weight, and fetal or perinatal death, some studies suggest that these outcomes can be prevented with complete coverage and a high quality of maternal care.¹¹ A study on maternal complications in teenagers showed that eclampsia, UTI, stillbirth, and neonatal mortality were more prevalent in cases of inadequate maternal care. In social terms, teenagers receiving inadequate maternal care were often those who were single and did not quit smoking during the first trimester.¹³

Primary care physicians play a key role in engaging with these adolescent patients in confidential, open, and non-threatening discussions of reproductive health and educating them on responsible sexual behavior. Their services must include contraceptive counseling that reinforces appropriate contraceptive practices, such as emergency contraception to

prevent unwanted pregnancies. Through greater understanding of the antecedents of teenage pregnancy, interventions in developing countries may be able to reduce the prevalence of this issue. We aim to use our findings to assist with the development of preventive measures that can be utilized by outreach programs to reduce the burden of teenage pregnancy. We look to integrate our results with university projects on reproductive adolescent health care, which will serve as platforms for disseminating information on sexual well-being.

Objective

To determine the prevalence of teenage pregnancy and compare its obstetric and perinatal outcomes with those of non-teenage pregnancies.

Materials and Methods

Study design

We conducted a retrospective study in the Department of Obstetrics and Gynaecology at Hospital Tuanku Jaafar Seremban by, with permission, obtaining data from Malaysian National Obstetric Registry (NOR) forms. The study received ethical approval from the International Medical University Joint Committee of Ethics and National Medical Research Register (NMRR ID: NMRR-16-845-30277).

National Obstetric Registry

The NOR is a clinical “disease” database that compiles obstetric data to enable healthcare planning, implementation, and evaluation in a defined population. The objective of the NOR is to develop a complete picture of patient care through a comprehensive database that helps track patient management and outcomes.

Sampling method

Between May 2015 and May 2016, a total of 5,800 pregnancies and deliveries of all ages were recorded there. We reviewed all NOR forms recorded during the 12-month period and selected all 164 teenagers (11–19) for participation in our study. The sample size for the study group was calculated using the “sample size for a proportion or descriptive study” calculator from Open Source Statistics for Public Health. Based on this sample size calculation, the number of participants proved to be more than sufficient ($n=136$).

To determine obstetric outcomes, we isolated the forms of females aged 20–30 and used simple random sampling to select an additional 168 adults as a control group. Women with high-risk conditions in pregnancy, such as diabetes, heart disease, hypertension, and thyroid disorders, were excluded from the study. The following variables were drawn from the NOR form and categorized into demographic and antenatal backgrounds (maternal age, marital status, social welfare, BMI, parity, type of antenatal care, hemoglobin status); pregnancy outcomes and complications (mode of delivery, type of cesarean section, term or preterm labor, antenatal complications, perineal tears); and perinatal outcomes (birth outcome, birth weight, APGAR score at the 5th minute, neonatal complications).

To protect patient anonymity, we did not include personal information aside from age in our study. Social welfare support is simply whether or not the patient receives welfare support from relevant governmental or non-governmental agencies. BMI is categorized as underweight (<18.5), normal (18.5–24.9), overweight (25.0–29.9), obese I (30.0–34.9), obese II (35.0–39.9), and obese III (>40). Anemia was defined as hemoglobin concentration <11g/dl. For the purpose of this study, stillbirth was defined as any death prior to expulsion from the mother beyond 24-week gestation; infant death was defined as any postnatal fatality during the first year of life. We categorized birthweight as low (LBW) if the birthweight was $\leq$ 2500 gm or normal (NBW) for all other birthweights. Prematurity was defined as any delivery before the 37th week of pregnancy.

Analysis

We analyzed our data using IBM SPSS Statistics v22. We used a chi-squared test to identify the statistical significance of the relationship between teenage pregnancy rates and obstetric outcomes. The 95% confidence intervals were generated and $P < 0.05$ was considered statistically significant.

RESULTS

Sociodemographic profile

Teenage pregnancies were selected from the NOR at Hospital Tuanku Jaafar Seremban. Of the 5,800 deliveries between May 2015 and May 2016, 164 were by teenagers (11–19), meaning there was a teenage pregnancy rate of 2.8%. The mean age of the teenagers was 17.9.

Table 1 shows the demographic and antenatal backgrounds of the teenage and non-teenage pregnancies. There was a significant difference ($p < 0.001$) between the groups in terms of marital status; 39 (23.8%) teenage mothers were unmarried at the time while just 1 (0.6%) non-teenage mother was unmarried. The teenage group had a significantly higher incidence of low BMI ($p < 0.05$) as well as a higher risk of anemia ($p < 0.05$) than the non-teenage group. A large majority (126; 80.3%) of teenagers were nulliparous, though 26 (16.6%) were para 1 and five (3.1%) were para ≥ 2 .

Obstetric complications: Antenatal and intrapartum

Regarding antenatal complications, the incidence of preterm birth is higher in the teenage group (26; 15.9%) than in the non-teenage group (9; 5.4%), and that difference is statistically significant ($p < 0.001$). Regarding medical disorders during pregnancy, about 1.2% of the teenage group had gestational diabetes while eclampsia, severe pre-eclampsia, antepartum hemorrhage, and pre-labor rupture of membrane were present in just 0.6% of teenage pregnancies. Regarding the mode of delivery, 111 teenagers (72.1%) delivered through spontaneous vaginal delivery, 15 (9.7%) through induced labor, 10 (6.5%) through instrumental labor, and 18 (11.7%) through cesarean section. There was no significant difference in the mode of delivery between the 2 groups. It is worth noting that both teenage and non-teenage pregnancies (83.3% and 69.7%, respectively) were more likely to result in an emergency cesarean section than an elective one. This information is displayed in **Table 2**.

Perinatal complications

Fifty-one of the teenagers (31.9%) delivered low-birthweight babies (≤ 2500 g) while just 27 (16.2%) did the same; this difference is statistically significant ($p < 0.001$). No significant differences were detected between the two groups in Apgar score at the 5th minute or neonatal complications. However, there were two intrauterine death (1.2%) in the teenage group but just one (0.6%) in the non-teenage group. A majority of babies in both the teenage group and the non-teenage group (143, 87.2% and 133, 79.2%, respectively) were discharged to their mothers; 8 (4.9%) live births from the adolescent mothers were admitted to NICU. This information is displayed in **Table 3**.

Table 1: Demographic and antenatal backgrounds of the study and control groups

Variable	Teenage group (11–19 years) n=164	Non-teenage group (20–30 years) n=168	p-value
Maternal Age (years), mean (±SD)	17.98 (±1.24)	26.42 (±2.63)	
Marital Status, n (%)			
Married	107 (65.2)	165 (98.2)	0.001
Unmarried	39 (23.8)	1 (0.6)	
Divorced	1 (0.6)	0	
Unknown	17 (10.4)	2 (1.2)	
Social Welfare, n (%)			
Yes	5 (3.0)	1 (0.6)	0.43
No	159 (97.0)	167 (99.4)	
BMI, n (%)*			
Underweight	28 (19.2)	14 (8.6)	0.32
Normal	59 (40.4)	52 (31.9)	
Pre-Obese	41 (28.1)	55 (33.7)	
Obese I	16 (11.0)	32 (19.6)	
Obese II	1 (0.7)	7 (4.3)	
Obese III	1 (0.7)	3 (1.8)	
Parity, n (%)*			
0	126 (80.3)	86 (52.8)	0.001
1	26 (16.6)	42 (25.8)	
≥2	5 (3.1)	35 (21.4)	
Types of Antenatal Care, n (%)			
Government	148 (90.2)	161 (95.8)	0.27
Private	4 (2.4)	4 (2.4)	
Government and Private	2 (1.2)	2 (1.2)	
Other	3 (1.8)	0	
No Care	7 (4.3)	1 (0.6)	
Booking Hb status, n (%)			
Normal	96 (58.5)	130 (77.4)	0.05
Anemia	47 (28.6)	31 (18.5)	
Missing information	21 (12.8)	7 (4.2)	

Data analyzed with X² test, p-value <0.05 set as significant

* Missing data in BMI and parity because the variables are not filled out in the NOR form

Table 2: Comparison between the outcomes and complications of teenage and non-teenage pregnancies

Variable	Teenage group (11–19 years) n=164	Non-teenage group (20–30 years) n=168	p-value
Preterm labor, n (%)			
Term	138 (84.1)	159 (94.6)	0.001
Preterm	26 (15.9)	9 (5.4)	
Antenatal Complications, n (%)			
Eclampsia	1 (0.6)	0 (0.0)	0.237
Gestational Diabetes	2 (1.2)	8 (4.8)	
Severe Pre-eclampsia	1 (0.6)	1 (0.6)	
Pre-labor Rupture of Membrane	1 (0.6)	0 (0.0)	
Antepartum Hemorrhage	1 (0.6)	0 (0.0)	
Cord Prolapse	0 (0.0)	1 (0.6)	
Mode of Delivery, n (%)			
Spontaneous Vaginal Delivery	111 (72.1)	101 (63.1)	0.05
Induced Labor	15 (9.7)	23 (14.4)	
Instrumental Labor	10 (6.5)	9 (5.6)	
Cesarean section	18 (11.7)	33 (16.9)	
If Cesarean, n (%)			
Elective	3 (16.7)	9 (27.3)	0.312
Emergency	15 (83.3)	24 (72.7)	
Perineal Tears, n (%)			
Intact	39 (23.8)	86 (51.2)	0.001
Tears	31 (18.9)	35 (20.8)	
Episiotomy	94 (57.3)	47 (28.0)	

Data analyzed with X² test, p-value <0.05 set as significant

Table 3: Perinatal outcomes in teenage and non-teenage groups

Variable	Teenage group (11–19 years) n=164	Non-teenage group (20–30 years) n=168	p-value
Birth Outcome, n (%)			
Live Birth	162 (98.8)	167 (99.4)	0.491
Still Birth	2 (1.2)	1 (0.6)	
Low Birth Weight, n (%)			
≤2500g	51 (31.9)	27 (16.2)	0.001
>2500g	109 (68.1)	140 (83.8)	
APGAR Score at 5 min, n (%)			
Good (≥7)	157 (98.1)	163 (99.4)	0.302
Bad (<7)	3 (1.9)	1 (0.6)	
Neonatal Complications			
Asphyxia	2 (22.2)	2 (28.6)	0.481
Transient Tachypnea of Newborn	0 (0)	4 (57.1)	
Prematurity	4 (44.4)	0 (0)	
Other Complications	1 (11.1)	0 (0)	
Intrauterine Death	2 (22.2)	1 (14.3)	
Baby Discharged to, n (%)			
Mother	143 (87.2)	133 (79.2)	0.44
NICU	8 (4.9)	11 (6.5)	
Nursery	11 (6.7)	23 (13.7)	
Mortuary	2 (1.2)	1 (0.6)	

Data analyzed with X² test, p-value <0.05 set as significant

Discussion

The rate of teenage pregnancy in our study was 2.8% and the mean incidence of adolescent pregnancy was 28 births per 1000 women. This is fairly lower relative to the rest of the world—teenage births accounts for 11% of all births worldwide.^{14,15} However, this figure may not reflect the reality of the situation, as there is a rise in Malaysia of illegal abortion and infant abandonment among teenage mothers. The National Registration Department of Malaysia has reported a rising trend of children born out of wedlock, from 8.1% in 2006 to 10.4% in 2010.^{16–18} This is supported by findings from Ruhaizan et al.¹⁹ In contrast, our study found that nearly a quarter of teenagers 23.8% were unmarried compared to just 1 of the adults. These pregnancies could be unplanned with a high reliance on natural contraception despite engaging in premarital sex. Poor education on matters of sexual health may be a contributing factor. A Malaysian analysis found that sex education in local schools was vague and inappropriately taught.²⁰ However, as unmarried teenage pregnancies frequently go unreported, this surprising rate may signify a shift toward a lesser degree of stigma and greater acceptance among parents. Almost none of the mothers from either group in this study were found to have social welfare support. While society is

beginning to show greater acceptance toward teenage pregnancy, Malaysians still hesitate to accept help on this matter, as they may still view it as shameful.²¹

Hemoglobin level was considered because previous studies have shown that anemia is more prevalent among adolescent mothers;²² our findings are consistent with those studies. This association stems from both physiological and social factors. During puberty, large growth spurts and the onset of menstruation deplete the body's iron levels. Furthermore, pregnant adolescents are less likely to be involved in profitable employment due to their low educational status or professional inexperience. Hence, we speculate that they may have limited access to nutritious food or a poor understanding of nutrition. This pairing between insufficient nutritional intake and inadequate iron levels predisposes adolescent mothers to anemia, which increases the risk of preterm labor, low birthweight, and infant mortality, especially during the first two trimesters.^{21,22}

Interestingly, our study found that there was no significant difference in the mode of delivery between the teenage and non-teenage groups, which is congruent with some previous studies. However, this finding contradicts the common belief that, due to physical immaturity, the

underdevelopment of the bony pelvis causes an increased risk of cephalopelvic disproportion in adolescent mothers.²³ One possible explanation is that the large majority of our teenage group was made up of late adolescents who have already reached full bone maturity—female pelvic structures are known to reach adequate maturation within two years after menarche.²³

Consistent with previous research, this study found that babies born of teenage mothers were more likely to have low birthweights (LBW; $\leq 2500\text{g}$). While the exact mechanisms that contribute to low birth weight remain unclear, it is one of the most significant drivers of neonatal morbidity. One physiological factor may be the low female biological maturation of teenage women.^{10,23,24} The maturation rate of the blood supply in both the uterus and cervix is not uniform across all women during pubescence. Hence, it may predispose adolescent mothers to a higher risk of subclinical infections, which, in turn, precipitate preterm birth, the proximal cause of LBW in infants.^{23,24} Another explanation may be that restricted blood supply to the uterus and cervix can stimulate prostaglandin production, which ultimately leads to preterm delivery. Additionally, a low level of gonadal hormones in young mothers may predispose them to premature vaginal contraction.²³ Studies have shown that post-menarchal adolescents need at least three years to establish a mature ovulatory cycle. Immaturity of the ovulatory cycle may compromise the secure attachment of a fetus to the uterine wall, resulting in vaginal bleeding and premature contractions.²⁵ Another possible physiological explanation for adolescent mothers being more prone to LBW babies is the competition of nutrients between a growing mother and a developing fetus.²³

Evidence indicates that a lack of prenatal care is a risk factor for preterm delivery.²⁵ Pregnant teens often receive inadequate prenatal care due to immature, negative, or ambivalent feelings surrounding a pregnancy.²⁶ Moreover, teens are often fearful, as they worry about their parents' reactions towards their pregnancies; they may not voluntarily access health care services.^{27,28} This postulation, however, contrasts with our findings. We found that the majority of the teenagers (95.7%) have gone for antenatal checkups in the public, private, or both sectors. Thus, our study suggests that society has become more liberal and tolerant and that teenage mothers now experience less stigmatization and psychological stress; they are able to obtain better support from their

family and community. Additionally, obstetric services in public hospitals are free of charge to all residents. Receiving adequate antenatal care may explain the lower-than-expected rate of adverse maternal and neonatal outcomes. Previous studies have suggested that adolescent pregnancy does not pose a high risk if good prenatal care is provided.^{23,29,30} This is backed up by our findings, as there were no significant differences in the antenatal or perinatal complications between the teenage and non-teenage groups.

Our study also found that the rate of episiotomy was significantly higher in the teenage group. Recent studies suggest that it is more common to have severe injuries involving the anal muscle if the perineum tears spontaneously rather than if an episiotomy is deliberately cut.^{31,32} This is supported by a case-control study of 1,282 teenage-pregnancy deliveries in Turkey, which states that episiotomy is performed to prevent the risk of further perineal tears in primiparous pregnancies.³³ However, a hospital-based retrospective cohort study of 4,101 deliveries in Nepal shows that the risk of delivery by episiotomy was significantly lower ($P < 0.05$) among teenage mothers.³⁴ However, this was due to the gynecologists' reluctance to perform surgical procedures on teenagers.

Outreach programs serve as good platforms to tackle social problems and spread knowledge about teenage pregnancy on a community and individual level. It is commendable that governmental and non-governmental healthcare organizations in Malaysia have made great strides in improving teenage-pregnancy outcomes. However, it is still crucial that pregnant teenagers receive antenatal care as early as possible. Primary care physicians must play a pivotal role in educating the adolescent population; contraceptive practices must be promoted among married adolescents in order to avoid pregnancy until they reach maturity.

Limitations

This study did not include private hospitals, as private-sector data is far less readily accessible. Additionally, most of our data collected from NOR forms is missing certain information—the details on patients' follow-up appointments and previous visits were not available. Thus, we were unable to make certain correlations. For example, we could not examine the association between past medical history and obstetric outcomes. These limitations highlight the fact that the NOR forms must be strengthened in

order to avoid missing data. The national registry is an important database for evaluating the management of obstetric outcomes; its reliability is crucial for developing guidelines that improve care.

Conclusion

Teenage pregnancies are associated with several adverse outcomes, such as increased risks of anemia, episiotomy, preterm labor, and delivering low-birthweight babies. The prevalence of teenage pregnancy is lower in this study than it is in most others. However, this study highlights the fact that a high proportion of pregnant teenagers were unmarried and did not receive social welfare. Sexual education in local schools requires revision; educators must be properly trained in order to enable the dissemination of accurate information. Finally, the social stigma of teenage pregnancy and the shaming that these teenagers face must be addressed.

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Conflicts of interest

The authors report no real or perceived vested interests relating to this article that could be construed as a conflict of interest.

Ethical Approval

The study was approved by the Medical Research and Ethics Committee (MREC). NMRR ID: NMRR-16-845-30277

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References

1. Ermisch J. Does a 'teen-birth' have longer-term impacts on the mother? Suggestive evidence from the British household panel survey. Institute for Social and Economic Research; 2003.
2. Said S. Exploring the causes of teenage pregnancy in M'sia. Bernama. 3rd January 2019. Available from: [Accessed].
3. Noordin N, Zakaria Z, Sawal MZHM, Hussin ZH, Ngah K, Nordin J. The voice of youngsters on baby dumping issues in Malaysia. *Global Journal of Health Science* 2013;3(1): 66–71.
4. Ministry of Health Malaysia. Annual Report 2012. [internet] [cited 1st November 2015]. Available from: <http://www.moh.gov.my/images/gallery/publications/MOH%202012.pdf>.
5. Berrington A, Diamond I, Ingham R, et al. Consequences of teenage parenthood: pathways which minimise the long-term negative impacts of teenage childbearing. Southampton: University of Southampton; 2005. Final report.
6. UNICEF. Child poverty and perspective: an overview of child wellbeing in rich countries. New York: UNICEF; 2007.
7. Office for National Statistics. Conception statistics in England and Wales. 2007.
8. Sedgh G, Finer LB, Bankole A, et al. Adolescent pregnancy, birth, and abortion rates across countries: levels and recent trends. *Journal of Adolescent Health*. 2015;56(2): 223–230.
9. Braine T. Adolescent pregnancy: a culturally complex issue. *Bulletin of the World Health Organization*. 2009;87(6): 410–411.
10. Hamilton, BE, Martin, JA, Osterman, MJK, Curtin, SC. Births: final data for 2014. Hyattsville, MD: National Center for Health Statistics; 2014.
11. Suan MAM, Ismail AH, Ghazali H. A review of teenage pregnancy research in Malaysia. *Medical Journal of Malaysia*. 2015;70(4): 214–219.
12. Kaplanoglu M, Bülbüla M, Koncab C, et al. Gynecologic age is an important risk factor for obstetric and perinatal outcomes in adolescent pregnancies. *Women and Birth*. 2015;28(3): 1–5.
13. Lukkari S, Hakko H, Herva A, Pouta A, Riala K, Räsänen P. Exposure to obstetric complications in relation to subsequent psychiatric disorders of adolescent inpatients: specific focus on gender differences. *Psychopathology*. 2012;45(5): 317–326.
14. World Health Organization. Adolescent Pregnancy Fact Sheet. [internet] Switzerland: WHO; 2014 [cited 1st November 2015]. Available from: http://apps.who.int/iris/bitstream/10665/112320/1/WHO_RHR_14.08_eng.pdf.
15. Leppälähti S, Gissler M, Mentula M, Heikinheimo O. Is teenage pregnancy an obstetric risk in a welfare society? A population-based study in Finland, from 2006 to 2011. *BMJ Open*. 2013;3(8): e003225.
16. Noordin N, Zakaria Z, Sawal MZHM, Hussin ZH, Ngah K, Nordin J. The voice of youngsters on baby dumping issues in Malaysia. *Global Journal of Health Science*. 2013;3(1): 66–71.
17. Ministry of Health Malaysia. Annual Report 2012. [internet] [cited 1st November 2015]. Available from: <http://www.moh.gov.my/images/gallery/publications/MOH%202012.pdf>.

18. Augustin S. Malaysia: birth out of wedlock 'alarming'. *New Straits Times*. 21st March 2011. Available from: LINK [Accessed DATE].
19. Ruhaizan H, Ravichandran J, Rozima S, et al. Unwed mothers from 14 tertiary hospitals in Malaysia 2011–2012. In: *7th National Conference for Clinical Research, 3–5 September 2013*. Kuala Lumpur; 2013.
20. Talib J, Mamat M, Ibrahim M, Mohamad Z. Analysis on sex education in schools across Malaysia. *Procedia - Social and Behavioral Sciences*. 2012;59: 340–348.
21. Saim NJ. Social support, coping, resilience and mental health in Malaysian unwed young pregnant women and young mothers: their experiences while living in a shelter home [dissertation]. Umeå: Umeå Universitet; 2013 [Accessed 26th July 2016]. Available from: <http://umu.diva-portal.org/smash/record.jsf?pid=diva2%3A662379&dsid=-3159>.
22. Banerjee B, Pandey GK, Dutt D, Sengupta B, et al. Teenage pregnancy: a socially inflicted health hazard. *Indian Journal of Community Medicine*. 2009;34(3): 227–231.
23. Brabin BJ, Hakimi M, Pelletier D. An analysis of anemia and pregnancy-related maternal mortality. *Journal of Nutrition*. 2001;131(2): 604S–615S.
24. Zimmermann MB, Hurrell RF. Nutritional iron deficiency. *The Lancet*. 2007;370: 511–520.
25. Thaitae S, Thato R. Obstetric and perinatal outcomes of teenage pregnancies in Thailand. *Journal of Pediatric and Adolescent Gynecology*. 2011;24(6): 342–346.
26. Roth J, Hendrickson J, Schilling M, Stowell DW. The risk of teen mothers having low birth weight babies: implications of recent medical research for school health personnel. *Journal of School Health*. 1998;68(7): 271–275.
27. Khashan AS, Baker PN, Kenny LC. Preterm birth and reduced birthweight in first and second teenage pregnancies: a register-based cohort study. *BMC Pregnancy and Childbirth*. 2010.
28. Centers for Disease Control and Prevention. *Preterm Birth*. Available from: <http://www.cdc.gov/reproductivehealth/maternalinfanthealth/pretermbirth.htm> [Accessed DATE].
29. Xi-Kuan Chen, Shi Wu Wen, Nathalie Fleming, et al. Teenage pregnancy and adverse birth outcomes: a large population based retrospective cohort study. 2007;36: 368–373.
30. Debiec KE, Paul KJ, Mitchell CM, et al. Inadequate prenatal care and risk of preterm delivery among adolescents: a retrospective study over 10 years. 2010;203(2): 122.
31. Sulaiman S, Othman S, Razali N, Hassan J. Obstetric and perinatal outcome in teenage pregnancies. *South African Journal of Obstetrics and Gynaecology*. 2013;19(3): 77–80.
32. NHS. *Episiotomy and Perineal Tears*. Available from: <http://www.nhs.uk/conditions/pregnancy-and-baby/pages/episiotomy.aspx> [Accessed DATE].
33. Demirci O, Ertugrul Y, Özgür T, et al. Effect of young maternal age on obstetric and perinatal outcomes: results from the tertiary center in Turkey. 2016;33(3): 344–349.
34. Yadav S, Choudhary D, Narayan KC, et al. Adverse reproductive outcomes associated with teenage pregnancy. 2008;11(2): 141–144.

