PST

PEDIATRIC SURGERY IN TROPICS

www.pediatricsurgeryintropics.com

Volume 1 Issue 4 October-December 2024

Official Journal of
The Association of Pediatric Surgeons in Tropics

Platinum Open Access Journal

Pediatric Surgery in Tropics

Official journal of the Association of Pediatric Surgeons in Tropics

October - December 2024 Volume 1 Issue 4

Address of the publisher

Postal: Dr. Vivek Gharpure, 13. Pushpanagari, Dr. Ambedkar Road, Aurangabad 431001
Email: editor@pediatricsurgeryintropics.com
Telephone: +91-9325212384

Published by the Association of Pediatric Surgeons in Tropics

Panel of Accredited Reviewers of PST

Pediatric Surgery in Tropics

About the journal

Aims and objectives

Pediatric Surgery in Tropics (PST) aims to improve scientific communication among pediatric surgeons of tropical countries. It is intended to be an author friendly journal. The journal focuses more on the content of manuscripts rather than on their presentation or formatting. We understand the compelling circumstances in which tropical doctors are working. Therefore, we sympathetically accept to publish even partial evidences when advanced facilities for complete work-up are not available. Socio-cultural milieu, economic inequality, limited resources, illiteracy of patients and lack of advanced training that are typical of tropical countries will be taken into account while accepting articles for publication.

What do we publish?

We publish all types of scientific manuscripts that are useful to practicing pediatric surgeons. We publish not only diseases peculiar to tropical countries but also on all aspects of pediatric surgery pertinent to tropical or resource poor settings. We are not limited to article types such as original articles, review articles, case reports, images, letters and viewpoints. Rather we consider every article as a useful scientific communication. Thus we publish even unconventional category of articles. The smallest article we publish is 'clinical tips' of 50-100 words and the largest manuscript is review articles of several thousand words. Opinions regarding legislations and policies that may affect the practice of pediatric surgery will also be considered. We also publish patient perceptions that may provide insightful information to doctors.

Copyright

Economic Model

PST is an e-journal. It is managed on frugality principles. We avoid printing the journal, thereby saving expenditure and remain eco-friendly. Print copies are made available only on special request. All the editors and reviewers offer voluntary free service. The minimal cost of running the journal is met out by charity donations and advertisements. PST is a platinum open access journal that is made available free of cost to all individual readers except institutions and libraries. We neither charge our authors; nor pay them any royalty.

Advertising policy

PST accepts voluntary advertisements from individuals, institutions and manufacturers. Advertisements may be placed in journal website or within journal pages. Advertisements should be useful to the practicing pediatric surgeons and should be pertinent to the professional services. Advertised material should not violate international laws, child safety, public decency, professional ethics and scientific principles. Editors of PST hold no responsibility for the authenticity of advertised materials. Institutional job vacancies, conference announcements, sale of clinics or pre-owned equipments, pharmaceutical products, surgical instruments, prosthetic materials, book releases and such items are accepted for advertisement. All the advertised products shall have direct relevance to the practice of pediatric surgery. Tariff for advertisements:
Web advertisement (renewed quarterly) - INR 2000
Journal pages (Institutions) - INR 2500 for half page
Journal pages (Individuals) - INR 500 for quarter page

Indexing

PST values practical usefulness of articles more than the impact factor of the journal. PST will shortly be indexed in data bases.

Compliance

PST subscribes to the norms of COPE, WAME and ICMJE.

Pediatric Surgery in Tropics

Author instructions

Pediatric Surgery in Tropics (PST) is an author friendly journal. Keeping in mind the limitations of authors from developing countries, we do not strictly look into the manuscript formatting. Rather we only see if there is a clinically useful message in the manuscript. Hence, we request our authors to concentrate more on the content rather than on the format. Those who prefer to use templates (available in PST website) may use it; other may simply type their text in plain Microsoft word or RTF file and send it by email attachment to the editor: **editor@pediatricsurgeryintropics.com**

Title page should include all the following components:

1. Title of the paper
2. List of all authors in preferred order
3. Institutional affiliation of each author
4. Address of the corresponding author (both postal and email) and telephone or mobile phone numbers
5. Three to four keywords for indexing
6. Source of funding
7. Conflicts of interest statement
8. Statement of compliance with ethical standards
9. Patient consent for publishing identifiable details.

Each manuscript should include the following details:

- Why was the paper written (introduction, the background, problem statement)
- What was done (methods and materials)
- What was observed (Results of studies or case report)
- What do the authors want to say on this topic (Discussion, opinions, recommendations, criticisms, conclusions)
- References to support the authors' statements

We do not bother our authors with stringent word limitations. We only expect them to write briefly and succinctly without losing the readability and usefulness of the text. Number of references should also be kept to minimum necessary number, but sufficient enough to support statements. If the articles with useful message are boringly lengthy, our editorial team will suggest places where trimming is required (Author compliance with editorial suggestions is a precondition for final acceptance).

Illustrations should really illustrate. We believe in the aphorism "one picture should replace 1000 words". Although there are no limitations on the number of illustrations, they should be kept to the minimum required numbers. We encourage colour figures without any charging. We request high quality figures which are preferably more than 300 dpi in pixel resolution.

Tables are better submitted as word file and not as Excel or other formats. Authors are requested to use table maker of Microsoft word.

During initial submission all figures and tables can be incorporated at appropriate places of the text file and the entire manuscript is submitted as a single word file. After peer review, we may ask to provide separate figure files if required for better clarity of reproduction. In such case, figures can be submitted as JPEG or TIFF files. Resolution must be more than 300 dpi.

References should be in Vancouver style. Copy-pasting of citation details from PubMed is desirable. List references at the end of main text in the order of their citation in the text. Refer to them in text by superscript serial numbers within brackets.

When the manuscript is ready, please run 'spelling and grammar check' of Microsoft word. Submit the article as email attachment along with the following documents to **editor@pediatricsurgeryintropics.com**. Documents to accompany the article submission: (1) Covering letter, (2) Author declaration signed by corresponding author on behalf of all authors.

PST does not tolerate any form of publication misconduct including plagiarism. Authors are urged to paraphrase their sentences to avoid embarrassment. PST will deal with publication misconducts on the established principles of COPE.

Address of the Chief Editor

Dr. Vivek Gharpure, 13. Pushpanagari, Dr. Ambedkar road, Aurangabad 431001. Telephone: 9325212384 Email editor@pediatricsurgeryintropics.com

PEDIATRIC SURGERY IN TROPICS (PST)

Author declaration

Title of paper _____

Authors list in order _____

_____

Date of submission _____

Corresponding author _____

I, Dr _____, the corresponding author of the above mentioned paper, hereby sign the following declaration which is legally binding.

1. I sign this document on behalf of all my co-authors noted above. It is my responsibility to keep my coauthors informed and get their concurrence regarding the publication of the above mentioned manuscript. In case of any dispute between authors, it is my responsibility to settle the matter.
2. I assure that the manuscript is exclusively submitted to **Pediatric Surgery in Tropics (PST).** It has not been previously published in any other journal, magazine, print media or electronic media of any kind.
3. I have declared in the manuscript if this paper has been presented orally at any conference or if the abstract (alone) is published previously. I have included the full details of such presentations in my editorial communication.
4. After publication in PST, I shall not publish the same manuscript in any other media unless it is permitted by the editor of PST.
5. All authors of the paper satisfy authorship criteria of ICMJE / WAME / CSE. I assure that there are no gift authorship / ghost authorship. I also assure that no eligible author is omitted from the author list. Addition, deletion or change of author order is not permissible after the final PDF proof is accepted by the corresponding author.
6. I assure that artificial intelligence was not used in writing this paper. If used, I have explicitly mentioned the role and degree of AI usage.
7. I assure that the text is not copy pasted from another source. I assure that the manuscript do not contain any copyrighted materials including texts, tables and figures.
8. I assure that figures are original and no image manipulation (other than trimming of size) has been done.
9. I assure that patient identity / privacy / rights are protected. When it is unavoidable to reveal identity, necessary written permission is secured from patients. International regulations on human experimentation (Helsinki declaration) and human rights (Geneva declaration) have been followed while conducting / reporting the study. When required I have also secured necessary permission from Institute Ethics Committee (IEC).
10. Although authors retain copyright, the manuscript published in PST shall not be reproduced for commercial purpose. When it is reproduced for educational activities, it shall be accompanied by a note of acknowledgement attributing it to PST.
11. Parts or full text of the articles may be reproduced for non-commercial activities provided the original source is explicitly acknowledged as PST.

12. Authors desirous of republishing PST articles for commercial purpose shall get prior permission of editor of PST and 5% of gross sale proceedings shall be paid to the PST journal fund. A special agreement shall be signed by all stakeholders for this purpose.
13. I acknowledge that PST does not pay any royalty to authors on publication of article.
14. I assure that the content of the article is authentic and original. I have the full control of all data presented in the paper and I shall submit raw data when required by editor PST.
15. I indemnify editors of PST against all legal, academic, administrative claims / disputes that may possibly arise from publication of this article.
16. Neither I, nor any of my coauthors have any conflicts of interest (COI) in publishing this article. I have explicitly mentioned in the manuscript of any such potential COI. I have also declared the source of any external funding of the study.
17. I acknowledge that editors of PST by their discretionary powers may retract our article even after publication if any violation of undertaking is detected or any publication misconduct is proved. Editors of PST may publish a note of concern / retraction mentioning the names of the authors. Editors may also approach the institutional heads of the authors notifying the publication misconduct of authors. I forfeit the right to sue PST or its editors/publisher on this ground.
18. Editor of PST has the right to modify the submitted article provided the proof of such modification is finally approved by the corresponding author (in liaison with all coauthors). Once the final proof is approved, thereafter the authors forfeit any right to ask for further modification of the article.
19. I grant editors of PST to publish / republish the article in any media (both print and electronic) and to store it in appropriate repository.
20. I agree that jurisdiction of any legal dispute of this publication shall be at the place of chief editor of PST.

Name _____ Signature _____ Date _____

(Signed on behalf of all coauthors)

Contents

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 203-205
DOI: 10.5281/zenodo.13893261

Editorial

Challenges and Safety of Women Surgeons during Residency and Practice

Jameel-Ahamed Sulthana Dhilras

Department of Pediatric Surgery, Government Mohan Kumaramangalam Medical College Hospital, Salem 636002, Tamilnadu, India.

