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CONTENTS

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EDITORIAL

The Need for a Vaccine Against Hand, Foot, and Mouth Disease in Malaysia

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Hand, foot, and mouth disease (HFMD) is a contagious viral infection, which commonly affects young children under five years of age (Yu et al., 2019). HFMD is caused by serotypes of the Enterovirus A species, particularly enterovirus 71 (EV71) and coxsackievirus A16 (CVA16), in the genus Enterovirus (Takahashi et al., 2016). Symptoms of HFMD usually include fever, sore throat and maculopapular or vesicular rashes on hands, feet and mouth, which resolve spontaneously. On the other hand, severe HFMD, which is often associated with EV71 infection, can lead to life-threatening cardiopulmonary and neurologic complications (Xu et al., 2015).

Both EV71 and CVA16 are non-enveloped viruses, containing a genome of single-stranded, positive-sense RNA with a single open reading frame (ORF) encoding a polyprotein. This polyprotein is further cleaved to form four structural proteins (VP1-VP4) and seven non-structural proteins (2A-2C, 3A-3D) (Guo et al., 2019). VP1 region is the main neutralising antigenic site and has higher genetic variability than the other capsid proteins, making it desirable for genotyping of enterovirus (Noisumdaeng et al., 2019). Based on the genetic diversity of the VP1 gene, EV71 is classified into seven genogroups (A-G), where genogroups B and C are more widely circulated throughout the world and consist of 11 genotypes, B0-B5 and C1-C5; and two subgenotypes, C4a and C4b (van der Sanden et al., 2016). Meanwhile, CVA16 is classified into two genogroups (A and B), where B is the predominant genogroup

circulating worldwide. Genogroup B is further classified into two genotypes, B1 and B2; and three subgenotypes, B1a-B1c (Zhou et al., 2021).

Studies have shown distinct geographic distribution for these enteroviruses and many genotypes emerge progressively with uneven global distribution. For instance, EV71 genogroups D, G and H were only circulating in South Asia, while genogroups E and F were only found in Africa (Xu et al., 2021). Even within the same region, such as Malaysia and Singapore, the circulating genotypes were different between these countries (Xu et al., 2021). Besides, the predominance of the genotype circulating in a country changes over time. Genotype B3 was the predominant type that was associated with fatal cases in the 1997 HFMD outbreak in Malaysia. Later in 2000, B4 emerged as the predominant genotype and from 2003 onwards, HFMD in Malaysia was mainly caused by genotype B5 (Fong et al., 2021). Genotype changes are also observed in CVA16. Between 1997 and 2014, the predominant genotype of CVA16 found in Malaysia was B2. However, a recent outbreak in Sabah in 2018 witnessed the emergence of subgenotype B1a as the predominant type, which has never been reported in Malaysia (Fong et al., 2021). Apart from that, a study by Ling et al. (2014) found another serotype of enterovirus, namely CVA6, to be the main type that caused HFMD in Seri Kembangan, Malaysia between 2012 and 2013.

Currently, there is no treatment for HFMD and no vaccine is available in Malaysia. Even though HFMD is usually mild and self-limiting, severe HFMD can be an economic burden to the country, society and families of patients. Moreover, the significant rise in HFMD cases in Malaysia (Fong et al., 2021; Fong, 2022; Kaos Jr et al., 2022) can further increase this burden. To date, the world's only EV71 vaccines are China's three inactivated monovalent EV71 vaccines, which are based on the prominent genotype, C4 in the country (Li et al., 2021). Although the vaccine strain belongs to genotype C4, studies have found good cross-neutralisation against other major genotypes (Li et al., 2021), suggesting that China's vaccines can protect people globally against HFMD caused by EV71. However, it was also reported that the three vaccines were not effective against non-EV71 serotypes (Tong et al., 2021). This is a concern since there is an increase in the emergence of non-EV71 serotypes, such as CVA6, CV10 and CVA16, as the main causative pathogens for HFMD. The ultimate goal is to have a globally representative HFMD vaccine that can protect against all major genogroups and genotypes of each enterovirus serotype. Efforts from local governments, international organisations and the pharmaceutical industry are needed to achieve this ultimate goal. Considering the changing epidemiology of HFMD, epidemiological studies are necessary to provide updated information on the current circulating strains, which is crucial for the development of multivalent vaccines.

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ORIGINAL ARTICLE

The Career Planning of Female Intern Doctors and Their Specialty Preference

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ABSTRACT

Internship time is a prime time for doctors to decide on their future career planning. It should be decided based on liking, work-life balance, and other factors. This was a cross-sectional study on 92 female intern doctors at Sylhet Women's Medical College Hospital, Sylhet, Bangladesh. Information was achieved by questionnaire. Data of preferred subjects in MBBS, preferred specialty selection, influencing factor, and preferred post-graduate degree were considered variables. A western degree (37.7%) was the most preferred, followed by a fellowship (26%). The most popular specialty was general surgery (29.3%), followed by obstetrics & gynaecology (OBG). Most participants (70%) decided on their own regarding future career planning. It is important to have a well-organized future career plan during the internship. Clinical subjects were preferable to basic subjects as a specialty. Western degrees were popular over national post-graduate degrees.

INTRODUCTION

Physicians are one of the noblest professionals in the world. In most countries, completing a bachelor's degree in medicine takes nearly 5 – 7 years. The career planning of a doctor usually starts from the first year of the studentship, which becomes more focused during the internship.

Their experience widely influenced the career planning of doctors during studentship, attitude, values, personal circumstances, and aptitude (Yang et al., 2016). The junior doctors of Europe also consider the residency or post-graduation course structure while deciding on their careers (Stilwell et al., 2000). In the last few decades, male and female doctors have considered their family lifestyle when selecting future specialties (Allen et al., 1994). General practitioners prefer lifestyle, working hours, and working environments more than doctors who work in a hospital setup (Ahmed et al., 2011).

A study on Bangladeshi medical students revealed that 50% of students preferred medicine specialties, and 48% preferred surgical specialties as career plans (Ahmed et al., 2011). More than two-thirds (67%) of Bangladeshi medical students want to do private services, followed by government jobs (25%), armed forces (5%), and non-government organisations (3%). Regarding reasons behind choosing the career plan, 38% considered a vast job option, 38% preferred to help others, and 35% felt an enjoyable lifestyle. Some (22%) wanted to deal with the patients directly, and few (13%) wanted to do the job for a fixed hour. More than half (51%) of students wanted to practice abroad, and the majority (90%) wanted to practice in big cities. This was a cross-sectional descriptive study. This study aimed to evaluate the career planning of Sylhet Women's Medical College Hospital intern doctors.

MATERIALS AND METHODS

This was a cross-sectional study to evaluate the intern doctors' career plans and their career choices. All the participants were female. A total of 92 participants were enrolled. The study was conducted at Sylhet Women's Medical College Hospital. Ethical approval was achieved by the institutional research board (IRB). All the participants were invited into the college auditorium. Informed consent was taken from the participants. The objectives of the study were explained to all the participants. When all the participants were confident about self-assessment, then questionnaires were distributed. Participants documented their own career choices and future career planning. All the data from the questionnaires were transferred to an Excel master sheet. Descriptive mathematical calculations were done to find out the means and percentages.

RESULTS

Table 1 shows that nearly ninety per cent of the participants were Bangladeshi, and the rest were from Nepal. The mean age of the participants was 24.7 years.

Table 1 Distribution of all participants (n = 92) according to their country of origin and their mean age.

Country	Number of participants	Percentage (%)	Mean age
Bangladesh	82	89.1%	24.7
Nepal	10	10.8%	

Table 2 illustrates participants' career choices during the first year and internship. In the first year, Anatomy and Biochemistry was a career plan for 2.1% and 1% of participants, but eventually, those changed after becoming a doctor. Participants who selected general physicians, paediatric surgery, and pharmacology in the first year also did not show interest in those subspecialties later on. Participants grew more interested in dermatology, neurology, paediatrics, medicine, and general surgery during their study period. Pathology is the only subspecialty where nobody was interested in the first year, but 2.1% developed interest during the internship.

Table 2 Career choice of participants in the first year and during the internship (n=92)

Subjects	1st year	1st year %	During internship	During internship %
Anatomy Biochemistry Cardiology Cardiac surgery Dermatology General physician OBG	02	2.1%	– – 02	– – 2.1%
Medicine Neurology Neurosurgery Oncology Paediatric surgery Paediatrics Pharmacology	01	1.0%	02	2.1%
Psychiatry Radiology General surgery Thoracic surgery Not decided	12	13.0%	04	4.3%
Pathology	02	2.1%	– 20	– 21.7%
			17	18.4%
	01	1.0%		
			04	4.3%
	01	1.0%		
			01	1.0%
	21	22.8%		
			01	1.0%
	09	9.7%		
			– 08	– 8.6%
	01	1.0%	– 01	– 1.0%
			01	1.0%
	03	3.2%		
			27	29.3%
	01	1.0%		
			01	1.0%
	01	1.0%		
			01	1.0%
	04	4.3%		
			02	2.1%
	01	1.0%		
	02	2.1%		
	01	1.0%		
	15	16.3%		
	01	1.0%		
	13	14.1%		
	–	–		

Table 3 shows the expected degrees participants want to achieve in their plan. More than one-third opted to earn western country degrees; the next favourite was fellowship (26%), followed by the master (17.3%). More than one-tenth of participants wanted a PhD degree, while the rest focused on diplomas.

Table 3 Distribution of all participants according to their aim of the degree (n=92)

Degree	Total	Percentage (%)
Diploma	09	9.7%
Fellowship	24	26%
Masters	16	17.3%
PhD	11	11.9%
Western country degree	32	37.7%

Table 4 demonstrates the subjects of the MBBS course that the participants liked more while studying. Surgery was the most popular subject (27.1%), followed by medicine (23.9%) and OBG (15.2%). Among the pre-clinical subjects, anatomy (7.6%) and pharmacology (5.4%) were liked most.

Table 4 Distribution of all participants according to their favourite subjects (n = 92)

Subject	Total	Percentage (%)	
Anatomy	07	7.6%	
Biochemistry	02	2.1%	
Cardiology	01	1.0%	
Forensic medicine		01	1.0%
OBG		14	15.2%
Medicine		22	23.9%
Microbiology		03	3.2%
Paediatrics		09	9.7%
Pathology		03	3.2%
Pharmacology		05	5.4%
General surgery		25	27.1%

Table 5 illustrates the persons who influenced the participants in choosing their careers. More than two- third participants chose by themselves, whereas family members influenced nearly one-fourth. Other doctors advised only 5% of participants. When making decisions alone, they mostly preferred general surgery as a specialty. On the other hand, medicine was advised by the family members mostly. When suggestions were received from other doctors, it was mostly general surgery.

Table 5 Distribution discipline preference according to influence/advice (n = 92)

	Total	Percentage (%)	Discipline advised	Percentage
By herself	64	70%	General Surgery OBG	28% 22%
Family member	21	23%	Medicine General surgery	28.5% 24%
Another doctor	05	5%	General surgery	40%
Other	02	2%	General surgery	100%

DISCUSSIONS

This cross-sectional study was performed on 92 intern doctors at Sylhet Women's Medical College Hospital. The career plan of the participants was evaluated. The results of this study were compared with those of international studies.

The career choice of physicians of Finland and their personalities were observed by Mullola et al. (2018) where a total of 2837 participants were enrolled. The standard three confounding factors were a medical specialty, patient contact, and employment sector in selecting a career. Other factors were gender and age. Participants with higher openness preferred working in the private sector. They also opted for psychiatry mainly as a career, enjoyed specialty switch, and did not like to practice with patients. High extraversion participants wanted paediatric as a career mostly. Low openness participants preferred ophthalmology and otorhinolaryngology

mostly. Among the participants, 1,838 were female with a mean age of 48. Nearly three-forth female doctors chose public hospital jobs over private. General practice was preferred by almost one-fifth (21.92%) of the lady doctors, where 14.68% selected medicine subspecialties, and 9.68% opted for public health. Other preferred specialties were psychiatry (12.56%), OBG (8.43%), paediatrics (6.47%), anesthesiology and critical care (6.52%), general surgery (4.35%), pathology (3.42%), ophthalmology/ ENT (3.69%) and radiology (2.99%). In this study, the highest number of participants preferred general surgery (29.3%) as a career plan, followed by OBG (21.7%) and medicine 18.4%). The preference for paediatrics (8.6%), pathology (2.1%), and radiology (1.0%) showed similarity with the study of Mullola et al. (2018).

Mone et al. (2019) published their study on the career and work-life balance of Bangladeshi lady doctors. Data were collected from six different divisions of Bangladesh. All lady doctors thought that increasing the number of medical colleges would only be helpful if the standard was maintained. Most participants concluded that their working conditions were uncomfortable due to low salaries, hospital staff policies, and extended working hours. The lady doctors also thought that job stress was higher in the private sector hospitals. One-third of the lady doctors opined that salary was good in the medical colleges. Two-thirds of the participants found that the communication level in the private sector is satisfactory due to quality control policies. All the lady doctors found the registration fees for post-graduation degrees were very high. Workplace security was a concern before selecting the hospital for most lady doctors. Few participants had chosen basic subjects to avoid confrontation with the patient's relatives and friends.

Darkwa et al. (2015) performed their study to find out the factors influencing doctors and nurses to retain in rural areas. Most doctors thought that big cities like Dhaka, Chittagong and Rajshahi are rich in training facilities compared to the rural areas. The training was even difficult in the district hospitals. Doctors showed a better relationship with the community members than the nurses. Most of the rural doctors and nurses demanded more incentives as working hours were more than in cities and also after working income was less. Electricity power cuts and drinking water facilities were concerns in rural areas. In this study, more than one-third of the participant (37.7%) wished to continue their studies in Western countries.

Zia et al. revealed that more than one-third of final-year students had a doctor in their families. More than half (58%) of final-year students wanted to build a career in medicine or sub-branches. Nearly one-third (34%) opted for surgery and sub-specialties as a profession. Less (7.9%) of students chose basic subjects as a career, and Radiology was the most popular among the basic subjects. Approximately one-fourth (24.9%) of participants considered working hours and emotional attachment as influencing factors for career decision-making. In this study, (27%) of the participants had a doctor in their family. Overall, (24.9%) of participants wanted to make a career in medicine and sub-specialties, (27.1%) in surgery, and sub-specialties (22.5%) in basic / pre-clinical subjects.

CONCLUSION

Most of the participants decided on their future careers independently, as opposed to being influenced by family members, professional colleagues, or others. Surgery and allied subjects were the most typical choice for a future career for the lady intern doctors. The aim of their professional degree for the most participants was to obtain a western post-graduation degree, followed by the second highest number of participants opting for a fellowship in their desired subjects.

CONFLICT OF INTEREST

The authors declare that they have no competing interests in publishing this article.