Validity and reliability of the Patient Assessment on Chronic Illness Care (PACIC) questionnaire among patients with type 2 diabetes mellitus in Malaysia: English version

Azam AF, Lai PSM, Abdullah A, Haidi Hanafi NS

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Authors:

Pauline Siew Mei Lai

(Corresponding author)

B Pharm, PhD

Department of Primary Care
Medicine, Faculty of Medicine
University of Malaya, 50603
Kuala Lumpur, Malaysia
Email: plai@ummc.edu.my

Ahmad Fithri Azam

MBBS, M Fam Med

Health Clinic Peringgit, Jalan Pantai
Peringgit, Malacca 75400, Malacca
Malaysia

Department of Primary Care
Medicine, Faculty of Medicine
University of Malaya, 50603
Kuala Lumpur, Malaysia

Adina Abdullah

MBBS, M Fam Med

Department of Primary Care
Medicine, Faculty of Medicine
University of Malaya, 50603
Kuala Lumpur, Malaysia

Nik Sherina Haidi Hanafi

M Fam Med, PhD

Department of Primary Care
Medicine, Faculty of Medicine
University of Malaya, 50603
Kuala Lumpur, Malaysia

Abstract

Introduction: The Patient Assessment on Chronic Illness Care (PACIC) was developed to assess patients' perspectives on the alignment of primary care to the chronic care model. The Malay PACIC has been validated; however, Malaysia is a multicultural society, and English is spoken by many Malaysians and expatriates. We sought to validate the English version of the PACIC among patients with diabetes mellitus in Malaysia, as Malaysians may interpret a questionnaire that was originally developed for Americans in a different way.

Method: This study was conducted between November and December 2016 at two primary care clinics that offered integrated diabetes care at the time. These sites were selected to assess the discriminative validity of the PACIC. Site 1 is a Malaysian Ministry of Health-run primary care clinic while site 2 is a university-run hospital-based primary care clinic. Only site 1 annually monitors patient performance and encourages them to achieve their HbA1c targets using a standard checklist. Patients with diabetes mellitus who understood English were recruited. Participants were asked to fill out the PACIC at baseline and two weeks later.

Results: A total of 200 out of the 212 invited agreed to participate (response rate=94.3%). Confirmatory factor analysis confirmed the 5-factor structure of the PACIC. The overall PACIC score and the score in two of the five domains were significantly higher at site 1 than at site 2. The overall Cronbach's alpha was 0.924. At test-retest, intra-class correlation coefficient values ranged from 0.641 to 0.882.

Conclusion: The English version of the PACIC was found to be a valid and reliable instrument to assess the quality of care among patients with diabetes mellitus in Malaysia.

Introduction

Chronic non-communicable diseases, such as diabetes mellitus (DM), hypercholesterolemia, and hypertension, collectively constitute the global leading cause of death; they were responsible for 38 million (68%) deaths worldwide in 2012.¹ According to the 2015 Malaysia National Health Morbidity Survey (NHMS-5), the prevalence of DM among adults ≥18 years was 17.5%,² marking a clear increase from 11.6% in 2006³ and 15.2% in 2011.⁴

In Malaysia, a large majority (79.3%) of DM patients receive treatment from government providers.² However, only 24% of patients have their DM adequately controlled.^{5,6} Consequently, the majority of DM patients suffer from microvascular complications (75%), macrovascular complications (28.9%), and severe late complications, including cataract (27.2%), microalbuminuria (7%), neuropathy

(45.9%), leg amputation (3.8%), and angina pectoris (18.4%).⁵ Despite efforts by the Malaysian Ministry of Health to improve diabetic care and glycemic control, the prevalence of diabetes-related complications in Malaysia has held steady.⁶ This may be because none of these efforts have empowered patients or enabled self-management, whereby patients are responsible for day-to-day decisions about and activities to control their illness. There is a dire need to transform the current health care system in Malaysia by adopting a new health care model that emphasizes and enhances self-management.

The Chronic Care Model (CCM) aims to optimize outcomes for patients with a chronic illness. This model requires an engaged and competent patient to partner with the healthcare system in order to receive the most effective care.⁷ The CCM consists of six elements to improve chronic-illness management: health care organization, clinical information system,

delivery system design, decision support, self-management support, and community resources.⁷ The model encompasses evidence-based medicine and patient-centered care (PCC; defined as “providing care that is respectful of and responsive to individual patient preferences, needs, and values, and ensuring that patient values guide all clinical decisions”).⁸ PCC has been proven to improve patient outcomes and health care quality as well as reduce patient burden. The CCM has successfully changed healthcare practices for chronic conditions and has increased the success of managing DM patients. Integrated care requires patients with DM to perform self-blood glucose monitoring and enact lifestyle changes.⁷

Several instruments have been developed and validated to assess PCC.⁹ Among them is the Patient Assessment of Chronic Illness Care (PACIC), which was selected due to its use by previous studies to assess patients’ perspectives on the alignment of primary care to the CCM⁹ and patient self-empowerment.¹⁰ Additionally, the instrument has good psychometric properties. In Malaysia, the Malay PACIC has been validated;¹¹ the aim of this study is to validate the English version of the PACIC for DM patients, as Malaysia is a multicultural society, and English is an important secondary language spoken by many Malaysians and expatriates. This re-validation is important because the standard English version of the PACIC, which was originally developed in the United States, may not necessarily be interpreted the same way in Malaysia on account of the cultural differences between the two countries.¹²

Methods

This validation study was conducted between November and December 2016 at two sites. Site 1 is a Malaysian Ministry of Health-run primary care clinic. It is serviced by one family medicine specialist, two medical officers (defined as those who have completed their housemanship, but have no specialist training), one diabetes educator, and one pharmacist. The infrastructure and staffing at site 1 are fairly limited; there are only three consultation rooms. Nevertheless, it provides integrated care for >1500 DM patients annually. The site-1 team delivers all elements of integrated care at one location except for diabetes retinopathy screening, which is done at a nearby clinic. Site 1 uses just one manual form to document all diabetes care; all team members have access to this record in order to ensure continuity of care. As with other public

health centers under the Ministry of Health, this clinic provides data for the National Diabetic Registry and is subject to regular audits. The diabetic team at site 1 annually monitors patient performance and encourages them to achieve their HbA1c targets using a standard checklist in order to achieve the care target set by the Ministry of Health.

Site 2 is a university-run primary care clinic in a teaching hospital under the Ministry of Education. It is serviced by 19 family medicine specialists, 35 family medicine trainees (currently undergoing their 4-year training to qualify as a family medicine specialist), and two medical officers. Site 2 has 32 consultation rooms. All DM patients have access to allied healthcare services (such as a diabetes nurse educator, a pharmacist, and a dietitian) and onsite diabetes retinopathy screening. Nevertheless, diabetes care in site 2 is delivered fragmentally, as all these services are located in different locations throughout the hospital. Providers split their time between patients in the primary care, endocrine, and geriatric clinics, among others; they may not always be well versed in the principles of primary care medicine. Site 2 uses electronic medical records, which likely aids continuity of care. However, access to these records may be limited to certain team members, which would limit its usefulness. Additionally, diabetes care is not regularly audited because there is no diabetes registry.

These sites were selected so we could assess the discriminative validity of the PACIC. We hypothesized that site 1 would have a higher PACIC score than site 2 because it monitors patients’ clinical outcomes and encourages them to achieve their HbA1c target at every consultation.

Participants

Participants were English-speaking DM patients who had been followed-up on for at least six months in the clinic. Patients with cognitive impairment or those who were too ill to participate were excluded.

Sample size

The sample size required to perform factor analysis was based on the number of items in the questionnaire multiplied by 10.¹³ Since the PACIC has 20 items, the minimum number of participants required was 200 (20*10).

Instruments used

Baseline demographic questionnaire

This instrument was used to collect the baseline demographic data of participants (age; gender; level of academic attainment; duration of diabetes; co-morbidities; diabetic medication).

The Patient Assessment of Chronic Illness Care (PACIC)

The PACIC consists of 20 items in five domains: patient activation (3 items), delivery system settings (3 items), goal setting/tailoring (5 items), problem solving/contextual (4 items), and follow up/ co-ordination (5 items). Each item was rated on a 5-point Likert scale from 1 (almost never) to 5 (almost always). Permission was obtained for its use through email on June 16th, 2016. Each domain is scored by the number of items within that domain. The overall PACIC score is calculated by summing the scores of all 20 items. A high PACIC score indicates that the care received was integrated and congruent with the CCM. A low PACIC score indicates that steps can be taken to improve health care services by improving the integration of delivered care.

Face and content validity

Face and content validity of the PACIC was determined by an expert panel consisting of two family medicine specialists, an academic experienced in instrument validation, and a family medicine trainee. The PACIC was then piloted in five adults with DM. Participants were invited to verbally express whether the items were easy to understand—no problems were reported. Hence, no changes were made.

Procedure

Participants were recruited through convenient sampling. All DM patients were screened for eligibility at the triage counter. Patients who fulfilled the inclusion criteria were informed of the voluntary study's purpose through the patient-information sheet. From those who agreed, written informed consent was obtained. Participants were then asked to fill in the baseline demographic form and the PACIC. Two weeks later, the PACIC was mailed to all participants. They were asked to mail the completed questionnaire back using the prepaid stamped envelope. If a reply was not received within a week, a reminder was sent.

Ethics approval was obtained prior to the study from the University Malaya Medical Centre Medical Ethics Committee (approval number: 20167-2615).

Data analyses

All analyses were performed using the Statistical Package for Social Sciences version 23.0 (Chicago, Illinois). Confirmatory factor analysis was done using Analysis of Moment Structure version 24.0 (Chicago, Illinois). Normality was assessed with the Kolmogorov–Smirnov test. Since normality could not be assumed, non-parametric tests were used. Continuous data was presented as median and interquartile range while categorical variables were presented as frequency and percentage.

Validity

Confirmatory factor analysis (CFA) was used to test whether our data fit the original 5-factor model. Various standard fit indices were used: normed chi-square, comparative fit index (CFI), and root mean square error of approximation (RMSEA).

The criteria of a good model of fit were as follows: normed chi-square < 3, CFI > 0.90, and RMSEA < 0.08. Each item should have a factor loading of > 0.4. Average variance (AVE) was calculated as the sum of the squared standardized factor loading divided by the number of items while the composite reliability (CR) was computed as a function of factor loading and error variance.¹⁴ CR and AVE values of more than 0.6 and 0.5, respectively, indicate good construct reliability.¹⁴

The Mann–Whitney U test was used to assess the discriminative validity of the PACIC by comparing the score between the two sites.

Reliability

The internal consistency of the PACIC was assessed using Cronbach's alpha. A Cronbach's alpha value of > 0.90 is said to be highly reliable with excellent internal consistency; 0.70–0.90 suggests that the scale has adequate internal consistency; < 0.70 indicates inadequate internal consistency.¹⁵ Corrected item-total correlation was used to determine the items that did not fit well in the questionnaire. Item-total correlation values must be > 0.20 to be considered acceptable. The effect of removing a single item on the Cronbach's alpha was also determined.¹⁵

Test-retest reliability was assessed using the intra-class correlation coefficient (ICC) to examine the strength of agreement between the repeated measures: >0.75 indicates excellent inter-rater agreement; 0.60–0.74 shows good agreement; 0.40–0.59 indicates fair to moderate agreement; <0.4 means poor agreement. Correlation was assessed using Spearman's rho correlation: <0.2 is poor; 0.21–0.40 is fair; 0.41–0.60 is good; 0.61–0.80 is very good; 0.81–1.0 is excellent.¹⁶

Results

A total of 200 out of the 212 invited agreed to participate (response rate=94.3%). Patients at site 2 were, relative to those at site 1, more likely to have a background in tertiary education and, on average, had a longer duration of DM (Table 1).

Table 1: Demographic characteristics of participants

Variables	Total (n=200) n (%)	Site 1: Ministry of Health-run district primary care clinic (n=95) n (%)	Site 2: University- run hospital-based primary care clinic (n=105) n (%)	p-value#
Median age in years (IQR)	59 (52.0-66.0)	58 (52.0-67.0)	60 (50.5-64.5)	0.445
Gender				
Male	81 (40.5)	40 (42.1)	41 (39.5)	0.660
Female	119 (59.5)	55 (57.9)	64 (61.0)	
Highest level of education				
Primary (6 years of education)	20 (10.0)	20 (10.0)	6 (5.7)	0.010*
Secondary (12 years of education)	109 (54.5)	109 (54.5)	50 (47.6)	
Diploma/tertiary (≥13 years of education)	71 (35.5)	42 (21.0)	49 (46.7)	
Median duration of DM (years; IQR)	7.0 (4.0-11.0)	5.0 (3.0-9.0)	8.0 (5.0-13.0)	<0.001*
Hypoglycemic agents prescribed				
Biguanide (e.g., metformin)	176 (88.0)	83 (87.4)	93 (88.6)	
Sulfonylurea (e.g., gliclazide)	96 (48.0)	53 (55.8)	43 (41.0)	
Insulin	54 (27.0)	24 (25.3)	30 (28.6)	
Alpha-glucosidase inhibitor (e.g., acarbose)	22 (11.0)	8 (8.4)	14 (13.3)	
DPP-4 inhibitor (e.g., sitagliptin)	21 (10.5)	5 (5.3)	16 (15.2)	
Number of patients followed-up on				
By the dietician	119 (59.5)	46 (48.4)	73 (69.5)	
For fundoscopy	141 (70.5)	53 (55.8)	88 (83.8)	
By the diabetic nurse educator	138 (69.0)	80 (84.2)	58 (55.2)	

The Mann–Whitney U test was used for continuous variables while the chi-squared test was used for categorical variables.

* Statistically significant; DPP-4=dipeptidyl peptidase-4

Validity

CFA showed that the PACIC had five domains (Table 2); the normed chi-square was 2.284, the CFI was 0.89, and the RMSEA was 0.08. When all 20 items were loaded into the five constructs, all items had standardized loading factors of >0.40 (except for item 16), average variance extracted (AVE) values >0.50 (except for the delivery system domain and the follow-up/coordination domain), and composite reliability (CR) values >0.60.

Table 2: Confirmatory factor analysis of the Patient Assessment of Chronic Illness Care

Item no.	Domains (factor loadings)					Average variance extracted	Composite Reliability
	Patient activation	Delivery system design/ decision support	Goal setting/ tailoring	Problem solving/ contextual	Follow-up/ coordination		
1	0.812					0.551	0.871
2	0.771						
3	0.632						
4		0.652				0.371	0.639
5		0.577					
6		0.597					
7			0.824			0.666	0.804
8			0.762				
9			0.512				
10			0.516				
11			0.716				
12				0.704		0.591	0.852
13				0.767			
14				0.829			
15				0.769			
16					0.207	0.373	0.722
17					0.471		
18					0.609		
19					0.768		
20					0.800		

Normed chi-squared=2.284; comparative fit index (CFI)=0.89; root mean square error approximation (RMSEA)=0.08.

The overall PACIC score, as well as the score of two domains (goal setting/tailoring and follow-up/coordination), were significantly higher at site 1—a district primary care clinic that monitored patient outcomes—than at site 2, a hospital-based primary care clinic that did not monitor patient outcomes (Table 3).

Table 3: Discriminative validity of the Patient Assessment of Chronic Illness Care

Domain	Site 1: Ministry of Health-run primary care clinic (n=95)		Site 2: University-run hospital-based primary care clinic (n=105)		Mann–Whitney U test	
	Median	IQR	Median	IQR	z-score	p-value
Patient activation	11.00	11.00	10.00	10.00	-1.693	0.090
Delivery system design/ decision support	11.00	11.00	11.00	11.00	-1.87	0.061
Goal setting/tailoring	18.00	18.00	16.00	16.00	-3.635	<0.001*
Problem solving/contextual	14.00	14.00	14.00	14.00	-0.728	0.467
Follow-up/coordination	16.00	16.00	15.00	15.00	-2.825	0.004*
Overall PACIC score	70.00	70.00	67.00	67.00	-2.148	0.032*

The Reliability

The overall Cronbach's alpha for the PACIC was 0.924, with each domain ranging from 0.639 to 0.850 (Table 4). All corrected item-total correlation values were >0.20. The deletion of item 16, "contacted after a visit to see how things were going," increased the Cronbach's alpha from 0.700 to 0.744.

At retest, 141 of the original 200 participants responded (response rate=70.5%); 59 participants were uncontactable (n=48) and 11 refused to answer the questionnaire again (n=11). Spearman rho correlation ranged from 0.597 to 0.858 while intra-class correlation coefficient values ranged from 0.641 to 0.882 (Table 4).

Table 4: Psychometric properties of the Patient Assessment of Chronic Illness Care

Domain	Item no.	Test (n=200)			Retest (n=141)	
		Cronbach alpha	Corrected item-total correlation	Cronbach alpha if item is deleted	Spearman rho correlation	Intraclass correlation coefficient
Patient activation	1	0.780	0.642	0.676	0.746	0.782
	2		0.663	0.657	0.737	0.762
	3		0.554	0.769	0.811	0.882
Delivery system design/ decision support	4	0.639	0.425	0.583	0.829	0.805
	5		0.491	0.490	0.597	0.660
	6		0.438	0.555	0.786	0.811
Goal setting/ tailoring	7	0.792	0.654	0.730	0.715	0.740
	8		0.622	0.741	0.629	0.641
	9		0.498	0.781	0.854	0.823
	10		0.515	0.776	0.851	0.829
	11		0.616	0.739	0.760	0.844
Problem solving/ contextual	12	0.850	0.640	0.830	0.795	0.805
	13		0.698	0.806	0.842	0.801
	14		0.751	0.782	0.743	0.847
	15		0.670	0.818	0.785	0.813
Follow-up/ coordination	16	0.700	0.225	0.744	0.801	0.785
	17		0.504	0.631	0.825	0.766
	18		0.445	0.656	0.838	0.827
	19		0.535	0.620	0.783	0.766
	20		0.606	0.585	0.858	0.773

Discussion

The PACIC was found to be a 5-factor model and a good model-of-fit with adequate psychometric properties. It was able to discriminate between two sites with different levels of integrated care for DM patients.

CFA confirmed that the English version of the PACIC was a 5-factor model, as per the original PACIC validation study.⁹ Some studies reported that the PACIC was a 1-factor,¹⁷ 2-factor,^{18,19} or a 3-factor model.^{11,20} The Malay PACIC found that their instrument was a 3-factor model

("patient healthcare interaction," "follow up/ coordination," and "delivery system design") after two items (10 and 16) were deleted.¹¹ "Goal setting" was not recognized as a domain but was integrated into a new domain ("patient health-health care interaction").¹¹ One possible explanation is that patients may have assessed their care differently as health care systems vary by site. The original PACIC validation also found that the five PACIC domains did not map perfectly to the CCM.⁹ Two of the CCM's components (delivery system design and decision support) merged to become a single domain in the PACIC, while the self-

management component of the CCM was further divided into three separate domains: patient activation, goal setting/tailoring, and problem-solving/contextual counseling.

The PACIC was able to discriminate the integrated care provided, thus confirming the discriminative validity of our instrument; this is in line with a previous study.¹⁹ Site 1 had a higher overall PACIC score because it monitored patients' clinical outcomes and provided a higher level of integrated care than did site 2. This may be because the diabetes care-delivery model at site 1 mirrored the CCM, as it encouraged self-management. Additionally, the advisory clinic panel in site 1 consisted of leaders from the local community, which collaborated with a non-communicable disease prevention community ("*Komuniti Sibat Pembina Negara*" [KOSPEN]),²¹ to enhance primary health care. The regular use and implementation of diabetes care audits using the National Diabetes Registry also enabled site 1 to closely monitor their DM patients (and easily trace those who default treatment) and ensure the delivery of coordinated care.

Site 2 patients were more likely to have a background in tertiary education than those at site 1. This may be because site 2 is an established hospital-based primary care clinic (>50 years) that is located next to a university, meaning it is populated by well-educated retired individuals. Site 2 also saw longer DM durations, likely because these patients tend to have more complications, and specialist referrals are easier at site 2 than at site 1.

The overall Cronbach's alpha of the PACIC was 0.924, which is similar to the findings of previous studies.^{10,18–20} This suggests that the PACIC has achieved adequate internal consistency. However, the "delivery setting" domain only had a Cronbach's alpha of 0.639. This may be due to item no. 4 ("Given a written list of things I should do to improve my health"). Ideally, doctors would provide patients with a written list to improve their health. Instead, however, doctors in site-2 generally advise patients verbally. The practice of contacting a patient after a visit is seldom practiced in Malaysia due to resource limitations (item no. 16: "Contacted after a visit to see how things were going"). However, deletion of item 16 only slightly increased the Cronbach's alpha from 0.700 to 0.744. This item was retained because a Cronbach's alpha value of ≥ 0.700 is adequate. It would be best to keep all the items in the questionnaire so that, when re-validating a questionnaire in another country,

results can be compared. Test-retest reliability is sufficient; Spearman rho values indicate good to excellent correlation while ICC values indicate good to excellent inter-rater agreement, showing that the PACIC has achieved stable reliability. Previous validation studies also found that the PACIC was a reliable instrument.⁹

Although no changes were made to the original English PACIC, it was important for the English PACIC to be re-validated in Malaysia to ensure that the interpretation of the questionnaire was similar to that of the original.¹² This was confirmed by the adequacy of its psychometric properties, which confirmed that the English PACIC can now be used in Malaysia to assess the quality and care provided by primary care physicians.

During the original development of the PACIC, the authors recruited 283 adults suffering from chronic illness to validate their instrument.⁹ The most common chronic illnesses among these participants were hypertension, arthritis, depression, diabetes, asthma, and pain.⁹ Diabetes is an archetypal chronic illness needing integrated care between patients and healthcare providers. Although we only recruited participants with diabetes, it is likely that most of them also have other chronic illnesses, such as hypertension and hyperlipidemia. Therefore, the results of our study may also be applicable to patients with other chronic conditions.

One limitation of this study is that we were unable to randomly recruit participants, as neither site had a registry of DM patients coming in for the day. Additionally, it was not possible to assess the convergent validity of the PACIC because there were no other validated instruments that assessed PCC when this study was conducted. A clear strength of our study is that the English PACIC underwent the process of validation, which provides evidence for the construct and discriminative validity of the English PACIC in Malaysia.

Conclusion

The English PACIC was deemed a 5-factor model and a good model of fit. It was found to be a valid and reliable instrument for patients to assess the quality and care provided by their primary care physicians based on the CCM. This instrument can be used to evaluate the care provided by various centers to better align them with CCM recommendations, ultimately improving the outcomes of DM patients.

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Abbreviations used

DM	: Diabetes mellitus
NHMS-5	: 2015 Malaysia National Health Morbidity Survey
CCM	: Chronic Care Model
PCC	: Patient-centered care

PACIC	: Patient Assessment of Chronic Illness Care
CFA	: Confirmatory factor analysis
CFI	: Comparative fit index
RMSEA	: Root mean square error of approximation
AVE	: Average variance
CR	: Composite reliability
ICC	: Intra-class correlation coefficient
KOSPEN	: “Komuniti Sihat Pembina Negara”

Conflicts of interest

All authors declare that they have no conflict of interest.

How does this paper make a difference to general practice?

- The English Patient Assessment of Chronic Illness Care (PACIC) instrument was found to be a 5-factor model and a good model of fit.
- It was found to be a valid and reliable instrument, as it had adequate psychometric properties.
- It can now be used to assess the quality of care among patients with diabetes mellitus in Malaysia and to determine whether the care aligns with chronic care-model recommendations.

