Malaysian Family Physician

www.e-mfp.org

Official Journal of the Academy of Family Physicians of Malaysia and Family Medicine Specialist Association

2016 Volume 11 No. 1

- ***Aetiological profile of paediatric stridor in a Malaysian tertiary hospital.***
- ***Prevalence of falls among community-dwelling elderly and its associated factors: A cross-sectional study in Perak, Malaysia***

PP2089/12/2012 (031677)
ISSN :
1985-207X (Print)
1985-2274 (Electronic)

About MFP

The Malaysian Family Physician is the official journal of the Academy of Family Physicians of Malaysia. It is published three times a year.

Circulation: The journal is distributed free of charge to all members of the Academy of Family Physicians of Malaysia and the Family Medicine Specialist Association. Complimentary copies are also sent to other organisations that are members of the World Organization of Family Doctors (WONCA).

Subscription rates:
Local individual rate: RM60 per issue
Local institution rate: RM120 per issue
Foreign individual rate: USD60 per issue
Foreign institution rate: USD120 per issue

Advertisements: Enquiries regarding advertisement rates and specimen copies should be addressed to the Secretariat, Academy of Family Physicians of Malaysia. Advertisements are subject to editorial acceptance and have no influence on editorial content or representation.

All correspondence should be addressed to:
The Editor
The Malaysian Family Physician Journal
Academy of Family Physicians of Malaysia,
Suite 4-3, 4th Floor, Medical Academies of Malaysia,
210, Jln Tun Razak, 50400 Kuala Lumpur, Malaysia
Email: afpm@po.jaring.my
Tel: +60340251900
Fax: +60340246900

MFP is indexed by:
DOAJ, EBSCOHOST, EMCARE, Google Scholar, Open J-Gate, MyAIS, MyCite, Proquest, PubMed Central, Scopus, WPRIM

Editorial Board

Information for Authors

The Malaysian Family Physician welcomes articles on all aspects of Family Medicine in the form of original research papers, review articles, case reports, evidence-based commentaries, book reviews, and letters to editor. The Malaysian Family Physician also welcomes brief abstracts of original papers published elsewhere but of interest to family physicians in Malaysia.

Articles are accepted for publication on condition that they are contributed solely to the Malaysian Family Physician. Neither the Editorial Board nor the Publisher accepts responsibility for the views and statements of authors expressed in their contributions. All papers will be subjected to peer review. The Editorial Board further reserves the right to edit and reject papers. To avoid delays in publication, authors are advised to adhere closely to the instructions given below.

SUBMISSION OF MANUSCRIPTS

All manuscripts must be submitted through the Open Journal System (OJS) at http://e-mfp.org/ojs

Format:

1. The manuscript should be submitted in electronic copy only and in Microsoft Word.
2. Please include a section on 'How does this paper make a difference to general practice' (for original article only). This section should be written in bullet points (up to five points) and must not exceed 100 words.
3. Please include all authors' email address.

Cover letter must be signed by the corresponding author on behalf of all authors. This letter must include this statement "this manuscript is my (our) own work, it is not under consideration by another journal, and this material has not been previously published."

All authors must sign the declaration form and submit it together with the manuscript and cover letter. Please download the form (http://e-mfp.org/wp-content/uploads/2014/02/MFP-author-declaration-form-v3.pdf).

PREPARATION OF THE MANUSCRIPT

The following information must be given in the manuscripts:

- Corresponding author's mailing address, designation, institution and contact details (email, telephone and fax numbers).
- The full names, professional qualifications (limited to two only) and institutions of all authors. In addition, a shortened name of author(s) should be written in the style of surname or preferred name followed by initials, e.g. Atiya AS, Rajakumar MK, Hee WJ, for future indexing.
- A statement indicating whether the study had received any funding support and ethical approval (if so, please provide the specific information).
- A declaration of conflicts of interest by all authors.
- In the preparation of your manuscript, please follow the Uniform Requirements for Manuscripts Submitted to Biomedical Journal as recommended by the International Committee of Medical Journal Editors (http://www.icmje.org/urm_full.pdf). In addition to the above, the suggestions below and a few "house rules" also apply.

Type and length of manuscript

1. ***Review (CME) article:*** A comprehensive review of the literature with synthesis of practical information for practising doctors is expected. Length should not exceed 4000 words with a maximum of 30 references. An abstract is required (may be in the form of key learning points). Please provide 3-5 keywords or short phrases (preferably MeSH terms).
2. ***Original article:*** The original research should be conducted in the primary care setting on a topic of relevance to family practice. Length should not exceed 3000 words with maximum of 5 tables or figures and 20 references. An abstract is required (preferably a structured abstract of no more than 250 words) together with the keywords. Both qualitative and quantitative studies are welcome.
3. ***Case report:*** Case reports should preferably be less commonly seen cases that have an educational value for practising doctors. Length should not exceed 1000 words and no more than 10 references. Before submitting the case report, the authors must ensure that the patient's identity is protected both in the text and pictures.
4. ***Evidence-based commentary:*** These are short reports based on a focused question arising from a clinical encounter, and accompanied with a summary of the appraised evidence.

Guide for the preparation of an original article

1. ***Text:*** Author(s) should use subheadings to divide the sections of the paper: Introduction, Methods, Results, Discussion, Acknowledgments, and References. Do not justify the paragraph of the text (i.e. no need to straighten the left margin).
2. ***Introduction:*** Clearly state the purpose of the article with strictly pertinent references. Do not review the subject extensively.
3. ***Methods:*** Describe the study in sufficient detail to allow others to replicate the results. Provide references to established methods, including statistical methods; provide references and brief descriptions of methods that have been published but are not well known; describe new or substantially modified methods, give reasons for using them, and evaluate their limitations. When mentioning drugs, the generic names are preferred (proprietary names can be provided in brackets). Do not use patients' names or hospital numbers. Include numbers of observation and the statistical significance of the findings. When appropriate, state clearly that the research project has received the approval of the relevant ethical committee.
4. ***Results:*** Present your results in logical sequence in the text, tables and figures. Tables and figures may be left at the respective location within the text. These should be numbered using Arabic numerals only. Table style should be "Simple" (as in Microsoft Word). Do not repeat in the text all the data in the tables or figures.
5. ***Discussion:*** Emphasise the new and important aspects of the study and conclusions that follow from them. Do not repeat data given in the Results section. The discussion should state the implications of the findings and their limitations and relate the observations to the other relevant studies. Link the conclusions with the aims of the study but avoid unqualified statements and conclusions not completely supported by your data. Recommendations, when appropriate, may be included.
6. ***Acknowledgements:*** Acknowledge grants awarded in aid of the study as well as persons who have contributed significantly to the study (but do not qualify for authorship).
7. ***Funding and Conflicts of Interest:*** The source of funding for the study, if available, must be cited. All authors must declare their conflicts of interest.

References. Number references consecutively in the order in which they are first mentioned in the text. Identify references in text, tables and legends by Arabic numerals (in superscript). Please use the citation style adopted by the National Library of Medicine, Maryland, USA (http://www.pubmed.gov), some examples are given below. For indexed journals, the short forms for the journal names can be accessed at the PubMed website (search within Journal Database). Avoid citing abstracts, personal communication or unpublished data as references. Include among the references manuscripts accepted but not yet published; designate the journal followed by "in press" (in parenthesis). When referencing website, please include the full title and accessed date.

Some examples of citations

- Standard journal article: List up to three authors only; when four or more list only the first three and add et al. For example, Connor EM, Sperling RS, Gelber R, et al. Reduction of maternal-infant transmission of human immunodeficiency virus type 1 with zidovudine treatment. Pediatric AIDS Clinical Trials Group Protocol 076 Study Group. N Engl J Med. 1994 Nov 3;331(18):1173-80.
  Standard journal article: Corporate Author International Committee of Medical Journal Editors. Uniform requirements for manuscripts submitted to biomedical journals. N Engl J Med. 1997 Jan 23; 336(4):309-16.
- Books and other monographs: Personal Author(s) Stewart M, Brown JB, Weston WW, et al. Patient-Centered Medicine: Transforming the Clinical Method. Thousand Oaks, California: Sage Publications; 1995.
  Books and other monographs: Corporate Author WONCA International Classification Committee. International Classification of Primary Care, ICPC-2. 2nd ed. Oxford: Oxford University Press; 1998.

Policy for using any published materials

Authors must seek approval from and acknowledge the MFP if they wish to use any published materials from this journal. You can write to the Editorial Manager at email: journal.mfp@gmail.com

CONTENTS

	i	**About MFP**
	ii	**Editorial board**
	iii	**Information for authors**
Editorial	1	**Moving Forward** Liew SM
Original Article	2	**Aetiological profile of paediatric stridor in a Malaysian tertiary hospital** Lum SG, Noor Liza I, Priatharisiny V, Saraiza AB, Goh BS
	7	**Prevalence of falls among community-dwelling elderly and its associated factors: A cross-sectional study in Perak, Malaysia** Yeong UY, Tan SY, Yap JF, Choo WY
Case Report	15	**Fever and rash in an 11-month-old girl** Jamani NA, Puteri Shanaz JKC
	18	**Multiple concurrent primary extramammary Paget's disease** Leelavathi M Norazirah MN, Nur Amirah AP
	22	**Pneumoperitoneum or Chilaiditi's sign** Teoh SW, Mimi O, Poonggothai SP, Liew SM, Kumar G
	25	**Isolated unilateral mucoid sixth cranial nerve palsy–a rare presentation of dengue fever** Mazliha M, Boo YL, Chin PW
	27	**Immune reconstitution inflammatory syndrome (IRIS) in a HIV-infected patient with disseminated tuberculosis** Koh KC, Pak JW
Test Your Knowledge	31	**An elderly with a 'bony' smile** Mohamad I, Nadarajah S
	33	**A teenager with mucoid discharge from the neck** Mohamad I

EDITORIAL

Moving Forward

Liew SM

Editor-in-Chief

This issue of the Malaysian Family Physician marks a change in the Chief Editorship. Professor Dr Ng Chirk Jenn has led the Board of Editors admirably during the last four years. Under his able stewardship, the journal achieved many firsts including indexing in PubMed Central. Professor Ng will continue to serve on the Board of Editors and help guide the journal towards further success.

Although the editorship has changed, the aim of the Malaysian Family Physician remains the same. The journal will continue to act as a platform for the publication of research, guidelines and reports relevant to family medicine. Our overarching aim is to improve the translation of research and experience to improve clinical practice. To achieve this, we have expanded the Board of Editors and reviewers. Last December, a workshop was conducted at the 5th Asia Pacific Primary Care Research Conference 2015 for reviewers to improve their skills. We will strive to ensure that the journal continues to maintain high standards of ethical, methodological and professional quality.

This will be the last issue of the Malaysian Family Physician that will be printed on paper. We aim to go fully electronic in the next issue. This decision was made by taking into account the views of the board of editors and readers via a pilot survey. Going fully electronic would mean substantial savings from printing and mailing of the print copies. An online journal would offer authors greater flexibility in terms of page allowances, image colour and quality, and hasten time to publication. 1 Readers will be able to readily access the journal using mobile technology, search past issues easily and lessen their carbon footprint.

In this issue, we have 2 original research articles and 5 case reports. Lum et al conducted a retrospective review of paediatric patients presenting with stridor to determine the aetiological profiles and management. Yeong et al reported that one in every 25 elderly Malaysian living in the community experienced falls in the past year. These articles highlight the diverse range of patients that we see in primary care in terms of age and medical conditions.

I hope that all our readers, contributors, reviewers and editors will continue to support the Malaysian Family Physician. Your support of the journal is greatly appreciated and indispensable for the continuing progress of the journal.

References

1. Berquist TH. To print or not to print: are we ready for online only? AJR 2008; 191:949–950.
2. Lum SG, Noor Liza I, Priatharisiny V et al. Aetiological profile of paediatric stridor in a Malaysian tertiary hospital. Malays Fam Physician. 2016;11(1);2-6.
3. Yeong UY, Tan SY, Yap JF, et al. Prevalence of falls among community-dwelling elderly and its associated factors: A cross-sectional study in Perak, Malaysia. Malays Fam Physician. 2016;11(1);7-14.

ORIGINAL ARTICLE

Aetiological profile of paediatric stridor in a Malaysian tertiary hospital

Lum SG, Noor Liza I, Priatharisiny V, Saraiza AB, Goh BS

Lum SG, Noor Liza I, Priatharisiny V et al. Aetiological profile of paediatric stridor in a Malaysian tertiary hospital. *Malays Fam Physician.* 2016;11(1);2-6.

Keywords:
Congenital laryngeal anomaly, laryngomalacia, paediatric, stridor, synchronous airway lesion

Authors:

Lum Sai Guan
(Corresponding author)
MD (UKM), MS ORL-HNS (UKM)
Department of Otorhinolaryngology, Hospital Serdang, Jalan Puchong, 43000 Kajang, Selangor, Malaysia
Email: lumsg728@gmail.com

Noor Liza Ishak
MB BCH BAO (NUI), MS ORL-HNS (UKM)
Otorhinolaryngology and Head and Neck Surgery Unit, Universiti Putra Malaysia, Selangor, Malaysia

Priatharisiny Velayutham
MBBS, MS ORL-HNS
Department of Otorhinolaryngology, Hospital Serdang, Selangor, Malaysia

Saraiza Abu Bakar
MD, MS ORL-HNS (UKM)
Department of Otorhinolaryngology, Hospital Serdang, Selangor, Malaysia

Goh Bee See
MD (UKM), MS ORL-HNS (UKM)
Department of Otorhinolaryngology-Head and Neck Surgery, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia

Abstract

Background: Conditions causing stridor in paediatric patients can range from minor illnesses to life-threatening disorders. Proper evaluation and correct diagnosis are essential for timely intervention. The objective of this study was to determine the aetiological profiles and the management of paediatric patients with stridor referred to the Otorhinolaryngology Department of Hospital Serdang.

Methods: Medical records of all paediatric patients presenting with symptom of stridor from January 2010 to February 2015 were reviewed retrospectively. The patients' demographic data, clinical notes, laryngoscope findings, diagnosis and management were retrieved and analysed.

Results: Out of the total 137 patients referred for noisy breathing, 121 patients had stridor and were included in this study. There were 73 males and 48 females—most were of Malay ethnicity (77.7%). The age of presentation ranged from newborn to 10 years, with a mean of 4.9 months. Eighteen patients (14.9%) had associated congenital pathologies. The majority were congenital causes (90.9%), in which laryngomalacia was the commonest (78.5%), followed by subglottic stenosis (5.0%), vallecular cyst (2.5%) and congenital vocal fold paralysis (2.5%). Twelve patients (9.9%) had synchronous airway lesion. The majority of the patients were managed conservatively. Thirty-one patients (25.6%) required surgical intervention, of which only one needed tracheostomy.

Conclusion: Laryngomalacia was the commonest cause of stridor among paediatric patients. A synchronous airway lesion should be considered if the child has persistent or severe symptoms. The majority of the patients were managed conservatively.