In recent decades, with an increasing number of women pursuing medical career, there is a striking contrast of women taking-up surgical specialties. Though, surgery is traditionally a macho field, the status of women surgeons has significantly improved in recent years. But, the basic challenges remain yet to be addressed.[1] Is surgery really appealing to women or is it appalling? Several issues drive them away from choosing a surgical carrier. These multifaceted factors include years of laborious, physically demanding, emotionally exhausting training which is compounded by male dominated work-environment, insufficient family time, poor quality of personal life, negligible prospects in career advancements and less financial perks. While most of the women doctors chose relatively cooler, family-friendly, less demanding or even 'feminine specialties', only a few (including me) take-up surgery with great passion.

Transition from Motherhood to Hardihood

Balancing between a surgical career and a family life is often a tight rope-walk for woman surgeons. In the current socio-cultural milieu of the SAARC countries, child-bearing and rearing undermine their surgical and academic performance.[1] Many of the woman surgical-residents postpone their marriage and pregnancy in favour of professional career, and as a consequence of this, few of them embarking on marital life in their fourth decade need assisted reproductive techniques. They often have the dilemma of "whether family first or practice first?" and face the dual guilt - the guilt of neglecting the career and the guilt of being an inadequate mother. While male surgeons are perceived as 'dedicated', ambitious female surgeons are frequently labelled as 'bossy and aggressive' at workplace and 'negligent mom' at home.[2] To be a successful 'she surgeons', it is essential to exhibit remarkable resilience in the arduous work-life journey.

Role of Robust Support System

There is a need to build a robust support system to empower female surgeons. Involvement of spouse in child rearing is crucial. Only a few are fortunate to have a strong parental and spousal support in child rearing. Many have to opt for the help of a paid maid. I have seen children of many female surgeons are being raised up not at home, but in the hospital corridors! A difficult child rearing can significantly alter the career trajectory of woman surgeons.

De-skilling, Re-skilling and Up-skilling

Due to the career breaks of pregnancy, women are perceived to be at the risk of 'de-skilling'.[3] Fear of incompetence, failures and repercussions together

with the lack of a good support system are the common causes of attrition. Women are often disgruntled in an isolated and male-dominated work-environment. A female surgeon has to pull her weight like a man, to receive the same privileges and respect as her male colleagues and be "re-skilled" to achieve professional competence. "Up-skilling" can be attained by attending trainings, workshops or fellowship programmes.

Gender Disparity and Equality

Gender plays a major role in career challenges faced by female surgeons.[1] Though gender gap is closing fast, there is a gender bias, with major a favouritism towards the males thereby limiting the opportunities of woman surgeons. Many of them face scepticism about their competence and end-up with professional frustration and dissatisfaction. It is important to work towards gender equality to attract brilliant women to surgery who will break the vicious cycle of lack of mentorship and leadership.

Pressing Safety Concerns

Workplace harassment, be it sexual or otherwise, prevails throughout a woman's career impacting her mental and emotional well-being. It is only the people around her most of the time, who create a hostile work-environment.[2] Many of the woman surgeons face micro-aggression from colleagues which affects self-esteem thereby leading to burn-outs, depression and a tremendous gap in the confidence level. Proper accommodation at the workplace and safety during travel and night calls are needed to protect the dignity of women in surgery.

Mentorship and Sponsorship

Proper guidance and hand-holding is crucial for woman surgeons to navigate through her career. Women prefer gender-concordant mentoring relationships, one for their admirable distinction in the surgical field and another for their advocacy on the integration of work-life balancing. 'Sponsorship' goes way beyond mentoring, where a senior colleague actively supports her career growth by providing recommendations and by promoting her in the highly competitive surgical field. This is hardly ever seen among the female surgeons and the struggle is real.[2] Incorporating mentorship and sponsorship programs will definitely help the brightest category of women who are stuck-up in mediocrity.

Pipelines and Pathways to Leadership

The increasing number of women in surgery are not been matched by the increasing number of women in leadership positions. This may be due to the 'pipeline effect'.[1] The numbers drop off at startling rates as one look at the ascending ranks. Male surgeons are able to take unilateral decisions, adapt to changes, explore new opportunities and move to the next level of career progression. But, women surgeons prefer 'safety cum security' and work-life stability. Apprehensive of working with a new team or workplace, they often resist changes, resulting in career stagnation. It is important to break the proverbial glass ceiling and enter the 'growth zone' to attain leadership positions.[3] Without restructuring the surgical career advancement scheme, the profession will leave women behind.[2]

Your network is your net worth

Building a strong social network is of utmost importance to break barriers, build confidence and to provide support system among the aspiring woman surgeons. The *'Wonder Women Pediatric Surgery'* group in the Whatsapp, the *'Association of Women Surgeons' (AWS)* group in the Instagram and the *'Association of Surgeons of India - Women in Surgery (ASI-Win-S)'* are few such social networking platforms that are of great help in connecting the woman surgeons around the globe. They play a crucial role in voicing out their needs and views.

Lastly, I remember one of my mentors saying, "Let your degree not be a jewel in your crown; let it be a passion in your heart".

REFERENCES

[1] Seemann NM, Webster F, Holden HA, Moulton CA, Baxter N, Desjardins C, Cil T. Women in academic surgery: why is the playing field still not level? Am J Surg. 2016 Feb; 211(2): 343-9.

[2] Stephens EH, Heisler CA, Temkin SM, Miller P. The current status of women in surgery: How to affect the future. JAMA Surg. 2020 Sep 1; 155(9): 876-885.

[3] Lai CS, Mundy JA. Equity, Inclusion and Diversity in Surgical Training. Indian J Surg 2022;84(Suppl 1): 45-51.

[4] Singh C, Loseth C, Shoqirat N. Women in surgery: a systematic review of 25 years. BMJ Leader 2021;5: 283-290.

Address for communication: *Dr. J Sulthana Dhilras, Email: dhilras.12285@gmail.com*

Received 20 Sep 2024; Accepted 27 Sep 2024

Acknowledgements : Views expressed are personal opinions of the author
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : None (Personal opinion)

Citation*: Dhilras JAS. Challenges and safety of women surgeons during residency and practice. Pediatr Surg Trop 2024; 1(4): 203-205.*

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 206-210
DOI: 10.5281/zenodo.13893282

View Point

Assault on Doctors in India – A Rising Unhealthy Trend

Patta Radhakrishna

Department of Surgical Gastroenterology and Minimal Access Surgery, Malar-MGM (Mahatma Gandhi Memorial) Hospital, Chennai 600020, Tamil Nadu, India

INTRODUCTION

A young lady postgraduate of the Department of Pulmonology at the Radha Gobinda Kar Medical College Hospital (Kolkata, India) had to sacrifice her life in the most gory and brutal way[1] to wake an entire nation and its rulers up to the fact that doctors working in India are extremely vulnerable to physical assault and are totally unprotected by the law as well as the institution where they work.

In India, assaults on doctors have become a growing concern in the recent years.[2-6] For obvious reasons, big hospitals as well as small clinics tending to sick children are more vulnerable to such assaults. Physical attacks not only hamper the professional and emotional well-being of healthcare providers but also undermine the integrity of the medical system. Doctors, who are considered as healers and expected to save lives, often face physical and verbal abuse from patients and their families. Various factors contribute to this disturbing trend, and understanding them is essential to develop practical solutions.

COMMON CAUSES OF ASSAULTS ON DOCTORS

Patient Dissatisfaction and Unrealistic Expectations

One of the most prevalent causes of assaults on doctors is patient dissatisfaction, often arising from unrealistic expectations regarding treatment outcomes more so in the emergency room and in the intensive care units. Patients and their families sometimes expect instant or guaranteed recovery, which is not always possible, particularly in cases of severe trauma, terminal illnesses, or critical medical conditions. When these expectations are unmet, patients and their relatives get frustrated and direct their anger toward the concerned healthcare providers. For instance, in June 2019, a junior doctor at the Nil Ratan Sircar (NRS) Medical College (Kolkata, India) was severely assaulted after a 75-year-old man succumbed to an illness.[7] The patient's relatives, upset by the death, blamed the doctors and initiated a violent attack on them. This incident sparked nationwide protests by medical professionals.

Overcrowded and Underfunded Public Healthcare System

India's public healthcare system is overburdened due to her large population and lack of adequate resources. Public hospitals are often understaffed underfunded and underequipped, leading to long waiting times and poor patient outcomes. The frustration caused by overcrowding in hospitals, delay in receiving care and insufficient communication between medical staff and patients often boils over into aggression toward doctors. For example, in July 2021, a doctor at a government hospital in Rajasthan, India was assaulted by the relatives of a deceased COVID-19 patient.[8] The patient's family, aggrieved by the non-availability of ventilators and other critical medical supplies, held the doctor responsible, even though the pan-

demic had strained healthcare resources beyond capacity.

Overburdened Public Healthcare System

India's public healthcare system is overburdened and understaffed. This often causes delays in treat ment which frustrates patients and their families. Overworked doctors are left to handle the brunt of these frustrations, often becoming targets of violence. The incident at the Safdarjung Hospital (New Delhi, India) in 2021 is a stark reminder. A junior doctor was brutally assaulted by the family of a COVID-19 patient who had died, leading to an uproar in the medical community.[9]

Poor Communication and Lack of Transparency

Communication gaps between healthcare providers and patients or their families are yet another common cause of violence. When doctors fail to convey the gravity of clinical condition, the limitations of medical treatment or the risks involved in certain procedures, patients' families may feel misled or uninformed. The resultant misunderstanding often leads to hostility toward doctors.

An incident in Hyderabad (Andrapradesh, India) in 2017 highlights this issue. After the death of a woman during childbirth due to complications, the family accused the hospital staff of negligence and attacked the attending doctor. It was later revealed that the doctor had failed to adequately inform the patient's family about the high-risk nature of the pregnancy, leading to their anger and violent reaction.[10]

Cultural and Emotional Factors

Cultural and emotional factors also play a significant role in assaults on doctors. In many Indian communities, doctors are often seen as authoritative figures with almost supernatural abilities to save lives. When a loved one dies or their health worsens despite medical treatment, families may feel an intense sense of betrayal and grief. This emotional response, coupled with the cultural attitude that views death as a failure of healthcare system rather than a natural part of life, can incite mob violence. In June 2020, a doctor in Assam was brutally beaten by a mob after a patient who had been undergoing treatment for chronic liver disease passed away. This incident reflects the deep impact of emotional and cultural perceptions on violent confrontations.[11]

Political and Media Sensationalism

In some cases, political and media influences exacerbate the problem. Sensational reporting by the media, particularly when it involves allegations of medical negligence, can fuel public outrage. Politicians sometimes take advantage of such situations and offer support to the aggrieved families, which can embolden them to seek retribution against doctors.