References

1. World Health Organization. Innovative care for chronic conditions: building blocks for action. Geneva: WHO; 2002 [cited 2nd August]. Available from: <https://www.who.int/chp/knowledge/publications/iccreport/en/>.
2. Institute for Public Health. The national health and morbidity survey 2015: non-communicable diseases, risk factors & other health problems. CITY: ORGANIZATION; 2015 [cited 2nd August 2019]. Available from: <http://www.moh.gov.my/moh/resources/nhmsreport2015vol2.pdf>.
3. Letchuman GR, Wan Nazaimoon WM, Wan Mohamad WB, Chandran LR, Tee GH, Jamaiah H, et al. Prevalence of diabetes in the Malaysian national health morbidity survey III 2006. *Medical Journal of Malaysia*. 2010;65(3): 180–186.
4. Institute for Public Health. National health and morbidity survey 2011. CITY: ORGANIZATION; 2012 [cited 2nd August 2019]. Available from: <http://iku.moh.gov.my/images/IKU/Document/REPORT/NHMS2011-VolumeIII.pdf>.
5. Mafauzy M, Hussein Z, Chan SP. The status of diabetes control in Malaysia: results of DiabCare 2008. *Medical Journal of Malaysia*. 2011;66(3): 175–181.
6. Mafauzy M, Zanariah H, Nazeri A, and Chan SP. DiabCare 2013: a cross-sectional study of hospital-based diabetes care delivery and prevention of diabetes-related complications in Malaysia. *Medical Journal of Malaysia*. 2016;71(4): 177–185.
7. Wagner EH. Chronic disease management: what will it take to improve care for chronic illness? *Experimental and Clinical Psychopharmacology*. 1997;1(1): 2–4.
8. Institute of Medicine. *Crossing the quality chasm: A new health system for the 21st Century*. Washington: National Academy Press; 2001.
9. Glasgow RE, Wagner EH, Schaefer J, Mahoney LD, Reid RJ and Greene SM, et al. Development and validation of the Patient Assessment of Chronic Illness Care (PACIC). *Medical Care*. 2005;43(5): 436–444.
10. Vrijhoef HJ, Berbee R, Wagner EH and Steuten LM. Quality of integrated chronic care measured by patient survey: identification, selection and application of most appropriate instruments. *Health Expectations*. 2009;12(4): 417–429.
11. Abdul-Razak S, Ramli AS, Badlishah-Sham SF, Haniff J, Jenn NC, Ludher I S, et al. Validity and reliability of the patient assessment on chronic illness care (PACIC) questionnaire: the Malay version. *BMC Family Practice*. 2018;19(1): 119.
12. Lai P. Validating instruments of measure: is it really necessary? *Malaysian Family Physician*. 2013;8(1): 2–4.
13. Gorsuch RL. *Factor analysis (2nd ed.)* Hillsdale: Lawrence Erlbaum Associates; 1983.
14. Fornell C, Larcker DF. Evaluating structural equation models with unobservable variables and measurement error. *Journal of Marketing Research*. 1981; 18(1): 39–50.

15. Cortina JM. What is coefficient alpha? An examination of theory and applications. *Journal of Applied Psychology*. 1993;78(1): 98.
16. Sullivan R, Kinra S, Ekelund U, Bharathi AV, Vaz M, Kurpad A, et al. Evaluation of the Indian Migration Study Physical Activity Questionnaire (IMS-PAQ): a cross-sectional study. *International Journal of Behavioral Nutrition and Physical Activity*. 2012;9(1): 13.
17. Aung E, Ostini R, Dower J, Donald M, Coll JR, Williams GM, et al. Patient Assessment of Chronic Illness Care (PACIC) in Type 2 Diabetes: a longitudinal study. *Evaluation & the Health Professionals*. 2016;39(2): 185–203.
18. Tušek-Bunc K, Petek-Šter M, Branko S and Petek D, et al. Validation of the Slovenian version of Patient Assessment of Chronic Illness Care (PACIC) in patients with coronary heart disease. *Collegium Antropologicum*. 2014;38(2): 437–444.
19. Taggart J, Chan B, Upali UW and Christl B, et al. Patients Assessment of Chronic Illness Care (PACIC) in two Australian studies: structure and utility. *Journal of Evaluation in Clinical Practice*. 2011;17(2): 215–221.
20. Wensing M, Lieshout J, Jung H and Hermesen J, et al. The Patients Assessment Chronic Illness Care (PACIC) questionnaire in the Netherlands: a validation study in rural general practice. *BMC Health Services Research*. 2008;8(1): 1.
21. Ministry of Health Malaysia. KOSPEN: Komuniti Sihat Pembina Negara. CITY: ORGANIZATION; 2017 [cited 18th June 2018]. Available from: <https://kpkesehatan.com/2017/11/01/kospen-komuniti-sihat-pembina-negara/>.

Pay-for-performance challenges in family physician program

Gharibi F, Dadgar E

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Authors:

Farid Gharibi

(Corresponding author)
PhD of Health Services Management,
Food Safety Research Center (safi),
Semnan University of Medical
Sciences, Semnan, Iran
email: gharibihsa@gmail.com

Elham Dadgar

PhD candidate in Health Care
Services Management
Department of Health Management
and Economics, School of Public
Health, Tehran University of Medical
Sciences, Tehran, Iran

Abstract

Objective: This study was conducted to investigate the challenges faced in the implementation of the pay-for-performance system in Iran's family physician program.

Study design: Qualitative.

Place and duration of study: The study was conducted with 32 key informants at the family physician program at the Tabriz University of Medical Sciences between May 2018 and June 2018.

Method: This is a qualitative study. A purposeful sampling method was used with only one inclusion criterion for participants: five years of experience in the family physician program. The researchers conducted 17 individual and group non-structured interviews and examined participants' perspectives on the challenges faced in the implementation of the pay-for-performance system in the family physician program. Content analysis was conducted on the obtained data.

Results: This study identified 7 themes, 14 sub-themes, and 46 items related to the challenges in the implementation of pay-for-performance systems in Iran's family physician program. The main themes are: workload, training, program cultivation, payment, assessment and monitoring, information management, and level of authority. Other sub-challenges were also identified.

Conclusion: The study results demonstrate some notable challenges faced in the implementation of the pay-for-performance system. This information can be helpful to managers and policymakers.

Introduction

The primary mission of health systems is to provide high-quality care and satisfactory health outcomes using their available resources. However, evidence indicates that the level of spending does not necessarily determine the quality of outcomes. For example, health expenditure in the US constitutes about 16 percent (USD 4 trillion) of the country's GDP.¹ However, despite spending twice as much as most industrialized countries, the US health care system is ranked last among industrialized countries in terms of quality, accessibility, and efficiency;² this low ranking is exacerbated by severe geographical, economic, and racial inequalities in terms of access to health services and health outcomes.³

Accordingly, many governments have adopted initiatives to manage health care performance indicators, especially in rural areas. Pay-for-performance (P4P) is one of the most prominent programs in this respect.^{4,5} P4P is designed to enhance health services through financial incentives;⁶ it aims to improve the quality and efficiency of services and overcome the shortcomings of conventional repayment systems, which provide financial incentives merely on the basis of the volume and complexity of services.⁷ Successful design and

implementation of P4P requires many elements, including performance indicators and standards for effective evaluation, training for surveyors/evaluators, appropriate external evaluation processes, educating for family physicians and their team members on evaluation requirements, linkages between performance-evaluation results and payment systems, and financial incentives to encourage high-quality services.⁸

Financial incentives in P4P achieve two major goals: one, they provide an economic incentive to change provider behavior by encouraging a high-quality, evidence-based performance;^{9,10} two, they eliminate the negative effects of existing repayment systems, such as those that consider volume rather than value.¹¹ Traditional payment approaches like fee-for-service (FFS) lead to inducing demand and overuse while controlled care results in the underuse of health services.¹² P4P is a system in which payments are based on the quality and efficacy of the provided care. This system is used as a complement to volume-based methods (FFS), case payments (payment for each discharge based on diagnosis-related group) and per capita (capitation) payments.¹³

Studies show that the number of P4P-related programs has dramatically increased (from 37 cases in 2003 to 170 cases in 2007).¹⁴ In the

United States, P4P is used by more than 100 private health-care programs as well as Medicaid and Medicare.¹⁵ England, Canada, and Australia are among the countries that use P4P as the basis of medical payments.^{16–18}

Of course, P4P is not without its flaws or negative consequences; in some cases, its implementation has resulted in the spread of inequality and a reduction in the quality of some services.^{19,20} This system has some major disadvantages, including inappropriate health outcomes, the spread of inequality in the health sector, and a potential increase in cost.²¹ Another negative consequence of the system is that service providers often neglect areas of care that, despite significantly impacting health outcomes, are un-measurable or attribute no rewards.²² This system can sometimes improve the quality of documentation rather than that of services and care, failing its main objective.²³

Using P4P, especially in areas such as primary health care (PHC) and family physician programs, may result in the inappropriate and unnecessary use of therapeutic procedures. For example, a hospital seeking better health outcomes may prescribe antibiotics for patients with pneumonia or other infectious diseases regardless of whether they are required.^{24,25} This system can also exacerbate inequality along racial, ethnic, gender, linguistic, or economic lines. For instance, P4P can lead to the exclusion of high-risk patients or patients from specific social groups;²⁶ it can also result in cream skimming, meaning that providers may choose individuals with a higher probability of better results and a higher performance.²⁷ It is worthwhile to note that P4P systems, which are currently used all around the world, vary significantly in terms of evaluation methods and payment mechanisms; consequently, there is significant outcome variance.²⁸

PHC is the core of any public health system. It is defined as “the health care services delivered in the first level of contact between individuals, families, and society with the health system.” The PHC system provides essential health care services in people’s major life environments, such as the home, school, and the workplace. The Iranian PHC system is a well-designed system that has expanded to provide its service package—which includes child and maternity health, environmental health, professional health, communicable health and immunization, non-communicable health, disaster management, health education and

promotion, healthy nutrition, and oral health—to all citizens, including those in rural areas. It is the largest component of the overall Iranian health care system with a high workload but a low share of the annual budget from the Iranian Ministry of Health and Medical Education (MOHME). Most of the services provided by the Iranian PHC system are free and those that are not are low in cost. Accordingly, this system should receive significant funding from the MOHME; however, it received just 10% of the main budget.

Generally, the family physician program is an effective and efficient plan that provides comprehensiveness, gatekeeping, a referral system, continual care, access to care, quality, and safety in the PHC system. The family physician program in the Iranian PHC system was launched more than a decade ago to meet the medical needs of and serve as a gatekeeper for the population. While family physicians mainly focus on primitive and preventive services, they usually do the same routine curative affairs done by all physicians outside the PHC system. Family physicians are medical doctors that have graduated from medical universities; they can work as general practitioners but they must first work, in return for their free education, in rural or urban areas as family physicians. However, medical doctors often remain in PHC as family physicians after their compulsory work.

Medical graduates willing to work in PHC can join FPP immediately after medical school without passing any additional training courses. These physicians will be the managers of PHC centers in rural or urban areas. This is a remarkable feat, as a successful performance in both occupations (family physician and health services manager) requires high knowledge and experience. Family physicians are paid a fixed monthly amount unrelated to performance, quality, or even quantity.

Given the importance and necessity of improving the quality, effectiveness, and efficiency of services provided in the Iranian family physician program through a P4P system, it is essential to understand and address the challenges involved in the implementation of P4P. This paper aims to investigate these challenges and provide appropriate solutions.

Methods

This study uses a qualitative approach. The data

was collected through interview sessions with 32 participants. We interviewed 23 family physicians earning their Master of Public Health (MD, MPH); four MPH instructors/academics (PhD in one of the following fields: social medicine, health services management, and epidemiology), and five current or former senior managers of the family physician program (MD, PhD) at the Tabriz University of Medical Science. A total of 17 individual and group interviews were conducted in an unstructured manner (non-directive approach). The MPH is an informal, non-mandatory educational qualification general practitioners can acquire to enhance their competencies in public health, PHC, and family medicine. The qualitative approach was used to meet the needs of this study due to the lack of any similar quantitative study and its related questionnaire.

This study was conducted by two researchers (one male and one female) with PhD degrees in health services management. Both authors currently work as faculty everyone invited to participate accepted the invitation. This research topic was selected because of its importance and relevance to the fields of public health and health services management.

Purposeful sampling was used to gather information from people who are able to provide rich views and experiences on the subject. This sampling method allowed the researchers to extract conceptual patterns from the minds of various individuals in an optimal manner.²⁹ For this study, the researchers conducted 17 individual and group face-to-face interviews in order to obtain participants' thoughts on the challenges in the implementation of P4P in the family physician program. Information was obtained through unstructured questions without directing their views to avoid bias.

The scheduled interviews were conducted in the health services management and social medicine departments at the Tabriz University of Medical Sciences. One of the researchers asked the determined interview questions and facilitated the discussion between participants while the other researcher took notes and sent feedback to the participants. The duration of the individual and group interviews ranged from 45 to 60 and 105 to 135 minutes, respectively. The interviews continued up to the point of data saturation level; there was eventually no new information coming from the participants. Finally, the received comments were spread among all participants and feedback was collected for confirmation and

correction. All interview sessions were recorded with a recorder device and then transcribed, meaning the researchers had two sources of data for analysis: the notes obtained from and approved by participants and the transcriptions of the recorded interviews on paper.

Content analysis was used to analyze the data obtained from the interviews. This means that the concepts and existing themes in the data were extracted, interpreted, and reported using a systematic approach (30). In the coding process, challenges expressed by the participants were presented in the form of a code or item and were nominated. Then, according to the overt and covert themes embedded in the codes, identical codes were categorized based on their meanings/contents and the sub-themes of the study were formed. Finally, the main themes of the study were created and labeled with the integration of sub-themes. Initially, 74 codes were obtained; then, similar codes were merged to form a total of 46 codes.

During the interpretation stage, which entails searching for patterns, communications, concepts, and interpretations in the aggregated data, the researchers developed and interpreted the ideas and their contents by examining the final themes.³¹ In order to enhance and confirm the rigor of the study, the two researchers analyzed the transcripts independently. An independent external assessor then compared the results of the two analyses and a conclusion was reached after a final discussion. The analysis results were given to all of the participants for their approval and agreement. Additionally, two experts confirmed the validity of the obtained items, subthemes, and themes.

Informed consent was obtained from all participants. All participants participated in the study freely and permission was obtained from all of the participants to record their interviews. Anonymity was guaranteed. This study was approved by the Ethics Committee of Tabriz University of Medical Sciences (IR.TBZMED.REC.1394.580).

Results

The age range of study participants was 29–48 with a mean age of 35. The majority of participants (65%) were male and worked for the government (mainly in PHC centers as family physicians) in collaboration with other health workers. Currently, family physicians in Iran receive fixed monthly payments that are not

affected by people covered (per capita), services delivered (per case), or quality of provided care (P4P).

This study identified 7 themes, 14 sub-themes, and 46 items related to the challenges in the implementation of P4P systems in the Iranian family physician program. The main themes are: workload, training, program cultivation, payment, assessment and monitoring, information management, and level of authority. Other sub-challenges were also identified (**Table 1**).

Table 1: Challenges in the implementation of the pay-for-performance system in Iran's family physician program

Main Themes	Sub-Themes	Related Codes
Workload	Heavy workload of family physicians	- Broadness of duties - Large number of covered people
Training	Lack of management skills in family physicians	- Lack of knowledge and skills related to leadership - Lack of knowledge and skills related to quality - Lack of knowledge and skills related to teamwork
	Lack of knowledge and skills related to preventive and social medicine in family physicians	- Medical students neglecting public health courses - Lack of newcomer and in-service trainings - Weakness of family physicians, especially in promotional and preventive affairs - Physician's activity in family physician team limited to the conventional therapeutic approach - Weakness of family physicians' attitude towards the nature and activities of this program
Program cultivation	Lack of awareness among people about the nature and importance of family physicians	- Poor education by the MOHME and mass media about the family physician program - Public failure to follow the referral system - Low public trust in the expertise and ability of family physicians - Use of physicians with little experience as a family physician
	Self-underestimation of status and importance among family physicians	- Giving little importance to family physicians relative to clinical specialties - View among family physicians that the job is a temporary one that they will leave soon
	Weak intra/extra-collaboration with family physician team	- Poor collaboration of institutions outside the health sector with family physician team - Lack of feedback about referred patients by specialists to the family physicians
Payment	Low PHC budget	- Low PHC budget relative to hospital services - Low wages for family physicians relative to specialists
	Lack of proper infrastructure for instituting P4P	- Insufficient infrastructure for performance-based payment system - Clinical and treatment views of the managers in charge of paying family physicians - Individual-centeredness of payments - Lack of competition among family physicians

Main Themes	Sub-Themes	Related Codes
Assessment and monitoring	Lack of criteria and scientific tools for qualitative assessment of the program	- Lack of suitable criteria and assessment tools even for routine monitoring programs - Lack of consideration of the conditions of different work environments and communities while monitoring - Lack of attention to the multiplicity of functions and results in evaluations
	Lack of a well-defined mechanism for assessment	- Government-owned monitoring and evaluation system and the lack of an independent entity for this task - Lack of proper accreditation system in the field of PHC - Overlooking doctor mistakes due to lack of physicians - Lack of organization and discipline in monitoring and evaluation
	Lack of appropriate assessors	- Lack of trained and experienced assessors - Inappropriate treatment of assessors by family physicians and their teams
Information management	Poor information infrastructure	- Weakness in software and hardware infrastructures - Lack of proper health records, especially in electronic form
	Poor systemic management of production cycle and information flow	- Poor management in the cycle of data collection and analysis as well as the production and flow of information - Poor and incomplete filling of existing files by physicians - Lack of appropriate databases related to health centers and their performances - Lack of proper information exchange between institutions and different levels in the provision of services - Neglecting the verification of documentations and reports provided by family physicians - Non-correspondence of the data and information created in the system with the real needs - Negligence of user-friendliness of the information provided for different users
Level of authority	Insufficient authority of family physicians	- Inadequate authority of family physicians to establish intra/extra sectoral relationships - Lack of sufficient supervisory power of family physicians over the health team and social workers - Inadequate authority of physicians in the selection or modification of the health team members

Workload

Heavy workload of family physicians

One challenge faced in the implementation of a P4P system by those in the family physician program is the broad range of responsibilities, which results in a heavy workload. They claim

that the workload is so heavy that they cannot perform many of their duties in a high-quality manner; when the number of people covered by a physician is too high, even their basic duties—such as providing proper health education, positively influencing patients, conducting research, and obtaining statistics—cannot be performed effectively. The following statements

represent this theme: “The range of defined duties for family physicians is very extensive and the workload is extremely high, and in such a situation there is not enough time for doing preventive and promotional activities” (MPH student, 34, man); “The implementation of P4P system will be effective only when the duties assigned to family physicians are reasonable and within their capabilities” (MPH instructor, 51, woman).

Training

Lack of management skills in family physicians

Another challenge is the lack of management knowledge among family physicians, especially regarding leadership and quality improvement. Family physicians do not receive theoretical and practical training, so they do not have skills in this field. This theme is exemplified by these statements: “Leadership training courses of the physicians are very limited, and, in many cases, the doctors do not receive any in-service training, especially in management affairs. Their in-service trainings are also inadequate and inefficient” (PHC manager, 52, man); “People in the family physician team have not been trained to carry out the affairs related to this program, and they have been working with the same mentality and incomplete skills for years” (MPH student, 39, man).

Lack of knowledge and skills in preventive and social medicine among family physicians

Another challenge is the limited capability of family physicians to deal with preventive and social medicine due to their lack of adequate knowledge, attitude, and skill in those areas. As a result, family physicians are not involved in areas such as medical prevention and family care. This exclusion results in the lack of community-based services and the provision of services in a defective, treatment-based manner. This theme stems from statements in the vein of the following: “Medical students do not receive proper education on how to play an effective role in family physician program at the university, and this leads to the formation of a treatment-based mentality in them” (PHC manager, 48, man); “Many doctors start their job as family physicians immediately after obtaining a degree and they do not receive proper trainings at the beginning of their service. Resultantly, family physicians take the same treatment-based approach acquired at the university and practice it” (MPH instructor, 37, woman).

Program cultivation

Lack of awareness among people about the nature and importance of family physicians

Another obstacle is the insufficiency of cultural programs from the MOHME and other responsible institutions aimed at educating the community about the nature, objectives, importance, and approach of the family physician program. This has left people unaware of the program, resulting in low levels of cooperation with family physician teams. The following statement embodies this problematic theme: “People are unfamiliar with the nature and philosophy of family physicians, because such responsible bodies as the MOHME and the mass media do not provide adequate and continuous information about family physician program and its importance to the people” (MPH student, 27, woman).

Self-underestimation of status and importance among family physicians

In addition to the community’s lack of awareness, many family physicians themselves consider the family physician program to be far less important than clinical specialties. This is simply incorrect. Family physicians and their teams in PHC provide the most cost-effective services bringing and the most benefits to society; they are strongly supported by international health organizations and academic institutions. This sub-theme has been inferred from statements like: Family physicians consider their duties as less important than clinical specialists and they are not aware of the important role of family physicians in creating a healthy society. Living in the countryside exacerbates this feeling” (MPH student, 32, man).

Weak intra/extra-collaboration with family physician team

Another aspect of cultivation is related to weak intra/extra-collaboration with family physicians. Solving many of the problems faced by communities through the family physician program requires the close cooperation of many institutions outside the health sector. Unfortunately, this is often difficult and is not achieved in many cases: “In many cases, in order to solve people’s health problems, we need the assistance of governorates and local authorities, water and wastewater organizations, road maintenance agencies, etc. And in some cases, this cooperation is not achieved because

external organizations are not justified with the importance of health issues or they claim to have a shortage in resources” (MPH instructor, 40, man).

Payment

Low PHC budget

Another challenge facing the implementation of P4P is the inadequacy of the PHC budget, which dramatically discourages qualified individuals from participating in the family physician program. Additionally, the inadequacy of financial resources reduces the feasibility of PHC-related interventions. Participants explained that: “The allocated fund to the PHC sector is much lower than that of hospital care and treatment affairs, which is in conflict with standard managerial and economic principles. This inadequate budget, which reflects the poor attitude of managers and policymakers of the MOHME and the lack of attention paid by health insurance system to the priority of prevention, will weaken the performance indicators of the health system, especially in the area of justice and access” (PHC manager, 47, man).

Lack of proper infrastructure for instituting P4P

The shortcomings of current management systems constitute one of the main challenges. Participants believe that the country’s current health system is excessively weak and faces serious problems in its daily operations. Moreover, the system is often unsuccessful in the implementation of new programs. They believe that the hardware and software facilities necessary for implementing such a system are not yet available. This theme is drawn from the following statements: “One of the main obstacles to the implementation of this program is the weak management of health care systems. We always run the world’s successful plans in the wrong way because we do not have enough studies on them. Moreover, we have poor localization in the implementation phase” (PHC manager, 62, man); “There is not a well-defined system for creating a P4P system because payments are often individual-based and in the form of salary. The managers responsible for payment systems of family physicians still have a hospital and treatment-based view” (MPH instructor, 41, man).

Assessment and monitoring

Lack of criteria and scientific tools for qualitative assessment of the program

The shortcomings of monitoring systems constitute a major problem for P4P implementation. According to the participants, there are copious shortcomings. They believe that the monitoring is not done objectively or scientifically; there is no suitable tool for regular monitoring. This theme stems from statements like: “Monitoring is not objective and documented, and personal tastes are applied to them” (MPH student, 35, man); “The performance of the family physician is not consistent with the monitoring questions. Moreover, monitoring is not based on actual performance” (MPH instructor, 38, man); “There are currently no clear criteria for measuring performance, and there is no direct relationship between good performance indicators and the amount of physician’s effort. Also, the conditions of different work environments are not considered in the monitoring process” (MPH student, 38, woman).

Lack of a well-defined mechanism for assessment

Another serious obstacle is the assessment system’s lack of appropriate mechanisms and processes. The current mechanisms are entirely state-owned; this is problematic, as a governmental mechanism is affected by political pressure and the consistency of the program will be hindered by changes in the government. Participants believe that: “An appropriate accreditation program needs to be defined and implemented for PHC. Afterward, many problems will be resolved” (MPH instructor, 56, man); “Monitoring family physician program is not done objectively and scientifically, but it is mainly fulfilled in a subjective manner. While assessing, the assessors are not very accurate and they do not get the needed scientific documents from the participants” (MPH instructor, 45, man); “If performance monitoring is done with an educational approach to help family physician team with their duties and enhance their skills, it can be useful. But the current system takes a top-down and detective-like view” (PHC manager, 42, woman).