Introduction

Noisy breathing is a common presenting symptom among paediatric patients to primary care.[1] Clinicians must be able to differentiate types of noisy breathing such as stridor, stertor, snoring and wheezing; each suggest different airway pathologies. Stridor is a musical, often high-pitched sound resulting from turbulent airflow through a partially obstructed upper airway.[1] There are various aetiologies of stridor ranging from minor illnesses to life-threatening disorders.[2] Unfamiliarity with the evaluation and management of a stridorous child may result in avoidable adverse outcomes. Therefore, proper evaluation and correct diagnosis are essential for timely intervention and management. Flexible nasopharyngolaryngoscopy (FNPLS) assessment in a stable patient provides a dynamic, real-time visualisation of the upper airway.[3] A stridorous child may be managed conservatively or by surgery depending on the severity and the underlying pathology. This study aims to determine the aetiological profiles and the management of all paediatric patients with stridor referred to the Otorhinolaryngology Department of Hospital Serdang.

Methods

From the electronic registry, the medical records of all paediatric patients aged 12 and younger who were referred for noisy breathing to the Otorhinolaryngology Department of Hospital Serdang from January 2010 to February 2015 were reviewed retrospectively. The sources of referral included primary care practitioners, district hospitals, emergency department and other departments in the studied hospital. The authors retrieved the clinical data from the electronic hospital information system (e-HIS). The patients' demographic data, clinical notes, laryngoscopy findings, diagnosis and management were reviewed. Patients without evidence of stridor or upper airway pathology after thorough assessment were excluded from

the study. All data were analysed with SPSS statistical software (SPSS Inc, version 22.0).

Results

A total of 137 paediatric patients were referred to the otorhinolaryngology department for noisy breathing during the study period. Out of the total number, 121 patients (81.8%) were diagnosed as stridor due to laryngeal pathology, and were then included in this study.

The patients consisted of 73 males and 48 females, with a male-to-female ratio of 3:2. Malay ethnic patients (77.7%) outnumbered those of Indian (14.9%), Chinese (4.1%) and other ethnicities (3.3%). The patients' age ranged from newborn to 10 years at presentation, with a mean age of 4.9 months (standard deviation ± 11.5 months). Most of the patients presented within the first 3 months of life (n = 72, 59.5%), of which 31 patients (25.6%) were neonate (Table 1). Fourteen patients (11.6%) were preterm at birth. Eighteen patients (14.9%) had underlying congenital pathologies such as Down syndrome, Pierre Robin sequence and Prader–Willi syndrome (Figure 1). All patients were assessed by FNPLS; 31 patients (25.6%) had direct laryngoscopy under general anaesthesia to confirm the diagnosis or exclude synchronous airway lesion.

In term of aetiology, congenital causes accounted for 90.9% (n = 110) of the cases and the remainder was of acquired origin (Table 2). Laryngomalacia was the commonest cause of congenital stridor (n = 95, 78.5%), followed by congenital subglottic stenosis (n = 6, 5.0%), vallecular cyst (n = 3, 2.5%) and congenital unilateral vocal fold paralysis (n = 3, 2.5%). The most common acquired aetiology was laryngopharyngeal reflux (n = 5, 4.1%), followed by laryngotracheobronchitis, acquired vocal fold paralysis and laryngeal foreign body (two cases, respectively). All the cases of vocal fold paralysis were unilateral, two of them acquired following open cardiac surgery. Besides the primary airway lesion, 12 patients (9.9%) were found to have synchronous airway lesions. Eleven of them had one synchronous airway lesion, one patient had multiple airway lesions namely laryngomalacia, vallecular cyst, laryngopharyngeal reflux and tracheomalacia (Table 3).

Table 1. Age distribution of patients at presentation

Age group	No. of patients (%)
<1 month	31 (25.6)
1–3 months	41 (33.9)
3–6 months	33 (27.3)
6–12 months	10 (8.3)
>12 months	6 (5.0)

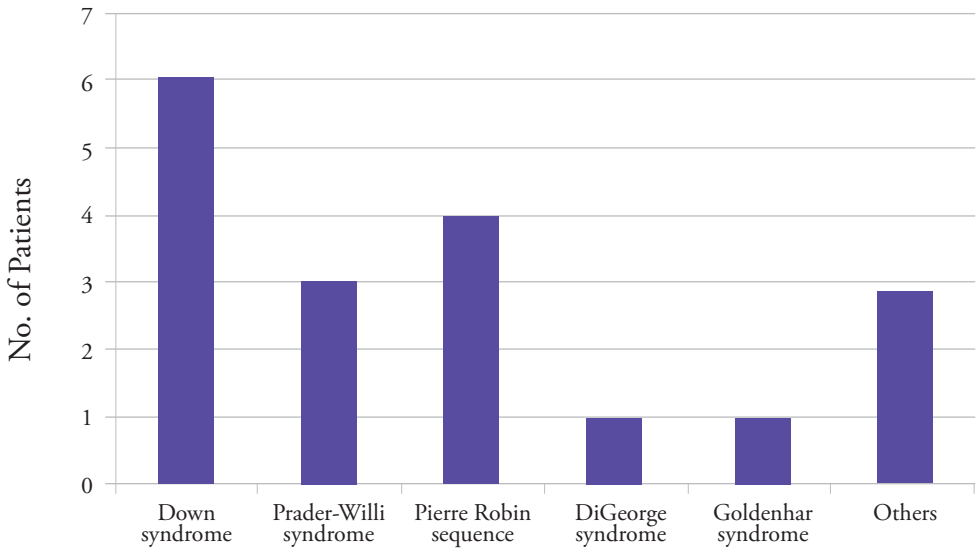

Figure 1. Number of patients with associated congenital pathologies

Table 2. Age distribution of patients at presentation

Diagnosis	No. of patients (%)
Congenital causes	
Laryngomalacia	95 (78.5)
Subglottic stenosis	6 (5.0)
Vallecular cyst	3 (2.5)
Unilateral vocal fold paralysis	3 (2.5)
Laryngeal web	1 (0.8)
Laryngeal dyskinesia	1 (0.8)
Posterior laryngeal cleft	1 (0.8)
Acquired causes	
Laryngopharyngeal reflux	3 (4.1)
Laryngotracheobronchitis	1 (1.7)
Unilateral vocal fold paralysis	2 (1.7)
Laryngeal foreign body	2 (1.7)

Table 3. Number of patients with synchronous airway lesion

Synchronous airway lesions	No. of patients
Laryngomalacia and laryngopharyngeal reflux	5
Laryngomalacia and tracheomalacia	4
Tracheomalacia and laryngeal dyskinesia	1
Laryngomalacia and type I laryngeal cleft	1
Laryngomalacia, vallecular cyst, laryngopharyngeal reflux and tracheomalacia	1

The majority of the patients were managed conservatively (n = 90; 74.4%), with the rest requiring surgical intervention. Out of all the patients with laryngomalacia, 18 (18.9%) required surgical intervention. The remainder had resolution of symptoms in 2–25 months (mean, 8.0 months), with a mean follow-up of 13.3 months (range, 3–30 months). From this study, only one patient with underlying spastic quadriplegic cerebral palsy and encephalomalacia needed tracheostomy to secure the airway. There was one death (0.8%) that occurred in a dysmorphic child with chronic lung disease, due to sepsis and multiorgan failure secondary to pneumonia.

Discussion

There are various aetiologies of stridor in paediatric patients that may be acute or chronic, congenital or acquired, intrathoracic or extrathoracic.[4] The incidence of stridor in the general paediatric population is unknown.[4] With increasing premature neonates survival due to improved intensive care, more airway lesions that would have been lethal previously are seen nowadays.[5] However, stridor must be differentiated from other noisy breathing such as stertor, snoring or wheezing. Careful and detailed history taking and clinical examination would help to assess the severity of the illness and to decide whether immediate intervention is necessary.

In this retrospective review, congenital stridor accounted for 90.9% of the cases, which is higher as compared to that found in other studies that range from 32.3% to 57.6%.[1–3] It was noted that the incidence of infective causes of stridor was significantly lower compared to another series that quoted 33.3%–35.0%.[1,2] This is probably due to the advent of antibiotics and the effectiveness of the current childhood immunisation programme that leads to the reduction of infective causes of stridor among paediatric patients such as epiglottitis and retropharyngeal abscess. Another reason is that in the hospital in which this study was carried out, the paediatric team manages most of the children diagnosed with laryngotracheobronchitis first, instead of direct referral to the otorhinolaryngology department.

Laryngomalacia (78.5%) was the most common congenital cause of stridor consistent with the results in other studies that ranged from 60.2% to 77.5%.[1,2] The majority of these patients (81.1%) were managed conservatively. The rest

(18.9%) required surgical intervention, mainly due to recurrent severe respiratory distress and failure to thrive. This is consistent with a large series reported by Richter and Thompson in which about 20% of the infants with laryngomalacia required surgical intervention.[6] Supraglottoplasty has been shown to improve the airway in more than 80% of the patients with severe laryngomalacia.[7]

Although the majority of the young children presenting with stridor have laryngomalacia, synchronous lower airway lesion should also be considered. Studies revealed that 21%–47.3% of patients had at least one other airway lesion that contributed to airway compromise.[5,8] This is found in 9.9% of the patients in our review. However, not every child with stridor warrants an invasive or extensive workup. Performing direct laryngoscopy and bronchoscopy in children with severe or persistent symptoms is a widely accepted approach.

This study revealed that 15.9% of the patients had other congenital pathologies, in particular Down syndrome. Sanchez et al. reported that 23% of patients with tracheobronchial anomalies have associated genetic disorder particularly Down syndrome.[9] In a study by Bent, 68.1% of patients had comorbidities and it was associated with poorer outcomes.[10]

Unilateral vocal fold paralysis is more common than bilateral paralysis and more likely to involve the left. All five cases of vocal fold paralysis in this study, including three congenital cases and two acquired cases were unilateral (four left, one right) so tracheostomy was not indicated. The two acquired vocal fold paralyses were left sided and occurred in patients who underwent open cardiac surgery for congenital heart anomalies. Green reported that 16% of paediatric patients developed upper airway obstruction after cardiac surgery due to recurrent laryngeal nerve injury. A third of these patients had complete recovery of vocal fold function by early childhood. Bilateral paralysis had the worst prognosis with only 52% recovering spontaneously.[11]

The majority of the patients (74.4%) were managed conservatively without the need for surgery. Thirty-one patients (25.6%) required surgical intervention, of which 18 were laryngomalacia patients. Out of it, only one patient (0.8%) needed a tracheostomy to secure the airway. The percentage of patients who required tracheostomy was significantly lower compared to previous studies such as Waalkens et al. in 1989 (33.3%) and Rupa et al. in 1991 (25%).[2,3] Advancement in endoscopy technology nowadays allows more detailed assessment of the larynx and therapeutic intervention at the same setting, thus avoiding unnecessary tracheostomy in these patients.

Conclusion

Stridor must be recognised as a symptom of upper airway obstruction, which can range from minimal to life threatening in severity. It is essential to establish the correct diagnosis for optimal management. Laryngomalacia was the commonest congenital cause of paediatric stridor referred to the Otorhinolaryngology Department in Hospital Serdang. Synchronous airway lesions should be considered if the child has persistent or severe symptoms. The majority of the patients were managed conservatively, and only one child required a tracheostomy to secure the airway.

Acknowledgement

This study received no specific grant from the government or any other funding agency.

Conflict of interest

The authors have no conflict of interest to disclose.

How does this paper make a difference to general practice?

- To expose family physicians and general practitioners to various aetiologies of stridor in paediatric populations, especially the common causes.
- To promote awareness among clinicians to recognise stridor as a symptom of upper-airway obstruction, which can be a life-threatening emergency.
- To highlight that early referral of a stridorous child to the otorhinolaryngology team is essential for optimal management of the patient.

References

1. Elango S, Htun YN. Aetiology of stridor in Malaysia children. *Ann Trop Paediatr*. 1994;14(4):329–32.
2. Rupa V, Raman R. Aetiology of paediatric laryngeal stridor in an Indian hospital. *Ann Trop Paediatr*. 1991;11(2):137–41.
3. Waalkens HJ, Wouters B, Zwans-Hesslink AG, et al. Stridor in children. *Ned Tijdschr Geneeskd*. 1989;133(40):1981–4. (Article in Dutch)
4. Boudewyns A, Claes J, Van de Heyning P. An approach to stridor in infants and children. *Eur J Pediatr*. 2010;169:135–41.
5. Zoumalan R, Maddalozzo J, Holinger LD. Etiology of stridor in infants. *Ann Otol Rhinol Laryngol*. 2007;116(5):329–34.
6. Richter GT, Thompson DM. The surgical management of laryngomalacia. *Otolaryngol Clin N Am*. 2008;41:837–64.
7. Sichel JY, Dangoor E, Eliashar R, et al. Management of congenital laryngeal malformations. *Am J Otolaryngol*. 2000;21:22–30.
8. Sakakura K, Chikamatsu K, Toyoda M, et al. Congenital laryngeal anomalies presenting as chronic stridor: A retrospective study of 55 patients. *Auris Nasus Larynx*. 2008;35(4):527–33.
9. Sanchez I, Navarro H, Mendex M, et al. Clinical characteristics of children with tracheobronchial anomalies. *Pediatr Pulmonol.* 2003;35(4):288–91.
10. Bent J. Pediatric laryngotracheal obstruction: Current perspectives on stridor. *Laryngoscope*. 2006;116:1059–70.
11. Green J, Walters HL, Delius RE, et al. Prevalence and risk factors for upper airway obstruction after pediatric cardiac surgery. *J Pediatr*. 2015;166(2):332–7.

ORIGINAL ARTICLE

Prevalence of falls among community-dwelling elderly and its associated factors: A cross-sectional study in Perak, Malaysia

Yeong UY, Tan SY, Yap JF, Choo WY

Yeong UY, Tan SY, Yap JF, et al. Prevalence of falls among community-dwelling elderly and its associated factors: A cross-sectional study in Perak, Malaysia. *Malays Fam Physician*. 2016;11(1);7-14.

Keywords:
Elderly, falls, Malaysia, older people, risk factors

Authors:

Yeong Uen Yea
(Corresponding author)
MBBS
Department of Social and Preventive Medicine, Faculty of Medicine, University of Malaya, 50603 Kuala Lumpur, Malaysia.
Email: yea0531@hotmail.com

Tan Soon Yee
MBBS
Department of Social and Preventive Medicine, Faculty of Medicine, University of Malaya, Kuala Lumpur, Malaysia.
Email: tansoonyee@hotmail.com

Yap Jun Fai
MBBS
Department of Social and Preventive Medicine, Faculty of Medicine, University of Malaya, Kuala Lumpur, Malaysia.
Email: junfai90@hotmail.comm

Choo Wan Yuen
PhD
Department of Social and Preventive Medicine, Faculty of Medicine, University of Malaya, Kuala Lumpur, Malaysia.
Email: ccwy@ummc.edu.my

Abstract

Introduction: Fall is a major cause of injuries and can increase the risk of early mortality among elderly. The objective of this study was to determine the prevalence of falls among community-dwelling elderly in rural Malaysia and its associated factors.