Money Matters

In most of the corporate hospitals, when patients die of incurable disease, their relatives indulge in violence just to escape paying the hospital bill for a treatment which was futile. Hospital administrators are inclined to disperse the crowd and cool tempers within the hospital premises. They are often willing to write off the bills of a deceased patient to buy peace. Relatives of patients tend to take advantage of this weakness of big corporate hospitals.

FREQUENCY OF ASSAULTS ON DOCTORS

Assaults on doctors are alarmingly frequent in India. According to a report by the Indian Medical Association (IMA), over 75% of doctors in India have faced some form of violence at work.[12] The frequency of these incidents has been steadily increasing, particularly in the emergency rooms of public hospitals, where the pressure on medical staff is often the highest. Although exact statistics are difficult to obtain due to underreporting and variations across the states, the frequency of violent attacks is a clear indicator of a widespread problem within the healthcare system.

TYPES OF ASSAULTS ON DOCTORS

Physical Assault

Physical violence is the most common type of assault faced by doctors in India. It can range from slapping, punching and kicking to more severe forms of violence like mob attacks using deadly weapons.[12] These physical assaults can cause serious injuries and permanent disabilities to doctors and hospital staff, leaving them mentally traumatized as well. In 2017 a renowned gastroenterologist in Mumbai was fatally assaulted by a mob when he refused to allow additional relatives to accompany a patient inside the emergency room. In a similar incident, a tea-estate doctor of Assam was killed in 2019.[13]

Verbal Abuse

Verbal abuse, though less physically harmful, can still have a profound psychological impact on doctors. Patients or their families often use derogatory language, threats and insults to express their anger or frustration. This constant exposure to verbal abuse can lead to stress, anxiety and burnout among the healthcare professionals.

Damage to Property

In addition to personnel assaults, angry relatives of patients often damage hospital equipment, shatter windows and destroy medical facilities in an outburst of violence. This not only endangers the lives of other patients but also hampers the overall functioning of the hospital. During the COVID-19 pandemic, hospitals in several states, including Maharashtra and Delhi, witnessed damage by mobs angry over the unavailability of oxygen cylinders, ventilators or hospital beds.

Cyber Bullying and Online Threats

With the rise of social media, many doctors now face online harassment and threats. Dissatisfied patients are using social media to publicly shame and threaten doctors, damaging their reputations and professional credibility. This type of assault can have long-lasting effects on a doctor's career and mental health.

POSSIBLE REMEDIES

Strengthening Security at Hospitals

One of the most immediate steps that can be taken is to improve security at hospitals, particularly in high-risk areas such as the emergency rooms and intensive care units. The presence of trained security personnel with weapons can act as a deterrent to potential assailants and helps in controlling the adverse situation before it escalates into violence. Many hospitals have started implementing security protocols like panic buttons, closed-circuit television (CCTV) cameras and rapid-response teams. For example, after the NRS Medical College incident, the Government of West Bengal deployed more police officers at its hospitals, reducing the frequency of such violent episodes. CCTV cameras installed in critical areas will enable recording the incidents of violence and hold the perpetrators accountable.

Public Awareness Campaigns

Public awareness campaigns aimed at educating patients and their families about the realities of medical treatment and the limitations of doctors could help in mitigating violence. Such campaigns should emphasize the importance of patience, cooperation and communication in achieving the best possible healthcare outcomes. Medical associations and governments can work together to change the unrealistic public perception of doctors as infallible miracle workers. In 2017, the state of Maharashtra initiated a campaign highlighting the dangers of violence against healthcare workers, urging people to respect doctors and follow protocols in hospitals. Such initiatives should be replicated nationwide.[14]

Legal Protection of Doctors

Despite the growing number of assaults, legal protections for doctors in India remain inadequate.

The government should consider passing stricter laws that impose harsher penalties on individuals who assault doctors. State level ordinances have been passed by Tamilnadu in 2008, Karnataka in 2009, Maharashtra in 2010, Bihar in 2010, Assam in 2011 and Delhi in 2021. Under these ordinances a non-bailable arrest warrant against culprits can be issued. Imprisonment up to 3 years and a fine of Rs.50,000 can be slapped and the money can be recovered from culprits who damage hospital properties. All other states are needed to follow suit.

Currently, the Epidemic Diseases (Amendment) Ordinance-2020, provides some protection to healthcare workers during pandemics.[15] But broader legislations are needed to protect doctors in all other situations. The Healthcare Services Personnel and Clinical Establishments (Prohibition of violence and damage to property) Bill, 2019[16] has been proposed by the central government of India recommending stricter punishment. Meanwhile, the Government of West Bengal has hastily passed a bill to this effect recommending capital punishment for certain heinous crimes committed against doctors, but has not yet been ratified by the concerned authorities.

Additionally, fast-track courts should be established to ensure quick resolution of cases involving violence against medical professionals. A lengthy legal process can dissuade doctors from pursuing legal action, thereby allowing perpetrators to go unpunished.

Training in Communication and Conflict Resolution

Improved communication skills and conflict resolution strategies can help doctors in managing difficult conversations with patients and their families. Medical curricula should include training on how to effectively convey bad news, manage patient expectations and de-escalate potentially volatile situations. In 2019, the All-India Institute of Medical Sciences (AIIMS) at New Delhi, India introduced workshops for doctors on handling agitated patients and families, aiming to reduce the risk of violent incidents.

Psychological Support for Doctors

Doctors who have been assaulted often suffer from psychological trauma.[17] Hospitals should establish support systems for healthcare workers, including counselling services and peer support groups, to help doctors cope with the emotional toll of violence. This will not only help them recover, but also prevent burnout and retain talent in the medical profession.

CONCLUSION

The increasing incidence of assaults on doctors in India is a multifactorial issue. It frequently stems from patient dissatisfaction, shortcomings of the system, poor communication and cultural factors. Physical and verbal violence against doctors is disturbingly common. A combination of legal remedy, better security measures, public education and improved doctor-patient communication can help in mitigating this problem. It is crucial to safeguard the well-being of doctors and nurses, who are at the forefront of providing essential healthcare services, and ensure that they can perform their duties without fear of violence. There is a need to hit the iron when it is hot and in the present milieu when there are continuous protests across the country against assaults on doctors, we need to bring in the toughest laws against this crime which to some extent will scare the potential culprits. It is next to impossible to improve the national psyche which breeds on impatience, immoral and indecent behaviour in public leading to frequent road rages, rapes, child molestations and crimes of all other types which were literally unknown in our society in the past. Beating up doctors is one of them.

REFERENCES

[1] Mahase E. Doctors in India launch nationwide strike after trainee is raped and murdered. BMJ. 2024 Aug 19; 386: q1827.

[2] Kumar R. Death of a doctor - Ek Doctor Ki Maut-Time to boost the ailing and failing public health system in India. J Family Med Prim Care. 2019 Dec 10; 8(12): 3771-3772.
[3] Kunnath R, Thayyil J, Suresh N, Soman S. Workplace Violence Faced by Medical Doctors in Kerala, India. Cureus. 2023 Nov 16; 15(11): e48887.
[4] Sarkar T. Harnessing Bourdieu's social theory to understand the deteriorating doctor-patient-nurse relationship in West Bengal government hospitals. Front Sociol. 2022 Oct 6; 7: 938734.
[5] Singh A, Ranjan P, Sarkar S, Kaur TP, Mathew R, Gora D, Mohan A, Jangra J. What do clinical resident doctors think about workplace violence? A qualitative study comprising focus group discussions and thematic analysis from a tertiary care center of India. J Family Med Prim Care. 2022 Jun; 11(6): 2678-2684.
[6] Samant M, Calnan M, Kane S. A critical analysis of news paper accounts of violence against doctors in India. Soc Sci Med. 2024 Jan; 340: 116497.
[7] Bhattacharjee K. Kolkata: Mob violently attacks doctors at NRS Hospital after Mohammed Sayeed's death, police mute spectators, allege students. OpIndia 11 June, 2019. {Available at: https://www.opindia.com/2019/06/kolkata-mob-violently-attacks-doctors-at-nrs-hospital-after-mohammed-sayeeds-death-police-mute-spectators-allege-students/} (Accessed on 20 Sep 2024)
[8] Arya D. India's Covid doctors demand action after attacks. BBC 6 July 2021 {Available at https://www.bbc.com/news/world-asia-india-57648320} (Accessed on 20 Sep 2024)
[9] Bandyopadhyay O. Protecting the doctors. British Safety Council. 01 July 2021 {Available at https://www.britsafe.in/safety-management-news/2021/protecting-the-doctors} (Accessed on 20 Sep 2024)
[10] Times News Network. 3 held for attack on doctor. Time of India (Hyderabad News) 17 Dec 2007, 02:06 IST {https://timesofindia.indiatimes.com/city/hyderabad/3-held-for-attack-on-doctor/articleshow/2626704.cms} (Accessed on 20 Sep 2024)
[11] Mob attacks on-duty doctor in Hailakandi hospital; medical fraternity demands action. Assam Tribune 22 Sept 2024 12:34 PM {https://assamtribune.com/assam/mob-attacks-on-duty-doctor-in-hailakandi-hospital-medical-fraternity-demands-action-1552244} (Accessed on 20 Sep 2024)
[12] Kumar SN, Kartik M. Violence against doctors: Indian perspectives. In: Martin CR, Preedy VR, Patel VB. (eds) Handbook of anger, aggression, and violence. Springer, Cham. 2023. pp 2661–2672.
[13] Saha A. Death of a doctor. The Indian Express. September 15, 2019 00:55 IST. (Available from https://indianexpress.com/article/north-east-india/assam/tea-plantation-hospitals-mob-lynching-death-of-a-doctor-5996015/} (Accessed on 20 Sep 2024)
[14] Ganapatye M. Maharashtra doctor's strike: Medical Teachers Association gives 48-hrs ultimatum. India Today Mar 23, 2017 21:47 IST. {Available at https://www.indiatoday.in/india/story/maharashtra-doctors-strike-967311-2017-03-23} (Accessed on 20 Sep 2024)
[15] Government of India, Ministry of Health and Family Welfare. The Epidemic Diseases (Amendment) Ordinance April 22, 2020. { Available at https://prsindia.org/billtrack/the-epidemic-diseases-amendment-ordinance-2020} (Accessed on 20 Sep 2024)
[16] Government of India, Ministry of Health and Family Welfare. Draft Legislation titled "The Healthcare Service Personnel and Clinical Establishments (Prohibition of violence and damage to property) Bill, 2019" {Available at https://prsindia.org/billtrack/draft-the-healthcare-service-personnel-and-clinical-establishments-prohibition-of-violence-and-damage-to-property-bill-2019} (Accessed on 20 Sep 2024)
[17] Singh A, Ranjan P, Agrawal R, Kaur T, Upadhyay AD, Nayer J, Chakrawarty B, Sarkar S, Joshi M, Kaur T, Mohan A, Chakrawarty A, Kumar KR. Workplace violence in healthcare settings: A cross-sectional survey among the healthcare workers of North India. Indian J Occup Environ Med 2023 Oct–Dec; 27(4): 303-309.