Lack of appropriate assessors

Another major barrier is the lack of appropriate assessors for objective and scientific evaluation of the family physician program. The Iranian health care system, especially in PHC, lacks

trained and experienced assessors. Resultantly, the assessors lack the knowledge, attitude, and performance necessary to properly conduct an accurate evaluation. Hence, we cannot expect their evaluations to improve performance. In this regard, a participant believed that: "Assessors are usually selected from the people who have not been involved in the health system, and in particular the family physician program. So, they have no administrative experience in this area, which leads to inappropriate assessments and lack of motivation in family physicians" (MPH student, 41, man).

Information management

Poor information infrastructure

Poor information infrastructure in the Iranian health care system, especially in the family physician program, is another major deficiency preventing the successful implementation of P4P. Neither the software nor the hardware infrastructure is sufficient for proper monitoring and evaluation, as most rural areas do not have reliable internet access or appropriate computer systems. Several statements exemplify this theme well: "We still do not have access to the internet and computer systems in many rural areas. The internet connection is also very slow and we have constant internet interruptions. Most of the health personnel in the villages are not skillful in using the internet and other related programs. Even family physicians themselves are unfamiliar with the existing programs" (MPH instructor, 50, man); "It is necessary to define and set up health records accurately based on health system's information requirements, especially family physician program, in both paper and electronic forms" (MPH student, 29, man).

Poor systemic management of production cycle and information flow

Collecting appropriate and timely data on the health status of people in the community and system performance is essential, as this data serves as the basis for management decisions. Accordingly, data collection, analysis, and presentation play a crucial role in the continual improvement of health care systems. Participants believe that: "Physicians and their health teams should receive the needed trainings on how to complete medical records and collect statistics related to their performance. It is also necessary to monitor the accurate and timely completion of medical records" (PHC manager, 55, man); "The family physician team must accurately identify

its information needs with the participation of all other parties, and have access to them. Unnecessary information should not be generated in the system and the information systems should cover all needs" (MPH instructor, 45, man).

Level of authority

Insufficient authority of family physicians

A fundamental factor in achieving a desirable job performance is a healthy balance between responsibility and authority. If they are tasked with many responsibilities, employers should also have sufficient authority, especially when managing their subordinates. Family physicians lack the authority necessary to influence their team or establish proper intra/extra-relationships. Participants believed that: "Family physicians lack adequate control and influence over their team and social workers, because they have little role in selecting, recruiting, or changing them" (MPH student, 30, woman); "Payments to health personnel are defined by the system and family physicians cannot change them. This is not logical because while the activities and performance of the health team members influence the performance of family physicians and their assessment scores, they do not have sufficient managing power over their subordinate groups" (MPH student, 42, man).

Discussion

This study was designed and conducted to identify the challenges in the implementation of a P4P system in Iran's family physician program. This study identified 7 themes, 14 sub-themes, and 46 codes related to these challenges. The main themes were found to be: workload, training, program cultivation, payment, assessment and monitoring, information management, and level of authority. Other sub-themes were also identified.

Kahn et al. (2010) investigated some challenges regarding the lack of tools and measures for assessing P4P. They suggested the following solutions to overcome the challenges: take appropriate measurements for the evaluation of high-quality performance; involve all parties in all stages of design, implementation, and evaluation; conduct appropriate research, especially for economic assessments.²¹ These results are similar to those of this study because both of them emphasize the need to develop an appropriate evaluation tool that covers all aspects of evaluation, including context, input, process, output, outcome, and impact. Additionally, both of these studies purport

that P4P could improve the quality of provided care and health outcomes.

Schatz et al. (2007) identified the following barriers to the implementation of P4P: outcome variance stemming from disease severity and problems associated with the alignment of patients during measurement and evaluation, immoral patient selection based on likelihood of improvement, lack of motivation, frustration, and low professional independence of physicians. They concluded that strengthening the management system and using scientific management rather than traditional management would help to overcome these obstacles.³² They also emphasized the clinical condition of the covered population and its effect on health outcomes and, in turn, on payment in P4P. This study suggests that the clinical condition of the population must be considered when determining payment. This major payment determinant was not mentioned by the participants of this study. This could be due to contextual differences; the study by Schatz et al. is about acute care while this study is about PHC. The last recommendation of Schatz et al. regarding the importance and necessity of good management in P4P is generally similar to the results of this study.

A study by Hart-Hester et al. (2008) raised significant challenges regarding the quality of health services provided by physicians with credible evidence and clinical guidelines. They proposed the following solutions: strengthen health-information systems by designating an electronic health record and inserting all clinical cases into it, strengthen the documentation of provided health services, use electronic tools to support clinical and management issues, and exchange health information among different specialists.² They pointed to the lack of a well-designed information-management system as the main challenge facing the implementation of a P4P system. These results are similar to those of this study, and both studies emphasize the importance of clinical- and information-management systems.

Brush et al. (2006) pointed out a number of challenges regarding the successful implementation of P4P. Some of their suggested solutions were as follows: compile quality measures based on valid evidence; create structures and tools to improve quality; recognize improvements in processes and outcomes; allocate adequate funds and incentives to improve physician performance; emphasize the role of data collection, data analysis, and the use of clinical and management data; define

functional goals based on a common consensus; establish objective and transparent means and rating health care providers; conduct research on related issues.¹² Their results are notably similar to ours; they emphasize the need to develop valid and evidence-based evaluation tools, design proper information-management systems, and define a suitable and effective evaluation process.

Of course, it is important to note that all of these mentioned studies were conducted in developed countries with high-quality PHC systems. They have all considered just two themes: assessment and monitoring mechanisms (Kahn 2010; Brush 2008) and information-management systems (Hart-Hester 2008; Schatz 2007); neither evaluated any of the other five themes. This indicates that there are challenges facing the implementation of P4P in Iran that do not exist in developed countries.

Many researchers have made suggestions and developed interventions to resolve the identified challenges, which will be discussed here.

In order to reduce the workload of family physicians, it is necessary to clearly define their responsibilities and assign a reasonable range of tasks based on appraisal, time, and epidemiologic and demographic characteristics of the covered population. The number of people under the supervision of each family physician should be carefully determined. Payment should be per capita, or based on the: number of people covered; patient characteristics and needs; quality of delivered services as determined by clinical protocols and guidelines; obtained health outcomes; caregiver satisfaction.³³

Providing family physicians with training on management skills as well as preventive and social medicine at university would improve their abilities and boost public confidence in them. Other training courses based on job description and public need, either before service or during service, should accompany this academic training. These training courses could be provided as formal and compulsory sessions, as is the case with other clinical specialties. The content of these courses should be determined by precise scientific studies. In order to cultivate the family physician program, extensive training on the importance and role of family physicians in community health should be provided to the public, intra/extra-sectorial organizations, and family physicians themselves.³⁴ In particular, the importance of the referral system must be explained to the public. In order to improve the payment system for family

physicians, payment mechanisms with a special look at PHC needs should be considered.³⁵

The requirements and infrastructure of P4P should be provided through experienced managers. Evaluation, assessment, and monitoring systems can be improved through the development of appropriate evaluation tools and the design of a proper evaluation process based on process-mapping rules and suitable mechanisms for assessors. Information systems should be reformed through the development of an electronic health record and a scientific mechanism for the preparation and publication of targeted and timely clinical and management information.^{36–38} Additionally, granting a reasonable level of authority to family physicians over their teams and establishing intra/extra-relationships would seriously enhance the feasibility of this program. It is necessary to identify the main organizational stakeholders, involve them in the program, and gather their views at all stages of development.³⁹

The main limitation of this study was the difficulty in managing interviews with the participants. The researchers resolved this problem by making appointments with the participants and encouraging them to participate.

Conclusion

There are numerous challenges in the implementation of P4P in the Iranian family physician program. First and foremost is the absence of a systematic post-graduate training

program for medical doctors interested in family medicine. Second, the range of duties for family physicians is both wide and unclear. This study also uncovered the absence of a robust evaluation-reward system or information-management system and the lack of family physician authority over their staff. Based on our findings and discussion, we made suggestions for strengthening the family physician program, especially in terms of training, evaluation, funding, and information management. Hopefully, the results of this study will be helpful for managers and policymakers in removing the identified barriers and improving the quality of the Iranian family physician program. The results of this study may also be useful for other countries attempting to implement P4P in their family physician programs.

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Conflicts of interest

The authors declare that there is no conflict of interest.

References

1. Berven S, Smith A, Bozic K, Bradford D. Pay-for-Performance: Considerations in application to the management of spinal disorder. *The Spine Journal*. 2007;32: 33–38.
2. Hart-Hester S, Jones W, Watzlaf VJM, Fenton SH, Nielsen C, Madison M, et al. Impact of creating a pay for quality improvement (P4QI) incentive program on healthcare disparity: leveraging HIT in rural hospitals and small physician offices. *Perspectives in Health Information Management*. 2008;5(14): 1–14.
3. Hood RG. Pay-for-performance; financial health disparities and the impact on health care disparities. *Journal of the National Medical Association*. 2007;99(8): 953–958.
4. Bokhour B, Burgess J, Hook J, White B, Berlowitz D, Guldin M, et al. Incentive implementation in physician practices: a qualitative study of practice executive perspectives on pay for performance. *Medical Care Research and Review*. 63(1): 73–95.
5. Rose J. Industry influence in the creation of pay-for-performance. *Quality Management in Health Care*. 2008;17(1): 27–34.
6. McGlynn EA, Asch SM, Adams J, Keesey J, Hicks J, DeCristofaro A, et al. The quality of health care delivered to adults in the United States. *New England Journal of Medicine*. 2003;348: 2635–2645.
7. Rosenthal MB, Frank RG. What is the empirical basis for paying for quality in health care? *Medical Care Research and Review*. 2006;63: 135–57.
8. Doran T, Fullwood C. Pay for performance: is it the best way to improve control of hypertension? *Current Hypertension Reports*. 2007;9: 360–367.
9. Cabana MD, Rand CS, Powe NR, AW W, Wilson MH, Abboud PA, et al. Why don't physicians follow clinical practice guidelines? A framework for improvement. *Journal of the American Medical Association*. 1999;282: 1458–1465.
10. Institute of Medicine. *Crossing the Quality Chasm: A New Health System for the 21st Century*. Washington: National Academy Press; 2001.
11. Fisher ES, Wennberg DE, Stukel TA, Gottlieb DJ, Lucas FL, Pinder EL. The implications of regional variations in Medicare spending. Part I: the content, quality, and accessibility of care. *Annals of Internal Med*. 2003;138: 273–287.

12. Brush JE, Krumholz HM, Wright JS. American College of Cardiology 2006 principles to guide physician pay-for-performance programs. *Journal of the American College of Cardiology*. 2006;48(12): 1–8.
13. Rosenthal MB. Pay for performance and beyond. *Expert Review of Pharmacoeconomics and Outcomes Research*. 2007;7(1): 351–355.
14. Sikka R. Pay for performance in emergency medicine. *Annals of Emergency Medicine*. 49(6): 756–761.
15. Epstein AM. Pay for performance at the tipping point. *N Engl J Med*. 2007;356:515–7.
16. Campbell S, Reeves D, Kontopantelis E, Middleton E, Sibbald B, Roland M. Quality of primary care in England with the introduction of pay for performance. *New England Journal of Medicine*. 2007;357: 181–190.
17. Scott IA. Pay for performance in health care: strategic issues for Australian experiments. *Medical Journal of Australia*. 2007;187: 31–35.
18. Millett C, Gray J, Saxena S, Netuveli G, Majeed A. Impact of a pay-for-performance incentive on support for smoking cessation and on smoking prevalence among people with diabetes. *Canadian Medical Association Journal*. 2007;176: 1705–1710.
19. Snyder L, Neubauer RL. Pay-for-performance principles that promote patient-centered care: an ethics manifesto. *Annals of Internal Medicine*. 2007;147: 792–794.
20. Hayward RA, Kent DM. 6 EZ steps to improving your performance: (or how to make P4P pay 4U!). *Journal of the American Medical Association*. 2008;300:255–256.
21. Kahn JM, Scales DC, Au DH, Carson SC, Curtis JR, Dudley RA, et al. An official American thoracic society policy statement: pay-for-performance in pulmonary, critical care, and sleep medicine. *American Journal of Respiratory and Critical Care Medicine*. 2010;181: 1–8.
22. Werner RM, Konetzka RT, Kruse GB. Impact of public reporting on unreported quality of care. *Health Services Research*. 2009;44: 379–398.
23. Roski J, Jeddeloh R, An L, Lando H, Hannan P, Hall C, et al. The impact of financial incentives and a patient registry on preventive care quality: increasing provider adherence to evidence-based smoking cessation practice guidelines. *Preventive Medicine*. 2003;36: 291–299.
24. Welker JA, Huston M, McCue JD. Antibiotic timing and errors in diagnosing pneumonia. *Archives of Internal Medicine*. 2008;168: 351–356.
25. Drake DE, Cohen A, Cohn J. National hospital antibiotic timing measures for pneumonia and antibiotic overuse. *Quality Management in Health Care*. 2007;16: 113–122.
26. Fiscella K, Franks P, Gold MR, Clancy CM. Inequality in quality: addressing socioeconomic, racial, and ethnic disparities in health care. *Journal of the American Medical Association*. 2000;283(19): 2579–2584.
27. Chien AT, Chin MH, Davis AM, Casalino LP. Pay for performance, public reporting, and racial disparities in health care: how are programs being designed? *Medical Care Research and Review*. 2007;64: 283S–304S.
28. Kirschner K, Braspenning J, Jacobs JEA, Kirschner RG. Design choices made by target users for a pay for performance program in primary care: an action research approach. *BMC Family Practice*. 2012;13(25): 1–11.
29. Graneheim UH, Lundman B. Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurse Education Today*. 2004;24(2): 105–12.
30. Lacey A, Luff D. Qualitative Data Analysis. London: The NIHR RDS for the East Midlands / Yorkshire & the Humber; 2009.
31. Zamanzadeh V, Valizadeh L, Lotfi M, Salehi F. Burn survivors' experience of core outcomes during return to life: a qualitative study. *Journal of Caring Sciences*. 2014;3(4): 227–237.
32. Schatz M, Blaiss M, Green G, Aaronson D. Pay for performance for the allergist-immunologist: potential promise and problems. *American Academy of Allergy, Asthma & Immunology*. 2007 120: 769–775.
33. Timony PE, Gauthier AP, Sanou B, Wenghofer EF. Workload of French-speaking family physicians in francophone rural and northern communities in Ontario. *Canadian Journal of Rural Medicine*. 2018;23(3): 76–84.
34. Tabrizi JS, Gharibi F. Primary healthcare accreditation standards: a systematic review. *International Journal of Healthcare Quality Assurance*. 2019;32(2): 310–320.
35. Esmaeili R, Hadian M, Rashidian A, Shariati M, Ghaderi H. Family medicine in Iran: facing the health system challenges. *Global Journal of Health Science*. 2014;7(3): 260–266.
36. Iezadi S, Tabrizi JS, Ghiasi A, Farahbakhsh M, Gholipour K. Improvement of the quality payment program by improving data reporting process: an action research. *BMC Health Services Research*. 2018;18(1):692–697.
37. Kondo KK, Wyse J, Mendelson A, Beard G, Freeman M, Low A, et al. Pay-for-performance and veteran care in the VHA and the community: a systematic review. *Journal of General Internal Medicine*. 2018;33(7): 1155–1166.
38. Gharibi F, Tabrizi JS. Development of an accreditation model for health education and promotion program in the Iranian primary healthcare system: a Delphi study. *Health Promotion Perspectives*. 2018;8(2): 155–162.
39. Behzadifar M, Behzadifar M, Heidarvand S, Gori HA, Aryankhesal A, Taheri Moghadam S, et al. The challenges of family physician policy in Iran: a systematic review and meta-synthesis of qualitative researches. *Family Practice*. 2018;35(6): 652–660.

Chronic non-healing ulcers as presenting sign of acquired immunodeficiency syndrome

Nasiri S, Barat T, Bidari- Zerehpooch F, Mozafari N

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Authors:

Nikoo Mozafari

(Corresponding author)

MD

Skin Research Center, Shahid Beheshti
University of Medical Sciences
Tehran, Iran

Department of Dermatology, Loghman
Hakim Hospital, Shahid Beheshti
University of Medical Sciences,
Tehran, Iran
Email: nikoo_md@yahoo.com

Soheila Nasiri

MD

Skin Research Center, Shahid Beheshti
University of Medical Sciences
Tehran, Iran

Tara Barat

MD

Skin Research Center, Shahid Beheshti
University of Medical Sciences
Tehran, Iran

Farahnaz Bidari- Zerehpooch

MD

Department of Pathology, Loghman
Teaching Center, School of Medicine
Shahid Beheshti University of Medical
Sciences, Tehran, Iran

Abstract

Atypical forms of herpes simplex virus (HSV) infections, which indicate severe impairment of cellular immunity can be challenging to diagnose. In this paper, we report the case of an atypical HSV infection presenting as chronic nonhealing wounds, which are the first sign of HIV, in a 50-year-old female patient. The lesions had emerged as two large, chronic, and painful ulcerations on the left buttock and labia major 8 months prior. The skin biopsy revealed multinucleated keratinocytes with ground glass nuclei and intranuclear Cowdry type A viral inclusions. A serologic test for HIV-1 was positive. Her CD4⁺ T-cell count was 42/mm³. Clinicians should be familiar with the dermatologic manifestations of HIV, as they are occasionally key to correctly suspecting an underlying HIV infection, allowing for early diagnosis and treatment.

Introduction

Herpes simplex virus (HSV) is a common and well-known viral skin disease. In cases of a typical lesion, the diagnosis is straightforward. However, atypical clinical manifestations of HSV can arise in immunocompromised patients presenting as chronic mucocutaneous ulcers and persist for more than a month.¹ Such an unusual presentation should lead the clinician to suspect immunodeficiency, acquired immune deficiency syndrome (AIDS) in particular.²

Since its 1981 discovery in the United States, AIDS has become an international crisis. The defective cellular immunity associated with AIDS puts infected persons at risk for a variety of opportunistic infections.² Individuals with HIV, including the one being discussed in this paper, often have more than one infection.¹ In an immunocompromised host, mucocutaneous lesions caused by infectious agents can be extensive, appear at unusual sites, and manifest atypically.¹

Here, we present the case of a 50-year old female who presented with chronic non-healing HSV ulcers on her genitalia and buttock as well as tinea corporis on her trunk, which led to an HIV diagnosis.

Case presentation

A 50-year-old woman came to our dermatology clinic with an 8-month history of persistent, non-healing vulvar ulceration. The initial lesion was a tender papule that gradually

progressed to form a large, painful ulcer. Three months after the emergence of the original lesion, she developed a similar ulcer on her left buttock which, similarly, grew worse through enlargement and increasing tenderness. A physical examination revealed swollen genitalia with superficial tender erosions on the patient's right labia major (Fig 1). On her left buttock was a shallow, well-demarcated erythematous ulcer 3–4 centimeters wide (Fig 2). Furthermore, a few erythematous annular plaques (Fig 3), which the patient was unaware of, were found in the left axillary region. The clinical differential diagnosis we considered for ulcers included skin malignancies, pyoderma gangrenosum, tertiary syphilis (gumma), deep mycosis, Crohn's disease, and chronic HSV and CMV infections.



Fig. 1 (Left): Labia major superficial ulcer with a scalloped border

Fig. 2 (Top Right): Shallow, well-demarcated erythematous ulcer on the left buttock

Fig 3 (Bottom Right): Annular erythematous plaques with elevated borders on the axilla

The histopathological findings of punch biopsies taken from the patient's buttock and labia major lesions indicated that they were epithelial ulcer with severe infiltration of inflammatory cells, multinucleated giant cells with a ground glass appearance, and keratinocytes with slate-gray nuclei and intranuclear Cowdry type A viral inclusions, which are compatible with herpetic infection as well (**Fig 4,a,b**). Another skin biopsy taken from the patient's axillary lesions showed pseudoepitheliomatous hyperplasia with fungal hyphae in the stratum corneum layer, compatible with dermatophytosis (**Fig 5a,b**).

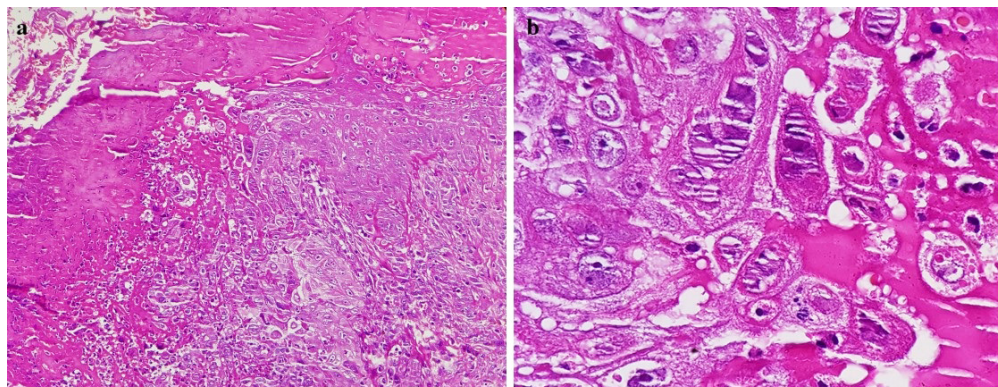


Fig. 4: Histologic examination showing ulcerated epidermis; keratinocytes with slate-gray nuclei and margination of chromatin containing eosinophilic intranuclear inclusion bodies surrounded by an artifact cleft (Cowdry type A inclusion). H&E (a):x20, (b):x100.

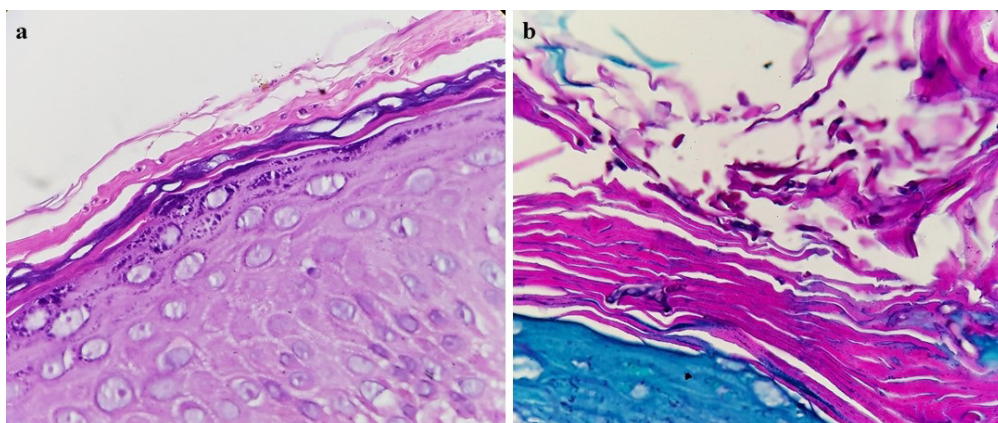


Fig. 5: Slightly hyperkeratotic stratum corneum containing numerous hyphae. (H&E stain x40) (a); fungi are made more apparent by PAS (periodic acid Schiff) stain x100 (b).

Given the unusual presentation of an HSV infection and the rarity of two concomitant skin infections in immunocompetent patients, we suspected an underlying HIV infection. ELISA testing revealed positive antibodies to HIV-1; the Western blot test confirmed the ELISA finding. The patient's total white blood cell count was $2.5 \times 10^3/\mu\text{L}$ (with CD4^+ cell count $42/\text{mm}^3$). The patient was treated with acyclovir 400 mg orally five times per day and terbinafine 250 mg once per day, which led to the complete resolution of the ulcers and annular plaques within two weeks. The patient was put on chronic daily suppressive therapy of acyclovir 400 mg twice per day for at least one year. The patient's husband was an IV drug user. He was tested for HIV-1 and HSV-2 infections and was found to be positive for both. After being counseled about HIV care

and treatment, he agreed to be referred with his wife to an infectious disease specialist for further evaluation.