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ORIGINAL ARTICLE

Hand Anthropometry: Baseline Data of The Major Ethnic Groups in Sabah

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ABSTRACT

Anthropometry, the quantitative interface between anatomy and physiology, is a scientific specialization concerned with applying measurement to appraise human size, shape, proportion, composition, development, and gross function. Anthropometric measurement is essential in many areas, including archaeology, anthropology, ergonomics, forensic sciences, anatomy, and nutrition. Although different researchers have studied the anthropometry of hand extensively in Malaysia, there is still a paucity of data among the major ethnicities (Kadazandusun, Bajau, Malay and Chinese) that exist in Sabah. The study was designed to create baseline data for normative values of hand length, handbreadth, middle finger length, second inter-crease length of the middle finger, and the hand span of major ethnic groups in Sabah. This cross-sectional study was conducted from February 2021 to January 2022 by applying a stratified random sampling method. At first, the students were stratified into ethnicities and were further stratified as males and females. The hand dimensions were measured using a digital calliper. The baseline data for Sabah's four major ethnic groups were compared for symmetry, gender, and ethnic differences. The right handbreadth was broader than the left hand ($p<0.01$). The result also demonstrated a statistically significant (0.001) difference between gender; however, there was no significant difference among

the four ethnicities. Further study at the community level is recommended for different age groups and ethnicities by addressing hand activity, hormones, and brain asymmetry to complement the findings of this study.

INTRODUCTION

Anthropometry, the quantitative interface between anatomy and physiology, is a scientific specialization concerned with applying measurement to appraise human size, shape, proportion, composition, development, and gross function. It is a fundamental discipline for problem-solving in growth, practice, performance, and nutrition. It puts an individual into objective focus and provides a precise appraisal of his or her structural status at any given time or, more importantly, provides for quantification of differential growth (Bláha, 2007). Anthropometry is commonly used to measure a person's body size and height. However, anthropometry may also include functional anthropometry such as measuring strength for a specific application (Nurul Shahida et al., 2016).

Anthropometric measurement is essential in many areas, including archaeology, anthropology, ergonomics, forensic sciences, anatomy, and nutrition. For instance, Body Mass Index (BMI) can be calculated by anthropometry, and it is critical in the treatment of malnutrition, including obesity and undernutrition. The stature of dead humans or parts of a dead gives characteristic features of a population for archaeological materials (Akman et al., 2006). Apart from that, people use various tools in everyday life, including hand tools, to complete various tasks. The equipment used must be compatible with the users' physical characteristics.

According to Mondol et al. (2009), upper limb bones and stature dimensions vary in different ethnicities, gender, and age groups, and it also varies with the opposite side of the body (Plato et al., 1980; Kulaksiz & Gözil, 2002). The difference needs to be addressed as it will affect the safety and comfort of workers when a hand device or tools (e.g., gloves) are designed from data for different gender. Yu et al. (2014) emphasized the importance of hand gloves fitting with the users' hands to prevent injury and increase working performance. Also, hand anthropometry is a necessary input for tool design that promotes task productivity and workers' health García-Cáceres 2012). In the context of ergonomics, these tools and equipment should be compatible with the physical characteristics of the workers for better productivity and work safety, and hand anthropometry is a fundamental parameter that must be utilized for designing hand tools (Shahriar et al., 2020). Therefore, various research has been done on anthropometrics and mentioned the importance of relevant standardized anthropometry data in designing equipment (Park et al., 2014; Yu et al., 2014).

Hand anthropometry was further emphasized using different variables, such as developing predictive models of other body dimensions and hand strength. Predictive models would be necessary when only the upper limb is available for any reason, such as explosions, train/plane crashes, or natural calamities (Chandra et al., 2015). Predicting stature from hand dimensions would narrow the search area to identify the missing person in a particular population. Hence, this study result may help identify the missing person, impersonation, and any accidental death, like (fire, navy or air accident and natural disaster). The hand normative values may be helpful to plastic and reconstructive surgeons in their reconstructions of different parts of the hands. They may also be helpful to garments, gloves, and artificial limb manufacturing companies for supplying products of different sizes for different frame sizes. Moreover, hand measurements were used to predict the handgrip strength of the elderly population for specific product design applications (Nurul Shahida et al., 2016). The study finding would be beneficial for designing ergonomics facilities (Dawal et al., 2015).

In Malaysia, several studies have been done on anthropometry study. A study on anthropometric measurement among adult populations of three different ethnicities was previously done to determine the differences between Malay, Indian, and Chinese in Malaysia (Karmegam et al., 2011). In the east part of Malaysia, a study on anthropometry was conducted to estimate stature from hand measurements among the Iban population. This study provided the first forensic anthropometry database for the Iban population in Sarawak that may be useful for dismembered body identification (Zulkifly et al., 2018).

Different ethnics may show different anthropometry dimensions (Widyanti et al., 2015). In considering Sabah Malaysia's multi-ethnic population, although different researchers have studied anthropometry of hand extensively in Malaysia, there is still a paucity of data among major ethnic groups of Sabah. Under these circumstances, the present study was conducted to create baseline data for the major ethnic groups in Sabah.

MATERIALS AND METHODS

This cross-sectional study was conducted from February 2021 to January 2022. The study was designed to create baseline data for normative values of hand length, handbreadth, middle finger length, second inter-crease length of the middle finger, and hand span.

Selection Criteria

Upon selecting the participants for the study for obtaining more valid information, specific inclusion criteria were imposed in selecting the participants. The participants qualified for being included in the research by fulfilling the following criteria:

(a) The age range must be 18 – 45 years old.
(b) They are from Kadazandusun, Bajau, Malay, or Chinese ethnicity.

The exclusion criteria were:

(a) Individuals who have medical conditions (for example, Rheumatoid arthritis, Parkinsonism, and any other conditions) that may affect hand anthropometry.
(b) Individuals having parents or grandparents who were not from the same ethnic group.

Sample Size

The sample size per cell has been determined using the procedure outlined in International Organization for Standardization (ISO) 15535: 2003. This is the general requirement for establishing an anthropometric database. Following the requirement, the sample size has been determined by using the following formula:
$n = (Z \times CV/a)2 \times 1.534 = (1.96 \times 2.87/1)2 \times 1.534 = 46$

Here, the coefficient of variance (CV) of hand length was 2.87% in a pilot study on the same population, a = 1 (The level of precision was chosen because that is the best level of interobserver error that experienced measurers have achieved), and the constant of 1.534 was based on converting the sample size formula from estimating confidence at the mean to estimating confidence at the 5th and 95th percentile (ISO 15535).

Therefore for each stratum, the study required a minimum of 46 subjects which amounts to total respondents of (46 × 2 × 4) 368 for four major ethnic groups' males and females.

Study Population

The Sabahan population consists of multi-ethnic groups. Kadazandusun, Bajau, Murut, and Malay ethnic groups are the majority among the ethnic groups in the Sabahan population. At the same time, the Chinese made up the largest non-indigenous group in Sabah (Sabah State Government, 2022). This study included all these ethnic groups to create baseline data of hand anthropometry for each of these ethnicities. The study

included subjects based on ethnic groups where the subjects' parents and grandparents were from the same ethnic groups. Ensuring this criterion required subjects coming from specific districts of Sabah, as mostly Bajau can be found at Kudat, Tuaran, Kota Belud, and Papar, while Kadazandusun and Malay can be found at Tuaran, Ranau, Papar, and Tamparuli, and Chinese can be found in Kudat. However, during the COVID-19 global pandemic, data collection at the community level was not permitted. The researchers carried out the study at the university campus among the students who fulfilled the inclusion criteria and completed vaccination for COVID-19.

Sampling Techniques

Before sampling, a list of names and their permanent address was obtained from the university's academic services division (Bahagian Perkhidmatan Akademik). The researchers applied a stratified random sampling method. There were two types of strata in the sampling: ethnicity and gender. At first, the students were stratified into four ethnicities and were further stratified as males and females. Then forty-six subjects weres randomly selected from each stratum. The randomization process was a list of metrics numbers picked randomly from a container until each gender and the ethnic group met the intended sample size.

Procedure

The study design, objective, and methodology were explained to the respondent, and informed consent was obtained from them. Five hand dimensions were measured using mechanical digital callipers with an accuracy of 0.01 mm. The researcher started data collection only after the measurements were reliable, precise, accurate, and valid (Table 1). The Intra-class Correlation Coefficient (ICC) for all the parameters ranged from 0.972 to 0.998. The values of the ICC indicate the significant ($p<0.001$) correlation between the two readings for each parameter taken by the researcher. Researchers suggested that the lower the technical error of measurement (TEM) obtained, the better the appraiser's precision to perform the measurement (Arroyo et al., 2010). The TEM of the breadth of the hands and wrist circumference was the lowest (0.071). However, all the instruments demonstrated an acceptable (<1.5%) relative technical error of measurement (rTEM) value for all measurements (Pederson & Gore, 2004). Again based on the coefficient of reliability (R values >0.95), the measurements were sufficiently precise (Ulijaszek & Kerr, 1999).

Table 1 Hand dimensions and reliability, precision, accuracy, and validity test of the measurements

Variable		Mean±SD (cm)	ICC	TEM	rTEM (%)	R
Hand length	Right	17.48 ± 0.76	0.998*	0.036	0.21	0.998
	Left	17.55 ± 0.82	0.998*	0.037	0.21	0.998
Handbreadth	Right	8.04 ± 0.48	0.997*	0.027	0.33	0.997
	Left	7.93 ± 0.52	0.996*	0.033	0.42	0.996
Middle finger length	Right	7.43 ± 0.39	0.995*	0.025	0.34	0.996
	Left	7.51 ± 0.38	0.994*	0.035	0.46	0.992
2nd Interphalangeal length	Right	2.45 ± 0.20	0.987*	0.022	0.90	0.987
	Left	2.50 ± 0.16	0.972*	0.026	1.05	0.972

* Significant at $p<0.001$ level, ICC: Intra-class Correlation Coefficient, TEM: Technical Error of Measurement, rTEM= Relative Technical Error of Measurement, R: Coefficient of Reliability

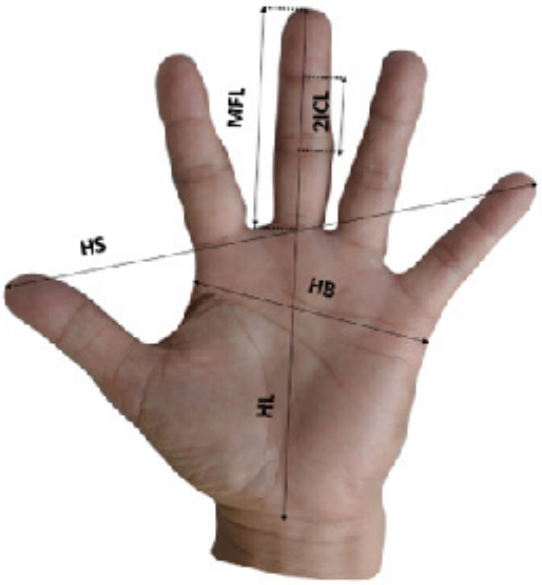

Figure 1 Hand measurement [HB: Handbreadth, HL: Hand length, HS: Hand span, MFL: Middle (third) 2ICL: finger length: 2nd inter crease length of the middle (third) finger]

Table 2 Operational definition for different hand dimensions

Hand dimension	Operational definition
Hand length (HL)	The hand's length was measured as the straight distance from the midpoint of the distal wrist crease to the most distal point of the middle finger (Sanli et al., 2005).
Handbreadth (HB)	The hand's breadth was measured as the hand's width from the lateral surface of metacarpal II to the medial surface of metacarpal V. The hand's breadth was measured at the level of the knuckles (Sanli et al., 2005).
Middle (third) finger length (MFL)	Measurement of the middle finger was taken from the proximal finger crease of the middle (third) finger to the tip of the middle (third) finger (Mollayousefi, 2008).
2nd inter crease length of the middle (third) finger (2ICL)	2nd inter crease length (middle phalanx) can be measured as the distance between the middle and distal finger creases (Hossain, 2009).
Handspan (HS)	Handspan was measured on the right hand from the tip of the thumb to the tip of the small finger, with the hand spreading as wide as possible (Ruiz et al., 2006).

Statistical Analysis

Data were analysed using Statistical Package for Social Science (SPSS) version 24.0. As the data were normally distributed (Table 3 and Figure 2), mean and standard deviation were used for describing the hand dimensions, paired t-test was performed to explore the difference between hand dimensions of both sides of the body, unpaired t-test was conducted to investigate the difference between the genders and one-way ANOVA was used to test the difference between ethnic groups.

Table 3 Descriptive statistics of hand dimensions

Variables (n = 368)	Min.	Lower fence	Q1	Median	Q3	IQR	Upper fence	Max.	Outlier
RHL	15.35	14.46	16.92	17.74	18.55	16.36	21.00	20.51	0
LHL	15.19	14.48	16.88	17.68	18.48	16.00	20.88	20.51	0
RHB	6.47	6.04	7.43	7.86	8.35	9.22	9.73	9.46	0
LHB	6.49	6.00	7.36	7.83	8.27	9.07	9.63	9.43	0
RMFL	6.43	6.21	7.25	7.63	7.95	6.94	8.96	8.93	0
LMFL	6.29	6.20	7.25	7.64	7.96	7.02	9.01	8.92	0
R2ICL	2.03	2.02	2.46	2.60	2.75	2.95	3.19	3.16	0
L2ICL	2.04	2.03	2.47	2.60	2.76	2.91	3.20	3.19	0
RHS	12.05	11.99	15.27	16.22	17.46	21.87	20.74	20.49	0
LHS	12.99	11.95	15.25	16.45	17.45	21.98	20.74	20.10	0

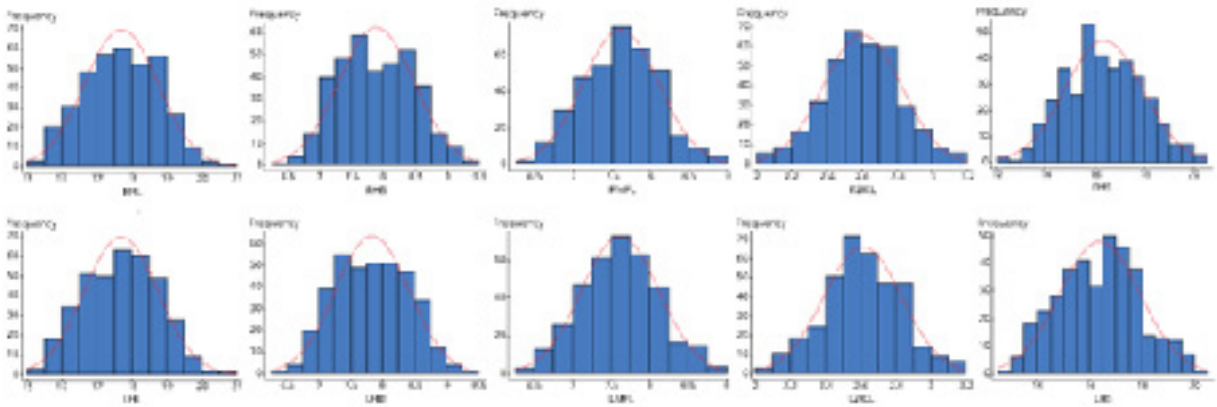

Figure 2 Histogram of the hand dimension data [RHB: Right handbreadth, RHL: Right-hand length, RHS: Right-hand span, RMFL: Right middle (third) finger length, R2ICL: Right 2nd inter crease length of the middle (third) finger, LHB: Left handbreadth, LHL: Left-hand length, LHS: Left-hand span, LMFL: Left middle (third) finger length, L2ICL: Left 2nd inter crease length of the middle (third) finger]

RESULTS

The mean and standard deviation of different hand dimensions are stratified according to ethnicity and gender in Table 4. The Malay males had the highest right and left-hand lengths and second inter-crease lengths, and Bajau males had the highest right and left handbreadths and hand spans. The Chinese males had the highest middle finger lengths. All the values of hand dimensions were noticeably higher for males than females, and there was no remarkable difference between the right and left side hand dimensions.