Address for communication: *Dr. Patta Radhakrishna, Email: patta_radhakrishna@yahoo.co.in*

Received 14 Sep 2024; Accepted 22 Sep 2024

Acknowledgements : Views expressed in this article are personal opinions of the author
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : None (Personal opinion)

Citation*: Radhakrishna P. Assault on doctors in India - A rising unhealthy trend. Pediatr Surg Trop 2024; 1(4): 206-210.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 211-215
DOI: 10.5281/zenodo.13893312

Clinical Study

Role of Enteral Hyperalimentation in the Management of Chemotherapy-Induced Neutropenia in Wilms Tumor

Priti Kashyap, Yogesh Kumar Sarin

Department of Pediatric Surgery, Maulana Azad Medical College and Lok Nayak Hospital, New Delhi-110002, India

Keywords
Cancer chemotherapy
Enteral hyperalimentation
Granulocyte colony-stimulating factor
Nephroblastoma
Febrile Neutropenia
Nutrition in cancer
Wilms tumor

Abbreviations
ANC - Absolute neutrophil count
GCSF - Granulocyte colony-stimulating factor
RDA - Recorded daily dietary allowance
WT - Wilms tumor

Abstract

Introduction: *Chemotherapy of Wilms tumor (WT) often induces neutropenia. This study explores whether enteral hyperalimentation can reduce the incidence of chemotherapy-induced neutropenia and the need for granulocyte colony-stimulating factors (GCSF) in these patients.*

Methods: *Eight patients with WT were prospectively enrolled and were given enteral hyperalimentation. Anthropometric parameters, serum albumin levels, frequency and severity of neutropenic episodes, need for therapy postponement or dose reduction and the need for GCSF administration of these 8 patients were compared with that of 7 historic controls that were treated with the same chemotherapy protocol sans enteral hyperalimentation. Statistical analysis was done using Chi-square test.*

Results: *Enteral hyperalimentation was found to reduce the frequency of chemotherapy-induced neutropenia, especially the febrile neutropenia in WT. The requirement of GCSF was also less in the study group. However, these differences were not statistically significant. With enteral hyperalimentation there was no need for therapy postponement or dose reduction of chemotherapeutic drugs.*

Conclusion: *This single-centre, small sample-size study of WT failed to conclusively show any benefit of enteral hyperalimentation in reducing the frequency of neutropenia or the need for GCSF administration.*

INTRODUCTION

Chemotherapy of Wilms tumor (WT) often causes neutropenia by myelosuppression, which may be associated with fever, sepsis and rarely death. The usual management of chemotherapy-induced neutropenia includes postponement of therapy schedule or reduction in the dose of chemotherapy drug and administration of granulocyte colony-stimulating factor (GCSF). Postponement of schedule or reduction of drug dosage may compromise therapeutic intend and may lead to tumor relapse, whereas GCSF is known to cause significant long-term morbidity.[1] It is also known

that patients undergoing adjuvant chemotherapy suffer loss of appetite and hence eat inadequately and expose themselves to multiple adverse effects of malnutrition. We hypothesized that enteral hyperalimentation may reduce the incidence of neutropenia and hence the need for GCSF in these patients.

MATERIAL AND METHODS

This prospective interventional study was done in a tertiary-care hospital between December 2015 and September 2017. Due approval was obtained from the Institutional Ethics Committee. Patients aged 0-12 years presenting with low- and intermediate-risk WT (UK-CCSG protocol) of stages I to III were prospectively enrolled. Stage-III high-risk tumors, stage-IV disease and those with inferior vena cava thrombus extending beyond hepatic veins were excluded.

Prospectively enrolled 8 patients formed the study group (Group B) and they received enteral hyperalimentation. They were compared with 7 historic controls (Group A) who were treated by a similar chemotherapy protocol sans enteral hyper alimentation in the preceding year of this study.

Demographic data, clinical features and diagnostic work-up of these patients were recorded in a predesigned proforma. Nutritional status of each patient was assessed fortnightly from the outset of treatment. The parameters include weight, height, mid-arm circumference, triceps skin-fold thickness and serum albumin.

The weight of WT accounted for as much as 10-20% of the total body weight. Hence, to know the effect of hyperalimentation, we considered only the post-operative weight. Preoperative chemotherapy period was not considered for analysis.

All patients in the Group B were administered enteral hyperalimentation postoperatively. The recommended daily dietary allowance (RDA) of each patient was calculated according to the guidelines of the Indian Council of Medical Research (ICMR)[2] and hyperalimentation diet was defined as 1.2 times that of the RDA. Patients were encouraged to take the calculated diet orally as per their choice. When the total oral intake was less than 60% of the calculated amount, forced nasogastric tube feeding was employed. Hyperalimentation was temporarily withheld during the times of therapy-related vomiting or diarrhea. Children were sent home between the scheduled cycles of chemotherapy and their parents were encouraged to continue hyperalimentation at home as per the dietician chart. They were also taught the method of recording oral intake at home, so that calorie and protein content could be calculated by the dietician on their return for the next cycle of chemotherapy.

Nutritional assessment and blood counts were done fortnightly. Neutropenia episodes, if any were graded according to the Common Toxicity Criteria of the National Cancer Institute.[3] GCSF was administered when neutropenia was grade-2 or more. GCSF was given at a dose of 5µg/kg and the dose was increased if neutropenia persisted. GCSF was stopped when absolute neutrophil count (ANC) exceeded 1500 x 10^9/l. During episodes of febrile neutropenia, antibiotics (third-generation cephalosporins and aminoglycosides) were also administered.

Table 1. Grading of Neutropenia*

Grade	Absolute Neutrophil Count
Grade 0	≥ 2,000/mm^3
Grade 1	1,500 - 1,999/mm^3
Grade 2	1,000 - 1,499/mm^3
Grade 3	500 - 999/mm^3
Grade 4	<500/mm^3

* *As defined by the Common Toxicity Criteria of the US National Cancer Institute.*[3]

Primary outcomes were defined as the frequency of febrile neutropenia and the total dose of GCSF

required per kg body-weight during neutropenia treatment. Secondary outcomes were defined as the total delay in the completion of chemotherapy and the total reduction in the dosage of individual chemotherapy agents.

Statistical analysis of discrete data was done using Chi-square test with Yates correction. Statistical significance was set at P-value less than 0.05.

RESULTS

The subjects of the two groups were matched for age and stage of WT. In the Group B, 75% of the children needed nasogastric feeding on and off throughout their therapy, as they were unable to tolerate oral take because of vomiting and feed refusal.

Weight of patients with stage-2 and stage-3 WT increased after hyperalimentation; but it was not statistically significant. However, paradoxically, one patient of stage-1 in Group B had weight loss despite hyperalimentation. There was marginal improvement in mean triceps skin-fold thickness and mid-arm circumference in group B, while this data was not available for group A.

Neutropenia (with and without fever) occurred 19 and 16 times in the groups A and B respectively. There were no episodes of grade-4 neutropenia in the group B. (Fig. 1) Although the febrile neutropenia was more frequent in the group A (Fig. 2), it was not statistically significant (P=0.613).

In the group A, 4 of the 7 patients, needed GCSF during chemotherapy, whereas in the group B, 5 of the 8 patients needed GCSF. (χ2=0.1004; P=0.75) The total dose of GCSF required in the Group A was 500µg/kg, whereas it was 360µg/kg in the Group B; the difference was not statistically significant (P=0.11).

Chemotherapy had to be temporarily deferred for 4 and 2 weeks respectively in two patients of the Group A because of febrile neutropenia. In group B, none had to skip the therapy schedule. Two-third dose reduction of individual component of chemotherapy regimen was needed for 5weeks in one patient of the Group A. Dose reduction of 3mg/m^2 was needed for vincristine, 90µg/kg for actinomycin-D and 100mg/m^2 for adriamycin. Chemotherapy dose reduction was not needed in any of the patients of the group B.

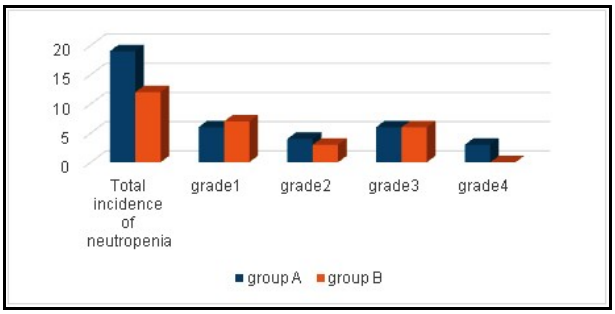


Fig 1. *Frequency of neutropenia.*

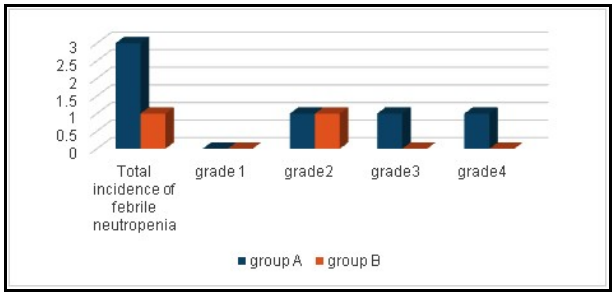


Fig 2. *Incidence of febrile neutropenia*

DISCUSSION

Impact of nutritional status on the outcome of cancer chemotherapy is a subject of much speculation, with limited evidence in the literature. Better nutritional status is presumed to be beneficial in two ways: firstly, by improving general immunity and secondly, by avoiding disruption of chemotherapy schedule from febrile neutropenia and low ANC. The present study was done to explore the effects of nutrition on chemotherapy-induced neutropenia.

Infectious complications are known to occur more frequently in malnourished children with cancer.