Discussion:

HSV is the most common sexually transmitted disease in patients infected with HIV. In relatively immunocompetent HIV-infected individuals, mucocutaneous HSV infections present in the typical fashion and severity with a recurrence rate similar to that of the general population.¹

Lesions may appear as painful, grouped, and often umbilicated vesicles on an erythematous base; these may evolve into pustules, erosions, and/or ulcers with characteristic scalloped borders.²

However, in severely immunosuppressed HIV-infected persons, lesions may appear at a higher recurrence rate² as chronic, non-healing, and deep ulcers that favor the perianal region, genitalia, and tongue.

Several case reports of herpes simplex lesions in patients with AIDS described atypical presentations consisting of nodular, tumoral, or verrucous growth clinically simulating malignancies.³

Orofacial lesions associated with nasogastric tube use⁴, periungual infections (herpetic whitlow)⁵, and chronic herpetic foot ulcers⁶ have been reported as presenting features of HIV infection.

Correlation has been found between certain HIV-associated cutaneous disorders and CD4⁺ cell count.⁷ For instance, large non-healing mucocutaneous HSV infections often emerge alongside CD4⁺ cell counts under 50/mm³, as shown in our case.

The diagnosis of herpes in immunocompromised patients can be difficult.⁸ Scrapings of the ulcer edge for Tzank smear has low sensitivity in HIV-infected patients. Multiple Tzank smears, a direct fluorescence assay (DFA), HSV Polymerase chain reaction (PCR), or a viral culture may be required for confirmation. If these are negative, a skin biopsy from the edge of the ulcer should be performed. The characteristic histopathologic features of an HSV infection, would be an intraepidermal vesicle with ballooning degeneration of keratinocytes and multinucleated giant cells.⁸

The first-line agent for treatment of HSV infections in HIV-infected patients is Acyclovir (400 mg Po 5 times a day)⁹. In HIV-infected patients, antiviral therapy should be extended until clinical resolution is evident. When patients fail to respond to this medication, alternative drugs (e.g, Foscarnet and Cidofovir) should be prescribed.⁹

Long-term anti-HSV suppressive therapy should be considered in HIV-1-infected persons, as symptomatic recurrent HSV-2 infection can be more severe and asymptomatic shedding can be more frequent in these patients.¹⁰ Treatment strategies for recurrent disease in HIV-infected persons include chronic suppressive therapy or episodic therapy based on frequency of recurrence and severity of outbreaks. According to 2015 guidelines on sexually transmitted

disease¹¹, patients with recurrent genital herpes, especially those with frequent, painful, or prolonged recurrences, should receive one of the following daily suppressive therapies: acyclovir 400–800 mg two to three times daily; famciclovir 500 mg two times daily; valacyclovir 500 mg two times daily. For episodic therapy, the CDC¹¹ recommends one of the following regimens for a minimum duration of five to ten days or until the lesion has healed: acyclovir 400 mg three times daily; famciclovir 500 mg two times daily; valacyclovir 1g two times daily. Once the patient's CD4⁺ count is ≥ 200 cells/mm, the ongoing need for suppressive antiviral therapy is evaluated annually based on disease activity.¹⁰ It is worth noting that, based on current evidence, suppressive anti-HSV therapy in persons with HIV does not reduce the risk for either HIV transmission or HSV-2 transmission to susceptible sex partners.¹²

HIV-infected patients often have mixed infections on account of their immunocompromised state. In our patient, the combined infection of HSV and dermatophyte suggested immunosuppression and an HIV infection.

Dermatophyte infections are common in HIV-infected individuals.¹³ Kaviarasan et al in a study on the prevalence and clinical variations in dermatophytosis in 185 HIV-infected patients found that more than 20% of all cases had a variant of dermatophytosis and *Tinea corporis* was the most common clinical form.¹⁴ In HIV-infected patients, dermatophytosis may manifest with unusual, multiple, or widespread lesions and may occur with less common etiologic agents.¹⁴ Moreover, a noteworthy incidence of anergic skin lesions with little to no inflammation can be seen in HIV-infected individuals.¹³

Conclusion

This report emphasizes clinical indicators suggestive of underlying immunodeficiency, particularly AIDS. Atypical clinical manifestations of HSV may arise in immunocompromised patients as chronic mucocutaneous ulcers that persist for more than one month. Such an unusual presentation should lead clinicians to suspect HIV.

Since skin diseases brought about by opportunistic infections are common in HIV-infected patients, and these lesions can be the first manifestation of underlying immunodeficiency, clinicians must be aware of the characteristics and presentations of HIV in order to make early diagnoses and avoid delays in therapy initiation.

How does this paper make a difference to general practice?

- Atypical forms of herpes simplex virus (HSV) infections can be challenging to diagnose.
- Lesions may appear as chronic, non-healing, deep, nodular, tumoral, or verrucous ulcers.
- Unusual presentation should lead clinicians to suspect immunodeficiency, AIDS in particular.
- Clinicians must be familiar with HIV's dermatologic manifestations, as they can be key to suspecting an underlying HIV infection.
- The first-line agent for treatment of HSV infections in HIV-infected patients is acyclovir (400 mg /5 times per day).

References

- Garman ME, Tying SK. The cutaneous manifestations of HIV infection. *Dermatologic clinics*. 2002;20(2):193-208.
- Lingappa JR, Celum C. Clinical and therapeutic issues for herpes simplex virus-2 and HIV co-infection. *Drugs*. 2007;67(2):155-74.
- Mosunjac M, Park J, Wang W, Tadros T, Siddiqui M, Bagirov M, et al. Genital and perianal herpes simplex simulating neoplasia in patients with AIDS. *AIDS patient care and STDs*. 2009;23(3):153-8.
- Genereau T, Lortholary O, Bouchaud O, Lacassin F, Vinceneux P, De Truchis P, et al. Herpes simplex esophagitis in patients with AIDS: report of 34 cases. The Cooperative Study Group on Herpetic Esophagitis in HIV Infection. *Clinical infectious diseases : an official publication of the Infectious Diseases Society of America*. 1996;22(6):926-31.
- El Hachem M, Bernardi S, Giraldi L, Diociaiuti A, Palma P, Castelli-Gattinara G. Herpetic whitlow as a harbinger of pediatric HIV-1 infection. *Pediatric dermatology*. 2005;22(2):119-21.
- Deluca J, Perino F, Maier J, Kluge R, Mazzoleni G, Eisendle K. Vegetating foot ulcer as presenting sign of acquired immunodeficiency syndrome. *Journal der Deutschen Dermatologischen Gesellschaft = Journal of the German Society of Dermatology : JDDG*. 2014;12(3):265-6, -7.
- Raju PV, Rao GR, Ramani TV, Vandana S. Skin disease: clinical indicator of immune status in human immunodeficiency virus (HIV) infection. *International journal of dermatology*. 2005;44(8):646-9.
- Langtry JA, Ostlere LS, Hawkins DA, Staughton RC. The difficulty in diagnosis of cutaneous herpes simplex virus infection in patients with AIDS. *Clinical and experimental dermatology*. 1994;19(3):224-6.
- Barde C, Piguet V, Pechere M, Masouye I, Saurat JH, Wunderli W, et al. Management of resistant mucocutaneous herpes simplex infections in AIDS patients: a clinical and virological challenge. *HIV medicine*. 2011;12(6):367-73.
- Johnston C, Wald A. Treatment of genital herpes simplex virus type 2 in HIV-infected patients [updated 10 February, 2020. Available from: <https://www.uptodate.com/contents/treatment-of-genital-herpes-simplex-virus-type-2-in-hiv-infected-patients>.
- Workowski KA, Bolan GA. Sexually transmitted diseases treatment guidelines, 2015. *MMWR Recommendations and reports: Morbidity and mortality weekly report Recommendations and reports*. 2015;64(RR-03):1.
- Smith C, Pogany L, Auguste U, Steben M, Lau T. Update on STIs: Does suppressive antiviral therapy for herpes simplex virus prevent transmission in an HIV-positive population? A systematic review. *Canada Communicable Disease Report*. 2016;42(2):37.
- Costa JE, Neves RP, Delgado MM, Lima-Neto RG, Morais VM, Coelho MR. Dermatophytosis in patients with human immunodeficiency virus infection: clinical aspects and etiologic agents. *Acta tropica*. 2015;150:111-5.
- Kavirasan PK, Jaisankar TJ, Thappa DM, Sujatha S. Clinical variations in dermatophytosis in HIV infected patients. *Indian journal of dermatology, venereology and leprology*. 2002;68(4):213-6.

Maternal obesity and its determinants: A neglected issue?

Nurul- Farehah S, Rohana AJ

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Authors:

Rohana Abdul Jalil

(Corresponding author)

Ph.D

Department of Community Medicine

Universiti Sains Malaysia, Malaysia

Email: rohanajalil@usm.my

Nurul Farehah Shahrir

MBBS, MPH

Department of Community Medicine

Universiti Sains Malaysia, Malaysia

Abstract

Maternal obesity is a global public health concern that affects every aspect of maternity care. It affects the short-term and long-term health of the mother and her offspring. Obese pregnant mothers are at an increased risk of developing complications during antenatal, intrapartum, and postnatal periods. Maternal complications include gestational diabetes mellitus, hypertensive disorder in pregnancy, pre-eclampsia and eclampsia, increased rate of cesarean delivery, pulmonary embolism, and maternal mortality; fetal complications include congenital malformation, stillbirth, and macrosomia. Moreover, both mother and infant are at an increased risk of developing subsequent non-communicable diseases and cardiovascular problems later in life. Several factors are associated with the likelihood of maternal obesity, including sociodemographic characteristics, obstetric characteristics, knowledge, and perception of health-promoting behavior. Gaining a sound understanding of these factors is vital to reaching the targets of Sustainable Developmental Goal 3—to reduce global maternal mortality and end preventable deaths of children under 5 years of age—by 2030. It is essential to identify pregnant women who are at risk of maternal obesity in order to plan and implement effective and timely interventions for optimal pregnancy outcomes. Importantly, maternal obesity as a significant pregnancy risk factor is largely modifiable.

Introduction

Around the world, obesity has become a significant public health concern, and the prevalence of obesity is growing at an alarming rate.¹ This epidemic presents a substantial challenge for non-communicable disease (NCD) prevention.² Obesity is a complex, multifactorial disease involving interaction between genetic, hormonal, behavioral, socioeconomic, and environmental conditions; if not controlled, it can result in devastating morbidity and mortality.³ Many countries have observed a rise in obesity following urbanization and industrialization over the last 30 years—some have seen obesity rates quadruple.⁴

Globally, obesity has nearly tripled since 1975. In 2016, the World Health Organization reported that more than 1.9 billion adults were overweight; among them, more than one-third were obese.

While the rate of obesity appears to be higher for women than for men⁵, obesity is increasing in both developed and developing countries. However, in developed countries, the peak prevalence is trending toward younger people.⁶

In Malaysia, the prevalence of obesity rose by 4.4% between 2006 and 2015, from 26.2%

to 30.6%.⁷ In fact, the country has the highest prevalence of obesity in Southeast Asia (SEA). According to the 2015 National Health Morbidity Survey, the prevalence of obesity among adults was 30.6%. The rate was higher for women (33.6%) than for men (27.8%), and higher for married women (33.8%) than for single women (22.7%). Moreover, those who are ethnically Indian were found to have the highest prevalence of obesity (43.5%). The highest rate was seen in the state of Wilayah Persekutuan Putrajaya (43%).⁷

The purpose of this narrative review is to identify modifiable and non-modifiable risk factors for maternal obesity that are considered relevant in the development of guidelines concerning maternal obesity as well as interventions aimed at preventing obesity among pregnant women in Malaysia.

Methods

Through a narrative review, we examined the factors associated with maternal obesity. We discussed results from recent studies and the potential underlying mechanisms of the observed associations. The search keywords used include maternal obesity, obesity in pregnancy, determinants, modifiable associated factors, and non-modifiable associated factors.

Defining obesity in pregnancy

Obesity is defined as a condition characterized by excessive accumulation of fat in adipose tissue that may cause impairment to health.⁸ One of the most commonly used indices of relative weight is the Body Mass Index (BMI), or body weight in kilograms divided by height in meters squared. The World Health Organization defines obesity as BMI ≥ 30 kg/m². However, Asian populations often see a lower BMI cut-off point, relative to Caucasian populations, for risk of complications. Few studies have been done on this difference, though some in Hong Kong⁹ and Singapore¹⁰ have shown an increasing prevalence of non-insulin dependent diabetes mellitus (NIDDM) and cardiovascular risk factors at cut-off points below 25 kg/m² for overweight and below 30 kg/m² for obesity.

In obstetric populations, the Royal College of Obstetricians and Gynaecologists defines maternal obesity as BMI ≥ 30 kg/m², similar to the general population. The National Institute for Health and Care Excellence (NICE) antenatal care guidelines recommend that maternal height and weight should be measured for BMI measurement in pregnant women by 10 weeks of gestation. In areas without the necessary equipment, self-reported height and weight can be used.¹¹ However, this method can often garner an overestimated height and an underestimated weight, especially in obese women, resulting in an inaccurate risk assessment for their pregnancy.¹² Regarding BMI cut-off points, a study in Singapore involving 8,843 mothers showed an increase in the prevalence of maternal obesity from 12.2% using the WHO cut-off points to 21.0% using the Asian BMI cut-off points.¹³ However, the prevalence of adverse feto-maternal outcomes associated with maternal obesity did not change with lower BMI cut off points.

Obesity among women in the reproductive age group and pregnant mothers

The most recent national population survey in Malaysia revealed that obesity is more prevalent among women than among men.⁷ A study found that 15.9% of women in the reproductive age group (20 - 49) were obese¹⁴, meaning that these women of childbearing potential (WOCBP) would start pregnancy with maternal obesity.

In 2014, there were approximately 38.9 million overweight and obese pregnant women globally, with a higher rate in both upper- and middle-income countries, including Malaysia.¹⁵ In India, the National Family Health Survey had shown

a rise in the prevalence of maternal obesity over seven years, from 10.6% in 1999 to 14.8% in 2006. In Malaysia, according to the 2016 National Health Morbidity Survey, the prevalence of maternal obesity was 14.6% with the highest rates (69.2%) among those of advanced age (45–49), ethnic Malays (16.8%), and ethnic Indians (15.6%).¹⁶ These findings have important implications for obstetric care, as maternal obesity is associated with a greater length of hospital stay.¹⁷ It has been estimated that maternal obesity accounts for 2.8% of a country's total healthcare expenditure.¹⁸

Maternal and perinatal health

Maternal health refers to the health of women during pregnancy, childbirth, and the postpartum period.¹⁹ According to the WHO, approximately 810 women died each day due to preventable causes related to pregnancy and childbirth in 2017. Globally, the five major direct causes of maternal morbidity and mortality include hemorrhage, infection, high blood pressure, unsafe abortion, and obstructed labor. In Malaysia, according to Confidential Enquiries into Maternal Death, the four leading causes of maternal death between 2009 and 2011 were associated medical conditions (31.2%), obstetric embolism (16.0%), hypertensive disorders in pregnancy (HDP) (15.8%), and postpartum hemorrhage (PPH) (11.6%). Three of these—obstetric embolism, hypertensive disorders in pregnancy, and postpartum hemorrhage—were associated with maternal obesity.^{20,21} Furthermore, a cross-sectional study by the Malaysian National Obstetric Registry of data from 88,837 pregnant women showed that maternal overweight and obesity were associated with stillbirths (OR 1.2; 95% CI 1.0, 1.4), shoulder dystocia (OR 1.9; 95% CI 1.2, 2.9), fetal macrosomia (OR 1.8; 95% CI 1.6, 2.0), and cesarean section (OR 1.9; 95% CI 1.8, 2.0).²²

Determinants of maternal obesity

Sociodemographic and obstetric characteristics

Ramonienè et al.²³, Gaillard et al.²⁴, and Callaway et al.¹⁷ have shown that older and multiparous women are at a higher risk of becoming obese. Similarly, Heslehurst et al.²⁵ found that advanced maternal age and multiparity were associated with maternal obesity after adjusting the confounders. Moreover, a prospective analysis of 2,923 non-obese women conducted by Rebholz et al. showed that the 5-year incidence of obesity was 11.3 per 100 multiparous women compared to

just 4.5 per 100 in nulliparous women.²⁶ However, this finding strayed from that in a study by Boudet-Berquier et al. which found that, among primiparous women, those between 25 and 29 were more likely to be obese than those between 30 and 34.²⁷

Ying Pang et al. found that maternal obesity was far more common among Malay (21.5%) and Indian (17.2%) women than among Chinese (4.6%) women.¹³ In contrast, some studies have shown that maternal obesity is associated with the minority group. In Queensland, Australia, the highest rate of delivery by overweight and obese mothers was found among indigenous women.²⁸ A study by Bahadoer et al. in Rotterdam involving 6,444 pregnant women found similar results, revealing a higher prevalence of maternal obesity among ethnic minority groups.²⁹ These racial/ethnic and nativity inequalities in the risk of maternal obesity were partially explained by living conditions, such as limited access to health-promoting environments like walkways, public transportation, and inexpensive healthy foods.³⁰ Ethnic differences may also be attributed to variance in genetic predisposition and developmental factors.³¹ Interestingly, one study conducted in the UK involving 36,821 pregnant women argued that ethnicity was not associated with maternal obesity. However, the interpretation of this finding is limited since the numbers representing the non-Caucasian populations were relatively low.¹¹

Low socioeconomic status has been linked to maternal obesity. A study in Middleborough, UK, of 36,821 pregnant women over 15 years found that pregnant mothers living in deprived areas (quintiles 1–3) were 2.5 times more likely to be obese than those living in the least deprived area (quintile 5).¹¹ This was consistent with Athukorala et al.³² and Nohr et al.³³, who found that low household income was significantly associated with maternal obesity.

Several studies have shown that a low level of maternal education level was associated with an increased risk of obesity among pregnant women. A study of 54,022 pregnancies in Flanders showed that high pre-pregnancy BMI was significantly more prevalent among women with lower levels of education.³⁴ Moreover, a study of 36,821 women in the UK revealed that a longer schooling period significantly reduced the odds of becoming obese while pregnant.¹¹ Similar findings were seen in a study by Boudet-Berquier et al., in which pregnant women with a higher level of education or in professional

workers were found to be less likely to be obese.²⁷ Other factors associated with high BMI among pregnant women include cigarette smoking and marital status.²⁴ Mkuu found that women who were married or living with a partner had a 73% higher risk of being overweight or obese.³⁵ In contrast, however, one study showed no significant association between marital status and maternal obesity.³⁶

Antenatal and postpartum physical activities

Adiposity is the result of positive energy balance; the decline in energy expenditure is more than the intake of calories, leading to accelerated fat deposition. The Institute of Obstetricians and Gynaecologists of the Royal College of the Physicians of Ireland recommends that pregnant women get at least 30 minutes of daily exercise. Meanwhile, The Royal College of Obstetricians and Gynaecologists (RCOG) recommends that women avoid a sedentary lifestyle and get at least 150 minutes of moderate-intense activity per week throughout pregnancy. Physical activity and exercise have numerous benefits for pregnant women and their fetuses, including the prevention of excessive weight gain and postpartum weight retention.³⁷

Restall et al. reported that women who reduced exercise levels by 14–16 weeks into their pregnancy were 50% more likely to develop excessive gestational weight gain (GWG); women with sedentary behaviors were more likely to gain weight to—based on Institute of Medicine (IOM) recommendations—an unhealthy extent, contributing to maternal obesity.³⁸ Nohr et al. reported that obese mothers were more likely to be physically inactive than normal-weight mothers.³³ A cross-sectional study of 491 overweight and obese women in North Carolina showed that nearly three-quarters of the respondents failed to meet the national physical activity recommendations during the postpartum period; women with a BMI ≥ 40 kg/m² spent more time doing sedentary activities than those with a lower BMI.³⁹

Many pregnant women become more sedentary, especially after reaching their third trimester.⁴⁰ According to Pereira et al., there was a significant reduction of women's physical activity from the antenatal period to six months postpartum; the prevalence of insufficient physical activity (≤ 150 minutes per week of activity) increased from 12.6% prior to pregnancy to 21.7% during the postpartum period.⁴¹ Caregiving duties, long working hours, limited knowledge of performing

physical activities, and concerns over pregnancy complications were among the barriers faced by pregnant women.⁴²

Physical activity makes up 15–30% of total daily energy expenditure, meaning it is the most modifiable component. Involvement in purposeful exercise (both short- and long-term) has been proven to influence resting energy expenditure⁴³, consequently reducing the likelihood of maternal obesity. Furthermore, a randomized control trial among 82 pregnant women found that antenatal physical exercise among overweight pregnant women resulted in less weight gain throughout the entire pregnancy.⁴⁴

Dietary intake during pregnancy and the postpartum period

A low healthy eating index (HEI) has been shown to be associated with overweight and obesity during pregnancy, as well as obesity prior to pregnancy.⁴⁵ There is some evidence to suggest that an overweight or obese woman is more likely to consume a poor-quality diet during pregnancy that worsens throughout the pregnancy and the postpartum period. A prospective observational study of 100 obese pregnant women found excessive intake of energy-dense processed foods with high fat and sugar content,⁴⁶ including crackers, cakes, preserves, confectionery, and savory snacks; creams, ice-cream, and chilled desserts; butter, spreading fats, and oils. Not only do these foods contribute to excessive weight gain during pregnancy and postpartum weight retention, but they also lack the essential micronutrients needed during pregnancy.⁴⁷

Between 50% and 88% of overweight and obese pregnant women have poor compliance with dietary guidelines for the intake of cereals and legumes, carbohydrates, n-3 and n-6 fatty acids, dairy, fruits, and vegetables.^{46,48} A study of 125 multi-ethnic women in the early-postpartum period showed that the dietary intake has low mean scores in fruits, total vegetables, and whole grains but had higher intake of sodium, saturated fats, and discretionary calories.⁴⁹ Oken et al. reported that every 0.5% increase in daily trans-fat intake resulted in a 1.3-times increase in the likelihood of retaining at least 5 kg weight in the postpartum period.⁵⁰ Similar results were found in Australia, where only 7% of respondents met vegetable-consumption recommendation and only 13% met fruit-consumption targets.⁵¹

Intake of excess energy above the requirement

for a growing fetus is stored as part of the anabolic state of pregnancy, increasing body fat composition further with each pregnancy.⁵² There are many barriers to healthy eating behaviors for underprivileged overweight and obese mothers, including low access to transportation, higher costs, low perceived self-efficacy, societal norms on food, low social support, and incorrect beliefs about the safe ways to manage weight gain during pregnancy.^{53,54}

Interpregnancy interval

Birth spacing, pregnancy spacing, or inter-pregnancy interval is the period between delivery of the previous infant and conception of the present pregnancy. Studies are scarce on the relationship between spacing and maternal BMI outcomes. However, short spacing may increase the risk of maternal obesity due to the weight changes during the interval resulting from weight retained from pregnancy or gained postpartum.⁵⁵

A retrospective cohort study of 38,178 women in Canada showed that short interpregnancy intervals were significantly associated with an increased risk of beginning the subsequent pregnancy obese (adjusted OR 1.61, 95% CI 1.05–2.45 for 0–5 months; adjusted OR 1.43, 95% CI 1.10–1.87 for 6–11 months).⁵⁶ However, more research is needed to have a good understanding of the impact of short spacing intervals on maternal obesity.

Pre-pregnancy BMI, excessive weight gain, and postpartum weight retention

Gestational weight gain (GWG) can be defined as the amount of weight gained between conception and before the birth of the infant. In 2009, the Institute of Medicine (IOM) released guidelines and recommendations for GWG based on pre-pregnancy BMI. These recommendations were released due to profound changes in population demographics—far more women were becoming pregnant while overweight or obese.