Methods: Data were obtained from a cross-sectional survey in five randomly selected districts in the state of Perak, Malaysia. A total of 250 households were randomly selected. A total of 811 individuals aged 60 years or more were recruited and interviewed using a structured questionnaire. Information about socio-demographic, history of falls in the past 1 year, medical history, drug history and physical activity level were enquired.

Results: The prevalence of falls in the past 1 year among community-dwelling elderly was reported to be 4.07%. Indigenous elderly (Adjusted odd ratio, AOR = 6.06, 95% CI = 1.10–33.55, p = 0.039) and living alone (AOR = 2.60, 95% CI = 1.04–6.50, p = 0.042) were shown to be factors associated with falls. Physical activity level, number of co-morbidities and number of medications used were not associated with falls.

Conclusion: Elderly of indigenous ethnicity and living alone are the main factors associated with falls in this population. Indigenous people may be at higher risk, which warrant further investigation with a larger sample to improve the precision of estimates.

Introduction

Malaysia is facing an aging population, as experienced by many high- and middle-income countries. An aging population poses a real challenge to society and healthcare systems, as the disease burden increases. Besides chronic diseases, fall-related injuries are common among the elderly, frequently resulting in disability, institutionalisation and even premature death.

Although estimates of fall rates vary widely based on the region, age and living arrangements of the elderly population, it is estimated that 28%–35% of people aged 65 years or more were found to experience fall each year. This estimate increases to 50% for those aged 85 years or more.[1] Falls are the underlying cause of 10%–15% of all emergency department visits and 31.6% of those who fall are admitted.[2] The high incidence of fall and its complications is an unnecessary healthcare burden, as it can be prevented.

Current evidence[3] has suggested that biological factors such as age and gender are risk factors for falls. Older adults are prone to falls largely due to the decline of physical, cognitive and affective capacities, and co-morbidities associated with chronic illnesses. Women are more likely than men to experience falls but fall-related mortality is higher in men.[3] The relationship between falls and ethnicity is still unclear.[3] The rate of falls and hospitalisations for fall-related injuries has been reported to be more than two times higher for Caucasians as compared to their Hispanics, Asians/Pacific Islanders and African counterparts.[4] However, more recent findings[5] suggest that the annual fall rate in Chinese older people is only approximately half of that found in white older people.

Despite the extensive epidemiological research on risk factors associated with falls, findings regarding several behavioural, environmental

and socio-economic factors associated with falls appear to be mixed. For example, behavioural factors such as physical activity and adequate dietary intake were found to have a protective effect against falls.[3] Studies have shown that fall risk is associated with the number of medications used, but only when at least one established fall risk-increasing drug was part of the daily regimen.[6] Association between environmental factors and falls have also been explored. A large-scale community study in Thailand reported that elderly who lived alone had a higher incidence of falls as compared with those who lived with others.[7] As literature on the association of living alone with risk of falls in Asia is limited, this study will contribute to the topic.

There is a scarcity of epidemiological research on falls in Asian populations. In Malaysia, only a limited number of large-scale community-based studies to find out the actual public health burden of falls have been carried out.[8,9] In a study among elderly seeking medical care in a primary care setting, it was reported that the prevalence of falls was 47%.[9] The prevalence was considerably higher compared to the 28% to 42% reported by other countries.[3] The present study was carried out to estimate the actual prevalence of falls among elderly living in a community setting and identify its associated factors. It is important to determine the magnitude, associated factors, and circumstances of falls so that appropriate falls prevention programmes could be formulated and implemented to suit the local setting and needs.

Methods

Study design

Data were collected from a cross-sectional study conducted in Perak, Malaysia in March 2011. Perak is the second largest state in Peninsular Malaysia. Located in the northern region, it borders Kedah to the north, Penang to the northwest, Pahang to the east and Selangor to the south. In 2010, the population in Perak was 8.3% (2.35 million) of the total Malaysian population with an ethnic composition of Malay (54.2%), Chinese (30.4%), Indian (12.3%) and others (3.1%). The socio-demographic background of the state population is similar to that of the whole country.[10] In this study, 50% of the districts located in the state of Perak (5 out of 10 districts namely Kampar, Kuala Kangsar, Parit Buntar, Gerik and Taiping) were randomly chosen. In each district, four villages were randomly selected from a list of villages provided by the State Department of Health. For each selected village, the Village Head provided a sampling frame consisting of the lists of available houses within their village. Each house was assigned a number and a total of 250 households were randomly selected based on the list of random numbers generated using the computer. The number of houses chosen from each village was proportional to the size of its population. The following analysis was performed based on a subset of a larger rural community health survey conducted. In total, 811 residents aged 60 years and above were included in the study. All residents were interviewed face-to-face in their respective houses during household visits by medically trained candidates and research assistants using a structured questionnaire to record information about socio-demographic details, history of falls, medical history, drug history and physical activity level. This study was approved by the Medical Ethics Committee of the University of Malaya Medical Centre. Written informed consent was obtained from the head of villages, as well as from all respondents.

Questionnaire

Each participant was required to complete a structured questionnaire based on the face-to-face interview. The questionnaire was based on the following:

Falls: Residents were asked if they had experienced falls in the past 1 year, irrespective of the number of falls. Fall was defined as an unexpected event in which the individual comes to rest on the ground, floor or lower level.[3] Those resulting from outside events, such as a motor vehicle accident or violence were excluded.

Socio-demographic variables: Demographic data include age, gender, race, religion, number of children and total income.

Physical activity: The level of physical activity of the participants was obtained using the International Physical Activity Questionnaire (IPAQ).[11] The items in IPAQ were structured to provide separate scores on walking; moderate-intensity; and vigorous-intensity activity, as well as a combined total score to describe overall level of activity. Computation of the

total score requires summation of the duration (in minutes) and frequency (days) of walking, moderate-intensity and vigorous-intensity activities. The levels of physical activity were then classified into three levels: low, moderate and high.

Number of co-morbidities: The diseases included were diabetes mellitus, hypertension, heart disease, stroke, hypercholesterolaemia, hearing impairment, urinary incontinence and obesity. Participants were asked if they had ever been informed by a doctor as having a diagnosis of diabetes mellitus, hypertension, heart disease, stroke and hypercholesterolaemia. Hearing impairment and urinary incontinence were self-reported.

Number of medications used: This includes medications for diabetes mellitus, hypertension, heart disease, stroke, hypercholesterolaemia and drugs that cause dizziness and sleepiness. Consumption of medications was self-reported and counter-checked by the interviewers during the house visit. They scored one point for consuming medication for a particular disease, regardless of the number of medications consumed for the same disease.

Living conditions: Information regarding participants' family structure (living alone or with spouse, children, family members), and number of family members living in the house were collected. Those living alone were defined as living by him/herself at home and independently able to carry out activities of daily living.

Statistical analysis

Prevalence was computed based on the self-reported falls in the past 1 year. Univariate logistic regression was used to compute crude odds ratios for all categorical variables. Adjusted odds ratio was computed using multivariate logistic regression models, including gender, age, ethnicity, total income, physical activity level, living alone, number of co-morbidities and number of medications used. The final model to determine the independent effect on falls was performed by adjusting for confounders. The associations were expressed as odds ratio and its 95% confidence interval. P-values were based on two-sided tests and were considered statistically significant at $p < 0.05$. All analyses were conducted using SPSS software (SPSS Inc, version 22.0).

Results

Prevalence of falls

A total of 811 elderly were included in the study. Table 1 shows the basic characteristics of the elderly population. The mean age of the participants was 70.21 ± 7.24 years. There were 10.4% more female participants compared to male. The distribution of the participants according to ethnic groups was comparable to the national census. Approximately 4.07% of the participants experienced a fall in the past 1 year. The majority of them (75.8%) fell in indoor settings. In the house, most of the fall incidents occurred at an outside compound (28%), followed by the bathroom (20%) and kitchen (20%) (Table 2).

Table 1: Basic socio-demographic characteristics of study participants

Variables	Number	Percentage
Gender		
Male	363	44.8
Female	448	55.2
Age (years)		
60–64	286	35.3
65–69	165	20.3
70–74	162	20.0
≥75	198	24.4
Ethnicity		
Malay	631	77.8
Chinese	140	17.3
Indian	28	34
Indigenous	12	1.5

Table 1: Basic socio-demographic characteristics of study participants (Continued)

Variables	Number	Percentage
Religion		
Islam	631	77.8
Christian	12	1.5
Buddhism	144	17.8
Hindu	14	1.7
Others	10	1.2
[a]No. of children		
≤5	406	50.4
6–10	363	45.1
>10	36	4.5
Total income (RM)		
≤ 300	462	57.0
301–500	104	12.8
501–1000	139	17.1
> 1000	106	13.1
Physical activity level		
High	263	32.4
Moderate	243	30.0
Low	305	37.6
**Living alone*		
Living alone	131	18.6
Not living alone	572	81.4

*Data with missing values.

Table 2: Location of falls in the house

Location	Frequency	Percentage
Bathroom	5	20
Kitchen	5	20
Living room	4	16
Bedroom	2	8
On the stairs	2	8
Outside compound	7	28

Factors associated with falls among elderly

Table 3 shows the crude odds ratio (COR) and adjusted odds ratio (AOR) of factors associated with falls. At bivariate level, there was no association between age and gender with the risk of falls among elderly. No significant increased risk of falls was found in elderly who have low physical activity level, multiple co-morbidities or consuming multiple medications.

Table 3: Correlates of falls among community-dwelling elderly

Variables	Percentage	Crude odds ratio (COR)	95% CI		[a]Adjusted odds ratio (AOR)	95% CI		*p-value
			Lower	Upper		Lower	Upper	
Gender								
Male	45.5	1.00	–	–	1.00	–	–	0.253
Female	54.5	0.97	0.48	1.96	0.60	0.25	1.44	
Age category (years)								
60–64	24.2	1.00	–	–	1.00	–	–	0.232
65–69	30.3	2.24	0.87	5.80	2.18	0.78	6.10	0.139
70–74	15.2	1.11	0.36	3.44	0.61	0.15	2.51	0.492
≥75	30.3	1.85	0.72	4.77	1.23	0.41	3.66	0.716
Ethnicity								
Malay	78.8	1.00	–	–	1.00	–	–	0.145
Chinese	12.1	0.68	0.24	1.99	0.61	0.20	1.86	0.386
Indian	3.0	0.86	0.11	6.59	0.77	0.10	6.16	0.808
Indigenous	6.1	4.65	0.97	22.33	**6.06**	**1.10**	**33.55**	**0.039**
Total income (RM)								
≤ 300	63.6	1.00	–	–	1.00	–	–	0.866
301–500	15.2	1.06	0.39	2.88	1.11	0.37	3.27	0.857
501–1000	12.1	0.62	0.21	1.84	0.68	0.21	2.21	0.525
>1000	9.1	0.61	0.18	2.09	0.70	0.18	2.66	0.597
Physical activity level								
High	24.2	1.00	–	–	1.00	–	–	0.623
Moderate	33.3	1.51	0.60	3.82	1.62	0.55	4.76	0.377
Low	42.4	1.53	0.63	3.72	1.58	0.57	4.38	0.383
Living alone	27.3	2.15	0.95	4.86	**2.60**	**1.04**	**6.50**	**0.042**
Number of co-morbidities	–	1.06	0.82	1.37	0.84	0.48	1.47	0.531
Number of medications used	–	1.17	0.90	1.53	1.25	0.68	2.32	0.472

[a] Final model from multiple logistic regression analysis including gender, age, ethnicity, total income, physical activity level, living alone, number of co-morbidities, number of medication used as shown in the table.

* *p*-value for adjusted odds ratio

In the multiple logistic regressions, the final model was simultaneously adjusted for gender, age, ethnicity, total income, physical activity level, living alone, number of co-morbidities and number of medications used. Living alone and ethnicity were found to be significant correlates for falls among the elderly. Elderly who lived alone had more than two-fold increase odds in the risk for falls (AOR = 2.60, 95% CI = 1.04–6.50, p = 0.042) compared with those living with family members/ relatives. Interestingly, indigenous elderly was found to have six times (AOR = 6.06, 95% CI = 1.10–33.55, p = 0.039) higher odds of experiencing falls.

Discussion

The prevalence of falls among the elderly in Malaysia's community setting, as determined by this study was 4.07%. This estimate appears to be fairly stable as the Third Malaysian National Health and Morbidity Survey in 2006 also reported a similar prevalence of 4%.[8] The prevalence of falls reported in Malaysia appeared to be lower when compared with other Asian countries such as Japan (15.8%),[12] Hong Kong (19.3%)[13] and China (11.1%).[14] The prevalence of falls is generally lower in this study, possibly reflecting a 'younger' elderly population in Malaysia

compared to these countries. More than half of the recruited participants (53.1%) in this study were aged 60–69 years. Falls occurred more commonly in indoor settings than outdoors. Location of falls was similar to those found in other studies.[7,8]

Living alone is a major factor associated with falls in this study. This finding has been reported by other recent studies, which suggested that the elderly who lives alone have twice or higher risk of falls and/or multiple falls compared to those who did not live alone.[7,15] The elderly who lives alone are likely to have lesser social support and fewer resources in time of needs. In addition, loneliness and social isolation are commonly experienced by elderly who lives alone, which may further exacerbate existing physical, cognitive and sensory limitations, and increase their risk of falls.[3] Another possible explanations of this increased risk of falls are psychological factors such as fear of falling. Older adults who live alone tend to have greater fear of falling compared to those who live with others.[15] Interestingly, Delbaere et al. reported that among elderly with excessive levels of perceived fall risk, 40% of them experienced multiple or injurious falls during the 1 year follow-up despite having low physiological fall risk.[16] The underlying mechanisms behind this relationship remains to be established, although the elderly with excessive levels of perceived fall risk in their study tend to show higher levels of depressive symptoms and decreased executive functioning, both of which are known risk factors for falls.[16] Psychological issues should also be considered managing for fall persons besides evaluating the physiological risk.

Another major factor associated with falls that deserves attention is the higher risk of falls found among the elderly of indigenous group. One in three indigenous people aged more than 65 years was reported to fall at least once per year.[17] There are a number of conditions that predispose indigenous elderly to higher risk of falling compared to other ethnic groups. Chronic diseases are more prevalent among indigenous people worldwide. These medical diseases increase the risk for fall by their complications such as diabetic retinopathy (which affects vision and predisposes to falls) and peripheral neuropathy (which leads to poor joint position sense and subsequently falls); poor nutrition; poor cardiovascular health and physical activity.[18] The economic and health status of the indigenous Malaysian population still lacks far behind that of other ethnic groups. This lack of parity covers life expectancy, childhood nutrition, and other well-being indicators.[19] It is therefore not surprising that minority indigenous elderly are more prone to falls due to interaction of these many risk factors. Furthermore, the majority of the indigenous Malaysian population lives in remote areas. As a result, access to healthcare services is limited.[19]

Both high and low levels of physical activity have been associated with increased fall risk in Caucasian older adults. A meta-analysis finding indicated that evidence increased or decreased physical activity in seniors as a major risk factor for falls and injurious falls is still inconclusive.[20] It is presumed that older adults with lower physical activity levels tend to be more frail due to muscle atrophy.[21] This directly restricts exposure to activities demanding higher neuromuscular and balance control and may lead to falls. Active older adults are more likely to fall outdoors due to a frequent exposure to risky environmental hazards (slippery floors, degraded pavements) or acute fatigue in daily life activities.[5] In a study conducted by a neighbouring country, the rate of falls among Singaporean elderly who practice Qigong (a form of physical activity engaged by the Chinese community) was lower compared to those who did not practice Qigong.[22] In this current study, there was no direct association between physical activity and risk of falls. The reason for this non-significant finding remains to be investigated. Possible explanations include underreporting of physical activity or fear of falling among elderly that may indirectly restrict elderly's activity. Clearly, more research is needed in this area to determine the various types and intensities of physical activity engaged by the elderly as they may serve as protective factors to falls.