Table 4 Hand dimension distribution based on gender and ethnicity

	Kadazandusun		Bajau		Malay		Chinese	
	Male (n = 42)	**Female** (n = 42)	**Male** (n = 42)	**Female** (n = 42)	**Male** (n = 42)	**Female** (n = 42)	**Male** (n = 42)	**Female** (n = 42)
	Mean (±SD)	Mean (±SD)	Mean (±SD)	Mean (±SD)	Mean (±SD)	Mean (±SD)	Mean (±SD)	Mean (±SD)
RHL	18.45 (±0.87)	16.97 (0.82)	18.12 (±0.78)	16.88 (±0.86)	18.63 (±0.72)	16.92 (±0.76)	18.48 (±0.64)	17.16 (±0.64)
LHL	18.42 (±0.86)	16.93 (±0.78)	18.15 (±0.73)	16.89 (±0.84)	18.65 (±0.73)	16.91 (±0.75)	18.48 (±0.64)	17.14 (±0.65)
RHB	8.41 (±0.43)	7.49 (±0.32)	8.38 (±0.40)	7.50 (±0.34)	8.26 (±0.45)	7.34 (±0.36)	8.29 (±0.36)	7.33 (±0.83)
LHB	8.32 (±0.41)	7.42 (±0.32)	8.28 (±0.43)	7.47 (±0.32)	8.24 (±0.41)	7.33 (±0.37)	8.21 (±0.33)	7.39 (±0.43)
RMFL	7.92 (±0.47)	7.30 (±0.39)	7.77 (±0.40)	7.31 (±0.45)	7.80 (±0.42)	7.34 (±0.46)	7.90 (±0.32)	7.40 (±0.37)
LMFL	7.91 (±0.50)	7.29 (±0.38)	7.84 (±0.39)	7.28 (±0.49)	8.00 (±0.40)	7.31 (±0.43)	7.90 (±0.34)	7.37 (±0.36)
R2ICL	2.70 (±0.25)	2.53 (±0.19)	2.60 (±0.23)	2.53 (±0.25)	2.71 (±0.22)	2.54 (±0.20)	2.68 (±0.15)	2.51 (±0.19)
L2ICL	2.71 (±0.25)	2.52 (±0.22)	2.63 (±0.23)	2.54 (±0.24)	2.75 (±0.21)	2.54 (±0.19)	2.69 (±0.15)	2.50 (±0.19)
RHS	17.13 (±1.20)	15.25 (±1.11)	17.56 (±1.23)	15.49 (±1.31)	17.06 (±1.16)	15.44 (±1.34)	17.15 (±1.32)	15.63 (±1.41)
LHS	17.16 (±1.20)	15.25 (±1.17)	17.57 (±1.44)	15.29 (±1.24)	16.97 (±1.13)	15.60 (±1.41)	17.12 (±1.45)	15.99 (±1.27)

RHL= Right hand length, LHL = Left hand length, RHB = Right handbreadth, LHB = Left handbreadth, RMFL = Right middle finger length, LMFL = Left middle finger length, R2ICL= Right second inter-crease length, L2ICL= Left second inter-crease length, RHS = Right hand span, LHS= Left hand span

The following hypothesis was tested to investigate the difference between right and left-hand dimensions using paired t-test was performed:

H_0: There is no difference between right and left-hand dimensions (length, breadth, middle finger length, second inter-crease length, and span).

H_1: There is a difference between the right and left-hand dimensions.

The participants were included in the study through stratified random sampling, and the number of male and female participants was more than 30 persons and was not more than 5% of the population. So, the observations were independent, and the sample fulfilled the assumptions for the intended t-test (Sullivan III, 2017).

Table 5 Paired difference between right and left-hand dimensions among the participants (n = 368)

	Mean difference (±SD)	**95% Confidence Interval of the difference**		**t**	**df**	**p-value**
		Lower	**Upper**			
RHL – LHL	0.006 (± 0.160)	–.010	.023	0.741	367	0.459
RHB – LHB	0.054 (± 0.161)	.038	.071	6.466	367	<0.001
RMFL – LMFL	–0.002 (±0.152)	–.018	.014	–0.240	367	0.811
R2ICL – L2ICL	–0.009 (±0.104)	–.020	.001	–1.692	367	0.091
RHS – LHS	–0.027 (±0.767)	–.106	.051	–0.686	367	0.493

The p-values for the t-statistics for the difference of means for hand length, middle finger length, second inter-crease length, and hand span were more than the level of significance, α = 0.05 (Table 5). Therefore, there was insufficient evidence to reject the null hypothesis. On the contrary, the p-value for the handbreadth difference was less than 0.05. So, the sample suggested sufficient evidence to conclude that the right handbreadth was significantly different from the left handbreadth.

As the data fulfilled the assumptions of normality, randomness, and independence, an independent sample t-test was performed to investigate the difference between the hand dimensions of male and female participants. The hypothesis was determined to start the investigation:

H_0: There is no difference between male and female hand dimensions (hand length, breadth, middle finger length, second inter-crease length, and span).

H_1: Male participants have higher hand dimensions than female participants.

Table 6 Difference between hand dimensions among gender

Variables	Mean Difference	Std. error difference	t	df	p-value	95% Confidence Interval of the Difference	
						Lower	Upper
RHL	1.44	0.08	17.868	367	<.001	1.28	1.60
LHL	1.46	0.08	18.467	367	<.001	1.30	1.62
RHB	0.92	0.05	19.092	367	<.001	0.83	1.02
LHB	0.86	0.04	21.853	367	<.001	0.79	0.94
RMFL	0.55	0.04	12.819	367	<.001	0.46	0.64
LMFL	0.60	0.04	14.001	367	<.001	0.52	0.69
R2ICL	0.15	0.02	6.573	367	<.001	0.10	0.19
L2ICL	0.17	0.02	7.652	367	<.001	0.13	021
RHS	1.77	0.14	13.160	367	<.001	1.51	2.04
LHS	1.67	0.13	12.440	367	<.001	1.41	1.93

The p-value for the t-statistics for the difference of means for hand dimensions of both sides is <0.001 (Table 6), which is less than the level of significance, α = 0.05. Therefore, the null hypothesis is rejected. Hence, there is sufficient evidence to state that males' hand dimensions were higher than females.

Finally, the difference in mean values of hand dimensions among the ethnicities was observed. The one-way ANOVA test was conducted to test the following hypothesis:

H_0: There is no difference in hand dimensions among the participants from Bajau, Kadazandusun, Malay and Chinese ethnicities.

H_1: At least one ethnicity have different hand dimensions than others.

Other than the samples being randomly selected and independent, the one-way ANOVA test requires that the populations from where the samples were obtained are normally distributed, and the populations must have the same variance (Sullivan III, 2017). As the assumption of normality is already satisfied, the assumption of having an equal variance and the standard deviations were compared. The largest standard deviation for each variable is smaller than twice the smallest. Thus, the requirement of equal population variances is satisfied.

Table 7 Difference between hand dimensions among ethnicities

Variables	Sum of Squares	df	Mean Square	F	p-value
RHL	5.480	3	1.827	1.649	0.178
LHL	4.763	3	1.588	1.440	0.231
RHB	1.369	3	0.456	1.312	0.270
LHB	0.611	3	0.204	0.615	0.605
RMFL	0.717	3	0.239	0.967	0.408
LMFL	0.548	3	0.183	0.695	0.556
R2ICL	0.192	3	0.064	1.269	0.285
L2ICL	0.213	3	0.071	1.359	0.255
RHS	6.434	3	2.145	0.874	0.455
LHS	6.492	3	2.164	0.921	0.431

As these p-values are more than the level of significance $\alpha = 0.05$, the null hypothesis is retained. There is insufficient evidence to conclude that there is a difference in hand dimensions among the participants from Bajau, Kadazandusun, Malay, and Chinese ethnicities.

DISCUSSION

The present study was conducted on the young adult Sabahan population from four major ethnic groups. The length of the hand, breadth of the hand, middle finger, second inter-crease length of the middle finger, and a hand span of both sides were measured by direct physical methods. The study was designed to get normative values of the variables for the young adult population of major ethnic groups of Sabah. Differences between right- and left-hand dimensions were observed. The study also investigated gender and ethnic differences in different hand dimensions. The present study was compared with the population of Egypt, Western Australia, India, Rajasthan, Bangladesh, Gujarat, Thailand, East Malaysia (Sarawak), and West Malaysia. Similarity and dissimilarity were compared with the researcher of other countries.

Comparison Between Males With Other Studies

The right hand's mean (±SD) length was 17.70 (±1.06) centimetres. Hossain Parash et al. (2022) also found similar hand lengths in their study. This result did not match with Khancan et al. (2010), Habib and Kamal (2010), Ishak et al. (2012), Dey and Kapoor (2013), Moorthy and Zulkifly (2015), Varu et al. (2016), Zulkifly et al. (2018), Kim et al. (2018), Asadujjaman(2019) and, Romphothong and Traithepchanapai (2019) where the mean (±SD) value of the length of hand was higher than the result of the present study. The left hand's mean (±SD) length was 17.69(±1.05) centimetres. This value was similar to the value found by Varu et al. (2016) and Hossain Parash et al. (2022). The left-hand length did not match with Khancan et al. (2010), Habib and Kamal (2010), Ishak et al. (2012), Dey and Kapoor (2013), Moorthy and Zulkifly (2015), Zulkifly et al. (2018), Kim et al. (2018), Asadujjaman(2019) and, Romphothong and Traithepchanapai (2019) where the mean (±SD) value of the length of hand was higher than the result of the present study.

The right hand's mean (±SD) breadth was 7.88 (±0.66) centimetres. This value coincided with that of Zulkifly et al. (2018). This result did not correspond to Khancan et al. (2010), Habib and Kamal (2010), Ishak et al. (2012), Dey and Kapoor (2013), Varu et al. (2016), Zulkifly et al. (2018), Kim et al. (2018), Asadujjaman(2019) and, Romphothong and Traithepchanapai (2019) where the mean (±SD) value of the length of hand was higher than the result of the present study. The value was not similar to Moorthy and Zulkifly (2015) and Hossain Parash et al. (2022), where the value was lower than the present study. The left hand's mean (±SD) breadth was 7.44 (±0.36) centimetres. This value did not match with Khancan et al. (2010), Habib and Kamal (2010), Ishak et al. (2012), Dey and Kapoor (2013), Varu et al. (2016), Kim et al. (2018), Asadujjaman(2019) and, Romphothong and Traithepchanapai where the mean (±SD) value of the length of

hand was higher than the result of the present study. The left handbreadth was not similar to Moorthy and Zulkifly (2015), Zulkifly et al. (2018), and Hosaain Parash et al. (2022), where the value was lower than in the present study.

The mean (±SD) length of the right and left middle finger was 7.60 (±0.51) and 7.61 (±0.51) centimetres. Both sides correspond to Zulkifly et al. (2018). This result did not match with Khancan et al. (2010), Habib and Kamal (2010), Ishak et al. (2012), Dey and Kapoor (2013), Moorthy and Zulkifly (2015), Varu et al. (2016), Zulkifly et al. (2018), Kim et al. (2018), Asadujjaman (2019), Romphothong and Traithepchanapai (2019) and, Hossain Parash (2022) where the mean (±SD) value of the length of hand was higher than the result of the present study.

The mean (±SD) second inter-crease length of the right and left middle finger were 2.60 (±0.22) and 2.61(±0.23) centimetres. This result did not correspond to Zulkifly et al. (2018), Jee and Yun (2015), and Habib and Kamal (2010), where the mean (±SD) value of the second inter-crease length on both sides was higher than the result of the present study.

The mean (±SD) of the right and left-hand span was 16.34(±1.57) and 16.37(±1.53) centimetres. The right-hand span was less than adult Bangladeshi males (Mostafiz 2011). The left-hand value could not be compared due to a lack of data.

Comparison Between Females From Other Populations

The right hand's mean (±SD) length was 17.0(±0.71) centimetres. The right hand's length was similar to Moorthy and Zulkifly (2015), Zulkifly et al. (2018), and Kim et al. (2018) and was dissimilar to Habib and Kamal (2010), Ishak et al. (2012), Dey and Kapoor (2013), and Romphothong and Traithepchanapai (2019) where the mean (±SD) value of the length of hand was higher than the result of the present study. This result did not match with Khancan et al. (2010), Varu et al. (2016) and Asadujjaman(2019) as the mean and SD(±**SD) were lower than the present study. The left hand›s mean (±SD) length was 16.96** (±0.76) centimetres. The left-hand length value corresponded to Khancan et al. (2010), Dey and Kapoor (2013), and Moorthy and Zulkifly (2015); however, it did not correspond with Zulkifly et al. (2018), Habib and Kamal (2010) and Ishak et al. (2012), where the mean and SD (±**SD) was higher than the present study. This result did not match with** Varu et al. (2016), Asadujjaman(2019), and Romphothong and Traithepchanapai (2019), where the mean (±SD) value of the length of the left hand was lower than the result of the present study.

The right hand's mean (±SD) breadth was 7.44 (±0.36) centimetres. This value coincided with Khancan et al. (2010) and Romphothong and Traithepchanapai (2019). This result did not correspond to Ishak et al. (2012), Dey and Kapoor (2013), Moorthy and Zulkifly (2015) and Kim et al. (2018), and Asadujjaman (2019), where the mean (±SD) value of the breadth of hand was higher than the result of the present study. The value was not similar to that of Varu et al. (2016) and Zulkifly et al. (2018), where the value was lower than the present study. The left hand's mean (±SD) breadth was 7.40 (±0.36) centimetres. This value match with Romphothong and Traithepchanapai (2019). This result did not correspond with Ishak et al. (2012), Dey and Kapoor (2013), Moorthy and Zulkifly (2015), Kim et al. (2018), and Asadujjaman (2019) here, the mean (±SD) value of the breadth of hand was higher than the result of the present study. This study did not match Khancan et al. (2010), Varu et al. (2016), and Zulkifly et al. (2018), where the value was lower than the present study.

The mean (±SD) length of the right and left middle finger was 7.33(±0.42) and 7.31(±0.42) centimetres. Both sides correspond to Rastogi et al. (2015). This result did not match with Ishak et al. (2012), Zulkifly et al. (2018), Asadujjaman (2019), and Romphothong and Traithepchanapai (2019), where the mean

(±SD) value of right and left middle finger were lower than the result of the present study.

The mean (±SD) of the right 2nd-inter-crease was 2.53 (±0.21). The length corresponded with Habib and Kamal (2010) and Zulkifly et al. (2018). This result did not match Jee and Yun's (2015), where the mean and SD (±SD) were higher than in the present study. The mean (±SD) of the Left 2nd inter-crease was 2.61(±0.23). This value did not match Habib and Kamal (2010), where the mean value was higher, and Zulkifly et al. (2018), where the mean value was lower than the present study.

The mean (±SD) of the right and hand span was 15.45(±1.29) and 15.53(±1.30) centimetres. Again, the paucity of data regarding the adult hand span contributed to the inability to discuss the data.