Pre-pregnancy BMI is the strongest predictor of GWG. A study of 4,619 African American and Caucasian women in Arkansas, US, revealed that overweight women were three times more likely than normal-weight women to exceed the IOM-recommended weight gain while obese women were four times more likely.⁵⁷ In Malaysia, a cross-sectional study of 180 pregnant women in the Batu Pahat district showed that women who were overweight or obese prior to being pregnant were seven times more likely than women with normal pre-pregnancy BMI to have an excessive

GWG rate (OR = 7.44, 95% CI = 2.07-26.66).⁵⁸ Similarly, a cross-sectional study of 422 pregnant women in a rural area of Gua Musang, Kelantan found that where pre-pregnancy overweight and obesity were significantly associated with excessive GWG.⁵⁹ Another cross-sectional study conducted in Selangor and Seremban found that the mean GWG rate for all pre-pregnancy BMI during the second and third trimesters was higher than the IOM recommendations; those with high pre-pregnancy body mass indices were two to three times more likely to have excessive GWG.⁶⁰

In general, the more weight women gain during pregnancy, the more weight they retain following pregnancy. GWG is positively and significantly associated with postpartum weight retention regardless of pre-pregnancy BMI. A study by Baker et al. found that weight retention was between 0.30 and 0.40 kg (with attenuation) per 1 kg of gained weight and persisted for at least 18 months.⁶¹ A separate study of 427 women in Atlanta revealed that the proportion of excessive weight gain was significantly higher among overweight and obese mothers; 52% of respondents retained more than 10 pounds one year postpartum.⁶² This resulted in 36.1% of the women moving to a higher BMI category, making 68.5% of them overweight or obese one year postpartum. Similarly, a cross-sectional study of 83 pregnant women in Seremban, Malaysia found that 32.5% of women retained at least 4.5 kg six months postpartum; this retention was significantly associated with higher GWG during pregnancy.⁶³

Hollis et al. highlighted the importance of adequate weight gain during pregnancy in order to minimize postpartum weight retention and prevent obesity in childbearing women.⁶⁴ They found that overweight (64.0%) and obese women (51.0%) were most likely to exceed the IOM GWG recommendations. Factors associated with postpartum weight retention include parity, socioeconomic status, marital status, race, GWG, breastfeeding, calorie intake, and physical activity.^{65,66}

Breastfeeding practice

Theoretically, the energy expenditure of lactation is 2.62MJ per day.⁶⁷ A biological rationale supports the hypothesis that breastfeeding promotes postpartum weight loss while a lack of breastfeeding contributes to weight retention and maternal obesity. A study of 2,571 women found that the odds of above-median postpartum weight retention among women who exclusively

breastfed for six to nine months dropped by 37%; they dropped by 46% for those who exclusively breastfed for twelve months.

Furthermore, the odds of early lactation cessation have been shown to rise alongside a rise in BMI.⁶⁸ Obesity is an independent risk factor for not breastfeeding at the time of postpartum discharge from hospital,⁶⁹ which contributes to increasing BMI with each successive pregnancy. This is in line with the findings of a cohort study of 700 expectant mothers in Selangor, Malaysia, which showed that obese women were more likely to delay breastfeeding initiation and cease breastfeeding earlier. Additionally, after six months post-delivery, greater weight retention was found among mothers who formula-fed their offspring (5.69 kg) compared to those who breastfed (1.45 kg).⁷⁰ In another study by Janney et al.⁷¹, weight loss was found to progress at slower rates when women ceased breastfeeding or switched from exclusive to non-exclusive breastfeeding. This could mean that breastfeeding protects against weight gain.⁷² Moreover, a study involving 726 women revealed that, among obese mothers, those who exclusively breastfed retained less weight six-years postpartum (-8.0kg) than those who never breastfed.⁷³

Gigante et al. reported that women who did not exclusively breastfeed or weaned at four months acquired higher fat mass and arm fat indices than those who exclusively breastfed.⁷⁴ A study by Dewey et al. found that formula-feeding mothers had gained triceps-skin-fold thickness while breastfeeding mothers acquired a net loss in this area (2.4 mm vs. -0.4 mm).⁷⁵ Thus, they concluded that exclusive breastfeeding for at least six months would enhance postpartum weight loss. This is in line with Endres et al., who found that women who met the recommendations from the American Academy of Pediatrics for six months of breastfeeding were significantly less likely to retain the weight gained during pregnancy twelve months postpartum.⁶⁵

In contrast, He et al. found that the continuation of breastfeeding beyond six months had minimal or no influence on weight change due to the heterogeneity between studies in intensity, duration of breastfeeding, population under study (small sample size, loss to follow up), assessment of weight retention, assessment on breastfeeding practices, employed statistical analysis, and the presence of confounders such as gestational weight gain, physical activity, and pre-pregnancy BMI.⁷⁶

Knowledge of pre-pregnancy BMI and gestational weight gain recommendation

Knowledge of pre-pregnancy weight status is associated with knowledge of GWG recommendations. Women who were aware of their pre-pregnancy weight status were twice as likely as those who were unaware of their pre-pregnancy weight status to know about the GWG recommendations.⁷⁷ Additionally, obese women tend to overestimate GWG recommendations. A longitudinal cohort study in San Francisco found that 24.1% of overweight women exceeded the IOM-guideline target weight gain limit while just 4.3% of normal-weight women did the same.⁷⁸ In another study, 74.0% of obese women underestimated their BMI category and 64.0% overestimated their recommended GWG.⁷⁹ Furthermore, women's knowledge of risks associated with excessive GWG and maternal obesity was poor; many reported incorrect beliefs about safe weight management in pregnancy.

Moreover, among both normal weight and overweight/obese women, those who misperceived their pre-pregnancy weight were more likely than those who knew their pre-pregnancy weight to exceed GWG recommendations.⁸⁰ Mehta-Lee et al. found that overweight/obese women who under-assessed their pre-pregnancy weight were 2.5 times more likely to gain weight than normal-weight women who accurately assessed their pre-pregnancy weight. Herring et al. found that the adjusted odds of excessive GWG were 2.0 (95% CI: 1.3, 3.0) in normal-weight over-assessors, 2.9 (95% CI: 2.2, 3.9) in overweight/obese accurate-assessors, and 7.6 (95% CI: 3.4, 17.0) in overweight/obese under-assessors compared to normal-weight accurate assessors.⁸¹

Existing guidelines in Malaysia

Comprehensive guidelines on the management of maternal obesity in primary care are essential to improve the quality of care. Currently, there are no such guidelines in Malaysia; the current national perinatal-care manual has no specific chapter on the management of obese mothers. Additionally, Malaysian obesity guidelines should be revised to include a section on managing obese women in the reproductive age group, as

they carry distinctive risks and require specific attention. Maternal obesity should be considered in light of high-risk pregnancy. Enhancing the co-management of clinical care between primary and tertiary centers during the antenatal, intrapartum, and postnatal periods is crucial for obese mothers regardless of the presence of other comorbidities. Furthermore, in the Malaysian Perinatal Care Manual (2013), green color coding is used to indicate women with a pre-pregnancy weight ≥ 80 kg and BMI ≥ 27 kg/m² and is only stated as criteria for screening for gestational diabetes mellitus (GDM). Maternal obesity is currently coded under a green color tag, which is manageable at primary care facilities by medical officers and nurses. However, maternal obesity should also be included in the context of shared care with other disciplines, including specific guidelines on assisting in management at primary care facilities as well as at the tertiary level. One example of a guideline in the management of obesity on pregnancy is available in Australia and endorsed by the Queensland Clinical Guidelines Steering Committee. Furthermore, Confidential Enquiries into Maternal Deaths (CEMD) in Malaysia is lacking in terms of assessing maternal obesity as a determinant of maternal death relative to CEMD in the UK. As a result, the magnitude and severity of this issue cannot be appreciated.

Conclusion

The global emergence of “maternal obesity” as a major public health concern is supported by a vast body of research findings, including some in Malaysia. Maternal obesity is a high-risk phenomenon requiring both surveillance and intervention. Gaining insight into the association between maternal obesity and its contributing factors would enable the development of a more targeted behavioral-change intervention that could be implemented before, during, and after pregnancy, as most determinants are modifiable. This is an essential step in ensuring the end of all preventable causes of maternal and child mortality (SDG 3) and severing the intergenerational chain of non-communicable diseases (NCD) by adopting a life-course approach.

References

1. Friedrich MJ. Global obesity epidemic worsening. *JAMA*. 2017;318(7): 603.
2. Bassett MT, Perl S. Obesity: the public health challenge of our time. *Am J Public Health*. 2004;94(9): 1477.
3. Chan RSM, Woo J. Prevention of overweight and obesity: how effective is the current public health approach *Int J Environ Res Public Health*. 2010;7(3): 765–783.

4. Agha M, Agha R. The rising prevalence of obesity: part A: impact on public health. *Int J Surg Oncol*. 2017;2(9): e42.
5. Case A, Menendez A. Sex differences in obesity rates in poor countries: evidence from South Africa. *Econ Hum Biol*. 2009;7(3): 271–282.
6. Ng M, Fleming T, Robinson M, et al. Global, regional, and national prevalence of overweight and obesity in children and adults during 1980–2013: a systematic analysis for the Global Burden of Disease Study 2013. *Lancet*. 2014;384(9945): 766–781.
7. Institute for Public Health. National health and morbidity survey. Malaysia Ministry of Health. 2015. Available from: <http://www.moh.gov.my/moh/resources/nhmsreport2015vol2.pdf> [Accessed 3rd May 2019].
8. World Health Organization. Obesity and Overweight. 2017. Available from: <http://www.who.int/mediacentre/factsheets/fs311/en/> [Accessed 3rd May 2019].
9. Ko GTC, Chan JCN, Chow C-C, et al. Effects of obesity on the conversion from normal glucose tolerance to diabetes in Hong Kong Chinese. *Obes Res*. 2004;12(6): 889–895.
10. Deurenberg-Yap M, Schmidt G, van Staveren WA, et al. The paradox of low body mass index and high body fat percentage among Chinese, Malays and Indians in Singapore. *Int J Obes*. 2000;24(8): 1011–1017.
11. Heslehurst N, Ells L, Simpson H, et al. Trends in maternal obesity incidence rates, demographic predictors, and health inequalities in 36,821 women over a 15-year period. *BJOG*. 2007;114(2): 187–194.
12. Gorber SC, Tremblay M, Moher D, et al. A comparison of direct vs. self-report measures for assessing height, weight and body mass index: a systematic review. *Obes Rev*. 2007;8(4): 307–326.
13. Ying Pang S, Loy E, Tan L-K. Maternal obesity: prevalence, outcomes and evaluation of body mass index cut-off values in a multi-ethnic Asian obstetric population. *Journal of Obesity and Diabetics*. 2016;1(1): 1–9.
14. Sidik SM, Rampal L. The prevalence and factors associated with obesity among adult women in Selangor, Malaysia. *Asia Pac Fam Med*. 2009;8(1): 2.
15. Chen C, Xu X, Yan Y. Estimated global overweight and obesity burden in pregnant women based on panel data model. *PLoS ONE*. 2018;13(8): e0202183.
16. Institute for Public Health. National health and morbidity survey maternal and child health. Bangsar, Kuala Lumpur: Malaysia Ministry of Health. 2016.
17. Callaway LK, Prins JB, Chang AM, et al. The prevalence and impact of overweight and obesity in an Australian obstetric population. *Med J Aust*. 2006;184(2): 56–59.
18. Withrow D, Alter DA. The economic burden of obesity worldwide: a systematic review of the direct costs of obesity. *Obes Rev*. 2011;12(2): 131–41.
19. World Health Organization. Maternal and Perinatal Health. 2018. Available from: http://www.who.int/maternal_child_adolescent/topics/maternal/maternal_perinatal/en/ [Accessed 24th May 2019].
20. Leddy MA, Power ML, Schulkin J. The impact of maternal obesity on maternal and fetal health. *Rev Obstet Gynecol*. 2008;1(4): 170–178.
21. Simpson EL, Lawrenson RA, Nightingale AL, et al. Venous thromboembolism in pregnancy and the puerperium: incidence and additional risk factors from a London perinatal database. *BJOG*. 2001;108(1): 56–60.
22. Ganesan M, Bujang MA, Soelar SA, et al. Importance of adopting BMI classifications using public health action points to delineate obstetric risk factors resulting in worsening obstetric outcomes among Asian population. *J Obstet Gynaecol India*. 2018;68(3): 173–178.
23. Ramonienė G, Maleckienė L, Nadišauskienė RJ, et al. Maternal obesity and obstetric outcomes in a tertiary referral center. *Medicina*. 2017;53(2): 109–113.
24. Gaillard R, Durmuş B, Hofman A, et al. Risk factors and outcomes of maternal obesity and excessive weight gain during pregnancy. *Obesity*. 2013;21(5): 1046–1055.
25. Heslehurst N, Lang R, Rankin J, et al. Obesity in pregnancy: a study of the impact of maternal obesity on NHS maternity services. *BJOG*. 2007;114(3): 334–342.
26. Rebholz SL, Jones T, Burke KT, et al. Multiparity leads to obesity and inflammation in mothers and obesity in male offspring. *Am J Physiol Endocrinol Metab*. 2012;302(4): E449–E457.
27. Boudet-Berquier J, Salanave B, Desenclos JC, et al. Sociodemographic factors and pregnancy outcomes associated with pre-pregnancy obesity: effect modification of parity in the nationwide Epifane birth-cohort. *BMC Pregnancy and Childbirth*. 2017;17: 273. Available from: doi:10.1186/s12884-017-1456-8.
28. Thrift AP, Callaway LK. The effect of obesity on pregnancy outcomes among Australian Indigenous and non-Indigenous women. *Med J Aust*. 2014;201(10): 592–595.
29. Bahadoer S, Gaillard R, Felix JF, et al. Ethnic disparities in maternal obesity and weight gain during pregnancy: the Generation R study. *Eur J Obstet Gynecol Reprod Biol*. 2015;193: 51–60.
30. Lovasi GS, Hutson MA, Guerra M, et al. Built environments and obesity in disadvantaged populations. *Epidemiol Rev*. 2009;31(1): 7–20.
31. Kumunyika SK. Obesity in minority populations: an epidemiologic assessment. *Obesity*. 1994;2(2): 166–182.
32. Athukorala C, Rumbold AR, Willson KJ, et al. The risk of adverse pregnancy outcomes in women who are overweight or obese. *BMC Pregnancy and Childbirth*. 2010;10(1): 56. Available from: doi:10.1186/1471-2393-10-56.
33. Nohr EA, Vaeth M, Baker JL, et al. Combined associations of prepregnancy body mass index and gestational weight gain with the outcome of pregnancy. *Am J Clin Nutr*. 2008;87(6): 1750–1759.
34. Bogaerts A. Obesity and pregnancy, an epidemiological and intervention study from a psychosocial perspective. Facts, Views & Vision in *ObGyn*. 2014;6(2): 81–95.
35. Mkuu RS. Prevalence and predictors of overweight and obesity among Kenyan women. *Prev Chronic Dis*. 2018;15:E44.
36. McCall S, Nair M, Knight M. Factors associated with maternal mortality at advanced maternal age: a population-based case-control study. *BJOG*. 2017;124(8): 1225–1233.

37. Farpour-Lambert NJ, Ells LJ, Martinez de Tejada B, et al. Obesity and weight gain in pregnancy and postpartum: an evidence review of lifestyle interventions to inform maternal and child health policies. *Front Endocrinol*. 2018;9: 546.
38. Restall A, Taylor RS, Thompson J, et al. Risk factors for excessive gestational weight gain in a healthy, nulliparous cohort. *J Obes*. 2014;2014(3):1-9
39. Durham HA, Morey MC, Lovelady CA, et al. Postpartum physical activity in overweight and obese women. *J Phys Act Health*. 2011;8(7): 988-993.
40. Huberty JL, Buman MP, Leiferman JA, et al. Trajectories of objectively-measured physical activity and sedentary time over the course of pregnancy in women self-identified as inactive. *Prev Med Rep*. 2016;3: 353-360.
41. Pereira MA, Rifas-Shiman SL, Kleinman KP, et al. Predictors of change in physical activity during and after pregnancy: Project Viva. *Am J Prev Med*. 2007;32(4): 312-319.
42. Evenson KR, Moos M-K, Carrier K, et al. Perceived barriers to physical activity among pregnant women. *Matern Child Health J*. 2009;13(3): 364.
43. Poehlman ET. A review: exercise and its influence on resting energy metabolism in man. *Med Sci Sports Exerc*. 1989;21(5): 515-525.
44. Nascimento S, Surita F, Parpinelli M, et al. The effect of an antenatal physical exercise programme on maternal/perinatal outcomes and quality of life in overweight and obese pregnant women: a randomised clinical trial. *BJOG*. 2011;118(12): 1455-1463.
45. Tsigga M, Filis V, Hatzopoulou K, et al. Healthy Eating Index during pregnancy according to pre-gravid and gravid weight status. *Public Health Nutr*. 2011;14(2): 290-296.
46. Lindsay KL, Heneghan C, McNulty B, et al. Lifestyle and dietary habits of an obese pregnant cohort. *Matern Child Health J*. 2015;19(1): 25-32.
47. Pick ME, Edwards M, Moreau D, et al. Assessment of diet quality in pregnant women using the Healthy Eating Index. *J Am Diet Assoc*. 2005;105(2): 240-246.
48. Rodríguez-Bernal CL, Ramón R, Quiles J, et al. Dietary intake in pregnant women in a Spanish Mediterranean area: as good as it is supposed to be? *Public Health Nutr*. 2013;16(8): 1379-1389.
49. Shah BS, Freeland-Graves JH, Cahill JM, et al. Diet quality as measured by the Healthy Eating Index and the association with lipid profile in low-income women in early postpartum. *J Am Diet Assoc*. 2010;110(2): 274-279.
50. Oken E, Taveras EM, Popoola FA, et al. Television, walking, and diet: associations with postpartum weight retention. *Am J Prev Med*. 2007;32(4): 305-311.
51. Wen LM, Flood VM, Simpson JM, et al. Dietary behaviours during pregnancy: findings from first-time mothers in southwest Sydney, Australia. *Int J Behav Nutr Phys Act*. 2010;7(13):1-7
52. King JC. Physiology of pregnancy and nutrient metabolism. *Am J Clin Nutr*. 2000;71(5): 1218S-1225S.
53. Chang M-W, Nitzke S, Guilford E, et al. Motivators and barriers to healthful eating and physical activity among low-income overweight and obese mothers. *J Am Diet Assoc*. 2008;108(6): 1023-1028.
54. Haynes-Maslow L, Parsons SE, Wheeler SB, et al. A qualitative study of perceived barriers to fruit and vegetable consumption among low-income populations, North Carolina, 2011. *Prev Chronic Dis*. 2013;10 (3): E34.
55. Davis EM, Babineau DC, Wang X, et al. Short inter-pregnancy intervals, parity, excessive pregnancy weight gain and risk of maternal obesity. *Matern Child Health J*. 2014;18(3): 554-562.
56. Hanley GE, Hutcheon JA, Kinniburgh BA, et al. Interpregnancy interval and adverse pregnancy outcomes. *Obstet Gynecol*. 2017;129(3): 408-415.
57. Krukowski RA, Bursac Z, McGehee MA, et al. Exploring potential health disparities in excessive gestational weight gain. *J Womens Health*. 2013;22(6): 494-500.
58. Chee WF, Si YM, Siew CY. Factors associated with rates of gestational weight gain among pregnant women in Batu Pahat district of Johor state, Malaysia. *Malaysian Journal of Medicine and Health Sciences*. 2019; 15(1): 33-39.
59. Farhana MN, Rohana A, Alina TT. Excessive and inadequate gestational weight gain among Malaysian pregnant women in rural area: are there any associated factors? *Pakistan Journal of Nutrition*. 2015;14(12): 854.
60. Yong HY, Mohd Shariff Z, Koo SJ, et al. Pre-pregnancy body mass index, height and physical activity are associated with rate of gestational weight gain among Malaysian mothers. *J Obstet Gynaecol Res*. 2016;42(9): 1094-1101.
61. Baker JL, Gamborg M, Heitmann BL, et al. Breastfeeding reduces postpartum weight retention. *Am J Clin Nutr*. 2008;88(6): 1543-1551.
62. Gould Rothberg BE, Magriples U, Kershaw TS, et al. Gestational weight gain and subsequent postpartum weight loss among young, low-income, ethnic minority women. *Am J Obstet Gynecol*. 2011;204(1): 52.e1-52.e11.
63. Yaw YH, Shariff ZM, J binti Shahdan J. Pre-pregnancy BMI and gestational weight gain are associated with 6 months postpartum weight retention. *International Journal of Public Health and Clinical Sciences*. 2017;4(4): 112-25.
64. Hollis JL, Crozier SR, Inskip HM, et al. Modifiable risk factors of maternal postpartum weight retention: an analysis of their combined impact and potential opportunities for prevention. *Int J Obes*. 2017;41(7): 1091-1098.
65. Endres LK, Straub H, McKinney C, et al. Postpartum weight retention risk factors and relationship to obesity at 1 year. *Obstet Gynecol*. 2015;125(1): 144-152.
66. Zanutti J, Capp E, Wender MCO. Factors associated with postpartum weight retention in a Brazilian cohort. *Rev Bras Ginecol Obstet*. 2015;37(4): 164-171.
67. Butte NF, King JC. Energy requirements during pregnancy and lactation. *Public Health Nutr*. 2005;8(7a): 1010-1027.
68. Baker JL, Michaelsen KF, Sorensen TI, et al. High prepregnant body mass index is associated with early termination of full and any breastfeeding in Danish women. *Am J Clin Nutr*. 2007;86(2): 404-411.
69. Ramji N, Quinlan J, Murphy P, et al. The impact of maternal obesity on breastfeeding. *J Obstet Gynaecol Can*. 2016;38(8): 703-711.

70. Hamid A, Bariah S. A cohort study of maternal obesity and breastfeeding outcomes for Malay women in Selangor, Malaysia [dissertation]. Perth: Curtin University; 2016 [Accessed 24th May 2019]. Available from: <https://pdfs.semanticscholar.org/6702/6592a423af5071ed386269836b98903bde32.pdf>.
71. Janney CA, Zhang D, Sowers M. Lactation and weight retention. *Am J Clin Nutr*. 1997;66(5): 1116–1124.
72. Krause KM, Lovelady CA, Peterson BL, et al. Effect of breast-feeding on weight retention at 3 and 6 months postpartum: data from the North Carolina WIC Programme. *Public Health Nutr*. 2010;13(12): 2019–2026.
73. Sharma AJ, Dee DL, Harden SM. Adherence to breastfeeding guidelines and maternal weight 6 years after delivery. *Pediatrics*. 2014;134(1): S42–S49.
74. Gigante DP, Victora CG, Barros FC. Breastfeeding has a limited long-term effect on anthropometry and body composition of Brazilian mothers. *J Nutr*. 2001;131(1): 78–84.
75. Dewey KG, Heinig MJ, Nommsen LA. Maternal weight-loss patterns during prolonged lactation. *Am J Clin Nutr*. 1993;58(2): 162–166.
76. He X, Zhu M, Hu C, et al. Breast-feeding and postpartum weight retention: a systematic review and meta-analysis. *Public Health Nutr*. 2015;18(18): 3308–3316.
77. Ledoux T, Van Den Berg P, Leung P, et al. Factors associated with knowledge of personal gestational weight gain recommendations. *BMC Res Notes*. 2015;8: 349.
78. Stotland NE, Haas JS, Brawarsky P, et al. Body mass index, provider advice, and target gestational weight gain. *Obstet Gynecol*. 2005;105(3): 633–638.
79. Shub A, Huning EYS, Campbell KJ, et al. Pregnant women's knowledge of weight, weight gain, complications of obesity and weight management strategies in pregnancy. *BMC Res Notes*. 2013;6: 278.
80. Mehta-Lee SS, Lischewski Goel JL, Xu L, et al. Perception of weight status and its impact on gestational weight gain in an urban population. *Matern Child Health J*. 2013;17(10): 1931–1939.
81. Herring S, Oken E, Rd J, Rich-Edwards J, et al. Misperceived pre-pregnancy body weight status predicts excessive gestational weight gain: findings from a US cohort study. *BMC Pregnancy and Childbirth*. 2008;8(1): 54. Available from: doi:10.1186/1471-2393-8-54.