Although there is evidence that medications are as risk factor for falls in Western elderly populations, this was not seen by this study. Fall risk has been found to be associated with the use of multiple medications, but only when at least one established fall risk-increasing drug (notably central nervous system drugs, psychotropic drugs and diuretics) was part of the daily regimen.[6] However, no respondent in this study (in the questionnaire) reported any usage of central nervous system drug or psychotropic drugs. A

larger sample is needed to address this issue. Similarly, no association was found between the number of co-morbidities and falls. The assessment of co-morbidities was dependent on the accuracy of the participants reporting their medical history – there may have been under-reporting. Future research should attempt to corroborate the self-reporting of diseases with medical records or caregivers' report.

There were some limitations in this study. Firstly, this study was cross-sectional in design and hence unable to determine the causal-effect relationship between falls and possible factors. Secondly, since screening for memory or cognitive testing was not performed among the participants, there was a possibility of recall bias, especially for the elders who tend to have poorer recall. Finally, the data collected did not ask the specific name of medications; hence fall risk-increasing drugs could not be defined. Other possible risk factors such as nutritional status, fall-related risk-taking behaviour and housing conditions could be included in future research to allow a more holistic assessment of risk factors associated with falls.

Conclusion

In conclusion, one in every 25 Malaysian elderly living in community setting experienced falls in the past 1 year. Living alone and being indigenous were major factors associated with falls among elderly. Preventive measures should be undertaken, which include providing social support and provision of health education to increase awareness among elderly. In addition, primary care physicians can provide comprehensive geriatric assessment for elderly including enquiry about living conditions and social support during screening or follow-up. Appropriate intervention to elderly living alone or with poor social support could be initiated, by referring them to social workers for welfare needs or to nurses for regular home visits.

Acknowledgement

The authors express their gratitude to the Department of Social and Preventive Medicine, Faculty of Medicine, University of Malaya for research administration and support. The work of Choo WY is supported by the University of Malaya Research Grant (RG468/12HTM).

Conflict of interest

None.

How does this paper make a difference in general practice?

- The experience of falls among community-dwelling elderly is quite common. One in every 25 elderly reported fall in the past 1 year.
- Primary care physician should enquire about living conditions and social support of elderly while providing geriatric assessment during their follow-up.
- Healthcare providers at the primary care level can provide regular visits to elderly patients or refer them to social workers, especially for those living alone and with poor social support.

References

1. Tinetti M, Doucette J, Claus E, et al. Risk factors for serious injury during falls by older persons in the community. *Home Healthc Nurse*. 1996;14(7):549.
2. Scuffham P, Chaplin S, Legood R. Incidence and costs of unintentional falls in older people in the United Kingdom. *J Epidemiol Community Health*. 2003;57(9):740–44.
3. WHO. *WHO Global Report on Falls Prevention in Older Age*. Geneva: World Health Organization; 2007.
4. Ellis A, Trent R. Hospitalized fall injuries and race in California. *Inj Prev*. 2001;7(4):316–20.
5. Kwan M, Close J, Wong A, et al. Falls incidence, risk factors, and consequences in Chinese older people: A systematic review. *J Am Geriatr Soc*. 2011;59(3):536–43.
6. Ziere G, Dieleman J, Hofman A, et al. Polypharmacy and falls in the middle age and elderly population. *Br J Clin Pharmaco*. 2006;61(2):218–23.

7. Sophonratanapokin B, Sawangdee Y, Soonthorndhada K. Effect of the living environment on falls among the elderly in Thailand. *Southeast Asian J Trop Med Public Health*. 2012;43(6):1537.

8. Lim KH, Jasvindar K, Normala I, et al. Risk factors of home injury among elderly people in Malaysia. *Asian J Gerontol Geriatr*. 2013;8(2):8.

9. Sazlina S, Krishnan R, Shamsul A, et al. Prevalence of falls among older people attending a primary care clinic in Kuala Lumpur, Malaysia. *Jurnal Kesihatan Masyarakat*. 2008;14(1):11–16.

10. Population distribution and basic demographic characteristics 2010 [Internet]. 1st ed. Putrajaya: Department of Statistics, Malaysia; 2011. Available at: http://www.statistics.gov.my/portal/download_Population/files/census2010/Taburan_Penduduk_dan_Ciri-ciri_Asas_Demografi.pdf. Accessed May 27, 2014.

11. PAQ International Physical Activity Questionnaire. Guidelines for Data Processing and Analysis of the International Physical Activity Questionnaire (IPAQ)—Short and Long Forms [Internet]. 2005. Available at: http://www.ipaq.ki.se/scoring.pdf. Accessed May 27, 2014.

12. Kitayuguchi J, Kamada M, Okada S, et al. Association between musculoskeletal pain and trips or falls in rural Japanese community-dwelling older adults: A cross-sectional study. *Geriatr Gerontol Int*. 2014;15(1):54–64.

13. Chu L, Chi I, Chiu A, et al. Incidence and predictors of falls in the Chinese elderly. *Ann Acad Med Singapore*. 2005;34(1):60–72.

14. Fang X, Shi J, Song X, et al. Frailty in relation to the risk of falls, fractures, and mortality in older Chinese adults: Results from the Beijing longitudinal study of aging. *J Nutr Health Aging*. 2012;16(10):903–7.

15. Elliott S, Painter J, Hudson S. Living alone and fall risk factors in community-dwelling middle age and older adults. *J Commun Health*. 2009;34(4):301–10.

16. Delbaere K, Close J, Brodaty H, et al. Determinants of disparities between perceived and physiological risk of falling among elderly people: Cohort study. *BMJ*. 2010;341:c4165.

17. Falls Prevention for Aboriginal People. A tool for Aboriginal Health Workers and Aboriginal Communities [Internet]. 1st ed. Australia; 2014. Available at: http://www.health.wa.gov.au/stayonyourfeet/docs/11827_SOYF.pdf. Accessed May 27, 2014.

18. Kay A. Aboriginal Injury Priorities: An overview of the evidence, best practice and prevention programs in Western Australia [Internet]. 1st ed. Australia; 2014. Available at: https://www.iccwa.org.au/useruploads/files/aboriginal_injury_priorities_review_and_consultation.pdf. Accessed May 27, 2014

19. Baer AS. *Health, Disease, and Survival*. Subang Jaya. Malaysia: Center for Orang Asli Concerns; 1999.

20. Health Quality Ontario. Prevention of falls and fall-related injuries in community-dwelling seniors: An evidence-based analysis. *Ont Health Technol Assess Ser.* 2008;8(2):1–78.

21. Chan B, Marshall L, Winters K, et al. Incident fall risk and physical activity and physical performance among older men the osteoporotic fractures in men study. *Am J Epidemiol*. 2007;165(6):696–703.

22. Wong ML, Goh LG, Chia HP, et al. Correlates of leisure-time physical activity in an elderly population in Singapore. *Am J Public Health*. 1999;89(10):1578–80.

Fever and rash in an 11-month-old girl

Jamani NA, Puteri Shanaz JK

Jamani NA, Puteri Shanaz JK. Fever and rash in an 11-month-old girl. *Malays Fam Physician*. 2016;11(1);15-17.

Keywords:
Fever, rash, roseola, diagnoses, children

Authors:

Nurjasmine Aida Jamani
(Corresponding author)
MD, M. Med (Fam Med)
Department of Family Medicine, Kulliyyah of Medicine, International Islamic University Malaysia, Jalan Sultan Ahmad Shah, 25200 Kuantan Pahang, Malaysia.
Email: minaida@iium.edu.my

Puteri Shanaz Jahn Kassim
MD, M. Med (Fam Med)
Department of Family Medicine, Faculty of Medicine and Health Sciences, Universiti Putra Malaysia, 43400 Serdang, Selangor, Malaysia.

Case Summary

An 11-month-old girl presented with a 3-day history of high fever, irritability and decreased appetite. However, there was no coryza, cough, diarhoea or fits. On the third day, a generalised erythematous rash appeared after the fever subsided abruptly. The rash was pinkish in colour, non-pruritic and discrete, which initially started on the trunk and later spread to the face, neck and extremities within 2 days. On further questioning, she was found to be appropriately immunised for her age according to the Ministry of Health's immunisation schedule. No similar illnesses existed in her family and the nursery she attended.

On examination, the infant appeared well, with a normal body temperature of 37°C and stable vital signs. A generalised erythematous maculopapular rash was observed extending from the head to her legs, which blanched with pressure. Involvement of the buccal mucosa was absent, with no palpable lymph nodes. She was prescribed paracetamol syrup regularly and care givers advised to maintain her hydration. The child remained well and active throughout the course of the illness. The rash subsequently subsided within a couple of days.

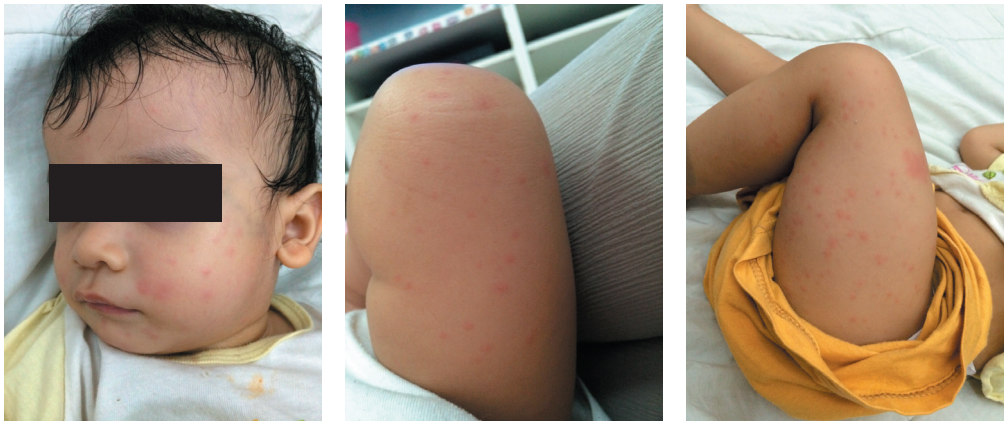

Figure 1. Erythematous maculopapular rash on the face and limbs

Questions

1. What is the most likely diagnosis?
2. What are the differential diagnoses?
3. What are the relevant investigations to be conducted?
4. What is the management?
5. Is there a need to notify the health authorities about this condition?

Answers

1. The most likely diagnosis for this presentation based on the clinical history and characteristic of the rash is roseola infantum or exanthema subitum. The characteristic of this condition is that the infant is febrile for 3 to 5 days with high body temperature that abruptly subsides with the emergence of a typical rash. The rash is macular or maculopapular in nature, starting from the neck and trunk and spreading to the face and limbs, as in this child. The rash also blanches with pressure, and usually clears up within 2 days.[1-3] While 93% of affected infants are symptomatic (fever, fussiness, rhinorrhoea), only 20% of those infected exhibit the rash of roseola.[2,5] In Asian infants, Nagayama spots (ulcers at the uvulopalatoglossal junction) can be observed.[4,5] However, in this child, no uvulopalatoglossal ulcers were noted. Also, 4% of affected children are reported to develop seizure during the febrile phase.[5]

The key feature of roseola is a rash presenting after the resolution of fever. It has been documented that by 2 years of age, 90% of children are infected, with a peak incidence occurring between 9 and 21 months of age.[5,6] Also known as the sixth disease, it is caused by human herpesvirus-6 (HHV-6). The exact mode of transmission of HHV-6 is yet to be established; it is presumed to be transmitted via the saliva of asymptomatic individuals to susceptible infants.[5,6] Only 1% of HHV-6 infection is acquired congenitally, without known sequelae.[5,6]

2. Several differential diagnoses can be made pertaining to fever and rash in children. As differentiating a rash in children is difficult based on appearance alone, it is very important that physicians should be able to recognise the characteristics of a rash to narrow down the possible diagnoses and aetiologies. Consideration should be made on the appearance and site of rash, associated symptoms and the clinical course. In this case, measles can be one of the differentials in view of the child's age and the duration of the fever. However, there were no accompanying symptoms such as cough, coryza and conjunctivitis. Measles can also be distinguished by the presence of Koplik's spots in the oral mucosa.[1] Other diagnosis may include rubella in view of the characteristic pink macular rash. In rubella, prodromal features such as fever and malaise are usually absent in children. The rash of rubella starts on the face, spreading downwards to the trunks and fading completely on day 3.[7] However, it causes less severe symptoms, and its exanthem typically has a shorter duration (2–3 days).[1] Other common infectious conditions in children include erythema infectiosum and scarlet fever.[1,2] However, erythema infectiosum, which is caused by human parvovirus B19, primarily affects older children between 3 and 12 years of age, and is usually accompanied by a viral prodrome followed by the "slapped cheek" facial rash.[7] The distinguishing feature of scarlet fever is that the rash typically starts from the upper trunk and then spreads out to the entire trunk, sparing the palms and soles, along with desquamation of the extremities after the rash fades.[7]

3. For classic roseola infection, laboratory investigation is seldom required,[4] as the diagnosis is made based on the signs and symptoms. However, if the child presents with complications such as febrile seizures, laboratory work up is necessary, which may include a full blood count, urinalysis, blood culture and cerebrospinal fluid examination. The illness can be confirmed with virus isolation or detection of viral DNA sequences in peripheral blood mononuclear cells.[7]

4. As for the management of this condition, only supportive treatment such as antipyretics and frequent hydration is required, as most of the cases are self-limiting.[4,7,8] Since transmission of this disease is through saliva, standard hygienic measures such as frequent and thorough hand washing is important to prevent the spread of infection.[4] The exact period when an infected child is contagious is unclear, but it most likely spreads during the febrile phase of the illness when there are no noticeable signs that the child is infected with the virus. In addition, there is no recommended period of isolation for a child with roseola.[4,8] However, it is best to keep the sick child at home during the febrile phase for close observation in addition to infectious control in day care centers.

5. Roseola infantum does not need to be notified because it is benign and self-limiting. In Malaysia, hand, foot and mouth disease and measles are the childhood communicable diseases that need to be notified within 24 hours and 48 hours, respectively, in order to prevent outbreak to other children.[9,10] This is important for surveillance as there are children who have not been vaccinated as scheduled.

Conflict of interest

None.

References

1. Tremblay C, Brady MT. RoseolaInfantum. *Up to Date Paediatric* [online]. Available at: http://www.uptodate.com/contents/roseola-infantum-exanthem-subitum. Accessed June 18, 2015.