Comparison From Other Population

This present study demonstrated lower in most hand dimensions as compared to the study done by Ishak et al. (2012) in western Australia. Historically, the Bangladeshi population has diverse origins from various communities that entered this region over many centuries. According to William (2018), between 1787 and 1868, approximately 168,00 convicts from Britain and Ireland were sent to Australia. Either parents of Australian mixed or British or Irish descent have contributed to this dissimilarity. This different origin and geographical location have a strong impact on anthropometric dimensions (**İşeri** & Arslan, 2009).

The present study found some similarities and dissimilarities with other populations. A study by Varu et al. (2016), for instance, on male respondents only found similarity with the left-hand length but not with the right-hand length. For females, both right and left-hand lengths were lower than in the present study. This different finding might be explained due to the sample of the study being cadavers aged 20 years and above. The exclusion criteria were any injury, disease or anomaly, decomposed, charred or mutilated dead bodies were also excluded from the study.

Symmetry

Results from Table 5 demonstrated that the right-hand length and right handbreadth were higher than the left-hand length and left handbreadth, whereas the left middle finger length, second inter-crease length, and handspan were higher than the right side. None of these differences was statistically significant except between the handbreadths of both sides. Kulaksiz and Gözil (2002) also had similar findings. On the contrary, Plato et al. (1980) found that all right-hand dimensions were higher than the left-hand. Although the difference between the handbreadths in the present study was statistically significant, it was not very important (upper limit: 0.071). Kulaksiz and Gözil (2002) investigated five groups based on hand usage strength strong right preferent, weak right preferent, ambidextrous, weak left preferent, and strong left preferent groups. The difference was evident in the strongly right preferent group. As the main objective of this study was to create baseline data, the subjects of this study were not categorized based on their degree of hand dominance. Not categorizing the subjects might not have yielded a practically significant difference in the present study, which was found in other studies.

Gender Differences

In this study, male-female differences were statistically significant ($p<0.01$) in all hand dimensions (Table 6). The hand dimensions of males were more prolonged and broader in all hand measurements than in females. This study's findings were in line with a previous study by Shahriar et al. (2020), where the male had a more extended and broader hand dimension as compared to the female ($p< 0.05$). On the contrary, Kulaksiz and Gözil (2002) did not find any asymmetry based on gender.

These gender disparities can be attributed to how the skeletal system grows and develops as a result of genetic and environmental influences (Frayer & Wolpoff, 1985). Males have an additional two or more years of skeletal growth than females due to hormonal influence, which causes early maturity and the cessation of bone growth in females (Krishan & Sharma, 2007).

Ethnic Variation

The present study also investigated the difference between the ethnicities of Sabah. The present study found no significant difference among the Kadazandusun, Bajau, Malay, and Chinese populations among ethnicities in their hand measurements, and this finding did not correspond with Karmegam (2011). That study's result demonstrated significant differences ($p<0.05$) in most of the measurements taken between the three ethnicities (Malay, Chinese and Indian). The Malay and Chinese populations within the sample frame of the present study have different origins than West Malaysia. Among the Chinese, the Hakkas are prominent in Sabah, Cantonese, Hokkien, Teochew, Hainanese, and Shantung (Pugh-Kitingan, 2015). On the other hand, Hokkien, Cantonese, Foochows, and other groups are the majority in West Malaysia (Tan, 2005). The Malays are mostly of Bruneian and Kadayan origin (Pugh-Kitingan, 2015) while Malay sub-ethnic groups in peninsular Malaysia are Melayu Kelantan, Melayu Minang, Melayu Jawa and Melayu Bugis (Hatin et al., 2011). This difference in their origin might have played a role in the dissimilarities in the findings of the studies. Meanwhile, Numan et al. (2013) studied the significant differences in Nigerian ethnics which were only found between Yuroba and Hausas but not between Hausas and Igbos and between Igbos and Yuroba ($p< 0.05$).

Kulaksiz and Gözil (2002) opined that environmental factors such as hand activity, hormones, and brain asymmetry might influence hand preference on hand anthropometric measurements. However, the present study was limited to hand symmetry, gender, and ethnicity. The data collection at the community level was not possible. Instead, the study included the students of the university who fulfilled the inclusion criteria, which may not be representative of the major ethnic groups of Sabah. Nonetheless, the subjects were from different parts and major ethnic groups of Sabah, and they had similar lifestyles, food habits, and cultures. Again, while selecting samples, the medical conditions that might influence hand anthropometry were screened using a questionnaire, and no investigation confirmed it.

CONCLUSION

This study has provided baseline data for hand dimensions that may be useful for designing hand tools and other equipment for the Sabahan population. The data showed significant differences exist between the genders in Sabah Malaysia and other populace from other countries. Practitioners must be aware of these differences in job performance, health, and safety in the work environment when a hand device is designed from data for different gender and different population. However, to obtain complete data on the Sabahan population, further study at the community level is recommended for different age groups and ethnicities by addressing hand activity, hormones, and brain asymmetry.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest in publishing this article.

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CASE REPORT

Retroperitoneal Extraskeletal Ewing's Sarcoma in Adult

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ABSTRACT

Ewing's sarcoma is a highly malignant tumour of osseous or non-osseous origin, typically seen in the paediatric and adolescent age group. However, extraskeletal Ewing's sarcoma is an uncommon tumour. It could arise from the soft tissue in the paravertebral area, chest wall, head and neck, and retroperitoneum. Retroperitoneal extraosseous Ewing's sarcoma is even rarer, with only a few reported cases in the literature. We describe the radiologic findings of this rare retroperitoneal extraskeletal Ewing sarcoma manifested in an adult patient. Our patient is a 32-year-old gentleman who presented with abdominal pain and constitutional symptoms for two months. Imaging shows a retroperitoneal tumour with local infiltration. It was confirmed via histopathological analysis as retroperitoneal Ewing's sarcoma. We discussed the diagnostic strategy as well as the literature review of this rare disease.

INTRODUCTION

Ewing's sarcoma is a family of tumours which includes: osseous Ewing sarcoma (OES), extraskeletal Ewing sarcoma (EES), primitive neuroectodermal tumour, and Askin tumour (Murphey et al., 2013). Histologically, these tumours demonstrate crowded sheets of small round blue cells (García-Moreno Nisa et al., 2007). These lesions share the same karyotype abnormality, which is a non-random reciprocal

translocation between chromosomes 11 and 22 (t [11;22][q24;q12]). The 22q12 locus codes for EWS, while the 11q24 site codes for FL1, resulting in the EWS-FL1 fusion transcript typically seen in 85% to 95% of Ewing sarcoma (Murphey et al., 2013). Extraskeletal Ewing's sarcoma was first described by Tefft et al. in 1969 and characterized by Angervall and Enzinger in 1975 (Nishino et al., 2003). It is rare in comparison with Ewing sarcoma of bone and usually manifests in young patients, with 85% of cases detected between 20 months and 30 years old (Murphey et al., 2013). Clinically, these patients will present with pain and a large, rapidly growing soft tissue mass. The mass is often deeply seated. The commonly reported locations include the paravertebral region (32%), lower extremities (26%), chest wall (18%), retroperitoneum (11%), and pelvic region (11%) (Murphey et al., 2013). Retroperitoneal Ewing's sarcoma often presents late as the retroperitoneum is a large potential space allowing the tumour to grow to a substantial amount before symptom manifestation.

CASE PRESENTATION

A 32-year-old male patient presented with complaints of abdominal discomfort and vague pain at the left hypochondrium for two months. It was associated with significant loss of appetite, causing 15 kg weight loss within this period. Clinical examination revealed a mass over the left upper abdomen. The initial abdominal radiograph (Figure 1) shows an ill-defined soft tissue mass with no clear border at the left hypochondrium, displacing the adjacent small and large bowels. Ultrasound abdomen (Figure 2) showed a huge heterogeneous hypoechoic mass, predominantly solid with small cystic components occupying the left hypochondrium and lumbar region, extending to the epigastric region. Calcification or internal vascularity was not seen within this mass. Computed tomography (CT) scan of the abdomen (Figure 3) was performed for further evaluation, which showed a large heterogeneously enhancing lobulated mass at the left retroperitoneal region, measuring approximately 18.6 cm × 20.0 cm × 26.6 cm. There are areas of non-enhancing hypodensities within this mass, suggestive of the presence of necrotic components. It also displaced the abdominal aorta, inferior vena cava, and their tributaries laterally to the right side; however, these vessels remained patent. It causes a mass effect on the adjacent organs, including the left kidney, pancreas, and small and large bowels. However, no evidence of distal metastasis to the lung, bone and other solid organs is seen. In magnetic resonance imaging (MRI) imaging, the lobulated mass is heterogeneously hypointense on T1-weighted (T1W) images and hyperintense on T2-weighted (T2W) images with enhancement in post-Gadolinium images. There are areas within, which are hypointense on T1-weighted (T1W) images and hyperintense on T2-weighted (T2W) images with no enhancement post-Gadolinium (Figure 4), suggestive of the presence of necrotic component. It is not suppressed on spectral attenuated inversion recovery (SPAIR) sequence to suggest the presence of fat components within. It has no clear plane with the left kidney, pancreas, and left psoas muscle. However, there are no abnormal signal intensities to suggest infiltration. Ultrasound-guided biopsy of this mass was performed. Histopathology examination showed monomorphic small blue round cells displaying round to oval hyperchromatic to vesicular nuclei, fine chromatin and small inconspicuous nucleoli with scanty to clear cytoplasm. There is the presence of glycogen highlighted by PAS and PASD stain. Immunohistochemistry shows positivity of strong diffuse membranous CD99 and nuclear FLI-1. It is negative for CKAE1/AE3, CK7, CK20, Melan A, HMB45, Chromogranin A, Synaptophysin, CD45, CD56, CD34 and CD10. These features are in favour of Ewing's sarcoma. The EWSR1 gene molecular analysis was not performed for this patient, due to the unavailability of this analysis at the cytogenetics laboratory at Hospital USM. The treating oncology team was satisfied with the HPE report and radiological findings of the tumour to initiate chemotherapy.

This patient is currently undergoing neo-adjuvant chemotherapy comprising a combination of vincristine, doxorubicin, and cyclophosphamide before tumour resection. Subsequent follow-up CT and MRI studies show a good response to chemotherapy, as evident by the tumour's size reduction.

Figure 1
Abdominal radiograph showing an ill-defined mass (white arrows) at the left hypochondrium displacing the small and large bowel loops

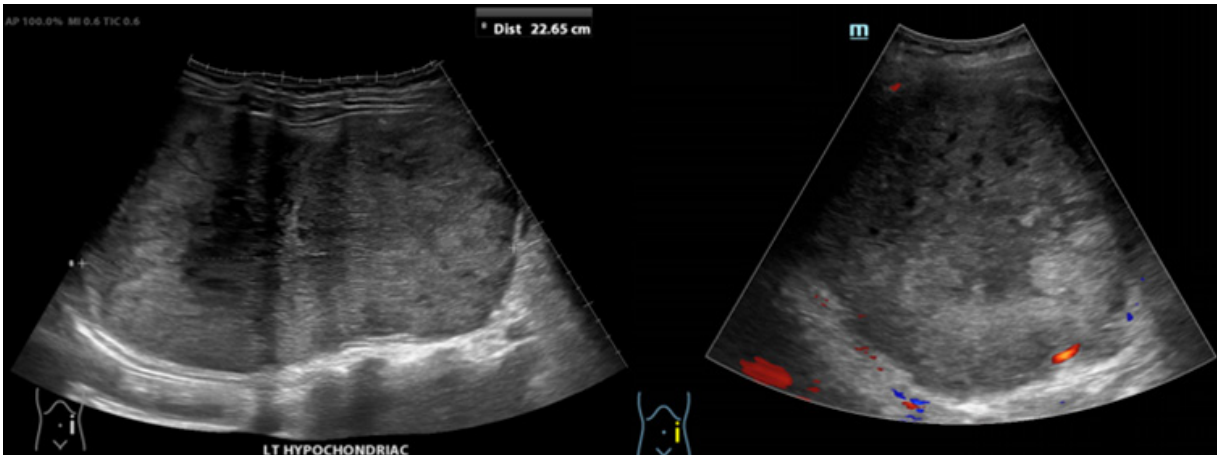
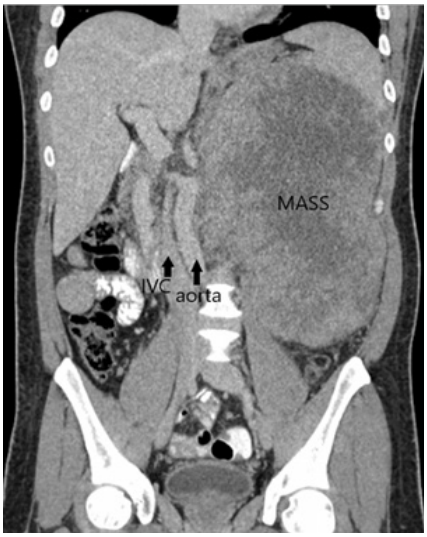


Figure 2 Ultrasound shows a retroperitoneal mass at the left hypochondrium. This mass is predominantly solid, with areas of necrosis within. No vascularity or calcification was seen within the mass.

Figure 3
A contrasted CT scan in coronal view showing a large soft tissue mass at the left retroperitoneal region. It causes a mass effect on the adjacent structures and displaces the inferior vena cava and the aorta to the right side. Areas of hypodensities that do not show contrast enhancement are in keeping with the necrotic component.



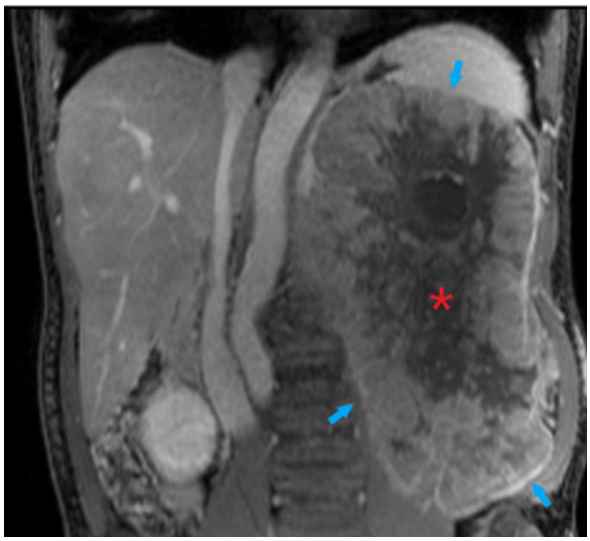


Figure 4
Post-Gadolinium coronal T1-weighted image showing the soft tissue component (blue arrows) demonstrating heterogeneous enhancement post-contrast. The non-enhancing region (labelled as *) within the mass represents the necrotic region.