The implication of stigma on people living with HIV and the role of social support – A case report

Hadi AM, Lee PY, Adibah HI

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Authors:

Abdul Hadi b. Abdul Manap

(Corresponding author)

MD (UPM), MMed (Family Medicine) (UPM)

Department of Family Medicine
Faculty of Medicine and Health Sciences, Universiti Putra Malaysia
Serdang, Selangor, Malaysia

Lee Ping Yein

MBBS (UM), MMed (Family Medicine) (UM)

Department of Family Medicine
Faculty of Medicine and Health Sciences, Universiti Putra Malaysia
Serdang, Selangor, Malaysia

Adibah Hanim Ismail@Daud

MD (USM), MMed (Family Medicine) (USM)

Department of Family Medicine,
Faculty of Medicine and Health Sciences, Universiti Putra Malaysia,
Serdang, Selangor, Malaysia

Abstract

Despite the advancements made in the knowledge and treatment of the human immunodeficiency virus (HIV) since it was first discovered, people living with HIV (PLWH) continue to be stigmatized. This paper presents the case of an HIV-infected patient who delayed the necessary treatment due to stigma and ultimately presented with AIDS. Through social support, however, he was able to overcome his internalized stigma; he was finally willing to start on antiretroviral treatment (ART). This case report addresses the effect of stigma on and the role of social support in the management of an individual with HIV.

Introduction

Patients infected with HIV can have internalized stigma, which is often compounded among men who have sex with men (MSM). Stigma can result in nondisclosure and delay the pursuit of treatment.¹ The case presented in this paper showcases the type and effect of stigma on a patient infected with HIV and how, through social support, he finally agreed to start on ART and improve his health outcome.

Case Presentation

A 32-year-old man presented with persistent diarrhea that started three weeks ago. He averaged nine episodes of diarrhea per day and had lost 10kg of his body weight since the problem emerged. There were no associated symptoms of fever, abdominal pain, joint pain, ulcers, or rashes. He described himself as a sexually active heterosexual. Three years prior, he was treated for latent syphilis; he denied being infected with HIV.

A physical examination revealed a normal hydration status, normal vital signs, and a normal body mass index. An abdominal examination was revealed no tenderness, mass, or hepatosplenomegaly. A per rectal examination revealed no presence of blood, melena, or mass.

An HIV rapid test was performed and came back as positive. A referral was made for further investigation and management of his chronic diarrhea. During the notification process, it

was noted that a notification had been done in 2014. He admitted to being diagnosed with HIV during his episode of latent syphilis. However, his CD4+ T cell count was 600 cells/mm³ and the patient was well after being treated for syphilis. He stopped attending his follow-up appointments after two months despite receiving multiple reminders from the clinic. He did this because he felt stigmatized for being infected with HIV despite never experiencing any form of external stigma. The severity of his internalized stigma was assessed using the Internalized HIV-Related Stigma Scale, on which he scored a three out of six. Despite being educated on the management, monitoring, and complications of HIV, he still ignored his follow-up appointments as a result of internalized stigma.

He was diagnosed with CMV colitis with a CD4+ T cell count of 171 cell/mm³. He was treated for the CMV colitis with ganciclovir for two weeks. He was initially hesitant to start on ART despite been counseled on its importance. This was due to the self-perceived stigma; he would feel ashamed if others knew his status. However, after disclosing his status to his roommates, they were supportive, helpful, and encouraging; afterward, he finally agreed to start on treatment. Once treatment was initiated, his diarrhea was completely resolved and his weight increased. He did not experience any side effects from the ART; his subsequent appointment showed an increase in his CD4+ T cell count and a reduction in his HIV viral load.

Discussion

The Joint United Nations Programs on HIV/AIDS (UNAIDS) defines stigma and discrimination as “a process of devaluation of people either living with or associated with HIV and AIDS, discrimination follows stigma and is the unfair and unjust treatment of an individual based on his or her real or perceived HIV status.” Stigma and discrimination have an even more profound effect on groups that are associated with HIV, such as commercial sex workers, MSM, intravenous drug users, and prisoners.² Chollier et al.³ detail how the experience of stigma ranges from effective stigma (whereby PLWH are the victims of an enacted stigma) to perceived stigma (whereby PLWH acknowledge that an enacted stigma is considered a norm) and internalized stigma (whereby PLWH acknowledge and validate their own stigma onto themselves).

Internalized stigma is the self-stigma, self-hate, and internalized shame felt by PLWH. It can cause self-loathing, isolation from society, fear of HIV testing, and poor adherence to treatment requirements.³ Internalized HIV-related stigma can be determined by the Internalized HIV-Related Stigma Scale. It has six statements with which participants can “agree = 1”, “disagree = 0,” or remain “neutral = 0.5.” Their marks are accumulated to assess the severity of their internalized stigma. The lowest possible score, 0, indicates low stigma while the highest possible score, 6, indicates high stigma.⁴ The presence of stigma has been shown to result in nondisclosure of status⁵; it is the most important factor behind a patient’s quality of life and adherence to treatment requirements.¹

Social support is crucial in the battle against stigma toward PLWH. It has been shown to result in a lower incidence of depression and

loneliness; ultimately, social support improves patients’ quality of life.¹ Emotional support also helps PLWH improve their coping skills and reduce their negative emotions.⁶ Support from friends and family helps PLWH rebuild their lives and even achieve success in their retroviral disease (RVD) treatment.⁷

Social support need not only comes from friends and family, however; it can also be obtained through support groups held by non-governmental agencies. Examples of such services available in Malaysia are MSM POZ, initiated by the Pink Triangle Foundation, and the Kuala Lumpur AIDS Support Services Society. Participation in support groups has been shown to increase adherence to ART.⁸ Support group participation also leads to improvement in sexual practices—the people in these support groups are more likely to practice safe sex than are those who did not participate.⁹ This can ultimately lead to a reduction in HIV-transmission rates. Support groups have also been shown to lead to an increased likelihood of treatment success, reduced morbidity, reduced mortality, and a higher quality of life in terms of confidence, self-esteem, coping skills, and a perceived reduction of stigma.⁸

Conclusion

Being infected with HIV can lead to stigma among patients. The ranges of stigma are effective, perceived and internalized stigma. Stigma has a significant negative effect on those affected such as poor quality of life and even poor adherence to treatment. Severity of stigma can be assessed using the Internalized HIV-Related Stigma Scale. Psychosocial management options, such as providing social support, are important in the management of HIV, as they can lead to better outcomes for stigmatized individuals.

References

1. Wu X, Chen J, Huang H, Liu Z, Li X, Wang H. Perceived stigma, medical social support and quality of life among people living with HIV/AIDS in Hunan, China. *Applied Nursing Research*. 2015;28:169–74.
2. Nyblade L, Stangl A, Weiss E, Ashburn K. Combating HIV stigma in health care settings: what works? *Journal of the International AIDS Society*. 2009;12:15.
3. Relf M, Rollins KV. HIV-related Stigma Among an Urban Sample of Persons Living With HIV at Risk for Dropping Out of HIV-oriented Primary Medical Care. *Journal of the Association of Nurses in AIDS care*. 2015 Feb;26(1):36–45.
4. Baugher AR, Beer L, Fagan JL, Mattson CL, Freedman M, Skarbinski J, et al. Prevalence of Internalized HIV-Related Stigma Among HIV-Infected Adults in Care, United States, 2011–2013. *AIDS Behav*. 2017 Sep;21(9):2600–8.

5. Miller CT, Solomon SE, Varni SE, Hodge JJ, Knapp FA, Bunn JY. A transactional approach to relationships over time between perceived HIV stigma and the psychological and physical well-being of people with HIV. *Social Science & Medicine*. 2016;162:97–105.
6. Xu X, Sheng Y, Khoshnood K, Clark K. Factors Predicting Internalized Stigma Among Men Who Have Sex with Men Living with HIV in Beijing, China. *J Assoc Nurses AIDS Care*. 2017 Feb;28(1):142–53.
7. Barroso J, Leblanc NM, Flores D. It's Not Just the Pills: A Qualitative Meta-Synthesis of HIV Antiretroviral Adherence Research. *J Assoc Nurses AIDS Care*. 2017 Aug;28(4):462–78.
8. Bateganya M, Amanyeiwe U, Roxo U, Dong M. The Impact of Support Groups for People Living with HIV on Clinical Outcomes: a systematic review of the literature. *J Acquir Immune Defic Syndr*. 2015 Apr 15;68(0 3):S368–74.
9. Qiao S, Li X, Stanton B. Social Support and HIV-related Risk Behaviors: A Systematic Review of the Global Literature. *AIDS Behav*. 2014 Feb;18(2):419–41.

An infant with kwashiorkor: The forgotten disease

Kamaruzaman NA, Jamani NA, Said AH

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Authors:

Nurjasmine Aida Jamani

(Corresponding author)

Department of Family Medicine,
Kulliyyah of Medicine, International
Islamic University Malaysia, Jalan
Sultan Ahmad Shah, 25200
Kuantan Pahang, Malaysia
Email: minaida@iiu.edu.my

Nor Azam Kamaruzaman

Department of Family Medicine,
Kulliyyah of Medicine, International
Islamic University Malaysia, Jalan
Sultan Ahmad Shah, 25200
Kuantan Pahang, Malaysia

Abdul Hadi Said

Department of Family Medicine,
Kulliyyah of Medicine, International
Islamic University Malaysia, Jalan
Sultan Ahmad Shah, 25200
Kuantan Pahang, Malaysia

Abstract

Undernutrition remains a major public health concern, especially in developing countries. Despite being rich in resources, Malaysia is still home to children suffering from severe undernutrition. This paper presents the case of a 5-month-old boy with kwashiorkor stemming from improper weaning which was overlooked. This case highlights the importance of recognizing the early signs of kwashiorkor to allow for early referrals for proper management and prevent its possible complications.

Introduction

Undernutrition remains a major public health concern, especially in developing regions such as Africa, in which three million children under the age of five die each year, constituting nearly half of global deaths in that category.¹ Undernourished infants and young children are at a greater risk of dying from common infections, as undernutrition not only increases the severity and frequency of such infections but also delays recovery.¹

Malaysia, a country that is rich with resources, simultaneously faces crises of overnutrition and undernutrition. Based on the National Health Morbidity Survey, eight percent of children under 5 suffer from undernutrition and wasting.²

Undernutrition is caused by an insufficient intake of carbohydrates, fats, proteins, and micronutrients (vitamins and minerals).³ Marasmus, kwashiorkor, and mixed marasmic kwashiorkor are the major forms of severe energy and protein undernutrition.

This paper reports the case of an infant with kwashiorkor stemming from improper weaning which was overlooked.

Case report

A 5-month-old Orang Asli baby boy was referred to the visiting Family Medicine Specialist by a nurse for failure to thrive because his serial weight gain was unsatisfactory. He was born full term with a birth weight of 2.42 kg. His weight continued to rise through the second month but began to fall below the -2SD in the third month and below the -3SD in the fifth month, as shown in **Figure 1**. Over this period, he was assumed to be thriving, as neither parent noticed any physical changes. He had a good appetite and normal bowel function. He was breastfed exclusively until he reached two months, at which point the mother stopped breastfeeding due to inadequate milk supply. He was then fed sweetened condensed milk, water, and occasionally plain rice. He had no known medical illness.

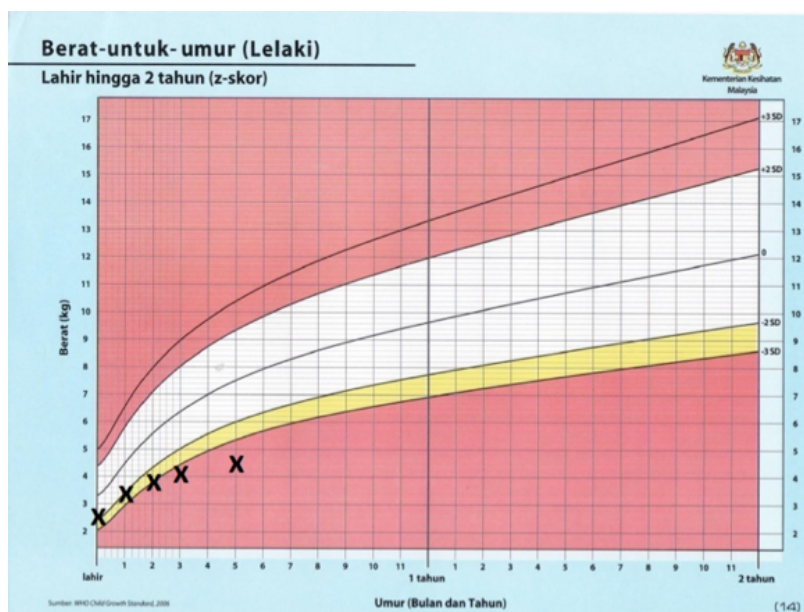


Figure 1: Serial weight-for-age measurement chart showing the baby's weight (-3SD).

After a clinical examination, his vital signs were found to be normal. His weight was 4.5kg (< -3SD) and his length was 57cm (< -3SD). He had prominent round cheeks resembling a cherubic appearance. (Figure 2). There were no signs of pallor, severe wasting, dehydration, or dysmorphic features. However, there was pitting edema visible in both lower limbs (Figure 3). Other examinations uncovered nothing of note. A developmental assessment showed that he is able to lie prone in the supine position and able to bear weight while standing. He exhibited no monosyllabic babbling.



Figure 2: “Cherubic appearance”



Figure 3 and Figure 4: Lower limb pitting edema

He was urgently referred to the hospital for severe acute malnutrition, as kwashiorkor was highly suspected. Laboratory investigations revealed a normal full blood count, but low serum protein and albumin levels. He was started on initial re-feeding with F-75, the “starter” formula, for one week before being given F-100, the “catch-up” formula. Emotional and sensory stimulation was provided throughout inpatient care. He was discharged after about one month to the outpatient clinic for a nutritional rehabilitation

program. His first monthly follow-up showed that he was doing well; within six months, his weight had normalized.

Discussion

Kwashiorkor is a syndrome of severe protein malnutrition. It is characterized by symmetrical peripheral pitting edema that starts in the most dependent regions, hypoalbuminemia and dermatitis. It then proceeds cranially as time progresses, sometimes with anasarca.^{3,4} It may also emerge alongside other micronutrient deficiencies, such as magnesium and zinc.⁵

Kwashiorkor has been reported in both developing and developed countries.⁶ In developing countries, most cases emerge due to poverty and a lack of knowledge about healthy feeding methods. Calvalho et al. reported a similar case in which kwashiorkor was caused by the substitution of non-dairy creamer for milk.⁶ In some cases, a diet for infants centered on rice milk, which is low in protein, has resulted in kwashiorkor.^{7,8} One study from Bangladesh found that faulty breastfeeding practices were a primary driver of undernutrition in children.⁹ Hence, the continuation of breastfeeding in infants under six months old is important.

The infant in our case was brought to the health clinic for a routine monthly check-up. Despite the fact that the infant’s height and weight were on the lower standard deviation on the growth chart, his “cherubic” appearance, caused by fluid retention led untrained eyes to overlook the possibility of kwashiorkor.

Other physical findings of kwashiorkor may include rounded cheeks, pursed lips, dry peeling skin, sparse hair, hepatomegaly, bradycardia, and hypotension.^{3,5,10}

The differential diagnosis of kwashiorkor in children includes congenital cardiac failure, glomerulonephritis, nephrotic syndrome, hepatic cirrhosis, hemolytic anemia, and protein-losing enteropathy.^{3,4}

Kwashiorkor cases are difficult to diagnose and in turn, are often overlooked. This difficulty stems from the fact that generalized edema can mask decreased muscle mass.^{11,12} If left untreated, kwashiorkor can lead to significant morbidity and mortality due to a greater susceptibility to and severity of infections.³

Therefore, it is crucial that all levels of primary

care are able to detect kwashiorkor. The World Health Organization has developed a strategy for reducing mortality and morbidity associated with major causes of childhood illness called Integrated Management of Childhood Illnesses;¹³ this is an integrated approach that addresses the overall health of a child, including nutritional status. This strategy requires those on the front lines of health care to, when working with children under 5 years of age, take weight-for-age, check for severe wasting, and check for edema in both feet. This strategy promotes the accurate identification of malnutrition so that appropriate referrals can be made and effective management strategies can be adopted.

Specific investigations are generally unnecessary in the vast majority of children, as kwashiorkor is a clinical diagnosis.⁵ Investigations are mainly done to look for underlying co-existing conditions, exclude other causes, and assess complications. Children with kwashiorkor usually have a very low plasma albumin concentration as a result of a lack of protein.⁵⁻⁷ However, new evidence has recently emerged that there are multifactorial causes behind edema in malnourished children, such as oxidative stress and intestinal microbiome changes.^{3,14}

The proper treatment for kwashiorkor is the gradual introduction of enteral feeds.¹⁰

Nasogastric feeding is often required for severely affected patients. In developing countries, the mainstay of dietary therapy for kwashiorkor involves cow's milk.⁶ While treating kwashiorkor, professionals should keep refeeding syndrome in mind, as it is a potentially lethal condition that can result from nutritional support.⁹ The World Health Organization has formulated a three-phase management approach for severely malnourished children in which they are 1) resuscitated and stabilized, 2) started on nutritional rehabilitation, and 3) followed up on for recurrence prevention.¹⁵

Conclusion

Undernourished infants and children are not uncommon in primary care. However, kwashiorkor can often be overlooked if it is not consciously kept in mind. The case presented in this paper highlights the importance of a high index of clinical suspicion towards kwashiorkor. Proper dietary histories and thorough physical examinations are crucial for making an accurate diagnosis and prompt referral. Information on healthy breastfeeding practices and proper nutrition should also be made available to parents by the healthcare professionals.

How does this paper make a difference to general practice?

- It increases awareness among family physicians, medical officers, and nurses of the need to recognize the early signs of kwashiorkor.
- It stresses the importance of educating parents on breastfeeding as the optimum source of nutrition for babies.

References

1. UNICEF Data. *Malnutrition*. Available from: <https://data.unicef.org/topic/nutrition/malnutrition/> [Accessed 27th March 2019].
2. Ministry of Health Malaysia. National health & morbidity survey 2015, volume 2. [internet] Putrajaya: Ministry of Health Malaysia; 2015 [Cited 20th March 2017]. Available from: <http://iku.moh.gov.my/images/IKU/Document/REPORT/nhmsreport2015vol2.pdf>.
3. Goday PS. *Malnutrition in children in resource-limited countries: Clinical assesment*. Available from: <https://www.uptodate.com/contents/malnutrition-in-children-in-resource-limited-countries-clinical-assessment> [Accessed 10th March 2019].
4. Williams CD, Oxon BM, Lond H. Kwashiorkor: a nutritional disease of children associated with a maize diet. 1935. *Bulletin of the World Health Organization*. 2004;81(12): 912-913.
5. Xavier MHSB, Magalhães E, Oliveira GF, Magalhães MK, Oliveira CPA, Oliveira, NB. A child with kwashiorkor misdiagnosed as atopic dermatitis. *Dermatology Online Journal*. 2017;23(5): 1-3.
6. Carvalho NF, Kenney RD, Carrington PH, Hall DE. Severe nutritional deficiencies in toddlers resulting from health food milk alternatives. *Pediatrics*. 2001;107(4): E46.
7. Diamanti A, Pedicelli S, D'Argenio P F Panetta, A Alterio, G Torre et al. Iatrogenic Kwashiorkor in three infants on a diet of rice beverages. *Pediatric Allergy and Immunology*. 2011;22(8): 878-879.

8. Grover Z, Ee LC. Protein energy malnutrition. *Pediatric Clinics of North America*. 2009;56(5): 1055–1068.
9. Sen S, Das Sharma J, Das D, Iqbal S, Badruddoza M. Faulty breast feeding practice: a risk factor in malnourished children. *Chattagram Maa-O-Shishu Hospital Medical College Journal*. 2015;14(2): 43–47.
10. Al-bar RH, Al-bar MH, Thigha RM. Case report: Kwashiorkor uncovered by Chickenpox. 2015;83(1): 561–566.
11. Liu T, Howard RM, Mancini AJ. Kwashiorkor in the United States: fad diets, perceived and true milk allergy, and nutritional ignorance. *Archives of Dermatology*. 2001;137(5): 630–636.
12. Schreiber R, Adelson JW. Kwashiorkor in an urban Canadian child. *Canadian Medical Association Journal*. 1985;133(9): 888–889.
13. World Health Organization. *Integrated Management of Childhood Illness (IMCI)*. Available from: https://www.who.int/maternal_child_adolescent/topics/child/imci/en/ [Accessed 27th March 2019].
14. Coulthard MG. Oedema in kwashiorkor is caused by hypoalbuminaemia. *Paediatrics and International Child Health*. 2015;35(2): 83–89.
15. World Health Organization. Management of severe malnutrition: a manual for physicians and other senior health workers. Geneva: WHO; 1999.

An unusual cause of acute abdomen and acute renal failure: Djenkolism

Sumitro K, Yong CS, Tan LT, Choo S, Lim CY, Shariman H, Anand J, Chong VH

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Keywords:

Djenkol; jering; djenkolism; acute renal failure; acute kidney injury; acute abdomen

Authors:

Sumitro Kosasih

(Corresponding author)

MMed

Gastroenterology and Hepatology

Services, Department of Internal

Medicine, Raja Isteri Pengiran

Saleha Hospital, BA1712

Brunei Darussalam

Email: Sumitro_kho@hotmail.com

Yong CS

MRCP

Neurology Unit, Department of

Renal Services, Raja Isteri Pengiran

Saleha Hospital, BA1712

Brunei Darussalam

Tan Lian Tat

MRCS

Department of Surgery, Raja Isteri

Pengiran Saleha Hospital, BA1712

Brunei Darussalam

Sandy Choo

MRCS

Department of Surgery, Raja Isteri

Pengiran Saleha Hospital, BA1712

Brunei Darussalam

Lim Chiao Yuen

FRCP

Department of Renal Services, Raja

Isteri Pengiran Saleha Hospital

BA1712, Brunei Darussalam

Abstract

The djenkol bean (*Archidendron pauciflorum*) is a native delicacy in Southeast Asia, though consumption can sometimes lead to djenkolism. Clinical features of djenkolism include acute abdominal pain, hematuria, urinary retention, and acute kidney injury (AKI). The pain can be severe, which often leads to a misdiagnosis of acute abdomen. In this paper, we report the case of an Indonesian migrant with djenkolism. Due to the short history and severity of the abdominal pain, medical professionals suspected acute abdomen and proceeded with a negative exploratory laparotomy. However, djenkolism was suspected once relatives informed the professionals that the patient had consumed djenkol beans hours earlier. The patient recovered through aggressive hydration and urine alkalization with bicarbonate infusion. We highlight the importance of being aware of this rare cause of AKI, especially in Southeast Asia, in order to provide early diagnoses and prompt treatments.

Introduction

The djenkol bean, or jering (*Archidendron pauciflorum*), is a native delicacy in the Southeast Asian countries of Indonesia, Thailand, Malaysia, and Myanmar. Djenkolism sometimes occurs, albeit infrequently after ingesting djenkol beans; clinical features include acute abdominal pain (especially suprapubic or flank pain), hematuria, urinary retention, and acute kidney injury (AKI). The acute presentation of the abdominal pain can mimic acute abdomen, resulting in misdiagnoses and unnecessary interventions. In this paper, we report the case of an Indonesian migrant with djenkolism who was suspected to have acute abdomen and underwent a negative exploratory laparotomy. The patient recovered through aggressive hydration and urine alkalization with bicarbonate infusion. This paper reports the case of an infant with kwashiorkor stemming from improper weaning which was overlooked.