2. Kliegman RM, Stanton BMD, St. Geme J, et al. *Nelson Textbook of Pediatrics*.17th ed. Philadelphia, PA: Elsevier Saunders; 2011. p. 1070

3. Hall CB. Herpes and the rash of roses: A new virus, HHV-6, as a cause of an old childhood disease, roseola. *Pediatr Ann*. 1990;19 (9):517.

4. Chua KB, Lam SK, Sazaly AB, et al. Uvulo-palatoglossal junctional ulcers-an early clinical sign of exanthema subitum due to human herpes vrius 6. *Med J Malaysia*. 1999;54 (1):32–6.

5. Tesini BL, Epstein LG, Caserta MT. Clinical impact of primary infection with roseola viruses. *Curr Opin Virol*. 2014;9:91–6.

6. Zerr DM, Meier AS, Selke SS, et al. A population-based study of primary human herpes virus 6 infection. *N England J Med*. 2005;352:768–76.

7. Wolff K, Johson RA. Fitzpatrick's Color Atlas and Synopsis of Clinical Dermatology. 6th edition. *McGraw Hill Medical*; 2009.

8. Gorman CR, James WD. Roseola infantum. *Medscape Drug and disease*.[online] Available at: http://emedicine.medscape.com/article/1133023-overview. Accessed July 1, 2015.

9. Ministry of Health Malaysia. Hand, Foot and Mouth Disease (HFMD) guideline; 2007.

10. Ministry of Health Malaysia. Measles prevention and control in Malaysia. Handbook for Healthcare Personnel; 2006.

CASE REPORT

Multiple concurrent primary extramammary Paget's disease

Leelavathi M, Norazirah MN, Nur Amirah AP

Leelavathi M, Norazirah MN, Nur Amirah AP. Multiple concurrent primary extramammary Paget's disease. *Malays Fam Physician.* 2016;11(1);18-21.

Keywords:
Axilla, extramammary, Paget's disease, scrotum, vulva

Authors:

Leelavathi Muthupalaniappen
(Corresponding author)
MMed (Fam Med)
Department of Family Medicine, Faculty of Medicine, 14 th Floor, Pre-clinical Block, Universiti Kebangsaan Malaysia Medical Center, Jalan Yaacob Latif, Bandar Tun Razak, 56000 Cheras, Kuala Lumpur, Malaysia.
Email: drleelaraj@gmail.com

Norazirah Md Nor
MBBCh(Cardiff) MRCP, ADVM Derm
Dermatology Unit, Department of Medicine, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Center, Kuala Lumpur, Malaysia.

Nur Amirah binti Amat Puad
MD
Department of Family Medicine, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Center Kuala Lumpur, Malaysia.

Abstract

Extramammary Paget's disease (EMPD) is a rare malignant disorder of the skin, which was described in as early as the nineteenth century. EMPD usually occurs as a single lesion in the apocrine sweat gland–bearing skin with abundant hair follicles. Here, we present an elderly man who suffered from a non-resolving chronic genital pruritus for 8 months. Initially, he was managed for recurrent fungal infection and eczema. Later, a diagnosis of the rare condition multiple primary EMPD was made based on the histopathology findings and appropriate treatment was given.

Case report

A 79-year-old Chinese man was first seen at a general outpatient clinic for itchiness of the skin over the genitalia for 8 months. Initially, he noticed a small red patch at the base of the penis, which gradually enlarged. The lesion was extremely itchy and occasionally became macerated and infected. He had consulted a number of doctors and was prescribed different treatments, without much improvement. His other medical problems included diabetes, dyslipidaemia and hypertension.

Clinically, a large, well-defined erythematous plaque was observed extending from the suprapubic area and involving the skin covering almost the whole of the scrotum and the shaft of the penis (Figure 1).

Incidental finding of a single well-demarcated brownish patch with central hyperpigmentation (4×5 cm) was noted on the left axilla (Figure 2). The axillary lesion appeared approximately at the same time but was less pruritic. Inguinal lymph and axillary nodes were not palpable bilaterally. Nipple, digital rectal, and other examination results were unremarkable. Symptoms of altered bowel movements, urinary symptoms or constitutional symptoms suggestive of any underlying visceral malignancy were also absent.

The patient was initially managed as a case of cutaneous candidiasis by the general practitioner. When he did not show any improvement with topical antifungal treatment, the diagnosis was revised to eczema with secondary bacterial infection. He was subsequently treated with oral

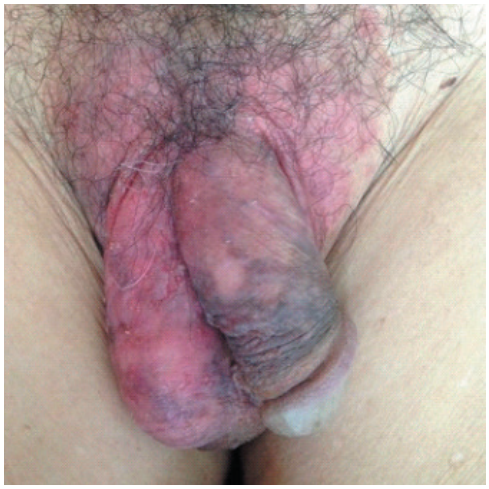

Figure 1. Genital lesion: erythematous plaque with focal areas of erosions involving the skin of the scrotum, the penis and the adjacent suprapubic area

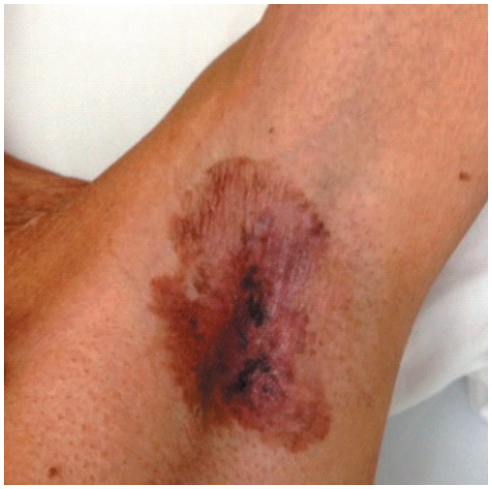

Figure 2. Left axilla: well-demarcated brownish patch with darker central areas

antihistamines, topical antifungals and topical steroids after different consultations without any improvement. The patient was eventually referred to a dermatology clinic for a second opinion and further management owing to poor response to therapy.

Differential diagnoses of EMPD, inverse psoriasis, candidiasis and Bowen's disease were considered. His full blood count was normal, but renal profile showed mild renal impairment. Serum calcium concentration was 2.45 mmol/L (2.09–2.42), whereas corrected calcium concentration was within normal values, that is, 2.31 mmol/L (2.14–2.58). Urine analysis results were normal. There was no evidence of primary or secondary lesions on chest radiographs. Computerized tomography (CT) of the neck, thorax, abdomen and pelvis did not reveal any abnormalities except for multifocal infarcts on brain CT. Histopathology of skin biopsy from the left axilla showed intra-epidermal proliferation of atypical, large (Paget) cells in nests and single distribution involving the follicular epithelium. These atypical cells had enlarged vesicular nuclei along with prominent nucleoli and pale cytoplasm containing mucin [periodic acid-Schiff (PAS)-positive, diastase resistant] extending throughout the full length of the epidermis. There was no invasion to the underlying dermis or subcutis. Histopathology of the lesion from the groin was similar to that of axilla. Immunohistochemically, the large cells were negative for S100 protein but positive for cytokeratin 7 (CK7) and carcinoembryonic antigen (CEA). Both lesions were also negative for p63 immunohistochemistry marker, which suggested that this was a primary EMPD.[1] Fungal elements were absent on PAS and Grocott stains. Based on the investigations and histopathology findings, the diagnosis was confirmed as primary EMPD of the left axilla and the groin. Figures 3–5 display the histopathology and staining of the inguinal lesion.

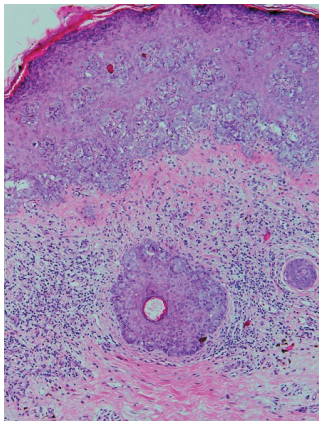

Figure 3. Intra-epidermal proliferation of Paget's cell in nests [10× magnification, haematoxylin and eosin (H&E)]

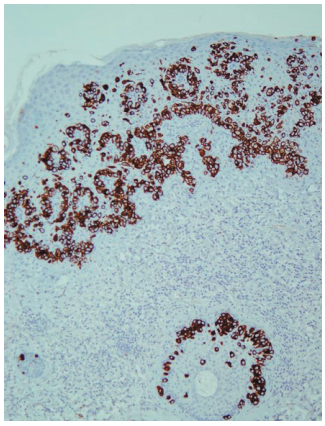

Figure 4. The Paget's cell stained positive for cytokeratin 7

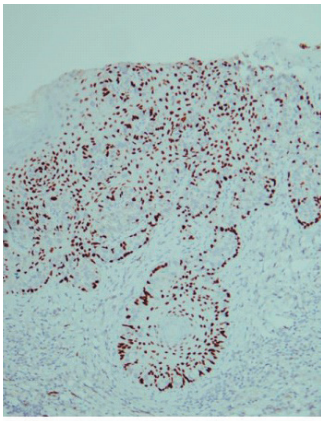

Figure 5. The Paget's cells stained negative for p63

A combination of surgery and radiotherapy was planned for the patient. The lesion on the left axilla was planned for excision because it was smaller, whereas radiotherapy was planned for the groin lesion owing to its extensive involvement.

Discussion

EMPD is a slow-growing adenocarcinoma developing in the apocrine gland–bearing skin, such as vulva, perianal, axilla and penoscrotal areas.[2] Rarely, the sites devoid of this gland, such as the back and umbilicus, have also been involved.[3,4] EMPD is similar to Paget's disease of the breast except that it occurs in sites away from the breast and is more common among the elderly. EMPD is a rare skin disorder usually occurring at a single site. Although multiple-site involvement of EMPD is uncommon, a number of cases have reported concurrent multiple EMPD, especially from Japan.[2,3]

The exact relationship between EMPD and an underlying malignancy is uncertain. It is believed that up to 42% of patients with EMPD may have an underlying primary or non-cutaneous malignancy.[5] EMPD is generally

classified as primary or secondary. Primary EMPD arises from the intra-epidermal cells and is generally not associated with any underlying carcinoma. In a recent finding, Toker cells were postulated as the precursor for primary EMPD.[6] Secondary EMPD, in contrast, has a non-cutaneous origin and is associated with underlying adnexal or visceral carcinoma, commonly of rectal or urothelial origin.[7] However, penoscrotal EMPD may have a lower incidence of internal malignancy (9%), mainly originating from the genitourinary system.[8] Axillary lesions may be associated with breast malignancy. Hence, the diagnosis of EMPD warrants the search for possible underlying malignancy, particularly in younger patients and those with perianal or invasive disease.[9,10]

The initial diagnosis of EMPD is often delayed or missed such as in this case, as its symptoms and morphology mimics those of various types of dermatoses, for example, flexural psoriasis, seborrhoeic dermatitis, eczema, candidiasis, tinea cruris and erythrasma. Heavy pigmentation of the lesion may be misdiagnosed as malignant melanoma.[11] In cases of multiple EMPD, two to four lesions may occur concurrently at different sites.[2,4,9]

The standard management strategy for primary EMPD is wide surgical excision; however, the recurrence rate is high (20%–50%).[12] Other options include radiotherapy, chemotherapy, photodynamic therapy or laser therapy depending on the site and extent of the lesion. Radiotherapy is more suitable for extensive involvement, for lesions located at a site where excision would be difficult or when the patient is unfit for surgery.[9]

Recently, topical imiquimod, which is an immune response modifier, used as first line therapy, showed an 80% cure rate with minimal side effects. It may be considered in situations where the lesion is extensive or in cases where surgical excision is not a suitable option, for example, advanced age or due to concurrent multiple medical conditions. However, data pertaining to recurrence after treatment with imiquimod are still lacking.[13]

In general, the prognosis for primary EMPD is favourable provided that it is detected early and given adequate treatment.[4] Ishihara et al. reported that some patients with both axillary and genital involvement did not develop invasive carcinoma even after many years of post-treatment follow-up.[14]

Conclusion

EMPD, although rare, must be considered as a differential diagnosis in elderly patients presenting with chronic dermatitis or candidiasis, especially when the condition does not respond to adequate therapy. Patients presenting with clinical suspicion of EMPD should be examined for the possibility of multiple site involvement, such as in this case. In suspected cases of secondary EMPD, investigation for primary malignancy in the gastrointestinal or genitourinary systems is necessary. Although primary EMPD is highly recurrent, the overall prognosis is promising, especially for smaller lesions and those detected early.

Acknowledgement

The authors would like to thank the patient for his consent to publish this case and the images. We also like to thank Dr. Suria Hayati and Dr. Fazarina from Department of Pathology of Universiti Kebangsaan Malaysia Medical Centre for the histopathology images and interpretation.

References

1. Yanai H, Takahashi N, Omori M, et al. Immunohistochemistry of P63 in primary and secondary valvular Paget's disease. *Pathol Int*. 2008;58:648–51.
2. Sawada Y, Bito T, Kabashima R, et al. Ectopic extramammary Paget's disease: Case report and literature review. *Acta Derm Venereol*. 2010;90(5):502–5.
3. Kawatsu T, Miki Y. Triple extramammary Paget's disease. *Arch Dermatol*. 1971;104:316–9.
4. Abe S, Kabashima K, Nishio D, et al. Quadruple extra-mammary Paget's disease. *Acta Derm Venereol*. 2007;87(1):80–1.
5. Lai YL, Yang WG, Tsay PK, et al: Penoscrotal extramammary Paget's disease: A review of 33 cases in a 20-year experience. *Plast Reconstr Surg*. 2003,112(4):1017–23.
6. Hashemi P, Kao GF, Konia T, et al. Multicentric primary extramammary Paget disease: A Toker cell disorder? *Cutis*. 2014;94(1):35–8.
7. Chanda JJ. Extramammary Paget's disease: Prognosis and relationship to internal malignancy. *J Am Acad Dermatol*. 1985;13(6):1009–14.

8. Park S, Grossfeld GD, McAninch JW, et al. Extramammary Paget's disease of the penis and scrotum: excision, reconstruction and evaluation of occult malignancy. *J Urol*. 2001;166(6):2112–7.

9. Li B, Ding Q. Extramammary Paget's disease involving the penis, scrotum and the axilla. *Australas J Dermatol*. 2011;52(1):e18–20.

10. Chiu TW, Wong PS, Ahmed K, et al. Extramammary Paget's disease in Chinese males: A 21-year experience. *World J Surg*. 2007;31(10):1941–6.

11. Hilliard NJ, Huang C, Andea A. Pigmented extramammary Paget's disease of the axilla mimicking melanoma: Case report and review of the literature. *J Cutan Pathol*. 2009;36(9):995–1000.