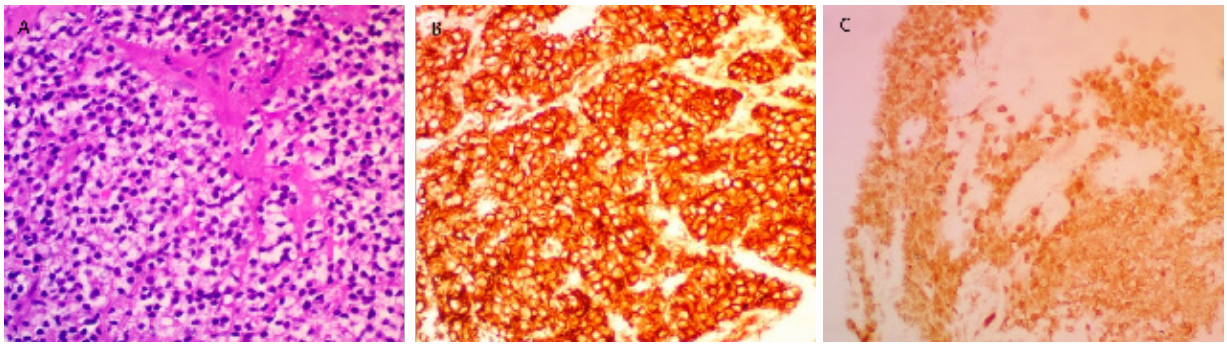


Figure 5 Histopathology examination showed monomorphic small blue round cells (A) displaying round to oval hyperchromatic to vesicular nuclei, fine chromatin and small inconspicuous nucleoli with scanty to clear cytoplasm. Immunohistochemistry shows positivity of strong diffuse membranous CD99 (B) and nuclear FLI-1(C).

DISCUSSION

OES is the second most common primary malignant tumour of bone in children and adolescents. EES is rarer compared to its osseous counterpart. The incidence of EES is 15% of that of OES. Only 11% of EES is seen in the retroperitoneal region (Murphey et al., 2013). Patients with retroperitoneal Ewing's sarcoma present with variable clinical manifestations. It depends on the size and site of the tumour as well as its invasion into adjacent structures. These tumours are made up of rapidly enlarging masses and commonly cause compression symptoms and abdominal pain, as in our patients. Often, a mass may be palpable on an abdomen examination (Javalgi et al., 2016). EES has been reported to have some different characteristics compared to OES as it does not have a predilection for male patients but rather is more equally distributed between both genders. Patients with EES are often slightly older by approximately 5 – 10 years, as in our patient. Meanwhile, OES is more common in the paediatric and adolescent age groups. EES also more commonly affects the trunk rather than the lower limbs (Murphey et al., 2013).

The initial diagnosis of EES is based on imaging modalities. In ultrasound, O'Keefe and colleagues reported that extraskeletal Ewing's sarcoma lesions are most frequently hypoechoic with anechoic areas within, likely to represent areas of necrosis. CT often demonstrates a soft tissue mass with similar

attenuation to the muscle (87% of cases). Areas of haemorrhage or necrosis can be seen within. MRI imaging is superior to CT for evaluating infiltration of the adjacent organs. MRI demonstrates a heterogeneous soft tissue mass with similar signal intensity to that of skeletal muscle on T1-weighted images (T1W) and intermediate to high signal intensity on T2-weighted images (T2W). Prominent contrast enhancement is seen in both CT and MR imaging (Murphey et al., 2013). Calcification and lymphadenopathy are rare, reported in less than 10% of the cases (Javery et al., 2011). It can displace, encase, or invade adjacent organs. However, unlike OES, it does not show osseous or marrow involvement (Murphey et al., 2013). Despite bulky disease at presentation, regional or distant metastases are uncommon (Javery et al., 2011).

Histopathology examination is essential for the definitive diagnosis of retroperitoneal Ewing's sarcoma. On histopathology, extraskeletal Ewing's sarcoma is similar to the classical Ewing's sarcoma of the bone. It shows closely packed small blue round cells arranged in sheets with round nuclei and indistinct nucleoli with a high nuclear-cytoplasmic ratio. Molecular analysis of these tumours demonstrates chromosomal rearrangements involving t(11;22) (q24; q12) (Javalgi et al., 2016).

Other more common primary retroperitoneal malignancies which should be considered as differentials include lymphoma, liposarcoma, undifferentiated pleomorphic sarcoma, leiomyosarcoma, and rhabdomyosarcoma (Mota et al., 2018). Lymphoma, the most common malignant retroperitoneal neoplasm, typically presents as a para-aortic mass encasing adjacent structures. Necrosis and calcifications are uncommon before treatment (Mota et al., 2018). Liposarcoma is the most common retroperitoneal sarcoma, affecting individuals in the fifth and sixth decades. On imaging, liposarcoma is a fat-containing lesion with multiple septa and enhancing soft tissue components. Meanwhile, undifferentiated pleomorphic sarcoma is a heterogenous lesion with a similar density to adjacent muscle, with enhancing soft tissue components and areas of calcification (Mota et al., 2018). Leiomyosarcoma is characterized by the presence of massive cystic components and the absence of fat or calcification within. It often shows the contiguous involvement of a vessel. Rhabdomyosarcoma is predominantly seen in the pediatric age group. It presents as an aggressive heterogeneous tumour with adjacent bone destruction (Mota et al., 2018).

EES is chemo and radiosensitive. Neoadjuvant chemotherapy is the standard of care before definitive surgery for localized disease. Chemotherapy consisting of vincristine, doxorubicin, and cyclophosphamide is used in most cases and has increased the long-term survival rate. This is followed by surgical resection of the primary tumour, which has been shown to improve local control and survival more than radiotherapy (García-Moreno Nisa et al., 2007).

CONCLUSION

Although rare, extraskeletal Ewing's sarcoma should be considered in young adults presenting with a large heterogeneous mass in the retroperitoneum. Imaging allows for early detection of these tumours, and histopathology examinations confirm the diagnosis. MRI imaging is essential for preoperative planning and evaluating response to treatment.

CONFLICT OF INTEREST

The authors have no competing interest to publish this case report.

CONSENT

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

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CASE REPORT

Binasal Hemianopia Due to Bilateral Internal Carotid Arteries Dolichoectasia: A Case Report and Literature Review

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ABSTRACT

Binasal hemianopia is rare. It is more commonly associated with ocular diseases than intracranial pathologies. Nevertheless, the proximity of internal carotid arteries (ICA) to the visual pathway may cause a variety of neurologic effects which may lead to visual loss. Although visual loss is uncommon, ICA aneurysm may compress the optic nerve causing visual field defect and reduced visual acuity. We report a 66-year-old male presented with asymptomatic binasal hemianopia detected during routine glaucoma screening. Computed tomography (CT) and magnetic resonance imaging (MRI) of the brain and orbit showed dolichoectasia in the cavernous segment of both ICA, which compressed the pre-chiasmatic optic nerve bilaterally. Humphrey's visual field test confirmed the diagnostic of binasal hemianopia respecting the vertical midline. This was the first reported case of binasal hemianopia without visual disturbances. This case highlights the importance of using CT and MRI as additional diagnostic tools to identify the true cause of binasal hemianopia (ICA dolichoectasia) in patients with glaucoma. A thorough assessment of any visual field defect helps to detect life-threatening intracranial pathologies effectively and is managed accordingly.

INTRODUCTION

Binasal hemianopia is a rare visual field defect with only a few reported cases (Hamann et al., 2015). Ocular and intracranial causes for binasal hemianopia have been reported with the ocular causes contributing a more substantial portion than the intracranial origin (Ashwin & Quinlan, 2006; Hamann et al., 2015). Documented reports of binasal visual field defects from the intracranial vascular disease are even rarer (Ashwin & Quinlan, 2006; Hamann et al., 2015; Salinas-Garcia & Smith, 1978). Despite its lower incidence, binasal hemianopia due to intracranial causes poses a more life and sight-threatening condition such as a cerebral aneurysm. The current report presents a complex case of asymptomatic binasal hemianopia which was discovered by confrontational visual field testing during glaucoma screening.

CASE PRESENTATION

A 66-year-old man, a high myope was detected to have mildly elevated intraocular pressure during a routine glaucoma screening. He was also found to have binasal visual field defect by confrontation visual field testing. He was then referred to our ophthalmology centre for further management. He denied any visual symptoms. Apart from high myopia, he was also pseudophakic following cataract surgery a few years prior. There was no family history of glaucoma. His systemic comorbidities include hypertension, hyperlipidaemia, and hypothyroidism.

Ocular examination showed his best corrected visual acuity was 6/7.5 in the right eye and 6/6 in the left eye. Pupillary reactions were normal. Intraocular pressures were 23 mm Hg and 24 mm Hg in the right and left eye respectively. Fundus examination showed bilateral tilted optic discs with prominent peripapillary atrophy. The right optic disc was diffusely pale. There was no obvious glaucomatous cupping (Figure 1).

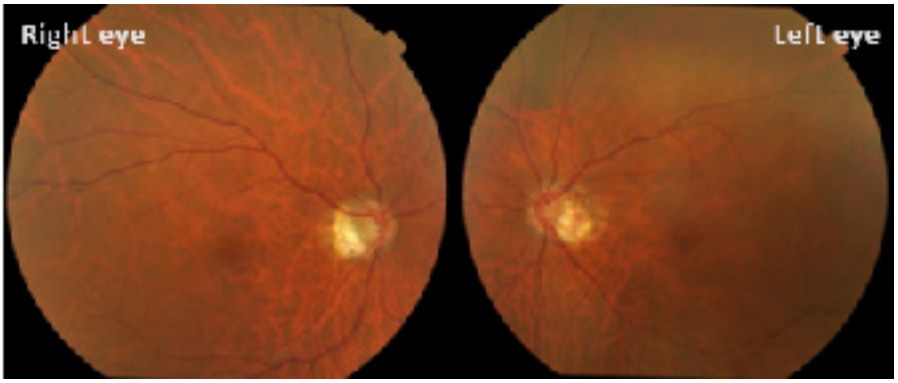

Figure 1 Fundus photographs showed a cup/ disc ratio of 0.7 (right eye) and 0.6 (left eye) with a bilateral tilted, mild pallor optic disc with thinning of the right inferotemporal rim

Humphrey visual field (24-2) test confirmed the binasal hemianopia respecting the vertical midline (Figure 2). Spectral-domain optical coherence tomography (OCT) scan showed the thinning of the retinal nerve fibre layer of the right eye, as compared to the left eye (Figure 3). Computed tomography (CT) scan of the brain and orbit revealed dolichoectasia of the supraclinoid portion of both internal carotid arteries with atherosclerotic calcifications closely related to the intracranial segments of both optic nerves. Magnetic resonance imaging (MRI) of the brain and orbit showed dolichoectasia

of the cavernous segment of both internal carotid arteries compressing onto the pre-chiasmatic optic nerve bilaterally (Figure 4 and Figure 5). He was counselled for microvascular decompression of both optic nerves. However, the patient opted for conservative management and was placed on regular neurosurgery follow-up. He was counselled on the risk of aneurysmal haemorrhage, its complications, and the potential sequelae of stroke and vision loss.

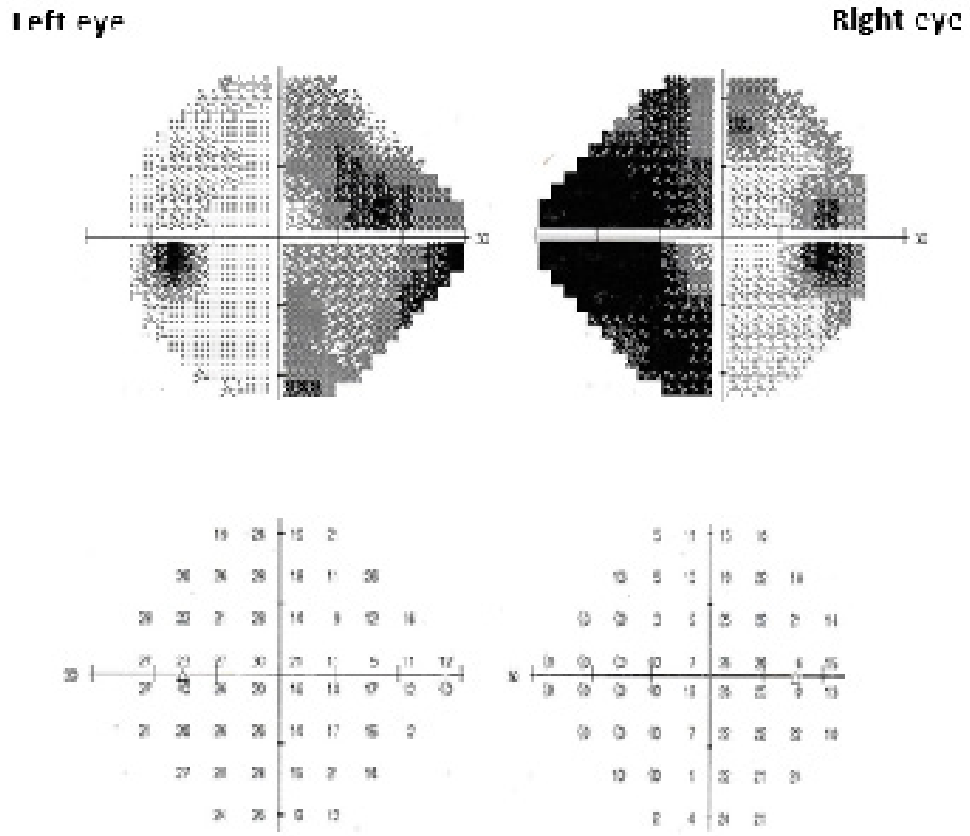

Figure 2 Humphrey's visual field showed binasal hemianopia respecting the vertical line

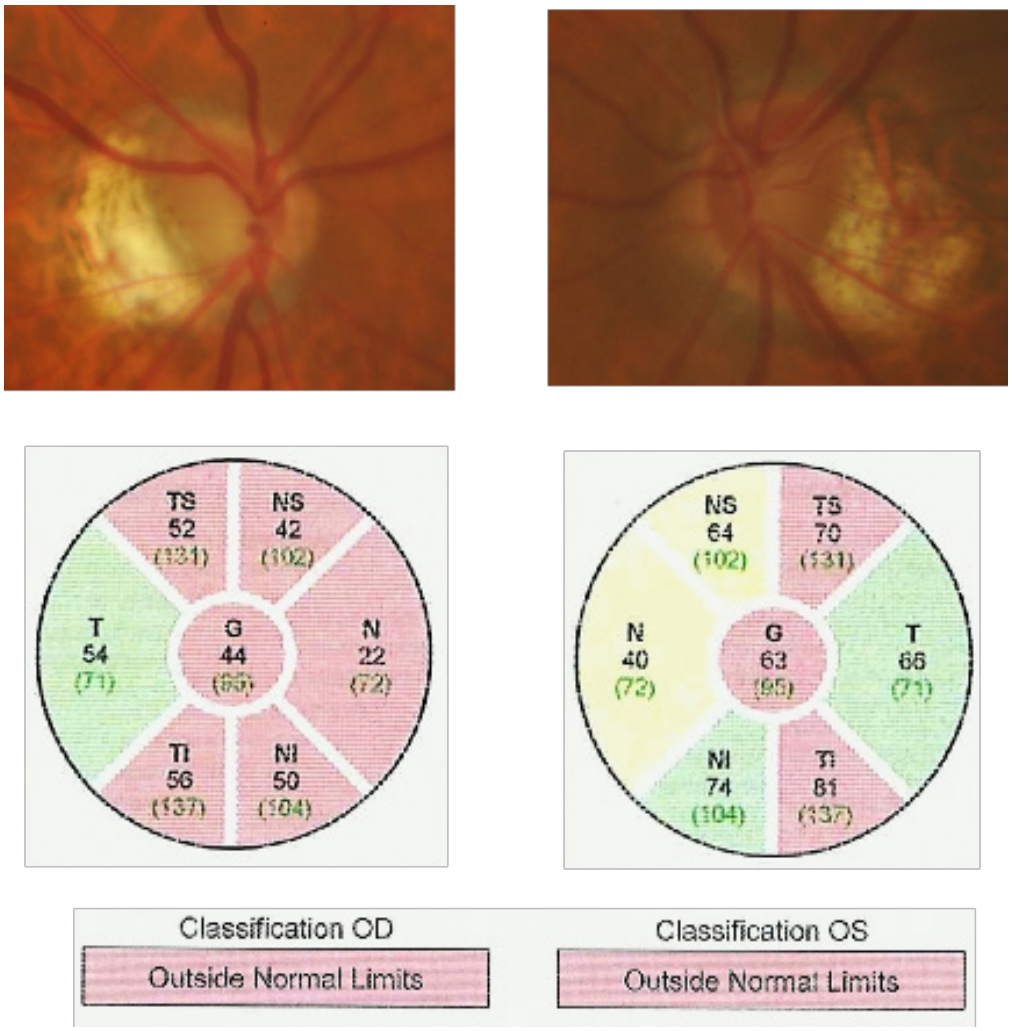

Figure 3 Fundus photographs and peripapillary retinal nerve fibre layer (RNFL) thickness using spectral optical coherence tomography (OCT). Bilateral peripapillary RNFL thinning especially on the right optic disc (upper panel). OCT revealed almost fully thinned RNFL of the right optic disc except for its temporal side with thinning on the superonasal and inferonasal of the left optic disc as denoted in red colour with thickness in μm (lower panel).