Several studies from low- and middle-income countries (LMIC) have shown a high prevalence of malnutrition at diagnosis and its adverse effect on the outcome of WT.[4,5]

In these children nutritional status can be improved by giving supplemental feeds through enteral or parenteral routes. Parenteral nutrition is associated with the risks of catheter infections, fluid-electrolyte imbalances and hepatopancreatic dysfunction.[6] Advantages of enteral feeding over parenteral route include prevention of bacterial translocation, preservation of normal flora, transit and histology of the gut, prevention of hyper-catabolic responses to stressful events and maintenance of gut immune function. Hence enteral nutrition is always preferable in terms of physiological response, local and systemic competence, quality of life, patient compliance and cost.[6,7] Nasogastric tube feeding can be a good option if oral acceptance is poor. Energy-enriched formula given through nasogastric tube feeding has been known to be effective in improving the nutritional status of children with cancer during the intensive phase of treatment.[8] Hyperalimentation is known to improve immune status, rate of wound healing and response to anti-neoplastic therapy.[9]

Israels et.al.[10] have noted grade-3 neutropenia in 59% of the WT and grade-4 neutropenia in 27% had grade-4 neutropenia. Grade-4 neutropenia occurred more commonly with 3-drug regimen than with 2-drug regimen (50% versus 15%). Nearly 60% of all patients and 85% of those receiving the 3-drug regimen had documented neutropenia of grade-3 or more. We also documented that 12 out of the 15 patients (80%) had a total of 35 episodes of neutropenia, of which 12 (34%) were of grade-3 and 8.6% were of grade-4. We observed less frequent neutropenic episodes in the patients receiving enteral hyperalimenta tion. Although grade-3 neutropenic episodes were equal in both the study group and historical controls, there were no episodes of serious grade-4 neutropenia in the group B. Disrupted therapy schedule and reduction in chemotherapy dosage was not needed in our patients who were given hyperalimentation. However, these differences did not reach the level of statistical significance, probably because of the small sample-size. A larger sample-size from multi centric study may provide the definitive answer in future.

CONCLUSION

This single-centre, small sample-size study failed to conclusively show the benefits of enteral hyper-alimentation in reducing the frequency of neutropenia or the need for GCSF administration in children receiving chemotherapy for WT.

REFERENCES

[1] Freedman MH, Bonilla MA, Fier C, Bolyard AA, Scarlata D, Boxer LA, Brown S, Cham B, Kannourakis G, Kinsey SE, Mori PG, Cottle T, Welte K, Dale DC. Myelodysplasia syndrome and acute myeloid leukemia in patients with congenital neutropenia receiving G-CSF therapy. Blood. 2000 Jul 15; 96(2): 429-36.

[2] Expert Group of ICMR. Nutrient requirements and recommended dietary allowances for Indians: A report of the expert group of the Indian Council of Medical Research. Hyderabad, National Institute of Nutrition, 2010.

[3] National Cancer Institute. Common toxicity criteria, version 2.0. {Available from URL: https://ctep.cancer.gov/protocoldevelopment/electronic_applications/docs/ctcv2nom-4-30-99-final3.pdf} [Accessed on June 5, 2024].

[4] Chukwu BF, Ezenwosu OU, Ukoha OM, Ikefuna AN, Emodi IJ. Nutritional Status of Children with cancer at the University of Nigeria Teaching Hospital, Ituku/Ozalla, Enugu, Nigeria. J Cancer Prev Curr Res 2016; 5(4): 00167. {DOI: 10.15406/jcpcr.2016.05.00167}

[5] Rahiman EA, Trehan A, Jain R, Menon P, Kakkar N, Srinivasan R, Sodhi KS, Saxena AK, Kapoor R, Bansal D. A higher tumor volume and under nutrition at diagnosis adversely affect the survival of children with Wilms tumor: A study of 200 patients. Pediatr Blood Cancer. 2022 Nov; 69(11): e29880.

[6] Seres DS, Valcarcel M, Guillaume A. Advantages of enteral nutrition over parenteral nutrition. Therap Adv Gastro enterol. 2013 Mar; 6(2): 157-67.

[7] Sala A, Wade L, Barr RD. Nutritional support for children with cancer. Indian J Pediatr. 2003 Oct; 70(10): 813-6.

[8] den Broeder E, Lippens RJ, vant Hof MA, Tolboom JJ, Sengers RC, van den Berg AM, van Houdt NB, Hofman Z, van Staveren WA. Nasogastric tube feeding in children with cancer: the effect of two different formulas on weight, body composition, and serum protein concentrations. JPEN- J Parenter Enteral Nutr. 2000 Nov-Dec; 24(6): 351-60.
[9] Karlberg HI, Fischer JE. Hyperalimentation in cancer. West J Med. 1982 May; 136(5): 390-7.
[10] Israels T, Chagaluka G, Pidini D, Caron H, de Kraker J, Kamiza S, Borgstein E, Molyneux L. The efficacy and toxicity of SIOP preoperative chemotherapy in Malawian children with a Wilms tumour. Pediatr Blood Cancer. 2012 Oct; 59(4): 636-41.

Address for communication: *Dr. Yogesh Kumar Sarin, Email: yksarin@gmail.com*

Received: 15 June 2024; Accepted: 9 August 2024

Acknowledgements : The authors acknowledge the constructive inputs of Drs AP Dubey and SK Sinha of the Departments of Pediatrics and Pediatric Surgery respectively. The authors also appreciate the services of the Dietary Department of Lok Nayak Hospital, New Delhi

Conflicts of Interest : None declared by the author

Source of Funding : None

Ethical concerns : Study approved by Institutional Ethics Committee of the Maulana Azad Medical College

***Citation**: Kashyap P, Sarin YK. Role of enteral hyperalimentation in the management of chemotherapy-induced neutropenia in Wilms tumor. Pediatr Surg Trop 2024; 1(4):211-215.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 216-248
DOI: 10.5281/zenodo.13893347

Tropical Surgery Review

Maggot Infestation (Myiasis) in Children

Venkatachalam Raveenthiran

Department of Pediatric Surgery, Government Cuddalore Medical College, Chidambaram 608002, Tamilnadu, India.

Keywords
Fly larva
Ivermectin
Maggot therapy
Myiasis (Myasis)
Turpentine
Parasitic infestation
Wound healing

Abbreviations
AMP - Adenosine mono-phosphate
ASEM - Alimentary secretions and excretions of maggots
ENT - Ear-nose-throat
MMP - Matrix metallo-proteinase
MM - Medicinal maggots
MT - Maggot therapy
WM - Wound myiasis

Abstract

Myiasis (myasis) is a parasitic infestation of fly (diptera) larvae. Maggots are both a pest and a therapeutic agent. Ayurvedic surgeons of ancient India (2000-600 BCE) have described Krimi-Karna (aural myiasis) and Krimija-Siroroga (nasal myiasis). They also used maggot therapy (MT) to heal wounds (Krimi Upattikara Chikitsa).

About 95% of pediatric myiasis is due to 80 species of flies belonging to 4 major categories: blowflies, flesh flies, botflies and house flies. Hence, myiasis is not one disease; but a group of diseases caused by various dipteran larvae. Cochliomyia, Chrysomya and Wohlfahrtia are the 3 important deadly maggots.

Myiasis is common in those who live in unhygienic environments or those who are unable to care for themselves. No age is immune and the youngest patient was a 1-day-old newborn. Virtually all body parts are affected with more frequent involvement of the intact skin, superficial wounds, body orifices and umbilical cord.

The diagnosis of surface myiasis is straightforward. Doppler ultrasound showing a hyperechoic wavy spindle-shaped shadow writhing in a hypoechoic tunnel (Bouer's criteria) is pathognomonic of maggots hidden in deeper tissues. Exact species identification is desirable, though it is not essential for the clinical management of myiasis. Retrieved maggots should be fixed in 70% alcohol rather than in formalin for entomological examination.

All infestation with wild maggots should be considered as a disease and treated by manual removal. Left over dead or ruptured maggots may evoke more troublesome inflammatory reaction than live maggots. Vermifuges (turpentine) Larvicides (ivermectin), asphyxiants (mineral oil), baits (bacon) and paralyzers (lignocaine, ether) are commonly used in the treatment of myiasis.

Laboratory cultured medicinal maggots are used to promote wound healing by debridement, disinfection and cellular proliferation. Current pediatric experience with MT is limited to anecdotal case reports. Some of the recent randomized controlled trials question the claimed benefits of MT.

INTRODUCTION

Parasitic infestation in which Dipterous larvae (maggots) spend a part of their life cycle in vertebrate (human or animal) host is called myiasis.[1] Diptera are two-winged insects, commonly known as flies (Greek: *Di* - two; *Ptera* - wings). This definition differentiates myiasis from the larval infestation of other insects (e.g. scabies, lice) and nematodes (e.g. ascariasis). The economic impact of maggot infestation in livestock is tremendous as they cost billions of dollars annually by affecting the yield and quality of milk, meat and leather.[2] But the actual impact of human infestation has not been well studied. Myiasis remains a neglected zoonosis.[3]

Myiasis is frequently misconstrued as a disease of unhygienic, invalid people of underdeveloped tropical countries. Perhaps, this could be the reason for poor funding of maggot research in the Western world. Contrary to the general notion, myiasis is the fourth commonest travel-associated disease noted in 7-10% of Western travelers.[3-5] Diaz rightly remarked, "Ectoparasitic diseases, including myiasis, are no longer infestations of children and socio-economically disadvantaged populations in tropical countries; they have re-emerged as unusual, but not uncommon, infectious diseases worldwide."[6]

In recent years, demonized imagery of maggots has undergone transfiguration. From being a 'foe of health' they have metamorphosed into a 'friend of therapy'.[7] Laboratory cultured maggots are now used in the treatment of chronic non-healing wounds.[8,9] Forensic importance of maggots and their ecological role as decomposers are also well appreciated.[10]

HISTORY

The status of maggots in human history has swung between 'enthusiastic receptions' and 'disdainful rejections'.[11] Prehistoric cave dwellers must have been annoyed by maggots despoiling their precious trophies of hunting, the uncooked meat.[11] At the same time, aboriginals of the Australian, Myanmar, Ngemban and Mayan tribes appear to have known the therapeutic potentials of maggots in wound healing.[12] Astonishingly, Mayan healers unwittingly knew a technique of culturing disinfected medicinal maggots. They dressed wounds with a piece of cloth soaked in animal blood and dried in sun light. Blood soaks must have attracted flies to oviposit and solar ultraviolet rays must have disinfected the eggs. With this dressing, they expected swarming maggots after a few days. It is worth remembering that until Francesco Redi (1626-97 CE) disproved the Aristotelian theory of abiogenesis[13] by his famous meat-jar experiments, no one knew the link between maggots and flies.