12. Tonguc E, Gungor T, Var T, et al. Treatment of recurrent vulvar Paget disease with imiquimod cream: A case report and review of the literature. *Arch Gynecol Obstet*. 2011;283:97–101.

13. Iavazzo C, Ntziora F, Karachalios C, et al. The clinical evidence and the role of imiquimod in the extramammary Paget disease. *Acta Dermato Venerol Croat*. 2014;22(2):103–9.

14. Ishihara K. Statistical study of extramammary Paget's disease in Japan. *Skin Cancer*. 1994;9:38.

CASE REPORT

Pneumoperitoneum or Chilaiditi's sign

Teoh SW, Mimi O, Poonggothai SP, Liew SM, Kumar G

Teoh SW, Mimi O, Poonggothai SP, et al. Pneumoperitoneum or Chilaiditi's sign. *Malays Fam Physician*. 2016;11(1);22-24.

Keywords:
Chilaiditi's sign, pneumoperitoneum, primary care Malaysia

Authors:

Teoh See Wie
(Corresponding author)
MBBS
Faculty of Medicine, University of Malaya, 50603 Kuala Lumpur.
Email: seewie@outlook.com

Mimi Omar
MFamMed
Klinik Kesihatan Kelana Jaya, Jalan SS 6/3A, 47301 Kelana Jaya, Selangor.
Email: drmimiomar@hotmail.com

Poonggothai S Periasamy
MBBS , LFOM
Klinik Kesihatan Kelana Jaya, Jalan SS 6/3A, 47301 Kelana Jaya, Selangor.
Email: drkothaiselva@yahoo.com

Liew Su May
MBBS, MMed (Fam Med) DPhil (Oxon)
Faculty of Medicine, University of Malaya, 50603 Kuala Lumpur.
Email: su_mayliew@um.edu.my

Gnana Kumar A/L P Gnanasuntharam
MBBS, FRCR MMed
Faculty of Medicine, University of Malaya, Kuala Lumpur.
Email: gnanak_rad@um.edu.my

Abstract

Chilaiditi's sign describes the incidental radiographic finding of the bowel positioned between the right diaphragm and the liver. This is often misdiagnosed as pneumoperitoneum or free air under the diaphragm, which may lead to unnecessary investigations or surgical procedures. Here, we report two incidental chest radiograph findings of air under the diaphragm in patients who were being screened for pulmonary tuberculosis. This case series highlights the importance of awareness of the diagnosis of Chilaiditi's sign to avoid unnecessary hospital referrals.

Introduction

The presence of air under the diaphragm is a surgical emergency until proven otherwise. However, Chilaiditi's sign is a rare exception. It describes the incidental radiographic finding of the bowel positioned between the right diaphragm and the liver. This is often misdiagnosed as pneumoperitoneum or free air under the diaphragm. The incidence of Chilaiditi's sign is 0.025%–0.28% globally.[1] It was first reported by Demetrius Chilaiditi, a radiologist, in 1910.[2] There have been two reported cases of Chilaiditi's sign in Malaysia to date.[3] In a primary care setting, clinical assessment can avoid unnecessary worry and referral, as this condition can be managed conservatively in asymptomatic patients.

Case report

Case 1

Ms. MAL, a 43-year-old Chinese woman, presented to a primary care clinic with cough and loss of appetite for 2 weeks. There was no weight loss, night sweat, haemoptysis or abdominal pain. MAL has Down syndrome and, according to her mother, only had a few minor ailments previously.

Physical examination results were normal. Her lungs were clear, and the abdomen was soft.

MAL was screened for pulmonary tuberculosis (PTB), for which a chest radiograph was performed (Figure 1). A large collection of air was found under the diaphragm on both sides, and the attending doctor suspected pneumoperitoneum. However, MAL was clinically well and did not exhibit any signs or symptoms of perforation. A radiologist was consulted regarding the abnormal radiological finding. The chest radiograph observation was reported as bilateral diaphragmatic interposition of the colon and diagnosed as Chilaiditi's sign. Other investigations for PTB yielded normal results, and the patient recovered after being treated for upper respiratory tract infection.

Case 2

Mr. TSF, a 78-year-old Chinese man, had productive cough with whitish phlegm for the past 3 weeks. He had PTB in his young age and had completed treatment. He was also undergoing treatment for hypertension and Parkinson disease, both of which were well controlled.

Physical examination results were normal. His lungs were clear, and the abdomen was soft and not distended.

Chest radiography was performed to exclude reactivation of PTB. It showed an old fibrotic scar at the right apex, with trachea deviation to the right and opacity at the left apex (Figure 2). A collection of air was noted separating the diaphragm and the liver at the right hypochondrium. The chest radiograph observation was reported as Chilaiditi's sign along with old PTB. Three different samples of sputum were analysed, which were negative for acid-fast bacilli. The patient responded to symptomatic treatment.

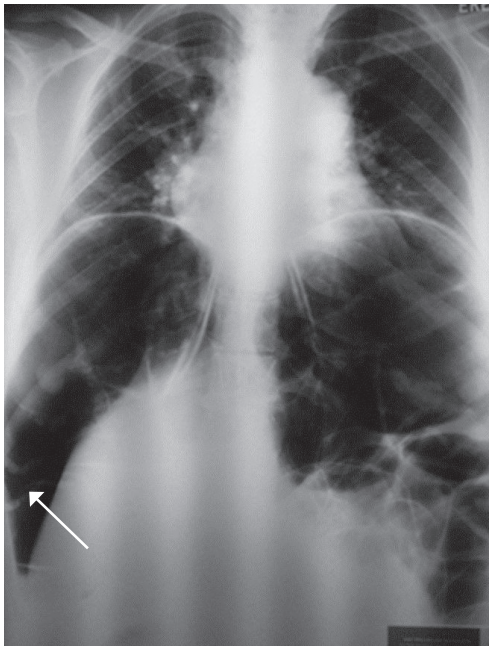

Figure 1. Chest radiograph of MAL showed diaphragmatic interposition of the colon. Note the haustra of the colon *(arrow)*

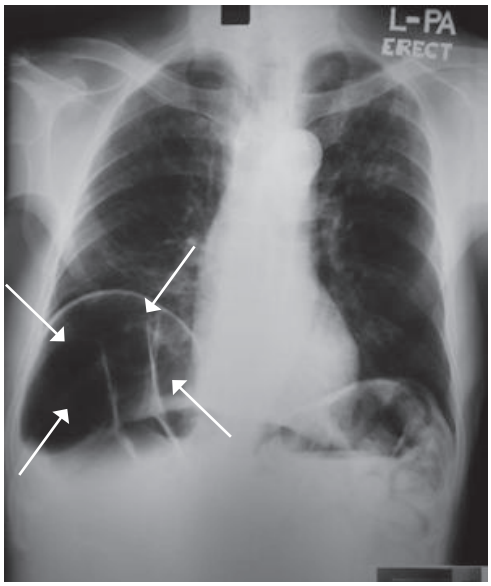

Figure 2. Chest radiograph of TSF showing the right hemidiaphragm *(arrows)* along with a distended colon and with air separating the diaphragm and the liver. Note the haustra of the colon *(arrow heads)*

Discussion

Chilaiditi's sign usually presents to the primary care practitioner as an incidental radiological finding as a result of investigations done for other diseases.[4] To differentiate Chilaiditi's sign from other differential diagnosis, three main elements that contribute to the presentation of the disease must be considered: the intestine, the liver, and the diaphragm.[5] There are three essential radiological characteristics to confirm the diagnosis.[6] First, the interposition must be significant enough to elevate the right hemidiaphragm. Second, the interposed colon (as seen by the haustral markings) must be distended with air and must lie between the diaphragm and the liver. Last, the liver must be displaced low enough that its upper margin lies lower than the left side of the diaphragm. These observations collectively are known as Chilaiditi's sign.[6] Both our patients fulfilled all the aforementioned criteria. If there is any doubt regarding the radiological findings, other differential diagnoses should be seriously considered. Pneumoperitoneum or free air under the diaphragm is most often caused by visceral perforation, which is a surgical emergency. Only 10% of the pneumoperitoneum cases are not surgical.[7] Other differential diagnoses not to be missed are perforated peptic ulcer disease, volvulus, intussusception, ischaemic bowel, and inflammatory conditions.[5]

History and physical examination play an important role in differentiating Chilaiditi sign from pneumoperitoneum. Patients who are asymptomatic and haemodynamically stable are likely to have Chilaiditi's sign.

The exact cause of Chilaiditi's sign is not clear, although a associations with schizophrenia and mental retardation, elderly male patients, obesity, chronic constipation, liver cirrhosis, chronic lung disease and multiple pregnancies have been reported.[5,6] Anatomical distortions involving the liver, diaphragm, and intestine arising from these disorders or congenital variations predisposed to the condition.

Although, according to the available literature, other imaging methods to confirm the diagnosis of Chilaiditi syndrome include the opaque enema technique and chest/abdominal computed tomography,[8] the availability of these resources in a primary care setting is limited.[9] In such a setting, the alternative is to repeat the abdominal radiograph with the patient in the left lateral decubitus position when the equipment required to perform this is available.[5] A change in the air location would indicate pneumoperitoneum and exclude Chilaiditi's sign.

Patients with Chilaiditi's sign are usually asymptomatic, but some cases may present

as Chilaiditi syndrome, a condition in which Chilaiditi's sign is accompanied by clinical symptoms. These symptoms can range from mild abdominal pain to acute bowel obstruction. The treatment for asymptomatic Chilaiditi's sign is generally conservative, which includes weight loss and change in decubitus position. For Chilaiditi syndrome, referral to a surgeon is mandatory if the patient has signs and symptoms of intestinal obstruction. Symptoms usually require hospital admission, although conservative management with bowel decompression and follow-up radiography is usually successful.[5] Surgical intervention would be the only option of treatment if the aforementioned options fail.

Conclusion

A radiographic finding of air under the diaphragm warrants careful history taking and physical investigation. Possible life-threatening emergencies must be excluded before the diagnosis of Chilaiditi's sign is made. Accurate diagnosis of Chilaiditi's sign in asymptomatic patient in primary care prevents unnecessary referrals or procedures.

Conflict of interest

None.

Source of funding

None.

Acknowledgement

We would like to thank the Director General of Health, Malaysia for permission to publish this paper.

References

1. Orangio GR, Fazio VW, Winkelman E, et al. The Chilaiditi syndrome and associated volvulus of the transverse colon. An indication for surgical therapy. *Dis Colon Rectum*. 1986;29(10):653–6.
2. Chilaiditi D. Zur frage der hepatoptose und ptose im allgemeinen im anschluss an drei fälle von temporärer, partieller leberverlagerung. *Fortschritte auf dem Gebiete der Röntgenstrahlen*. 1910;16:173–208.
3. Hisham AN, Gunn A, Jamil AA. Chilaiditi's syndrome presenting as acute abdomen. *Med J Malaysia*. 1995;50(3):281–3.
4. Alva S, Shetty-Alva N, Longo WE. Image of the month. Chilaiditi sign or syndrome. *Arch Surg*. 2008;143(1):93–4.
5. Moaven O, Hodin RA. Chilaiditi syndrome: A rare entity with important differential diagnoses. *Gastroenterol Hepatol (N Y)*. 2012;8(4):276–8.
6. Lekkas CN, Lentino W. Symptom-producing interposition of the colon. Clinical syndrome in mentally deficient adults. *JAMA*. 1978;240(8):747–50.
7. Mularski RA, Sippel JM, Osborne ML. Pneumoperitoneum: A review of nonsurgical causes. *Crit Care Med*. 2000;28(7):2638–44.
8. Nagem R, Rachid Guimarães N, Henrique Leite F. Síndrome de chilaiditi: Relato de caso chilaiditi's syndrome: A case report. *Radiologia brasileira*. 2011;44(5):333–35.
9. Prassopoulos PK, Raissaki MT, Gourtsoyiannis NC. Hepatodiaphragmatic interposition of the colon in the upright and supine position. *J Comput Assist Tomogr*. 1996;20(1):151–3.

CASE REPORT

Isolated unilateral sixth cranial nerve palsy: A rare presentation of dengue fever

Mazliha M, Boo YL, Chin PW

Mazliha M, Boo YL, Chin PW. Isolated unilateral sixth cranial nerve palsy: A rare presentation of dengue fever. *Malays Fam Physician.* 2016;11(1);25-26.

Keywords:
Dengue, sixth cranial nerve, abducens nerve

Authors:

Yang Liang Boo
(Corresponding author)
MD, MRCP
Department of Medicine, Hospital Enche' Besar Hajjah Khalsom, Jalan Kota Tinggi, 86000 Kluang, Johor Darul Takzim, Malaysia.
Email: coolrontin@gmail.com

Mazliha Mashor
MBChB, MRCP
Department of Medicine, Hospital Enche' Besar Hajjah Khalsom, Jalan Kota Tinggi, 86000 Kluang, Johor Darul Takzim, Malaysia.
Email: ammz83@yahoo.com

Pek Woon Chin
MBBS, MRCP
Department of Medicine, Hospital Enche' Besar Hajjah Khalsom, Jalan Kota Tinggi, 86000 Kluang, Johor Darul Takzim, Malaysia
E-mail: chinpw1964@yahoo.com

Abstract

Dengue fever is a common mosquito-borne viral infection endemic in tropical and subtropical countries. Neurological manifestations in dengue infection are relatively uncommon, and include encephalitis, encephalopathy, neuromuscular disorders and neuro-ocular disorders. Cranial mononeuropathy is a rare manifestation of dengue infection. A 40-year-old man was diagnosed with isolated, unilateral sixth cranial nerve palsy complicating dengue infection. The patient was managed accordingly, and full ocular recovery was observed. This was the first reported case of isolated sixth cranial nerve palsy associated with dengue fever in Malaysia. It is important for clinicians to consider dengue as a differential diagnosis in patients presenting with fever and sixth cranial nerve palsy.

Introduction

Dengue fever is a mosquito-borne viral infection endemic in tropical and subtropical countries.[1] It has four distinct, but closely related, serotypes (DEN-1 to DEN-4).[1,2] Its clinical presentations can vary from asymptomatic to life-threatening infections.[2] The involvement of the central nervous system has been classified as severe dengue in the recent guidelines published by the World Health Organization (WHO).[2] These neurological manifestations can be broadly categorized into encephalitis, neuromuscular, and neuro-ophthalmic complications with possible overlapping in certain cases.[3] To our best knowledge, this was the first reported case of dengue-related unilateral, sixth cranial nerve palsy in Malaysia.

Case report

A 40-year-old man presented with acute-onset binocular diplopia for 2 days associated with fever for 4 days. No other neurological symptoms or bleeding tendency was observed. Also, no significant past medical illness or recent traveling history was found.