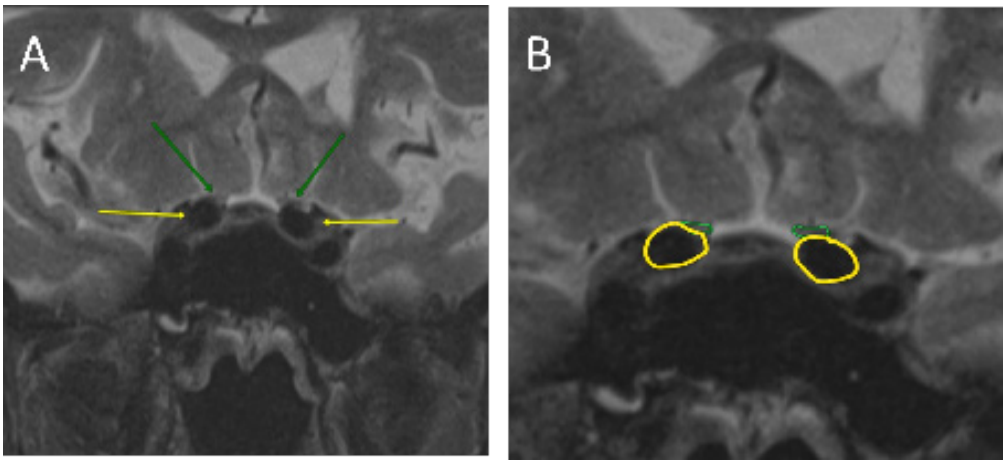

Figure 4 MRI T2WFS in Coronal view (A, B) at the level of prechiasmatic optic nerve. Bilateral prechiasmatic optic nerves (outlined green) are seen abutting the C6 segment of the internal carotid artery (ICA) bilaterally (outlined yellow)

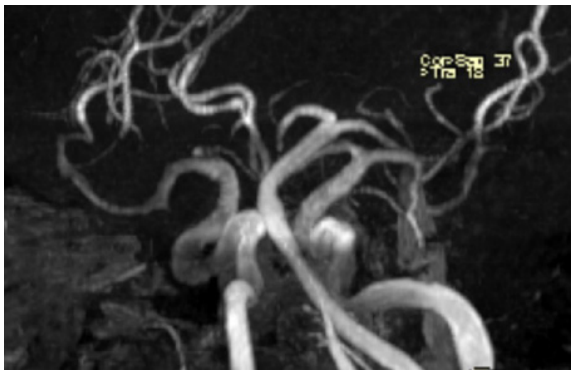

Figure 5 Magnetic Resonance Angiogram in TOF (Time of flight) – Non-contrasted in 3D reconstruction showing tortuous extracranial and intracranial ICA

DISCUSSION

Isolated binasal hemianopia is exceptionally uncommon. Various causes of ocular or neurological conditions have been reported and found to be commonly associated with ocular pathology (Hamann et al., 2015). Ashwin and Quinlan (2006) reported an incidental finding of binasal hemianopia caused by keratoconus. Another study by Salinas-Garcia and Smith (1978) found that 8 out of 100 patients referred for neuro-ophthalmologic examination had binasal visual field defects, six of them had ocular causes while the remaining two were of intracranial origin. Ischaemic optic neuropathy, optic nerve drusen, glaucoma, congenital bilateral optic nerve pits, and retinitis pigmentosa were the ocular pathologies reported. The remaining two cases were caused by congenital hydrocephalus. The mechanism involved is the distended third ventricle compresses the intracranial optic nerves laterally against the supraclinoid portion of the internal carotid arteries, giving rise to binasal visual field defect (Salinas-Garcia & Smith, 1978).

Despite the low incidence of binasal hemianopia associated with intracranial aetiologies, awareness of these neurological causes is essential due to its severe sequelae. The first documented report in 1912 reported that binasal hemianopia was found in a patient with a brain tumour (Cushing & Walker, 1915). Intracranial congenital anomalies, hydrocephalus, intracranial mass lesions, pneumosinus dilatans of sphenoid sinus, neurosyphilis and vascular causes such as pituitary apoplexy and internal carotid artery atherosclerosis were among the reported causes for binasal hemianopia (Bryan et al., 2014; Hamann et al., 2015; Kawahigashi & Nishiguchi, 2018; Kim & Kim, 2019; Lestak et al., 2011; Pringle et al., 2004; Salinas Garcia & Smith, 1978).

Cerebral aneurysms cause variable effects on the eye and visual system depending on their size and location. Bilateral aneurysm or atherosclerosis of the internal carotid arteries compresses adjacent non-crossing fibres of both optic nerves resulting in binasal hemianopia (Smith, 1905). Literature on this cause-effect relationship of binasal hemianopia with evidence of neuroimaging is still scarce. In the past, the anatomical relationship in binasal hemianopia was mostly discovered from post-mortem findings through autopsy reports with corresponding tangent screen perimetry, fundoscopy and X-rays when the patients were still alive. This occur before the advent of advanced radiological imaging such as CT or MRI scans (Knapp, 1932; Smith, 1905). Table 1 summarizes reported cases of binasal hemianopia due to intracranial vascular pathologies, demonstrating the scarcity of reports on binasal hemianopia visual field defects caused by vascular pathologies (Hamann et al., 2015; Kawahigashi & Nishiguchi, 2018; Rebolleda et al., 2015).

The hallmark of visual field defect respecting the midline seen in our patient is the tell-tale sign of a more posterior lesion in the visual pathway. At the same time, the patient demonstrated raised intraocular pressure and disc changes suggestive of glaucoma. Hence, one of the immediate differential diagnoses was primary open angle glaucoma. However, binasal hemianopia respecting the vertical midline is not a characteristic feature

of glaucomatous visual field defect. Thus, vascular abnormalities or space-occupying lesions causing binasal hemianopia became the main concern in this patient.

Peiris and Russell (1980) demonstrated a group of supraclinoid aneurysms presented with bilateral visual field defects alongside other features, especially headaches. This patient did not report any visual or neurologic disturbance suggestive of intracranial abnormalities as reported by previous authors (Table 1).

Table 1 Cases of binasal hemianopia due to intracranial vascular pathologies with neuroimaging described in previous literature

	Case Reports		
Clinical Features	**Kawagihashi et al. (2018)**	**Rebolleda et al. (2015)**	**Hamman et al. (2015)**
Age (years)	67	73	70
Gender	Male	Male	Male
Risk factors	Nil	Nil	Arterial hypertension and on a pacemaker
Symptoms	Headache, retro-orbital pain, diplopia and left ptosis	Progressive vision loss	Reduced night vision with photophobia
Fundus examination	Nil	• OD: temporal optic disc pallor • OS: normal	OU: Diffuse pallor of optic discs
Ocular findings	Left oculomotor nerve palsy evidenced by left blepharoptosis, incomplete ophthalmoplegia, slightly dilated pupil, BNH	BCVA RE 6/24, LE 6/6), positive right relative afferent pupillary defect (RAPD), BNH	Bilateral severe dyschromatopsia, BNH
OCT	Nil	Nil	• OD: Thinning of temporal peripapillary RNFL • OS: Thinning of nasal and superior peripapillary RNFL
Brain CT or MRI and other imaging	MRI findings: Pituitary apoplexy with pituitary adenoma	MRI findings: Elongation of right supraclinoid ICA compressing the right optic nerve and chiasm with right optic disc atrophy	• CT brain findings (MRI was not performed as the patient was on a pacemaker): Dolichoectasia of both ICA to optic nerves, anterior to the optic chiasm • Carotid artery duplex scan findings: 30% calcification of bilateral cervical segment of the left internal carotid artery

Due to the patient's atypical clinical features, determining the diagnosis was challenging, thus CT, MRI and MRA of the brain and orbit greatly facilitated us to identify the ectatic ICA in this patient. Diagnosis could have been different as the raised intraocular pressure suggestive of possible glaucoma could mask a more serious underlying aetiology. This neuroimaging pointed out the dolichoectasia of the cavernous segment of the internal carotid arteries which compressed the pre-chiasmatic optic nerve bilaterally. This finding led towards the true cause of binasal hemianopia which would eventually enable the appropriate management of this case.

CONCLUSION

The current report describes a rare case of binasal hemianopia secondary to bilateral internal carotid arteries dolichoectasia. Clinical features vary from acute visual loss secondary to ischaemic optic neuropathy or chronic visual loss from compressive optic neuropathy with other neurological manifestations. It can also remain silent without any visual disturbance as in the reported case. Diligent assessment of any visual field defect affecting the vertical midline may help to detect life-threatening intracranial pathologies.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest to publish this case report.

CONSENT

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

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CASE REPORT

Transient Brugada-Like Electrocardiogram Configuration Provoked by COVID-19 Fever

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ABSTRACT

There are varieties of cardiac complications which can occur in COVID-19 encompassing acute coronary syndrome, myocarditis to arrhythmia. We report a case of COVID-19 infected patient who presented with a high fever which provokes transient Type 1 Brugada-like ECG configuration. Brugada syndrome is a potentially life-threatening arrhythmia which could lead to sudden cardiac death secondary to ventricular arrhythmias. Being commonly asymptomatic, there are many triggers which could lead to its manifestations and subsequent complications. While fever, being one of the triggers, is the most common presentation of COVID-19. Besides, it could be multifactorial, either from the variant systemic complications in COVID-19 or from the management itself, especially common drugs used in resuscitation which could provoke the Brugada pattern. Looking at this association, ECG monitoring then becomes important in COVID-19, especially in those with chest pain and fever. Proper treatment for fever in COVID-19 is prudent to avoid malignant arrhythmia.

INTRODUCTION

Brugada syndrome was first published in 1992 by Pedro and Josep Brugada with a characteristic Electrocardiogram (ECG) pattern of right bundle branch block (RBBB) and persistent ST = segment elevation. It commonly presents with syncope and sudden

cardiac death due to ventricular arrhythmias (Brugada & Brugada, 1992). Mutations in SCN5A and SCN10A genes account for more than 50% of Brugada syndrome (Mizusawa & Wilde, 2012). Though its inheritance form is an autosomal-dominant trait with incomplete penetrance, most patients are undiagnosed until middle age and the average age is 40 (ranging from 1 – 77 years). It is more endemic in the Southeast Asia population and males are more prone to have this syndrome than females (8:1) (Hermida et al., 2000). Hallmark of ECG pattern in leads V_{1-3} for Brugada syndrome type 1 is coved ST-segment elevation ≥2mm followed by T wave inversion, type 2 is ≥2 mm of saddleback shaped ST-elevation or ≥1 mm of J-point elevation and type 3 is < 1mm of ST-segment elevation without any underlying structural heart disease, ischemia or electrolyte imbalance (Vohra & Rajagopalan, 2015). Fever is a very well-known cause to unmask Type 1 Brugada pattern in symptomatic patients which can lead to ventricular arrhythmia (Adler et al., 2013). COVID-19 is the current pandemic disease which is known to result in many systemic complications, including the cardiovascular system, in which there are many reports of cardiac complications encompassing acute coronary syndromes, myocarditis to arrhythmias (Wu et al., 2020). Fever is one of the most common presentations of COVID-19 infection. We report a case of COVID-19 infected patient who presented with a high fever which provokes a transient Type 1 Brugada type ECG pattern.

CASE PRESENTATION

A 49-year-old Chinese man was admitted to the hospital after presenting with a high fever, non-productive cough, breathlessness and sharp central chest pain for six days. He did not have any history of syncope, light-headedness or palpitation. He has a background history of bronchial asthma, hypothyroidism and hypercholesterolemia. He regularly takes Levothyroxine 75 micrograms once a day, Omeprazole 20 mg once a day, Simvastatin 40 mg once a night orally, Beclomethasone 100 micrograms twice a day and Salbutamol 100 micrograms four times a day metered dose inhalers. He is an ex-smoker and occasionally drinks alcohol less than 14 units per week. There is no family history of sudden cardiac death at a young age. He was born in the United Kingdom whereas his parents were originally from Hong Kong. On examination, he was fully conscious and not in respiratory distress. His initial vital signs were as follows: temperature of 38.6°C, blood pressure of 132/89 mm Hg, heart rate of 107 beats per minute, respiratory rate of 20 per minute, and oxygen saturation of 91% under room air. He needed 2 L oxygen support via nasal prong oxygen to maintain saturation above 96%. Lung examination showed coarse crackle in bilateral lower lobes. Heart sounds were normal and there was no murmur. Other system examinations were unremarkable.

SARS-CoV-2 RNA polymeric change reaction test was positive on admission. Chest radiograph and computed tomography of the pulmonary artery showed moderately extensive peripheral patchy ground glass opacities in all lobes and segments with upper lobe and posterior predominance which is consistent with COVID-19 pneumonitis (Figure 1). Initial resting ECG showed sinus tachycardia with RSR' pattern RBBB morphology, >2 mm cove-shaped J point ST-elevation with T wave inversion in V_{1-2} (Type 1 Brugada pattern) and borderline prolonged QT_C interval of 0.45 sec [normal range for male: 0.36 – 0.44 sec] (Figure 2). The subsequent ECGs were similar to admission ECGs during the intermittent spiking of fever. After three days of admission when temperature sided below 38°C, repeated ECG showed a resolution of coved-shaped ST-elevation but persistent RSR' pattern, T wave inversion in V_1 and prolonged QT_C interval of 0.49 sec (Figure 3). Initial blood investigation showed a white cell count of 5.6 (4 – 11 × 10^9/L), haemoglobin of 153 (115 – 160 g/L), platelet count of 162 (150 – 400 × 10^9/L), lymphocyte count of 1.2 (1.0 – 4.5 × 10^9/L), prothrombin 12.9 (10 – 14 s), INR 1.2 (1), D-dimer 257 (0 – 250 ng/mL), ferritin 3127 (12 – 200 mcg/L),

sodium 132 (135 – 145 mmol/L), K 3.7 (3.5 – 5.3 mmol/L), Urea 2.4 (2.5 – 6.7 mmol/L), creatinine 87 (70 – 100 μmol/L), eGFR 81 (>90), albumin 43 (35 – 50 g/L), LDH 958 (70 – 250 U/L), troponin <13 (<13 ng/L), CRP 32 (<10 mg/L), free T4 14.0 (9 – 22 pmol/L), TSH 7.46 (0.5 – 4.2 mU/L). A transthoracic echocardiogram revealed normal biventricular function with a normal size of all four chambers, and no regional wall motion abnormalities or valvular abnormalities.