The oldest textual reference to human myiasis is found the Ayurvedic medical texts of ancient India. Both Charaka and Sushruta (circa 1200-600 BCE) have mentioned 20 different types of *Krimis* (worms) including maggots. Ayurvedic physicians classified pathogenic *krimis* into *rakthaja* (blood), *kaphaja* (mucus) and *malaja* (fecal) origin. *Romaja* (hair) *krimi* described by Sushruta grossly resembles *Dermatobia* of the modern medicine. Sushruta's description of *Krimi-granthi, Krimi-karna* and *Krimija Siro-roga* are unmistakably that of orbital, aural and nasal myiasis respectively.[14]

> *"The disease of the head in which a pricking and tingling pain is felt inside the head as if being stung by some poisonous insect, and which is accompanied by a watery discharge mixed with blood from the nose, should be attributed to the existence of local parasites. This disease is a dangerous one and is known as the Krimija Siro-roga. The patient should be made to snuff in a quantity of animal blood. The worms or parasites, lured with the smell of the blood, would greedily come down into the passages of the nostrils when they should be carefully extracted by means of a tong" [14]*
>
> *(Sushruta Samhita - Bhishagratna Translation)*

Sushruta described two different techniques of maggot extraction: (1) Application of goat meat (*mamsa achadana*) to bait the worms out; (2) Applying vermifuges such as cow urine and herbal

decoctions in the form of fumigants, *nasyas* (snuff) or *avapidas* (liniments).[14,15] Sushruta was much ahead of his times when he advocated application of clarified butter or mustard oil to bring out maggots from deep burrows. This is principally similar to the asphyxiant therapy of modern days.

Ayurvedic physicians also used maggots to treat incurable cancers (*Kaphaja Arbuda*) and non-healing ulcers (*Dushtavrana*).[15] They applied *Dooshya Mamsa* (animal flesh) to wounds which would attract flies to lay eggs o it.[16] This technique, referred to as *Krimi Upattikara Chikitsa* has recently been rediscovered as 'bacon therapy'.[17] Probably, Hindu surgeons used obligate maggots to eat away live tumor tissues and facultative maggots to digest necrotic debris.[14] In colonial India, myiasis of the nose was known as *peenash* (Sanskrit: *Pee* - fetid, *Nash* - nose).[18,19] It is not clear if maggots were the cause of foul breath or *vice versa*.

Maggot infestation is well described in several mythologies. According to Homer's Iliad, the Greek hero Achilles requested his mother Thetis to protect his wounded friend Patroclus from being devoured by maggots.[20] *Garuda Purana* of Hinduism warns that in the nether life maggots will eat away the sinners (*Krimi-Bhojanam*).[19] The same theme is also found in the Old Testament, wherein Job lamented, "My body is covered with worms and scabs, my skin is broken and festering"(Job 7:5). The Exodus of the Old Testament also described an epidemic of myiasis (fly plague) in Egypt (Exodus 8:21).[11] King Herod, according to the Bible, was said to have died of myiasis-induced gangrene.[11] Flies and maggots are considered a symbol of *Nergal*, the Mesopotamian god of death.[11] Consequently, in ancient Babylonia, amulets were designed in the form of maggots. In Nordic mythology, *Leki* - the God of death - was believed to enter a house in the form of maggot creeping through a keyhole.[11] A wish-note buried with a Giza Mummy reads, "The maggots will not turn into flies within you".[11] Thus, Egyptians knew about the metamorphosis of maggots in to flies, a phenomenon that was rediscovered by Redi after several millennia. In hieroglyphics, double headed arrows represent maggots and flies.[11] Perhaps, ancient Egyptians symbolically meant that maggots are double edged swords!

Aulus Cornelius Celsus (circa 25 BCE – 50 CE) in his *De Medicina* described ear infestation due to *Wohlfahrtia magnifica* maggots and its treatment in great details.[21] Ambroise Pare (1510-90 CE) was the first to note the beneficial effects of maggots on wound healing.[12] However, he did not deliberately advise maggot therapy (MT). Similar observations were also made by Dominique Jean Larrey (1766–1842, the personal physician of Napoleon Bonaparte) and William Williams Keen (1837–1932, the army surgeon of the North States). John Forney Zacharias (1837–1901) is credited with the first intentional use of MT during the American Civil Wars of 1860s.[12] William Stevenson Baer (1872–1931), the founding chairman of orthopedics at the Johns Hopkins University,[22] successfully used MT in 1930s to treat more than 60 children with chronic osteomyelitis and bedsores.[23](Fig. 1) When two of his patients died of tetanus following MT, Baer realized the need of using disinfected maggots. His student Livingston popularized MT. However, the discovery of antibiotics in 1940s dampened the enthusiasm on MT. The pendulum swung back and the interest in MT was rekindled in 1970s when antibiotic resistance emerged as a great threat. Ronald Sherman and Edward Pechter rediscovered and popularized MT in 1983.[7,8,] In 1995, the International Biotherapy Society (IBS) was founded to sponsor annual conferences on MT.[24,25] In January 2004, the Food and Drug Administration of America (US-FDA) approved medicinal maggots under the category of medical devices rather than merely as a drug.[24] Since then, several randomized controlled trials on MT have been published.[26]

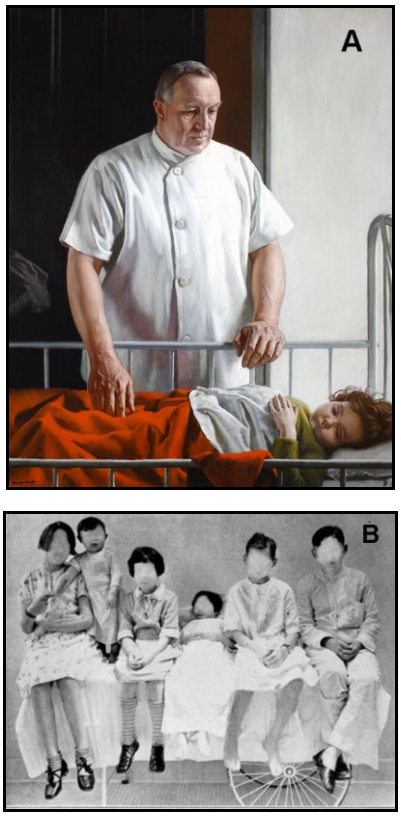


Fig 1. *(a) William Stevenson Baer (1872–1931), the pioneer of maggot therapy (Painting on display at the Orthopedic department of the Johns Hopkins University); (b) Photograph of Baer's patients (From Baer's original publication in the Journal of Bone and Joint Surgery 1931) (Public domain pictures)*

ETYMOLOGY AND NOMENCLATURE

In 1815, Kirby and Spence first used the term *Scholechiasis* to describe infestation by larva of any insect.[2] In 1837 Frederick William Hope, an English clergyman, Oxford professor and entomologist, coined the term *Myasis* (sic) to differentiate dipterous fly larvae from other the larvae of other insects (Greek: *Myia* - fly; *sis* - disease).[27-29] He suggested, the term *Scholechiasis* be restricted to infestation of *Lepidoptera* larvae and *Canthariasis* to *Coleoptera* larvae.[28] The term *Myiasis* is restricted to wild infestation while *'maggot therapy'* refers to iatrogenic infestation.[30] Myiasis is also known by a variety of vernacular names.[31,32] (Table 1)

Table 1. Synonyms of myiasis

Ancudo †
Bekuru †§ (Brazil) [32]
Berne † (Brazil) [50]
Bicherio § (Latin America)[Bapat]
Borro† (Bolivia) [32]
Colmoyote † (Mexico) [32]
Flystrike ‡
Gusano de mosquito / zancudo † (Venezuela) [32,50]
Gusano de monte † (Venezuela) [32,50]
Gusano de peludo † (Bolivia) [32]
Gusano macaco † (Venezuela) [32,50]
Ikitugu † (Brazil)
Kitudn † (Brazil)
Krimija ‡ (India)
Kturn † (Brazil)
Macaco †§ (Guyana) [32]
Maggot infestation ‡
Mberuaro † (Brazil)
Mirunta † (Peru) [32]
Miruta †[54]
Mosquito worm † (Trinidad)[4,32]
Moyocuil (Moyocutli) † (Mexico)[4,32]
Muskieten worm † (Suriname) [32]
Myiasis (Myasis) ‡#
Nuche † (Columbia)[Goldman]
Peenash *§ (India)[Center]
Suglacuru † (French Guyana)[4,32]
Suylacuru † (French Guyana)[4,32]
Torcel † (Central America)[Hunter]
Torsalo † - (Costa Rica)[Goldman]
Tupe † (Ecuador) [32]
Ura † (Argentina, Paraguay) [32]
Ver macaque † (French Guyana)[(32]
Verme de mata † (Venezula) [32]
Vermes Nasi * (India)[31]
Warble † (veterinary term)

Source: Center[18], Doss[31], Francesconi[4], Goldman[50], Hunter[32], Bapat[41], Quintanilla-Cedillo[54]
† Vernacular synonyms of furuncular myiasis.
** Synonym of nasal myiasis*
§ Spelling variantions in English: (Peenash, Pinash, Penash), (Bekuru, Bikuru), (Macaco, Macaw)
‡ General terms that are independent of the affected anatomical site.
 # Frederick William Hope originally spelt it as 'Myasis'(sic)

Table 2. Modified anatomical classification of human myiasis §

ECTOPARASITE

Non-inhabitants

Sanguinivorous (Blood sucking) *

Dermal inhabitants (Cutaneous myiasis)

Follicular
Wound (Traumatic)
Migratory (Creeping or Subdermal)
Umbilical †

ENDOPARASITE

Endoluminal (Orificial)

Nasal
Aural
Oro-pharyngeal
Orbital (Ophthalmomyiasis externa) ‡
Tracheal
Genitourinary
- Female - vaginal
- Male - Penile, preputial, scrtoal
- Urethrovesical

Gastrointestinal
- Anorectal
- Intestinal - Gastric, Enteric

Parenchymal (Visceral)

Cerebrospinal
Ocular (Ophthalmomyiasis interna) ‡
Pulmonary (Lower respiratory)

§ Combines the classifications of Bishopp [34], James [33] and Zumpt [1]

** These free living larvae in the environment approach hosts for just blood meals rather than infesting their body*

† Umbilical myiasis is exclusvely occur in neonates

‡ A distinction is made between orbital (eyeball socket) and ocular (eyeball chambers) myiasis.

The nomenclature of skin infestation (cutaneous or dermal myiasis) varies according to the location of maggots. It may be *furuncular myiasis* (dermis), *migratory myiasis* (subcutaneous plane) or *wound myiasis* (surface of an already existing wound).[29] The term *'traumatic or wound myiasis'* was first used by James in 1947.[33] The adjective 'traumatic' is inappropriate as it is also known to occur in ulcerated malignant tumors and in non-traumatic neuropathic ulcers.