Case Presentation

A 45-year-old Indonesian man with no history of medical illness arrived at an emergency department with acute colicky lower abdominal pain that started two hours prior. The pain was accompanied by gross hematuria and urinary retention. A physical examination revealed tenderness in the suprapubic and bilateral costovertebral regions. His vital signs were blood pressure 160/110mmHg, heart rate 114 bpm, and afebrile. A laboratory investigation revealed an elevated white cell count of $15.3 \times 10^9/L$,

a hemoglobin count of 13.2gr/dL, a platelet count of $301 \times 10^9/L$, an elevated urea level of 11.1mmol/L (2.5–7.1mmol/L), and a creatinine level of 282mmol/L (60–110mmol/L). An abdominal radiograph (KUB) did not show any urological stones or free air.

He was admitted to the urology service for suspected renal stones. He was treated with intravenous hydration, analgesics, and antibiotics. An urgent computed tomography (CT) was taken of the patient's abdomen; it revealed mild ascites and no hydronephrosis. A microscopic examination of the patient's urine revealed amorphous crystals. The patient's symptoms continued to become more severe; the level of abdominal distension, tenderness, and guarding exceeded that seen in cases of ureteric colic. In view of the rapidly worsening symptoms, the medical professionals suspected a sealed perforated viscus. After discussing with the patient and his family, they decided to proceed with an exploratory laparotomy. However, this only showed a fecally loaded right colon and a small number of ascites without any bowel perforations.

A family member later revealed that the patient had consumed a plateful of fried djenkol beans five hours before his pain started. With this insight, the patient was treated for djenkolism with intensive hydration and a urine alkalization using bicarbonate infusion with a pH aim of >7. His kidney function improved gradually over five days, alongside the resolution

Shariman H

MRCS

Department of Surgery, Raja Isteri
Pengiran Saleha Hospital, BA1712
Brunei Darussalam

Anand J

MD

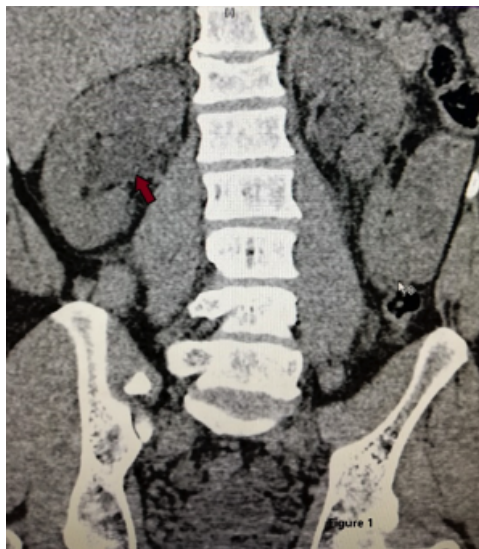
Gastroenterology and Hepatology
Services, Department of Internal
Medicine, Raja Isteri Pengiran
Saleha Hospital, BA1712
Brunei Darussalam

Chong VH

FRCP

Gastroenterology and Hepatology
Services, Department of Internal
Medicine, Raja Isteri Pengiran
Saleha Hospital, BA1712
Brunei Darussalam

of his hematuria. A retrospective review of his abdominal CT scan reveals mild hydronephrosis of the right system, with crystal deposit in the left ureter (**Figures 1 & 2**).



Figures 1: Red arrow points to dilated pelvic calyces of the right kidney, consistent with signs of right hydronephrosis.



Figures 2: Red arrow points to a left ureteric crystal deposit.

Case Discussion

Djenkolism is an uncommon clinical result of djenkol bean consumption that occurs sporadically. Djenkolism is typically reported in Southeast Asia, where the djenkol tree is a native plant. However, the onset of globalization and the ease of modern travel will likely boost the rate of cases elsewhere. Djenkolism is a predominantly male issue with a male-to-female ratio of 7:1.¹ The number of cases rises during rainy season (September to January), coinciding with the blooming of the djenkol tree.^{1,2}

Djenkol beans contain a large amount of djenkolic acid (0.3–1.3 gm/100gm wet weight), and 93% of it exists in a free state.^{2,3} In most cases of djenkolism, patients have consumed a large amount of djenkol beans. One study in Thailand reported a higher incidence of hematuria in children long-term djenkol bean-consumption patterns.⁴ Preventive measures for djenkolism are lacking, largely because the incidence is low and sporadic. However, one study proposed boiling djenkol beans in dilute alkali to remove the djenkolic acid prior to consumption.⁵

The pathogenesis of AKI from djenkol beans is not well understood. However, it is thought to occur due to either hypersensitivity or a direct toxic effect of djenkol bean metabolites resulting in AKI and/or urinary tract obstruction by djenkolic acid crystals, sludge, and/or possible ureteral spasms. In most severe cases, this could cause post-obstructive renal failure.⁶ The pain stems from ureteric colic from crystal precipitation and stones. However, as in our case, the severity of the pain can exceed that seen with ureteric colic; the pain may be a more severe form of ureteric colic.

The two main clinical syndromes of djenkolism are characterized as follows: a) mild presentation of flank/suprapubic pain and hematuria resulting from transient ureteral obstruction due to djenkolic acid crystal; and b) severe presentation in which pain and hematuria are accompanied by hypertension, oliguria, and azotemia.⁷ Our patient belonged to the latter syndrome which, in some of the most severe cases, leads to anuria and even death.⁸ The syndrome is likely determined by the amount of djenkol beans consumed.

Regardless, early recognition is of the utmost importance. Therefore, awareness of the potential consequences of djenkol beans and inquiry into patients' djenkol bean-consumption patterns are very important in general practice. The mainstay of treatment for djenkolism is aggressive hydration and alkalinization of the urine in order to clear the crystal and relieve pain. In severe cases, renal replacement therapy may be required. Our patient recovered within five days of aggressive hydration and alkalinization of the urine using bicarbonate infusion, though he did also go through a negative laparotomy. However, there are reports of surgical interventions being used to relieve obstruction caused by the crystal, sludge, or calculi.^{6,9}

Our case is interesting in that acute abdomen was suspected and the patient received an exploratory

laparotomy. The symptoms were severe and exceeded the severity of those of ureteric colic. In our case, there were two missteps that led to an unnecessary laparotomy: (1) incomplete history taking of djenkol bean consumption (2) improper review of the initial abdominal CT scan. This makes sense, as the consumption of djenkol beans is quite common and is generally innocuous; djenkolism only occurs in rare circumstances. Its occurrence probably depends on the amount of djenkol beans consumed. Furthermore, most public and healthcare professionals are unaware of the problematic association.

Conclusion

It is important to keep uncommon causes in mind when evaluating patients with symptoms of acute abdomen in order to avoid unnecessary invasive interventions. Djenkolism is a prime example of such an uncommon cause, typically the resultant pain is consistent with ureteric colic pain. Inquiry into patients' djenkol bean-consumption patterns is crucial when defining symptoms of djenkolism are seen.

How does this paper make a difference to general practice?

- It recognizes various potential causes of colic abdomen.
- It recognizes various potential causes of hematuria.
- It emphasizes history taking as a tool of the utmost importance to avoid diagnosis delays and unnecessary invasive interventions.

References

1. H'Ng PK, Nayar SK, Lau WM, Segasothy M. Acute renal failure following jering ingestion. *Singapore Medical Journal*. 1991;32(2): 148–149.
2. Areekul S. Djenkol bean, djenkolic acid and djenkolism. *Journal of the Medical Association of Thailand*. 1979;62(10): 530–531.
3. Barceloux DG. Djenkol bean [Archidendron jiringa (Jack) I. C. Nielsen]. *Disease-a-Month*. 2009;55(6): 361–364.
4. Vachvanichsanong P, Lebel L. Djenkol beans as a cause of hematuria in children. *Nephron*. 1997;76(1): 39–42.
5. West CE, Perrin DD, Shaw DC, Heap GH, Soemanto. Djenkol bean poisoning (djenkolism): proposals for treatment and prevention. *The Southeast Asian Journal of Tropical Medicine and Public Health*. 1973;4(4): 564–570.
6. Wong JS, Ong TA, Chua HH, Tan C. Acute anuric renal failure following jering bean ingestion. *Asian Journal of Surgery*. 2007;30(1): 80–81.
7. Bunawan NC, Rastegar A, White KP, Wang NE. Djenkolism: case report and literature review. *International Medical Case Reports Journal*. 2014;7: 79–84.
8. Segasothy M, Swaminathan M, Kong NC, Bennett WM. Djenkol bean poisoning (djenkolism): an unusual cause of acute renal failure. *American Journal of Kidney Diseases: The Official Journal of the National Kidney Foundation*. 1995;25(1): 63–66.
9. Areekul S, Muangman V, Bohkerd C, Saenghirun C. Djenkol bean as a cause of urolithiasis. *The Southeast Asian Journal of Tropical Medicine and Public Health*. 1978;9(3): 427–432.

A hard left supraclavicular mass in a young boy—is it cancer?

Ramasamy K, Saniasiaya J, Gani NA

Ramasamy K, Saniasiaya J, Gani NA. A hard left supraclavicular mass in a young boy—is it cancer?. *Malays Fam Physician*. 2020;15(2);53–55.

Keywords:

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Authors:

Kuganathan Ramasamy

(Corresponding author)
MBBS(Malaya) MRCSI(ENT)
Department of Otorhinolaryngology-
Head & Neck Surgery, Hospital
Tuanku Ja'afar Seremban
Malaysia
E-mail: kuganathan.ram@gmail.com

Jeyasakthy Saniasiaya

MMed(ORL-HNS)(USM)
Department of Otorhinolaryngology-
Head & Neck Surgery, Hospital
Tuanku Ja'afar Seremban
Malaysia

Norhaslinda Abdul Gani

MMed(ORL-HNS)(UKM)
Department of Otorhinolaryngology-
Head & Neck Surgery, Hospital
Tuanku Ja'afar Seremban
Malaysia

Case Summary

A 12-year-old boy was referred to our ENT clinic with an incidentally discovered left supraclavicular mass that was suspected to be malignant. He was asymptomatic and had no prior illnesses. Neck examination found a fixed, hard, non-pulsatile, and non-tender mass measuring 2 cm in diameter in the left supraclavicular fossa. Rest of the ENT assessment, chest and upper limb neurovascular examinations were unremarkable. The patient's full blood count was within the normal range. A plain chest radiograph was taken as part of the work-up (**Figure 1**).

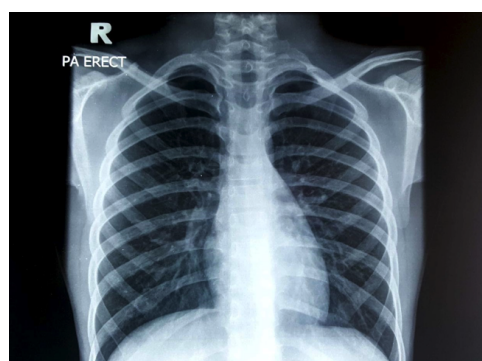


Figure 1. Plain chest radiograph

Questions

1. What abnormality is seen in the radiograph?
2. What differential diagnoses arise from a hard supraclavicular mass in a young boy?
3. What complications can potentially arise from the above condition?
4. List the management options for the above condition.

Answers

1. Description of the chest radiograph:

There are bilateral cervical ribs arising from the seventh cervical vertebra. The cervical ribs are asymmetric; the left one is more prominent. There is no obvious fusion of the cervical rib with the first rib.

2. Differential diagnoses for a hard supraclavicular mass in a young boy:
 - a) Reactive lymphadenopathy
 - b) Lymphoma/leukaemia
 - c) Metastatic lymphadenopathy
 - d) Rhabdomyosarcoma
 - e) Neuroblastoma
 - f) Cervical skeletal anomalies (i.e., cervical rib, transverse mega-apophysis)

A hard mass in the supraclavicular fossa must always be thoroughly investigated to exclude any sinister pathology, particularly malignant metastasis of the head and neck and thoracic neoplasms. Malignancies common in paediatric patients, such as lymphoma and leukaemia, should be ruled out as well. Reactive lymphadenopathy is the most common aetiology, and it is imperative to rule out serious endemic infections, such as tuberculosis. Nevertheless, the presence of a bony hard mass in an asymptomatic child without any risk factors should raise suspicions of a cervical skeletal anomaly, such as a cervical rib. There is a well-described clinical test called the “springing test” that can reliably differentiate between a cervical rib and a supraclavicular lymph node.¹ To perform this test, the clinician applies pressure to the point corresponding to the surface marking of the cervical rib shaft in a springing fashion and assesses whether there is any transmission of the movement to the swelling—while the tip of a cervical rib would move to and fro, a lymph node would remain stationary. A simple radiograph or ultrasound depicting calcified mass can confirm the diagnosis of cervical rib and avoid unnecessary invasive diagnostic procedures, such as fine needle aspiration biopsies or even open biopsies.² Notwithstanding the diagnosis of cervical rib, an essential practice point is to always perform a complete examination with baseline laboratory investigations to avoid missing serious pathologies, as mentioned above.

3. Thoracic outlet obstruction (vascular and/or neurogenic)

Cervical rib is a congenital anomaly characterised

by the emergence of an accessory rib usually from the seventh cervical vertebra, though there have been some cases of cervical rib emerging from the fourth, fifth, and sixth cervical vertebrae. This anomaly stems from a mutation of the *Hox* gene. It is a rare occurrence with a rate below 1%, and it is often asymptomatic.³ However, in about 10% of the cases, there is compression on the subclavian vessels and/or the brachial plexus from the cervical rib or the fibrous band that connects the cervical rib to the first rib. In these cases, patients present with neurogenic and vascular symptoms that suggest thoracic outlet syndrome. The neurogenic symptoms include radiating pain to the upper limb with paraesthesia, weakness, and muscle wasting in the affected limb and hand. The vascular symptoms include coldness, pallor, cyanosis, and swelling of the limb.⁴ In rare cases, cerebral embolus has been reported due to retrograde flow from subclavian artery compression, which can lead to stroke.⁵ There are few commonly used provocative manoeuvres that can help to identify thoracic outlet obstruction, such as Adson's test and the elevated arm stress test (EAST). A positive Adson's test is characterised by diminution of the radial pulse when the neck is fully extended and the head is tilted towards the symptomatic side while the patient is holding in a deep breath. EAST is performed by abducting the arms to 90 degrees with the elbows flexed in the "hands-up" position. The patient is then asked to repeatedly open and close their hands for three minutes; the test is positive when symptoms are reproduced or the patient is unable to do the hand movements for the full three minutes. The use of both of these tests reported diagnostic

specificity of about 82%, higher than either of them used independently. Nevertheless, in cases of suspected thoracic outlet obstruction, a CT or MRI scan is warranted for diagnostic confirmation and surgery planning.⁶

4. Management options:

a) Reassurance

- Asymptomatic patients do not require treatment. They should be reassured and advised to seek treatment later if symptoms of thoracic outlet obstruction emerge.

b) Conservative approach

- This is the first-line management option for symptomatic patients. It comprises analgesics (NSAIDs), muscle relaxants and physiotherapy.

c) Surgery

- If conservative approaches fail or there are acute neurovascular manifestations, surgical treatment is indicated.
- The surgical procedure entails the excision of the cervical rib and, often, the first rib as well to alleviate the compression on neurovascular structures.⁷

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No conflicts of interest reported by any of the authors.

How does this paper make a difference to general practice?

- Neck masses in children are encountered regularly in daily general practice and can often be perplexing.
- This paper highlights the importance of considering cervical rib as a differential diagnosis for children with a hard supraclavicular mass that are otherwise asymptomatic with no risk factors.
- When a cervical rib is suspected, a simple chest radiograph can be used to confirm the diagnosis.
- This paper details several bedside tests that can be used to assess potential complications stemming from a cervical rib.
- In addition to avoiding unnecessary invasive diagnostic procedures, a prompt diagnosis provides patients and parents with reassurance and prevents unnecessary anxiety over the possibility of malignancy.

References

1. Venkatesh Rao P. Springing test for differentiating a cervical rib tip from a supraclavicular lymph node. *Jpn J Surg.* 1988;18(5): 606–607. Available from: doi:10.1007/bf02471498.
2. Pepper T, Singh M, Brennan P. The transverse mega-apophysis—An unusual neck lump. *Br J Oral Maxillofac Surg.* 2010;48(3): 201–202. Available from: doi:10.1016/j.bjoms.2009.08.026.
3. Spadliński Ł, Cecot T, Majos A, et al. The epidemiological, morphological, and clinical aspects of the cervical ribs in humans. *Biomed Res Int.* 2016;2016:8034613. Available from: doi:10.1155/2016/8034613.
4. Jiang S, Shen H, Tan W, Lu H. Arterial thoracic outlet syndrome caused by cervical ribs—an unusual case report. *Medicine (Baltimore).* 2019;98(11): e14778. Available from: doi:10.1097/md.00000000000014778.
5. Shucksmith H. Cerebral and peripheral emboli caused by cervical ribs. *BMJ.* 1963;2(5361): 835–837. Available from doi:10.1136/bmj.2.5361.835.
6. Povlsen S, Povlsen B. Diagnosing thoracic outlet syndrome: current approaches and future directions. *Diagnostics.* 2018;8(1): 21. Available from: doi:10.3390/diagnostics8010021.
7. Leong SC, Karkos PD. A “hard” neck lump. *Singapore Med J.* 2009;50(4): e141–e142.

Letter to the editor regarding the article, “young lady with bilateral yellowish lesions on her eyelids”

Hisham A, Md. Mydin Siddik NSF, Ibrahim S, Mohd Yussof SJ

Hisham A, Md. Mydin Siddik NSF, Ibrahim S, et al. Letter to the editor regarding the article, “young lady with bilateral yellowish lesions on her eyelids”. *Malays Fam Physician*. 2020;15(2);56–57.

Keywords:

Eyelids surgery, Surgical excision, Xanthelasma, Xanthelasma Palpebrarum

Authors:

Shah Jumaat Mohd Yussof

(Corresponding author)
MRCS (Ire), MS Plastic Surgery (USM)
Faculty of Medicine, Universiti
Teknologi MARA, Cawangan
Selangor, Kampus Sungai Buloh Jalan
Hospital, 47000, Selangor, Malaysia
Email: shahjumaat@gmail.com

Akmal Hisham

MRCSed, MS Plastic Surgery (USM)
Faculty of Medicine, Universiti
Teknologi MARA, Cawangan
Selangor, Kampus Sungai Buloh
Jalan Hospital, 47000, Selangor
Malaysia

Nur Shazwani Farah Md. Mydin Siddik

MS Plastic Surgery (USM)
Faculty of Medicine, Universiti
Teknologi MARA, Cawangan
Selangor, Kampus Sungai Buloh Jalan
Hospital, 47000, Selangor, Malaysia

Salina Ibrahim

MD (UKM), FRCS (Edinburgh)
Department of Plastic and
Reconstructive Surgery, Hospital
Sungai Buloh, Selangor, Malaysia

Dear Editor-in-Chief,

We read the “test your knowledge” article by Ishak et al.¹ with great interest. We commend the authors for their educative discussion about the case of a 26-year-old woman with bilateral xanthelasma palpebrarum (XP).¹ We frequently encounter patients with XP in our plastic surgery clinic and feel that it is important to add several points regarding the treatment of this condition.

The authors correctly state that “Previously common practices used to remove this type of lesions were cautery and laser ablation. Nowadays, chemical cauterization using TCA is being practiced by most physicians.” However, they omitted surgical excision from the list of treatment options for XP despite it being the method preferred by most plastic surgeons,^{2–4} including those in our clinic. The advantages of surgical excision include a more inconspicuous scar, the prevention of eyelid deformation, a specimen for histopathological analysis, the complete removal of the XP, the minimization of clinic visits, and a lower recurrence rate.^{3,4}

Surgery is warranted for lesions more than 5 mm in height, lesions involving the deep dermis or the underlying muscle, and longstanding lesions with an onset exceeding a year; of course, patient preferences must also be considered.³ There are several surgical excision techniques currently available for XP management, including *en toto* excision in an elliptical fashion, staged serial excision, excision with blepharoplasty incision, and “uncapping surgery,” in which the lesion is “uncapped” and the cholesterol deposit is removed.² A combination of simple excision and local flaps or skin grafts are also available for more advanced stages of XP.³ All of these techniques can be performed as an office-based procedure under local anesthesia.

Specifically, regarding the titular case, the patient received three treatments of trichloroacetic acid (TCA). We applaud the authors for their partial success (40% improvement) without any reported complications. However, chemical cauterization with TCA is generally reserved for lesions under 3–4 mm in diameter, as emphasized by some of the authors cited by Ishak et al.^{2,5} We feel that the use of TCA in cases with large xanthelasma such as this one could lead to suboptimal treatment and, more severely, increase the likelihood of excessive scarring and ectropion. These complications can be far more disfiguring and problematic than the original lesion and require a more complicated reconstructive treatment.

The various therapeutic methods available to treat XP all have advantages as well as limitations. However, we argue that, for large or refractory XP, surgical management should be the mainstay of treatment.

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References

1. Ishak A, Mohd Yusoff SS, Wan Abdullah WNH. Young lady with bilateral yellowish lesions on her eyelids. *Malaysian Family Physician*. 2018;13(3): 44–46.
2. Rohrich RJ, Janis JE, Pownell PH. Xanthelasma palpebrarum: a review and current management principles. *Plastic and Reconstructive Surgery*. 2002;110(5): 1310–1314.
3. Lee HY, Jin US, Minn KW, et al. Outcomes of surgical management of xanthelasma palpebrarum. *Archives of Plastic Surgery*. 2013;40(4): 380–386.
4. Wang KY, Hsu KC, Liu WC, et al. Relationship between xanthelasma palpebrarum and hyperlipidemia. *Annals of Plastic Surgery*. 2018;80: S84–S86.
5. Cannon P, Ajit R, Leatherbarrow B. Efficacy of trichloroacetic acid (95%) in the management of xanthelasma palpebrarum. *Clinical and Experimental Dermatology*. 2010;35(8): 845–848.

COVID-19, family medicine, and primary care centers

Mungmungpuntipantip R, Wiwanitkit V

Mungmungpuntipantip R, Wiwanitkit V. COVID-19, family medicine, and primary care centers. *Malays Fam Physician*. 2020;15(2);58.

Keywords:

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Authors:

Rujitika Mungmungpuntipantip

(Corresponding author)

Private Academic Practice

Email: rujitika@gmail.com

Viroj Wiwanitkit

Honorary professor, Dr DY Patil

University, Pune, India

visiting professor, Hainan Medical

University, Haikou, China

Dear Editor,

We read the article “We have to write and share valid and reliable information on COVID-19” with great interest.¹ We would like to share some ideas from our country, Thailand, where the disease emerged relatively early.² We agree that the reliability of data is of the utmost importance, and local data is oftentimes not updated. The officially reported number of COVID-19 patients in might be an underestimation. In fact, some of those with the disease are asymptomatic, meaning the virus may be imported from countries with unknown risk levels.³

Family physicians play a significant role during this pandemic, as they are often where patients initially go to address medical problems. Therefore, family physicians should have up-to-date COVID-19 information in order to properly manage cases. Of course, they should also know how to protect themselves by following universal precautions. The question of whether patients should visit their local family physician to test for COVID-19 is an interesting one. Fear of disease is common in Thailand and people usually visit big hospitals for consultations specialists. However, the infection-control guidelines for COVID-19 in Thailand assert that those who suspect they have the disease should go to the primary care clinics. Only after testing positive are patients directed to the local hospital; referral to tertiary care centers is done in severe cases. Nevertheless, Thai citizens are free to choose between the free government healthcare system and paid private medical services. The promotion of trust in the primary care system and family physicians is crucial during this crisis.

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References

1. Liew SM, Khoo EM, Cheah WK, Peariasamy K, Goh PP, Ibrahim HM. We have to write and share valid and reliable information on COVID-19. *Malaysian Family Physician*. 2020;15(1): 1–2.
2. Yasri S, Wiwanitkit V. Editorial: Wuhan coronavirus outbreak and imported case. *Advance Tropical Medicine and Public Health International*. 2019;9: 1–2
3. Joob S. Bleeding presentation among the patients COVID-19 infection: an observation on 4 cases in Thailand. *Advance Tropical Medicine and Public Health International*. 2020;10: 3–4.