On arrival, his vital signs were stable. He displayed left-sided convergent squint and restriction of lateral gaze, consistent with left sixth cranial nerve palsy (Figure 1). Otherwise, neurological examination of other cranial nerves, visual acuity and fundoscopy examination were unremarkable. No evidence of plasma leakage was observed

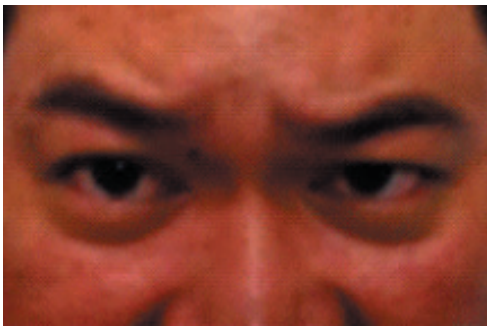

Figure 1. Neutral position of the patient's eyes showed convergent squint of the left eye

The initial laboratory investigations revealed haemoconcentration (haematocrit, 56%) along with normal white cell counts (7×10^9/L) and thrombocytopenia (12×10^9/L). His liver function test showed elevated levels of aspartate transaminase (107 IU/L) and alanine transaminase (62 IU/L). His renal profile and chest radiograph were normal. Both dengue non-structural protein 1 (NS-1) antigen and dengue-specific immunoglobulin (IgM) results were positive. Contrast-enhanced computed tomography (CECT) of the brain was performed, and reported as normal. Fasting blood sugar, human immunodeficiency virus (HIV), rapid plasma reagin (RPR), anti-nuclear antibody (ANA), anti-neutrophil cytoplasmic antibody (ANCA), and complement C3 and C4 test results were negative. Lumbar puncture was not carried out due to bleeding risk in the presence of severe thrombocytopenia. Nerve conduction study and electromyography were not performed.

He was diagnosed with dengue fever complicated with isolated left abducens nerve palsy and managed accordingly. His diplopia started to improve on the following day. He was discharged well, and on follow-up visit after 1 month, his convergent squint had completely resolved.

Discussion

Dengue fever is a global public health problem in endemic countries.[1] The complexity of various manifestations of dengue is attributed to the dynamic and systemic nature of the disease.[2] The ophthalmic complications of dengue infection were once thought to be rare; however, the incidence increased with more reported cases.[3] Most of the literature had identified maculopathy as the most common neuro-ocular complication, whereas less common presentation include retina vasculopathy, optic neuropathy and cranial nerve palsy.[4] Dengue-associated cranial neuropathy, especially abducens nerve, was relatively rare, with only a few reported cases.[5,6] Other cranial neuropathies included facial nerve and oculomotor nerve involvements.[7,8] Visual blurring is the most common presenting eye symptom, while other visual symptoms include conjunctival redness, retro-orbital pain, scotoma, colour vision abnormality and double vision.[4]

The pathogenesis in dengue-related neuro-ophthalmic complication was believed to be immune mediated, although the exact mechanism is not completely understood.[3] The delayed onset of visual symptoms of up to 1-week following dengue infection favours the immune-mediated theory.[3] Our patient presented with convergent squint 2 days after the onset of dengue fever, and this was consistent with the delayed presentation of ocular manifestation reported previously. Lesions that cause raised intracranial pressure with false localising signs such as tumour and intracranial haemorrhage, demyelinating diseases such as multiple sclerosis, infections such as meningitis and encephalitis, trauma and stroke need to be excluded accordingly.

The management of dengue-related abducens nerve palsy was mainly supportive, with improvement observed in previously reported cases.[5,6] Our patient had improved with expectant management. Similarly, patients with other dengue-associated cranial nerve palsies, such as Bell's palsy or oculomotor nerve palsy, have been reported to improve without any specific therapy such as steroid or intravenous immunoglobulin.[7,8] The overall prognosis for dengue-related ocular complications was good, and complete recovery coincided with improved platelet counts.[3,4]

Conclusion

With the cumulative number of dengue cases, it is important for clinicians to include dengue fever as one of the differential diagnoses for patients presenting with fever and isolated sixth cranial nerve palsy.

Acknowledgement

The authors would like to thank the Director General of Health Malaysia for permission to publish this paper.

Funding and conflicts of interest

None.

References

1. Murray NEA, Quam MB, Wilder-Smith A. Epidemiology of dengue: Past, present and future prospects. *Clini Epidemiol*. 2013: 299–309.
2. WHO. Dengue: guidelines for diagnosis, treatment, prevention, and control. Special Programme for Research and Training in Tropical Diseases. 2009. 147 p.
3. Carod-Artal FJ, Wichmann O, Farrar J, et al. Neurological complications of dengue virus infection. *Lancet Neurol*. 2013;12(9):906–19.
4. Yip VC-H, Sanjay S, Koh YT. Ophthalmic complications of dengue fever: A systematic review. *Ophthalmol Ther*. 2012;1(1):1–19.
5. Shivanthan MC, Ratnayake EC, Wijesiriwardena BC, et al. Paralytic squint due to abducens nerve palsy : A rare consequence of dengue fever. *BMC Infect Dis*. 2012;12(1):156.
6. Mishra A, Shukla S, Aggarwal S, et al. Lateral rectus palsy in a case of dengue fever. *Med J Armed Forces India*. 2013;1:10–2.
7. Peter S, Malhotra N, Peter P, Sood R. Isolated Bell's palsy–An unusual presentation of dengue infection. *Asian Pac J Trop Med*.2013;6(1):82–4.
8. Biswas NM, Pal S. Oculomotor nerve palsy in dengue encephalitis—a rare presentation. *Indian J Med Res*. 2014;140(6):793–4.

CASE REPORT

Immune reconstitution inflammatory syndrome in a HIV-infected patient with disseminated tuberculosis

Koh KC, Pak JW

Koh KC, Pak JW. Immune reconstitution inflammatory syndrome in a HIV-infected patient with disseminated tuberculosis. *Malays Fam Physician*. 2016;11(1);27-30.

Keywords:
Immune reconstitution inflammatory syndrome, HIV, tuberculosis, antiretroviral therapy

Authors:

Koh Kwee Choy
(Corresponding author)
MMed, MBBS
Department of Medicine, International Medical University, Jalan Rasah, 70400 Seremban, Negeri Sembilan, Malaysia.
Email: kweechoy_koh@imu.edu.my

Pak Jun Wee
MBBS
Hospital Raja Permaisuri Bainun, Jalan Hospital, 30990 Ipoh, Perak, Malaysia.
Email: joewee_12@hotmail.com

Abstract

Immune reconstitution inflammatory syndrome (IRIS) is the paradoxical worsening of pre-existing infectious processes after commencement of anti-retroviral therapy (ART) in HIV-infected patients. Its manifestations are dependent on the underlying opportunistic infections. We report a case of an HIV-infected patient with disseminated tuberculosis, who responded to anti-tuberculosis therapy but suffered from paradoxical worsening after commencement of ART.

Introduction

Immune reconstitution inflammatory syndrome (IRIS) occurs as a result of successful anti-retroviral therapy (ART), in which there is a sudden 'inflammatory flare' towards pre-existing infections that were previously subclinical due to immunosuppression.[1] The clinical features are closely linked to the type and location of the pre-existing infection. IRIS is usually self-limiting and is rarely fatal, especially if the pre-existing infection is effectively treated. Failure to recognize IRIS may lead to mismanagement and detrimental outcomes.

Case Summary

MF, a 35-year-old man, was first diagnosed in 2008 with human immunodeficiency virus (HIV) and hepatitis C virus (HCV) co-infection acquired from intravenous drug use with a history of needle sharing. He first presented on the 3rd of May 2013 with complaints of progressively enlarging painful neck swelling for more than 3 weeks associated with fever, night sweats and productive cough. In addition, he complained of significant weight loss and poor appetite for the past 6 months.

On examination, multiple tender and immobile cervical lymph nodes measuring 1–2 cm in diameter were observed. Although his chest radiograph results were normal, sputum analyses on the 7th and 8th of May 2013 showed the presence of acid-fast bacilli (AFB). Based on this, he was diagnosed with pulmonary tuberculosis along with tuberculous lymphadenitis and underlying HIV-HCV co-infection. At this point, fine-needle aspiration cytology (FNAC) and biopsy of the enlarged neck nodes were performed.

Oral first-line anti-tuberculosis (anti-TB) therapy was commenced for the patient, consisting of isoniazid, rifampicin, pyrazinamide and ethambutol with supplementation of pyridoxine and cotrimoxazole for prophylaxis against Pneumocystis jiroveci pneumonia (PJP), on the 10th of May 2015. When the patient was discharged on the 17th of May 2015, his symptoms of cough, fever and night sweats had resolved, and the neck swelling was noticeably reduced.

He returned for a review at the infectious diseases clinic on the 31st of May 2013. The neck swelling had completely disappeared. His CD4 cell count, on the 12th of May 2013, was 444 cells/mm^3. Taking into account the poor constitution of the patient at the time, ART regime was commenced consisting of stavudine, lamivudine and efavirenz for HIV infection, and the patient was advised to continue with the anti-TB therapy. Stavudine was chosen over the usual zidovudine, as the patient had anaemia at the time (haemoglobin level of 8.5 g/dL).

On the 24th of June 2013, MF presented with complaints of progressive painful enlargement of neck swelling for approximately 1 month, associated with intermittent fever for a week. Physical examination revealed multiple enlarged, tender, erythematous matted lymph nodes at the submental, submandibular and anterior cervical chain of the lymph nodes (Figures 1 and 2). Apart from that, examination of other systems yielded unremarkable results.

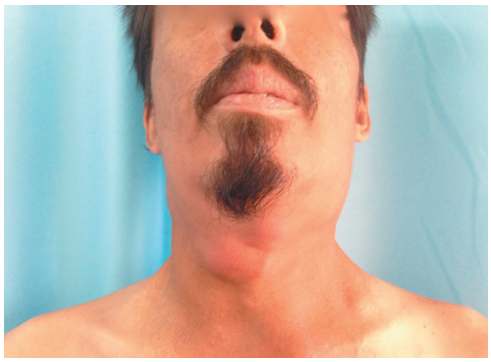

Figure 1. Anterior view of the neck swelling

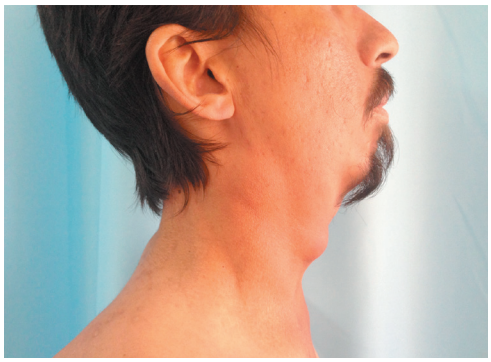

Figure 2. Lateral view of the neck swelling

Investigations showed elevated erythrocyte sedimentation rate (ESR) of 120 mm/h, C-reactive protein (CRP) level of 71.6 mg/L and lactate dehydrogenase (LDH) level of 292 U/L. Lymph node aspiration performed on the 26th of June 2013 was positive for AFB. A lymph node biopsy performed on the 27th of June 2013 was subsequently reported to show evidence of chronic granulomatous inflammatory process consistent with tuberculous lymphadenitis. However, special stains for AFB and fungal bodies were negative.

At this point, the diagnoses of drug-resistant *Mycobacterium tuberculosis*, non-tubercular *Mycobacterium* infection and IRIS were considered. Retrospective tracing of the initial sputum culture and sensitivity showed first-line anti-TB therapy–sensitive *M. tuberculosis*, ruling out the first two differential diagnoses. Careful history-taking revealed a temporal association between commencement of ART and the subsequent appearance of neck node enlargement.

A final diagnosis of IRIS was made. The same ART regimen, oral cotrimoxazole and anti-TB therapy were continued along with the addition of oral prednisolone at 50 mg daily (1 mg/kg/day). The cervical lymphadenopathy rapidly resolved in 2 weeks, with resolution of fever. The patient was discharged well on the 17th of June 2013, with continuation of the same ART regimen and oral cotrimoxazole; his anti-TB therapy regimen was converted to maintenance phase consisting of oral isoniazid and rifampicin.

Discussion

The incidence of IRIS in patients in whom ART is started varies with the pre-existing illness. Studies have reported IRIS in 63% of HIV-infected patients with cytomegalovirus retinitis, 30%–34% of those with inactive cryptococcus, and 30% of those with *M. tuberculosis* infection.[1,2]

Major risk factors for the development of IRIS include a CD4 cell count <100 cells/mm^3 (although IRIS may occur at any CD4 cell count), a heavy pathogen burden before ART initiation, and a short duration between treatment of the infection and commencement of ART.[3-8] IRIS usually presents within the first 4–8 weeks after the initiation of ART, although IRIS has been reported to occur as early as 3 days and up to several years after ART initiation.[5,9]

The immunopathology of IRIS is largely determined by the provoking pathogen.[10] In M. tuberculosis and other mycobacterial infections, granulomatous inflammation usually predominates,[2,10] as was the case in this patient. The IRIS phenomenon is presumed to be the result of partial reconstitution of the host immune response and a transient increase in inflammation.[11]

Although, no universally accepted diagnostic criteria for IRIS exist, a diagnosis of IRIS should be considered when all or most of the following features are present: HIV infection with low pretreatment CD4 count (one important exception to this rule is tuberculosis, in which IRIS can occur at any CD4 count), positive virological and immunological responses to ART, clinical features of inflammatory response, and a temporal association between the commencement of ART and the onset of clinical features of the illness.[12]

In tuberculosis-associated IRIS (TB-IRIS), manifestations include worsening of pulmonary symptoms or chest radiograph findings suggestive of worsening tuberculosis, enlarging lymph nodes, and meningeal symptoms.[5] TB-IRIS may develop as early as 10 days after initiation of ART to as long as 180 days, but usually occurs within 2 months of ART initiation.[13,14] Our patient had

cervical lymphadenopathy that resolved following anti-TB therapy only to recur and worsen after commencement of ART. This is known as *paradoxical worsening*, which is defined as the exacerbation or recurrence of symptoms from a previously known or treated disease after initiation of ART.[1]

The risk of IRIS is higher among patients in whom ART is initiated at earlier points than later within the course of TB treatment.[15,16] Increasing the duration of time between the treatment of known tuberculosis and the initiation of ART has been shown to decrease the incidence of IRIS.[17] Nevertheless, the benefits of early initiation of ART in these patients far outweigh the risks of morbidity associated with TB-IRIS. Although the recommended timing for initiation of ART in HIV-infected patients with tuberculosis and a CD4 cell count >350 cells/mm^3 is after the completion of anti-TB treatment, the decision to initiate ART earlier in this patient was taken on account of his severely malnourished state on presentation.[18,19]

Differential diagnoses that must be considered in such patients include anti-TB resistance, non-adherence to medication, drug hypersensitivity reaction and development of new occult infection. Effort must be made to rule these out before considering IRIS as the final diagnosis.

Patients should be treated for the underlying infection as soon as possible. Except in severe cases, ART should not be interrupted during the management of IRIS. Several case series have described the efficacy of corticosteroids in reducing the inflammatory response associated with IRIS.[10] Oral prednisolone at 1–2 mg/kg/day can be prescribed for 1–2 weeks followed by a taper. The potential benefits of corticosteroid therapy should be weighed against its risks, including worsening glycaemic control, hypertension, altered mental status, acute flare of infections and predisposition to new infections.

Conclusion

The clinician should be alerted to the possibility of IRIS after initiation of ART in HIV-infected patients treated with anti-TB therapy. Prevention of complications associated with IRIS requires careful monitoring, particularly in patients with low CD4 cell counts and a history of co-infections.