CLASSIFICATION

Clinical myiasis is classified differently by various authors.[2,4] In 1915, Bishopp classified it based on the affected organ.[34] James in 1947[33] and Zumpt in 1965[1] modified Bishopp's anatomical classification. (Table 2) Patton[35] considered that the anatomical classification is inappropriate because, a given species can cause disease in many anatomical sites and the same anatomical site may be infested by many species of maggots. Based on the host-parasite interaction, he proposed an ecological classification as *'Specific (obligate)'*, *'Semi-specific (facultative)'* and *'Accidental'*.[35] Hall[2] modified it by adding *'primary'*, *'secondary'* and *'tertiary'* subcategories to the *facultative myiasis*. (Table 3) Zumpt [1] renamed *accidental myiasis* as *pseudomyiasis*. Anatomical classification is clinically more practical, while ecological classification gives better understanding of the pathogenesis.[2] Sometimes, myiasis is also classified into *benign* and *malignant* depending upon the tissue invasion aggressiveness of the infesting maggots and the clinical outcome of the patient.[36]

ENTOMOLOGY

Flies are universal and diverse. Approximately, they consist of 150,000 species, 10,000 genera and 150 families.[4,37] The Order *Diptera* has two major Suborders, the *Nematocera* (Greek: *nema* - thread-like; *cera* - antenna) and the *Brachycera* (*brachy* - short). The former consists mostly of blood sucking insects such as the mosquitoes that act as vectors for many viruses and protozoa.[37] Rarely, *Nematocera* larvae can cause *accidental myiasis* in children.(Table 4) Human myiasis is caused by the 4 families of Brachycera: the *Muscidae* (house fly), *Oestridae* (botfly), *Calliphoridae* (blowfly) and *Sarcophagidae* (flesh fly).(Table 5) Just about 80 species of these 4 families are responsible for 95% of human myiasis.[2]

Table 3. Ecological classification of human myiasis

Category	Definition†
Obligate myiasis	Aggressive maggots that depend on a host for completing the life-cycle. (They are capable of penetrating intact skin and feed on healthy tissue)
Facultative myiasis*	
Primary myiasis	Free living maggots* that can grow in host tissues only when they are damaged
Secondary myiasis	Free living maggots* that depends on another larva to establish infestation in host tissue. (It is a form of mixed infestation.)
Tertiary myiasis	Free living maggots* that grow in hosts that are near death
Accidental myiasis (Pseudomyiasis)	Free living or dead maggot that pass through a host without undergoing further developmental changes

† These definitions are as per the original descriptions of Hall.[2] Later authors have distorted the definitions in many different ways. Some of them have used 'obligate myiasis' synonymous with 'primary myiasis' and 'facultative' equal to 'secondary'.

** Free living maggots may complete their life cycle either on dead decaying matters or on a living host tissue*

Table 4: Rare accidental myiasis in children*

Anatomical location	Name of the maggot species
Intestine	*Eristalis tenax* *Megaselia scalaris* *Telmatoscopus albipunctatus* *Hermetia*
Genitourinary tract	*Eristalis tenax* *Megaselia scalaris* *Piophila casei* *Psychoda albipennis* *Scenopinus*
Wounds	*Megaselia scalaris*
Eye, ENT	*Drosophila melanogaster* (Fruit fly)
Nosocomial	*Megaselia scalaris*
Skin-furuncular	*Hermetia*

** Source: Francesconi [4]*

Therefore, it must be emphasized that myiasis is not one disease; but a group of diseases caused by varius dipteran larvae. Clini-cal features of each species differ significantly that they should be considered as separate disease entities.

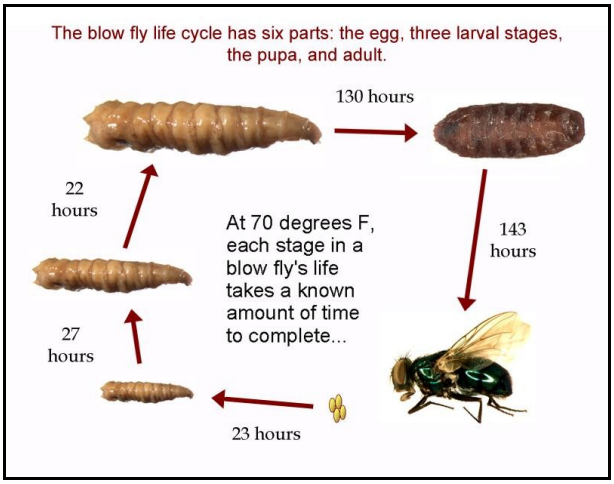


Fig 2. *Life cycle of flies (Courtesy of National Library of Medicine and Cleveland Museum of Natural History)*

Life span of many adult flies ranges between 1 to 10 days.[2,33,37] (Fig. 2) Therefore, they are in a great hurry to complete their reproductive life. A majority of flies oviposit, while some species like *Sarcophaga* larviposit.[30] Depending on the type of species, gravid flies are capable of laying 150 to 2000 eggs per batch.[2] An average female fly will

lay 4 to 5 batches of eggs in its life span. Flies oviposit either directly on the host tissue or on leaves and garbage from where they are transferred to the host. Fly eggs are very resistant to chemicals and hence can easily be disinfected using 5% formalin, 60% alcohol, mercury bichloride or acetic acid. This property is exploited in laboratory culturing of medical maggots.[38] From preventive medicine point-of-view, this is a disadvantage that they cannot be destroyed by even strong chemicals.

Dipteran eggs usually hatch in warm, moist, putrid substrate. They may remain dormant for several months until a suitable environment is available. Some species (e.g. *Cuterebra*) require a sudden fluctuation in environmental temperature for hatching. Usually such temperature change occurs in the months of March-April, June-July, and September-October. These periods coincide with the peak incidence of clinical myiasis.[2]

Larvae usually take 5-10 days to mature.[2,37] They pass through 2 or 3 stages of instars in the human hosts.[2,33] After 7-14 days of maturation, pupae fall off from the host and develop in soil. Maggots of different species vary in their shape and physical dimension (Fig. 3); but they are generally 2-30 mm in length and 1 to 7 mm in diameter. They are mostly spindle shaped with the oral end narrower than the posterior end. The paired, black respiratory spiracles at the caudal end are often mistaken for eyes.

Table 5: Entomology of common flies causing pediatric myiasis

Binomial name of the fly	Common name	Parasitism	Natural reservoir	Affected organ §
Alouattamyia baeri [2]	-	Obligate	Primates	(Rare) Lung, Throat, Skin
Auchmeromyia senegalensis [3]	Congo floor maggot	Obligate*	Man	External Blood suckers
Calliphora hilli	-	Facultative	Decomposed flesh	Eye
Calliphora vicina	Blowflies	Facultative	Decomposed flesh	ENT, GIT, TW, GUT
Chrysomya albiceps [3]	-	Facultative	Garbage, Feces	TW, Nose
Chrysomya bezziana [3]	OWSW	Obligate,	Sheep	TW, ENT
Chrysomya megacephala [3]	Oriental latrine fly	Facultative	Decaying flesh, Feces	TW, Ear
Chrysomya rufifacies [3]	Hairy Maggot fly	Facultative	Garbage, Feces	Tw, Nose
Cochliomyia hominivorax [3]	NWSW	Obligate	Mammals	ENT, Mouth, TW
Cordylobia anthropophagia [3]	African Tumbu fly, Mango fly	Obligate	Mammals, Chicken, Soiled linen, Feces	Skin
Cordylobia rodhaini [3]	Lund's fly	Obligate	Wild mammals, Soiled linen	(Rare) Skin
Cuterebra [2]	Rodent botfly	Obligate	Rodents, Grass	Skin, Viscera, Eye, RT
Dermatobia hominis [2]	Human botfly	Obligate	All mammals, Few birds	Skin
Eristalis tenax	Rat-tailed maggots	Facultative	Sewage, Polluted water	(Rare) GIT
Fannia canicularis [5]	Little house fly	Facultative	Decaying matter	Nasopharynx, GIT
Fannia Scalaris [5]	Latrine fly	Facultative	Decaying matter	GUT

(Table Continued)