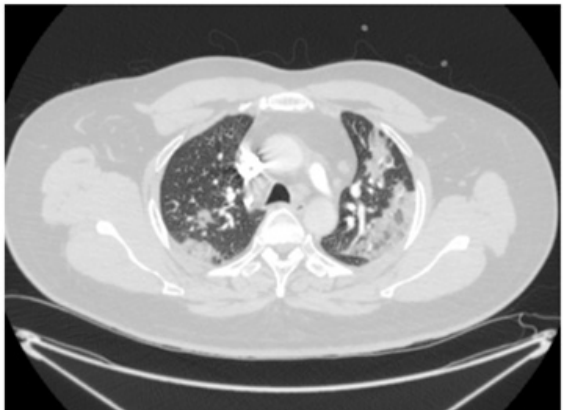

Figure 1 Computed tomography of the pulmonary artery showed moderately extensive peripheral patchy glass ground opacities in both lungs which is consistent with COVID-19 pneumonitis

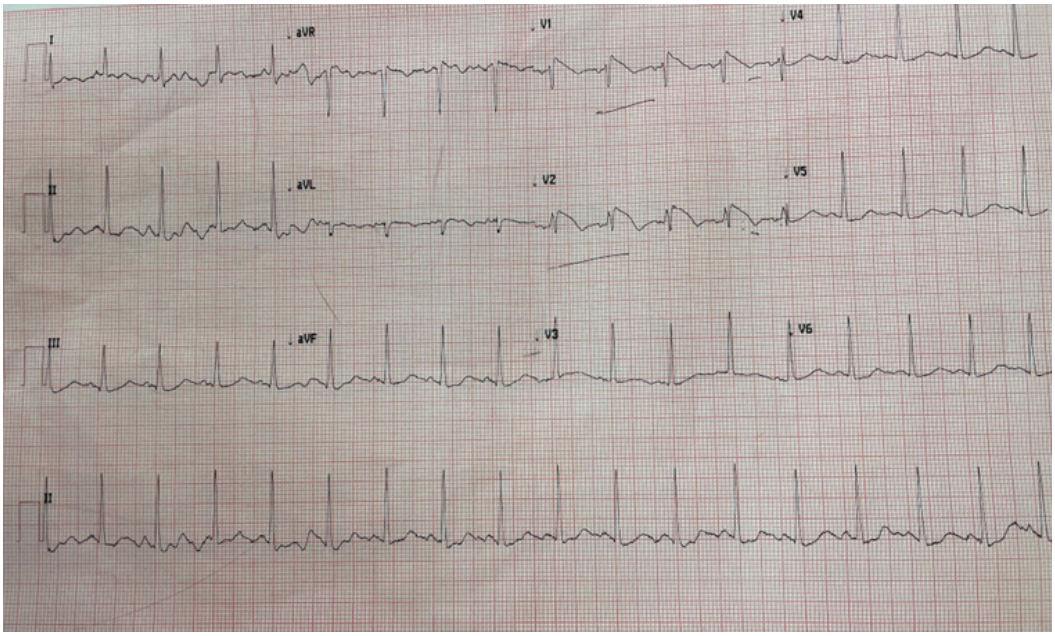

Figure 2 ECG shows sinus rhythm with partial right bundle branch block (RBBB) pattern and cove-shaped ST-elevation followed by T wave inversion in V_{1-2} (Type 1 Brugada pattern)

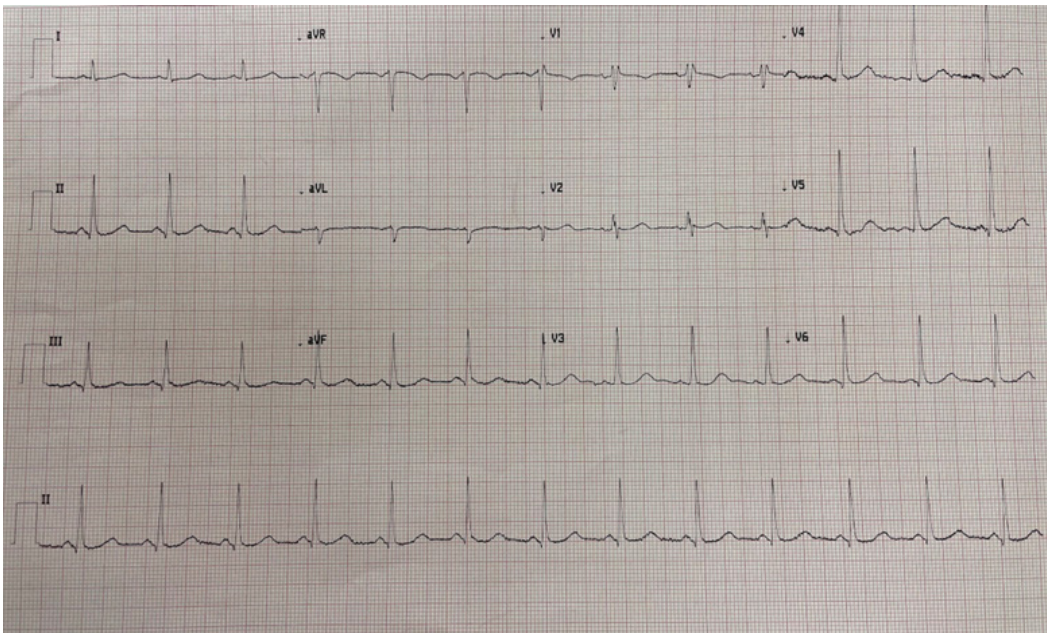

Figure 3 ECG shows normal sinus rhythm with RBBB morphology and borderline prolonged QT interval

He received intravenous antibiotics of amoxicillin 1 gram three times a day, methylprednisolone 80 mg once a day, Remdesevir 100 mg once a day, oral clarithromycin 500 mg twice a day and paracetamol 1 gram four times a day. He was put on a cardiac monitor to observe heart rhythm especially when the fever rises above 38°C. After 10 days of admission, he recovered from COVID-19 infection and was discharged with electrophysiologist follow-up in view of the absence of risk factors. He has been advised to treat fever aggressively and to avoid excessive alcohol. He was provided with a list of medications that he needs to avoid which might trigger arrhythmia. He has been explained the incidental ECG finding, the small risk of dangerous arrhythmias and sudden cardiac death.

DISCUSSION

A literature review conducted by Roomi et al. (2020) revealed that the Brugada ECG pattern (RBBB with cove or saddle-shaped ST-segment elevation in the right precordial leads) is commonly associated with pneumonia (30%) who has a fever (83%), cough (21%), sore throat (10%), and chest pain (7%). Fever can alter the ionic currents during action potential at the cardiac sodium channel during higher temperatures which can unmask the Brugada pattern (Adler et al., 2013). Brugada-like ECG pattern often disappears when the fever subsides (Baranchuk et al., 2012). With COVID-19 becoming a global pandemic and is known to cause high temperatures with respiratory symptoms, it is a new finding that fever associated with COVID-19 can unmask Brugada pattern ECG findings. A study by Wang et al. (2020) showed that arrhythmias occurred in 17% of hospitalized COVID-19 patients, which is quite significant for all clinicians to look into. The relationship between COVID-19 and arrhythmias could be attributed to multiple pro-arrhythmic factors in COVID-19, namely fever, electrolyte imbalance, stress and antiviral use (Korlipara et al., 2021). Brugada syndrome is one of the possible arrhythmias to happen in COVID-19, which should be highlighted more as it could be fatal and especially when most of the pro-arrhythmic factors are possible provoking factors for manifestation of Brugada syndrome. There are few similar case reports in patients with COVID-induced fever found to have Brugada syndrome (Kim et al., 2020;

Korlipara et al., 2021). Our case demonstrates that high fever (>38.5°C) due to COVID-19 pneumonia can induce tachycardia, Brugada pattern ST elevation and prolonged QT interval which warrants the clinician to further investigate for Brugada syndrome. One study thus recommended that in the management of COVID-19 patients, there should be a low threshold for ECG monitoring for any arrhythmias (Van de Poll & van der Werf, 2020). Wu et al. (2020) demonstrated that the risk of developing fever-induced life-threatening arrhythmia in Brugada syndrome is associated with a symptom which is syncope as well as age, in which particularly young and old patients have a higher risk. It has been recommended that patients with Brugada pattern ECG during COVID-19 infection, patients should be monitored intensively until the fever is resolved (Pasquetto et al., 2020). Also, it is important to monitor body temperature regularly and treat it appropriately with regular antipyretics to prevent fatal cardiac arrhythmia (Wu et al., 2020). Nevertheless, asymptomatic patients with fever-provoked Brugada patterns do not need antiarrhythmic drugs or intracardiac defibrillator placement.

CONCLUSION

In conclusion, a history of chest pain in COVID-19-infected patients is crucial as it may indicate COVID-19-related cardiac complications. COVID-19 fever in a middle age male is thought to be the provocative trigger for the Brugada ECG pattern, possibly due to the direct effect of the virus on the myocardial ion channel. ECG monitoring is important in COVID-19 infection, especially during high fever and should be continued monitoring till the fever subsides. Fever should not be taken lightly and should be treated aggressively and promptly in COVID-19 pneumonitis to avoid malignant arrhythmia triggered by Brugada syndrome.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest to publish this case report.

CONSENT

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

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CASE REPORT

Trichotillomania with Trichophagia and Trichobezoars: A Case Report

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Keywords: *trichotillomania, trichophagia, trichobezoar*

ABSTRACT

Trichotillomania is a condition when an individual is unable to resist the urge to pull out the hair on their head or in other places. Trichophagia is the repetitive swallowing of hair most usually connected with compulsive hair pulling. Trichotillomania with trichophagia is a rare psychiatric disease. Trichobezoar, on the other hand, has been documented in several surgical studies. It is unusual to see these symptoms appear at the same time. It can affect anyone at any age, but it is more common in adolescents, with a strong female predominance. This is a case report of an unusual case in an 11-year-old girl with all three disorders occurring at the same time and presenting with intestinal obstruction. This case report aims to identify signs and symptoms of trichobezoar, mainly in adolescents, to discover similar incidents that may link to psychiatric disorders like trichotillomania and trichophagia in medical practices.

INTRODUCTION

When compared to the previous DSM – IV, where it was classified under Impulse – Control Disorders Not Classified Elsewhere, trichotillomania is a condition in which an individual is unable to resist the urge to pull out the hair on their head or other places (DSM-4 Diagnostic and Statistical Manual, 2000). It is covered under the section on Obsessive-Compulsive and Related Disorders.

The criteria of an increased sense of stress shortly before ripping out the hair or when attempting to resist the activity were deleted in DSM-5. It has been superseded by repeated attempts to reduce or eliminate hair pulling (Christenson et al., 1991). The disease primarily affects females and adolescents, and they are subject to some but not all of the DSM-V criteria (DSM-5 Diagnostic and Statistical Manual, 2013). It is worth noting that hair-pulling can be automatic, occurring when the patient is unaware or attentive or completed when the patient is engaged in the pulling action (Grzesiak et al., 2017). Our goal is for early detection and precise diagnosis so that prompt action can be taken.

CASE PRESENTATION

An 11-year-old indigenous girl presented at the primary care clinic with acute epigastric discomfort and abdominal distension for four days. She had a history of haematemesis once but no alterations in bowel habits. She did not have a history of fever, a urinary tract infection, or an upper respiratory illness. She did not have melaena or loose stool. On examination, the abdomen was distended, with an epigastric mass measuring 4 × 5 cm and a spleen measuring 2 cm, but no hepatomegaly. Iron deficiency anaemia was discovered in initial blood testing, with haemoglobin of 8 g/dL and iron levels of 2.0 mmol/L. The blood result showed no evidence of infection with a white blood count of 10×10^3 per microlitre, neutrophils count of 50% and lymphocyte count of 40%. She was referred to the hospital for further management and plain abdominal imaging revealed the stomach and hepatic flexure appeared prominent, but there was no evident bowel dilation. Pneumoperitoneum was absent. As a result, an ultrasound abdomen was performed, which revealed an abundance of gas in the epigastric region, possibly due to gas in the pylorus or transverse colon. The CT abdomen and pelvis with oral contrast were then used to confirm the diagnosis. The CT abdomen and pelvis revealed a stomach distended with the presence of a mottled gas-patterned intragastric mass suspicious of a bezoar and a linear calcification was noted within. There was circumferential intramural wall thickening of the wall seen at the junction between the pylorus and duodenal cap suspicious of stenosis (Figure 1). The duodenum was also distended with a similar lesion which appeared to extend into the proximal jejunum (Figure 2). The CT scan concluded that the likely presence of gastric and proximal small bowel bezoar with features of gastric outlet obstruction is associated with long-standing chronic inflammation.

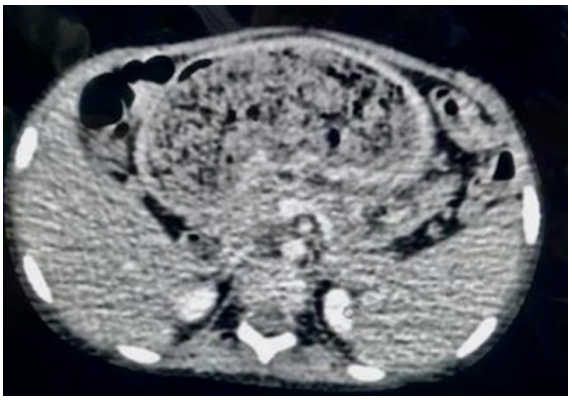

Figure 1 Axial view of abdominal computed tomography showed a hairball in the stomach

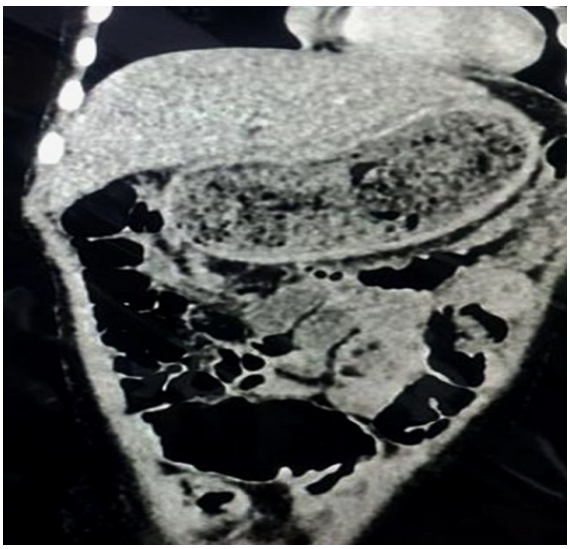

Figure 2 Coronal view of abdominal computed tomography showed the extension of a hairball from the stomach to the pylorus and duodenum

Figure 3 Sagittal view of abdominal computed topography

Pre-operatively, the patient's behaviours that may cause bezoars were thoroughly investigated. The mother noticed hair-pulling and hair-eating tendencies three years ago and tried to shave her hair to stop the behaviour but failed. The patient did not consume non-food items except for the hair. Then, an exploratory laparotomy was performed on the patient. A midline incision was made at the supraumbilical area, a gastrostomy and an enterostomy were performed, and a trichobezoar was delivered gently from the gastrotomy incision. A ball of hair from the stomach to the pylorus and duodenum (Rapunzel Syndrome) measuring 8 cm × 5 cm with an approximately 5 cm tail. Eight worms measuring 10 – 18 cm were evacuated through an enterostomy that was performed 30 cm from the duodenojejunal junction. The duodenum appeared normal, and stomach dissection revealed a 0.5 cm × 1.0 cm benign-looking ulcer on the body of the stomach with no biopsy performed. The incisions were repaired, and the rectus muscle and skin were closed with sutures. The parasitology findings of the worms resulted as *Necator americanus* is a species of hookworm infection.