In cases of paradoxical worsening, treatment of the underlying infection should be continued to suppress replication of the provoking pathogen and reduce antigen load.[10] ART should only be ceased if the disease is life-threatening. Corticosteroids may be useful in reducing the inflammatory response associated with IRIS.[12]

Acknowledgement

There was no funding provided by any other resources. Ethics committee approval was not applicable.

Conflict of interest

None.

References

1. French MA, Price P, Stone SF. Immune restoration disease after antiretroviral therapy. *AIDS*. 2004;18:1615–27.

2. Phillips P, Bonner S, Gataric N, et al. Nontuberculous mycobacterial immune reconstitution syndrome in HIV-infected patients: Spectrum of disease and long-term follow-up. *Clin Infect Dis*. 2005;41:1483–97.

3. Muller M, Wandel S, Colebunders R, et al. Immune reconstitution inflammatory syndrome in patients starting antiretroviral therapy for HIV infection: A systematic review and meta-analysis. *Lancet Infect Dis*. 2010;10(4):251–61.

4. Manabe YC, Campbell JD, Sydnor E, et al. Immune reconstitution inflammatory syndrome: Risk factors and treatment implications. *J Acquir Immune Defic Syndr*. 2007;46:456–62.

5. Shelburne SA, Darcourt J, White AC Jr, et al. The role of immune reconstitution inflammatory syndrome in AIDS-related *Cryptococcus neoformans* disease in the era of highly active antiretroviral therapy. *Clin Infect Dis*. 2005;40:1049–52.

6. Breton G, Duval X, Estellat C, et al. Determinants of immune reconstitution inflammatory syndrome in HIV type 1–infected patients with tuberculosis after initiation of antiretroviral therapy. *Clin Infect Dis*. 2004;39:1709–12.

7. Lawn SD, Myer L, Bekker LG, Wood R. Tuberculosis-associated immune reconstitution disease: Incidence, risk factors and impact in an antiretroviral treatment service in South Africa. *AIDS*. 2007;21:335–41.

8. Burman W, Weis S, Vernon A, et al. Frequency, severity and duration of immune reconstitution events in HIV-related tuberculosis. *Int J Tuberc Lung Dis*. 2007;11:1282–89.

9. Aberg JA, Chin-Hong PV, McCutchan A. Localized osteomyelitis due to *Mycobacterium avium* complex in patients with human immunodeficiency virus receiving highly active antiretroviral therapy. *Clin Infect Dis*. 2002;35:E8–E13.

10. French MA. Immune reconstitution inflammatory syndrome: A reappraisal. *Clin Infect Dis*. 2009;48:101–7.

11. Navas E, Martin-Davila P, Moreno L, et al. Paradoxical reactions of tuberculosis in patients with the acquired immunodeficiency syndrome who are treated with highly active antiretroviral therapy. *Arch Intern Med*. 2002;162:97.

12. Murdoch DM, Venter WD, Feldman C, et al. Incidence and risk factors for the immune reconstitution inflammatory syndrome in HIV patients in South Africa: A prospective study. *AIDS*. 2008;22(5):601–10.

13. Shelburne SA 3rd, Hamill RJ, Rodriguez-Barradas MC, et al. Immune reconstitution inflammatory syndrome: Emergence of a unique syndrome during highly active antiretroviral therapy. *Medicine (Baltimore)*. 2002;81:213.

14. Shelburne SA, Montes M, Hamill RJ. Immune reconstitution inflammatory syndrome: More answers, more questions. *J Antimicrob Chemother*. 2006;57:167.

15. Abdool Karim SS, Naidoo K, Grobler A, et al. Integration of antiretroviral therapy with tuberculosis treatment. *N Engl J Med*. 2011;365:1492.

16. Abdool Karim SS, Naidoo K, Grobler A, et al. Timing of initiation of antiretroviral drugs during tuberculosis therapy. *N Engl J Med*. 2010;362:697.

17. Schiffer JT, Sterling TR. Timing of antiretroviral therapy initiation in tuberculosis patients with AIDS: A decision analysis. *J Acquir Immune Defic Syndr*. 2007;44:229–34.

18. Ministry of Health Malaysia. Management of tuberculosis 3rd edition. Putrajaya: Malaysia Health Technology Assessment Section (MaHTAS); 2012. Available from: http://www.moh.gov.my. Accessed 6 January 2016.

19. Blanc FX, Sok T, Laureillard D, et al. Earlier versus later start of antiretroviral therapy in HIV-infected adults with tuberculosis. *N Engl J Med*. 2011;365:1471.

An elderly with a 'bony' smile

Mohamad I, Nadarajah S

Mohamad I, Nadarajah S. An elderly with a 'bony' smile. *Malays Fam Physician.* 2016;11(1);31-32.

Keywords:
Osteonecrosis, maxilla, biphosphonate

Authors:

Irfan Mohamad
(Corresponding author)
M.D, M.Med (ORL-HNS)
Department of Otorhinolaryngology Head & Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia Health Campus, 16150 Kota Bharu, Kelantan, Malaysia.
Email: irfankb@usm.my

Sanjeevan Nadarajah
M.D, FRCS (C)
Department of Otorhinolaryngology Head & Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia Health Campus, 16150 Kota Bharu, Kelantan, Malaysia

Case summary

A 62-year-old Caucasian man presented with progressive painless loss of the upper dentition and discolouration of the upper gums. He was referred by a dentist after undergoing dental treatment with debridement twice before his presentation to the otolaryngology department. He had a long history of hypertension and was diagnosed with osteoporosis 3 years ago. He was on regular antihypertensive medication and had been taking oral bisphosphonate for more than 2 years for osteoporosis. He denied steroid use in any form. No history of malignancy, radiation to the head and neck region or pre-existing dental or oral cavity issues was found. He was a smoker of 30 pack-years and an occasional alcohol user.

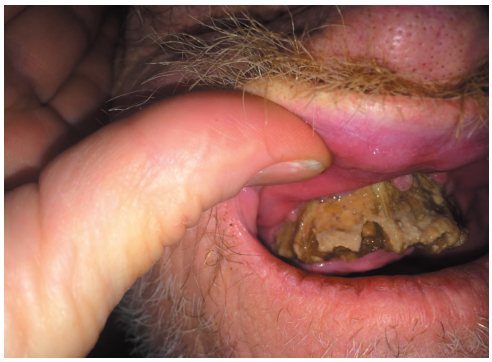

Figure 1. Maxilla can be seen upon smiling

Questions

1. Describe the abnormality?
2. What is the diagnosis?
3. What are the risk factors?
4. What are the options of management?

Answers

1. The oral cavity shows exposed maxillary alveolar bone, some residual gingival granulation tissue and the loss of upper dentition.

2. This is a classical appearance of 'medication-related osteonecrosis of the jaw (MRONJ)'[1]. In this case, it happened at the alveolar process of the maxilla.

3. Bisphosphonate treatment, treatment with other antiresorptive [e.g. receptor activator of nuclear factor-κB ligand (RANKL inhibitors)] or antiangiogenic agents (tyroxine kinase inhibitors), monoclonal antibody therapy against vascular endothelial growth factor (VEGF), prolonged duration of osteoporosis treatment, chronic corticosteroid treatment, chronic dental infection conditions such as periodontal disease and dental procedures (often the trigger) are some of the known risk factors.

The association between bisphosphonate and osteonecrosis of the jaw/maxilla has long been established.[2] However, previously it was predominantly observed after intravenous use of bisphosphonate for hypercalcaemia of malignancy and bone metastasis. A worrying trend has emerged recently as oral bisphosphonate is used for the treatment and prophylaxis of osteoporosis and osteopenia.[3]

The incidence of osteopenia and osteoporosis is bound to increase with increasing life expectancy. It is inevitable that treatment complications of the aforementioned conditions will be on the rise. The risk of new cases of MRONJ varies between 1/100,000 and 1/10,000 per year of therapy.[4,5] Duration of treatment has a negative impact. Thus, it is important that primary care physicians, specialist physicians and dentists are aware of this condition so that prompt and early action can be taken. If a patient has risk factors such as a history of cancer, chemotherapy, radiation therapy or steroid use, the implementation of dental screening and appropriate dental measures before initiating antiresorptive therapy can lower the risk.[1]

4. When a patient is diagnosed with MRONJ, the subsequent treatment aim is to eliminate pain, control the secondary infection to the soft and hard tissues and minimise the progression of bone necrosis.[1] The treatment can be non-surgical or surgical depending on the severity at presentation.

References

1. Ruggiero SL, Dodson TB, Fantasia J, et al. American Association of Oral and Maxillofacial Surgeons position paper on medication-related osteonecrosis of the jaw—2014 update. *J Oral Maxillofac Surg*. 2014;72(10):1938–56.

2. Marx RE. *Oral and Intravenous Bisphosphonate-Induced Osteonecrosis of the Jaws*. Hanover Park, IL:Quintessence Publishing Co; 2007.

3. López-D'alessandro E, Mardenlli F, Paz M. Oral bisphosphonate-associated osteonecrosis of maxillary bone: A review of 18 cases. *J Clin Experimental Dent*. 2014;6(5):e530–4. doi:10.4317/jced.51694.

4. Advisory Task Force on bisphosphonate-related osteonecrosis of the jaws, American Association of Oral and Maxillofacial Surgeons. American association of oral and maxillofacial surgeons position paper on bisphosphonate-related osteonecrosisof the jaws. *J Oral Maxillofac Surg*. 2007;65:369–76.

5. Ruggiero SL, Dodson TB, Assael LA, et al. American Association of Oral and Maxillofacial Surgeons position paper on bisphosphonate-related osteonecrosisof the jaws-2009 update. *J Oral Maxillofac Surg*. 2009;67:2–12.

A teenager with mucoid discharge from the neck

Mohamad I

Mohamad I. A teenager with mucoid discharge from the neck. *Malays Fam Physician.* 2016;11(1);33-34.

Keywords:
Neck, Mass, Congenital

Authors:

Irfan Mohamad
(Corresponding author)
M.D, M.Med (ORL-HNS)
Department of Otorhinolaryngology Head & Neck Surgery, School of Medical Sciences, Universiti Sains Malaysia Health Campus, 16150 Kota Bharu, Kelantan, Malaysia.
Email: irfankb@usm.my

Case summary

A 15-year-old boy presented with a history of recurrent discharge from an opening located at the lower anterior part of the neck since childhood. Occasionally, the surrounding area became swollen, and the amount of discharge increased. Examination revealed a non-tender fullness at the right lower one-third of the sternocleidomastoid region. A punctum with colourless mucoid discharge was also observed. The amount of discharge increased on applying pressure (Figures 1 and 2).

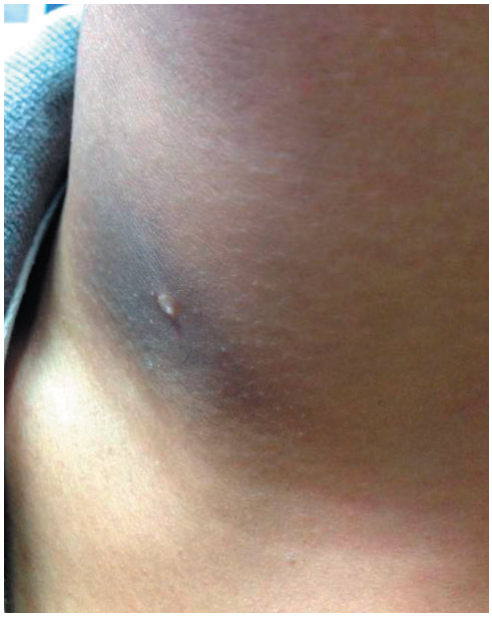

Figure 1. A punctum with colourless mucoid discharge

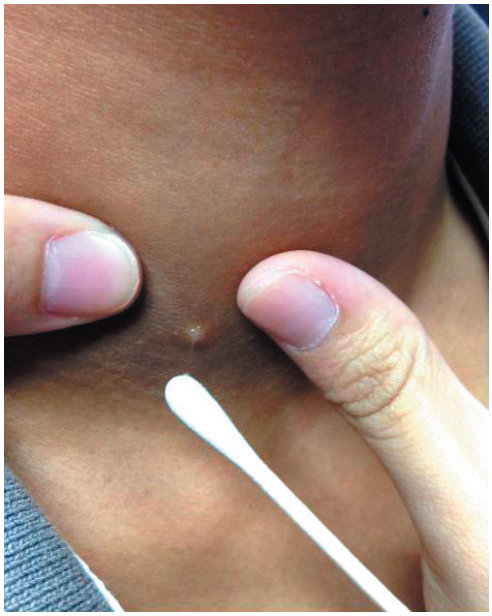

Figure 2. Increased discharge on applying pressure

Questions

1. What are the differential diagnoses?
2. State the required investigation.
3. Outline the management.

Answers

1. The presence of punctum indicates a communication between the deeper structure or space and the skin. In this case, the problem began in childhood. The most probable diagnosis is a branchial sinus, which is congenital in origin. The patients commonly present in the first 2 decades of life.[1] A complete branchial fistula (internal and external openings) needs to be ruled out. Sebaceous cyst, cervical abscess and suppurative thyroid lesions are differential diagnoses.[2]

2. A discharging sinus in the neck must be imaged to establish whether the communication ends in a blind sac (a sinus) or connects to the aerodigestive tract (a complete fistula). A fistulogram should be performed by injecting a contrast dye into the tract, and the flow should be captured on fluoroscopy.[2] In this case, the study showed a small loculated contrast collection at the anterolateral aspect of the right lower neck, approximately 1 cm beneath the surface of the tract opening. No communication with the internal lumen was observed.

   During the 2nd to 6th week of embryological life, six branchial arches develop to form the important structures in the head and neck region. Even though the underlying pathogenesis of branchial anomalies remains uncertain, the most

widely accepted theory is the incomplete obliteration of the branchial apparatus, resulting in the formation of cyst, sinus or fistula.[3] While most of the cases are unilateral, bilateral and multiple-arch abnormalities in a single patient have been reported.[4]

3. The patient can be reassured that this lesion is benign. However, the treatment of choice is complete surgical excision of the fistulous tract.[1,3] In this case, the surgery was indicated because the reservoir of mucoid content was present. This lesion may get infected and abscess can develop. Incision and drainage alone will not be a sufficient treatment strategy, as the fluid-containing sac will still be present under the skin.

References

1. Shankar VG, Babu TA, Swami HB. A rare case of complete second arch branchial fistula in a 7-year-old child. *Natl J Maxillofac Surg*. 2012;3(2):226–8.
2. Dench JE, Erdozain S, Nagendran R, et al. A complete second arch branchial fistula mistaken for a right-sided sebaceous cyst. *Ann R Coll Surg Engl*. 2004;86(6):W10–1.
3. Teo NW, Ibrahim SI, Tan KK. Distribution of branchial anomalies in a paediatric Asian population. *Singapore Med J*. 2015;56(4):203–7.
4. Singh AP, Kumar V, Narula V, et al. Bilateral first and second arch anomalies: A rare presentation. *Singapore Med J*. 2012;53(4):74–6.