During her psychiatric evaluation, she maintained a distance from the healthcare professionals while remaining close to her parents. Her assessment involved full background history, home environment, and mental state examination. On the social skills assessment, she did not interact with the doctor and appeared anxious when separated from her parents. She had poor eye contact and mostly talked to her mother. On further history, she has refused to attend school since she was eight years old for an unknown cause, and her mother has noted that she has acquired hair-pulling and chewing behaviour since then. Apart from the hair tugging and eating behaviours, the mother observed no other abnormality. A Goodenough Draw-a-Man test was performed on her with a score of 5.25. It indicated her mental age despite her actual age of 11. Her scalp was shaved with many coin-sized bald spots visible upon examination of her scalp. After the evaluation, she was diagnosed with trichotillomania, trichophagia, and borderline intellectual disability. She was started on a psychotherapy regime involving a psychiatrist, clinical psychologist, and occupational therapist, which included cognitive and behavioural therapy, reversal habit, and skill stimulation therapy. Her parents showed full cooperative support and affection throughout the process.

She and her parents attended all scheduled follow-ups throughout her follow-up and monitoring. She gradually reduced hair pulling, with no medicine prescribed for her. She was also able to restrain herself from eating the hair after pulling them with the help of close supervision from her parents. Her scalp examination showed old bald spots covered with minimal fine hair; no new bald spots were seen.

DISCUSSION

Trichotillomania commonly develops between 12 and 13 years old and is difficult to manage (Klobucar et al., 2018). The patient's atypical

symptoms delayed recognising the correct diagnosis and addressing the fundamental cause. Due to intellectual disability, acquiring an accurate history of the patient's disease is challenging. Her intellectual impairment, which hinders her from verbalising her feelings, may contribute to her.

Trichophagy has been found in more than half of trichotillomania cases (Malhotra et., 2008). In extreme trichotillomania cases, trichophagy and trichobezoar should be ruled out. In this case, a thorough history and examination were required to rule out trichophagy, and a USG abdomen revealed no trichobezoar. However, a CT scan of the abdomen was performed due to a mass over the stomach. It was revealing trichobezoar, allowing for the appropriate therapy for this patient. Rapunzel Syndrome, a rare form of trichobezoar extending from the stomach to the pylorus and duodenum, manifested late with gastrointestinal obstruction due to a low index of suspicion by healthcare personnel.

This girl's laparotomy revealed trichobezoar and eight worms ranging from 10 to 18 centimetres. Blood loss at the worm's intestinal attachment site caused this patient's iron deficiency anaemia. Iron and protein deficiency impairs growth and inhibits cognitive function which caused learning difficulties in this patient (Bagaskoro et al., 2017).

Worms and trichobezoar may cause intestinal obstruction. Pica is one cause of worm infestation. Pica causes people to consume non-food items like wormy dirt, causing worm infestations and iron deficiency anaemia (Al-Sharbati et al.,2003). However, this patient exhibited no signs of pica. Thus, her worm infection was likely attributable to low socioeconomic levels and water supplies. Worm infestation is still prevalent among indigenous Malaysians (Lim et al., 2019).

Trichotillomania is associated with intellectual impairment, depression, anxiety disorders, learning difficulties, enuresis, trichophagia, trichobezoar, and scalp abscess (Rehm et al., 2015). She also had comorbidities like learning disabilities and intellectual disabilities; however, she was not under any follow-up (Rothbart et al., 2013).

Habit reversal treatment (HRT) may comprise acceptance and commitment therapy and dialectical behaviour therapy for trichotillomania and trichophagia (Stanley et al., 1997). There are no first-line pharmacotherapies for trichotillomania. A recent Cochrane analysis found no evidence of a therapeutic effect for selective serotonin reuptake inhibitors (SSRIs) (Van et al., 2010). For this patient, her condition improved after behavioural treatment without pharmacological treatment.

In the treatment of trichotillomania, parents and primary care providers are critical. Early detection of intellectual disability should be improved at the primary care level to prevent the disorder from developing. As a result, the treating physician should be actively involved from the start (Woods et al., 2010).

This case demonstrates an unusual presentation of abdominal discomfort induced by trichotillomania and trichophagia complicated with trichobezoars, which should be considered while treating an adolescent with abdominal pain. Early diagnosis, intervention, and management, which include collaboration between a family medicine specialist, a paediatrician, a surgeon, and a psychiatrist, not to forget the patient's carers, as well as ongoing follow-up of the underlying ailment, are critical for averting disorder complications.

CONCLUSION

Trichotillomania should be evaluated as a differential diagnosis in a patient with sudden onset abdominal pain and a history of learning deficit. Focus history and examination are essential to aid in patient diagnosis and avoid delays in correct diagnosis and appropriate therapy, particularly with indigenous patients with a learning disability.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest to publish this case report.

CONSENT

Written consent was obtained from the patient to publish this case report. A copy of the written consent is available for review by the Chief Editor.

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FIGURE LEGENDS:

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SHORT COMMUNICATION

Whole Genome Sequence Analysis of a *Mycobacterium Iranicum*, a Newly Identified Non-Tuberculosis Mycobacteria, Strain SBH312 Isolated in Sabah, Malaysia

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ABSTRACT

In 2021, tuberculosis (TB) incidence partially rebounded but remained 12% lower compared with 2019, in which 10 million new TB cases have been reported worldwide. In Malaysia, 20 – 30% of the total TB cases were reported in Sabah. In this communication, we are reporting the whole genome sequence of *Mycobacterium iranicum* strain SBH312, isolated from a 49-year-old male suffering from a pulmonary infection. This is the first report of *M. iranicum*, non-tuberculous mycobacteria infecting humans, from Sabah, state of Malaysia. Phylogenetic analysis showed that the *M. iranicum* strain SBH312 was closely related to strains from Iran and Peninsular Malaysia.

INTRODUCTION

Mycobacterium iranicum is a newly identified non-tuberculous mycobacteria first reported from Iran in 2013 (Shojaei et al., 2013). Recently it has been proposed to move this species to the new genus *Mycolicibacterium* (Gupta et al., 2018). Till now, this pathogen has been reported in Russia (Lyamin et al., 2020), the USA (Kalra et al., 2022), Spain (Balakrishnan et al., 2013), France (Grandjean et al., 2017), Germany (Becker et al., 2018), Italy, Sweden, Netherlands, Greece (Shojaei et al., 2013), Saudi Arabia (Varghese et al., 2017), China (Zhang et al., 2019), Japan (Inagaki et al., 2016), and Malaysia (Tan et al., 2013) in patients

with pulmonary infections. However, it is not restricted to pulmonary infections but has also been detected to cause septic arthritis and tenosynovitis, bacteremia (Grandjean et al., 2017), skin granuloma (Becker et al., 2018), and peritonitis (Inagaki et al., 2016). Here, we report a strain of *M. iranicum* for the first time isolated in Sabah, its whole genome sequence analysis and relatedness with strains isolated from other places.

MATERIALS AND METHODS

A 49-year-old male from Kota Kinabalu, Sabah, Malaysia had suspected pulmonary tuberculosis in April 2017 detected through a TB mobile programme after the patient showed general symptoms of tuberculosis infection, including feelings of sickness or weakness, weight loss, fever, night sweats, coughing and chest pain. Sputum was collected and the patient was positive for *Mycobacterium tuberculosis* complex by GeneXpert MDR/RIF (Cepheid, Sunnyvale, CA, USA). The sputum was cultured in 7H9 Middlebrook medium using BACTEC MGIT 320 (Becton-Dickinson, Oxford, United Kingdom).

For further understanding and identification of the strain infecting the patient, whole genome sequencing was performed. Genomic DNA was extracted using Masterpure Complete DNA and RNA purification kit (Epicenter, Inc., Madison, Wisconsin, USA) according to the manufacturer's instructions. The concentration and quality of the extracted DNA were determined by Nanodrop 2000c spectrophotometer (ThermoFisher Scientific, USA) and Qubit® 2.0 fluorometer (Invitrogen, ThermoFisher Scientific, USA), respectively. The genome DNA was sequenced using the Illumina HiSeq 4000 paired-end sequencing platform.

Genomic bioinformatics analysis was performed, including genome assembly, annotation, and phylogenetic analysis based on a few genomes to identify closely related strains. The quality of the sequence read was checked by FastQC. All the raw reads were pre-processed using BBMap version 38.43 tools (Bushnell et al., 2017), whereby the adapters were trimmed and the reads <50bp were removed, based on the Phred with a quality below Q30 using BBDuk.sh. *De novo* assembly was performed using SPAdes version 3.11.1 (Bankevich et al., 2012) software. NCBI Prokaryotic Genome Annotation Pipeline (PGAP) (Tatusova et al., 2016) was utilized to annotate the generated contigs.

A total of 11 genomes of *M. iranicum* strains were extracted from GenBank for SNP-based phylogenetic analysis, and core-SNP was generated by the kSNP3 package (Gardner et al., 2015). The entire SNP matrix was used for phylogenetic analysis, using the maximum likelihood method available in MEGA X (Sudhir et al., 2018). The analysis used a general time-reversible (GTR) model, nucleotide sequences were aligned and the maximum likelihood method was used. The *Mycobacterium tuberculosis* strain H37Rv was used as an outgroup. The significance of branching was assessed by bootstrap analysis of 1,000 replicates. The numbers adjacent to nodes represent the bootstrap values (%); values less than 70% have not shown in this figure The scale bar shows the genetic distance, which is expressed as nucleotide substitutions per site. This study was approved by the ethics committee at the Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah [JKEtika 2/16 (6)].

RESULTS

The genome was sequenced until 99% completion of the genome using 332 times sequencing coverage reads. A total of 4,886,925 paired reads (~1GB) of a 300-bp insert-size library was generated from Illumina HiSeq 4000. The data sequence was deposited in the Sequence Read Archive (accession number SRR25526524) under biosample accession number SAMN32036409 and bio project accession number PRJNA908658. This whole-genome shotgun project was deposited at DDBJ/ENA/GenBank under Accession No. JAPQYE00000000.1.

De *novo* assembly generated 90 contigs of 99% of the draft genome, with a genome size of 6,516,536 bp and an N_{50} value of 404,830 bp. The GC content of the genome was 66.07%. The genome consisted of a total of 6,331 genes, including 6.275 predicted coding sequences (CDSs) and 56 RNAs (46 tRNAs and 10 rRNAs).

Phylogenetic analysis showed that *M. iranicum* strain SBH321 clustered with strains DSM45541 (LQPC00000000.1) from Iran and UM_TJL (AUWT00000000.1) from Malaysia (Figure 1).

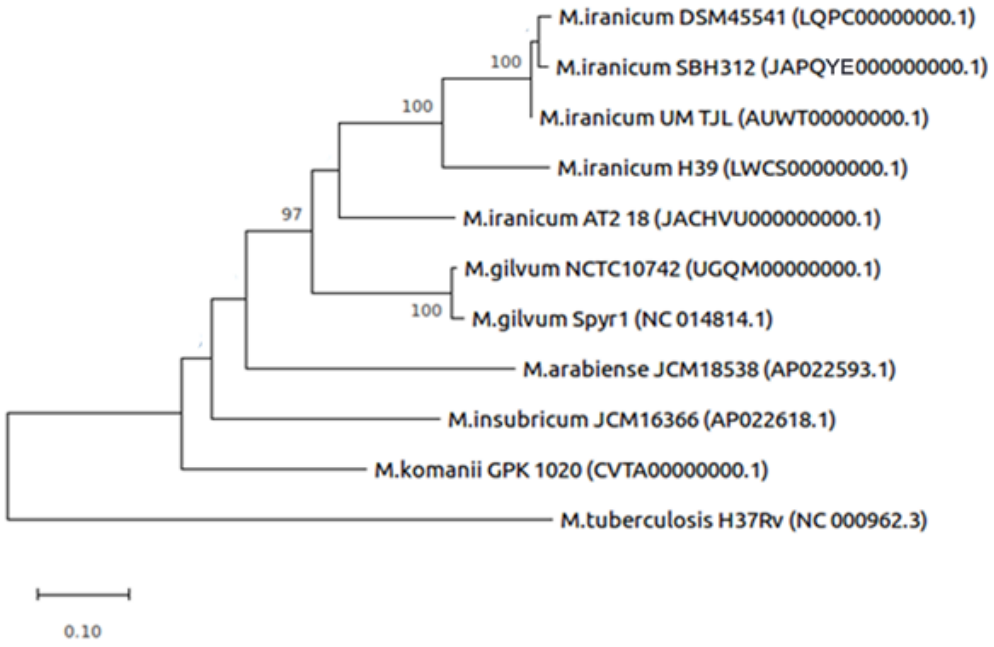

Figure 1 Phylogeny of Mycobacterium species including *M. iranicum* SBH312 strain clustering with strain DSM45541 (LQPC00000000.1) from Iran and UM_TJL (AUWT00000000.1) from Malaysia

DISCUSSION

Genus *Mycobacterium* has been further categorized into strict pathogens (*Mycobacterium tuberculosis* complex) and potential pathogens (non-tuberculous mycobacteria, NTMs) (Chai et al., 2018). NTMs, also known as environmental mycobacteria, are widely distributed in the environment and are passed on to humans by ingestion, inhalation, and inoculation from such sources. *M. iranicum* is a species of the phylum actinobacteria (Gram-positive bacteria with high guanine and cytosine content, one of the dominant phyla of all bacteria), and belongs to the genus *Mycobacterium*.

The comparative genomic analysis of Malaysian clinical isolate against representative mycobacterial species suggests its environmental origin that might have evolved into a consequential human pathogen (Tan et al., 2014). *M. Iranium* not only has been isolated from the environment (Lymperopoulou et al., 2017) but has also recently been isolated from animals (Dibaj et al., 2018), indicating the possible emergence of a new zoonosis. There are a few strains of *M. iranicum* reported as clinical isolates from clinical specimens such as M05 (HQ009482) (Shojaei et al., 2013) and UM_TJL (AUWT00000000.1) from Malaysia (Tan et al., 2013; Tan et al., 2014). This report shows that *M. iranicum* is a new species of NTM, which may infect people by producing similar symptoms as *M. tuberculosis*. More studies are needed to determine whether our strain is from zoonotic or environmental sources.

CONCLUSION

A new strain of *M*. iranicum, SBH312 was isolated from a suspected pulmonary tuberculosis patient from Sabah, Malaysia. This strain was closely related to the strains from Iran and Peninsular Malaysia. This *M. iranicum* needs further clarification of its status as an emerging human pathogen. Also, the clinical significance of this *M. iranicum* is still unclear.

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