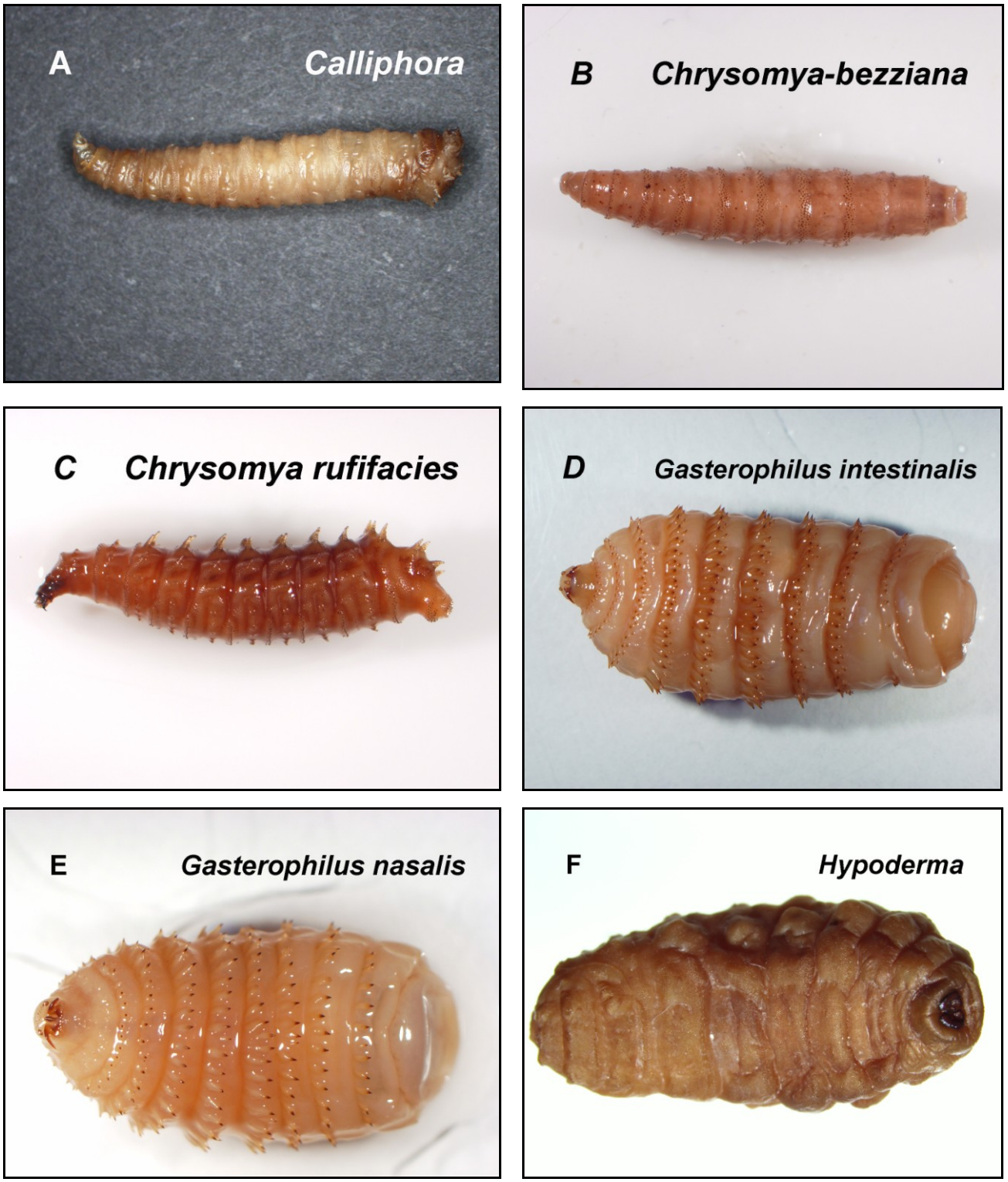
Gasterophilus intestinalis [2]	Horse botfly	Obligate	Wild mammals	Migratory, Eye, ENT, Lung
Hermetia illucens	Black soldier fly	Facultative	Animal wastes	GIT
Hypoderma bovis [2]	Cattle botfly	Obligate	Cattles	Migratory, Mouth, Throat
Hypoderma lineatum [2]	Common warble fly	Obligate	Cattles	Migratory, Brain
Hypoderma tarandi [2]	Reindeer botfly	Obligate	Raindeer, Caribou	Eye, Mouth, Throat, Skin
Lucilia cuprina [3]	Sheep blowfly	Facultative	Decaying matter	TW
Lucilia sericata [3]	Greenbottle blowfly	Facultative	Decaying matter	Nose, TW
Musca domestica [1]	House fly	Facultative	Decaying matter	GIT, GUT, TW, ENT
Oestrus ovis [2]	Sheep nasal botfly	Obligate	Sheep, Goat	Eye, ENT
Parasarcophaga crassipalpis	Flesh fly	Facultative	Decaying matter	Ear
Pharyngomyia picta	Deer throat botfly	Obligate	Wild mammals	Eye
Phormia regina [3]	Black blowfly	Facultative	Decomposed flesh, Cattle	TW
Piophila case	Cheese skipper fly	Facultative	Barks of trees	Rectum, GIT
Protophormia terranovae [3]	-	Facultative	Decomposed flesh, Cattle	TW
Rhinoestrus purpureus	Nasal botfly	Obligate	Horse	Eye
Sarcophaga peregrina [4]	-	Facultative	Excreta, Garbage	GIT
Sarcophaga ruficornis [4]	-	Facultative	Excreta	TW, Ear
Sarcodexia lambens [4]	Flesh fly	Facultative	Excreta	GIT
Sarcophaga haemorrhoidalis [4]	Fecal fly, Filth fly	Facultative	Excreta	TW, GIT, Ear
Tubifera tenax	-	Facultative	Sewage	GIT
Wohlfahrtia magnifica [4]	Spotted flesh fly	Obligate,	Livestock	ENT, Mouth, TW, Skin
Wohlfahrtia meigeni [4]	-	Obligate	Livestock	TW (infants)
Wohlfahrtia opaca [4]	-	Obligate	Livestock	TW
Wohlfahrtia vigil [4]	Fox maggot fly	Obligate	Livestock	TW (children), Skin

Source: Azami [(40)] , Hall [(2)]
Family attribution: [1]Muscidae, [2]Oestridae, [3]Calliphoridae, [4]Sacrophagidae, [5]Fannidae

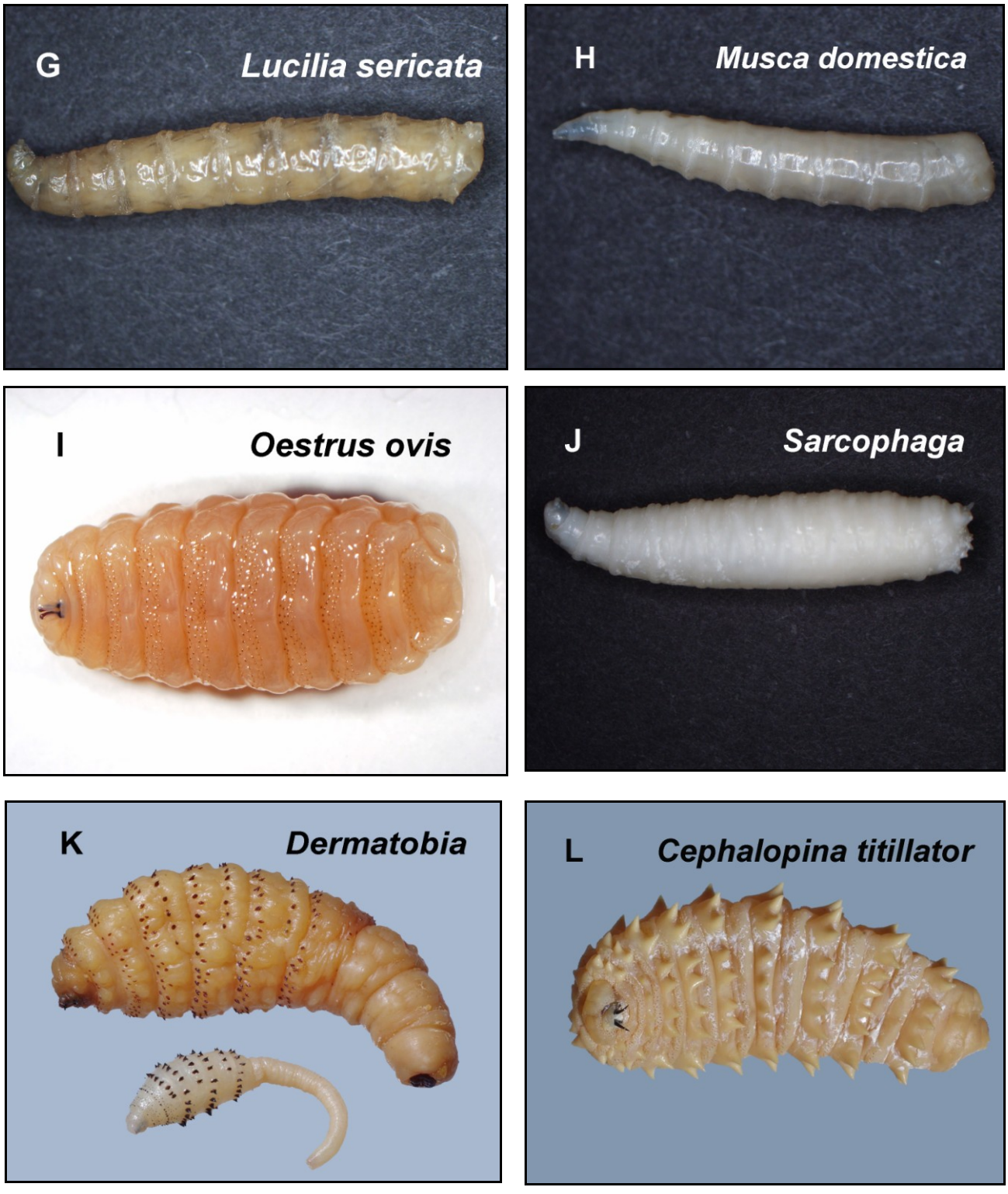
ENT - Ear, nose and throat, RT - Respiratort tract, GIT - Gastrointestinal tract, GUT -Genitourinary tract, TW- Traumatic wounds, OWSW - Old World screw-worm, NWSW - New World screw-worm

Lucilia was previously known as Phaenicia; Sarcophaga haemorrhoidalis as S. cruentata

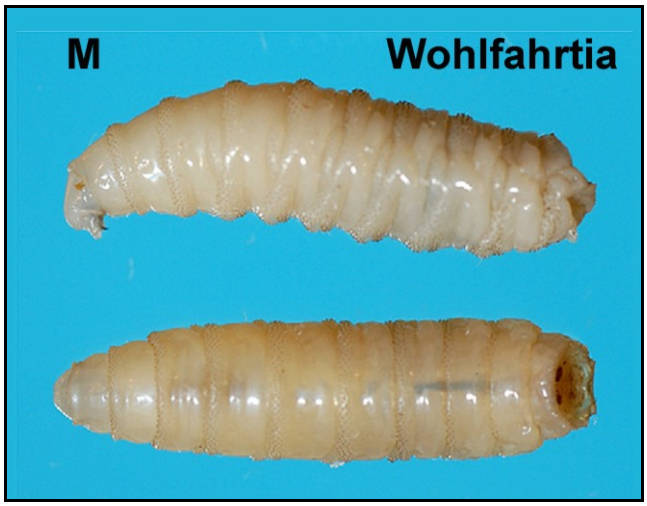
** Sanguinivorous (live in soil but approach host for blood meal); §The term skin refers to furuncular myiasis*

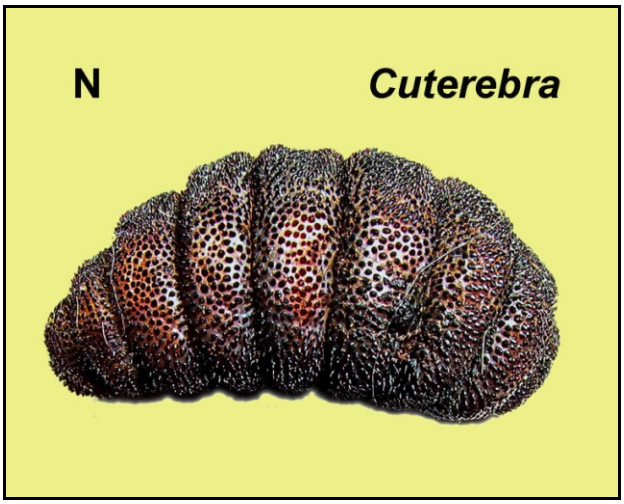


(Fig 3 Continued)



(Fig 3 Continued)





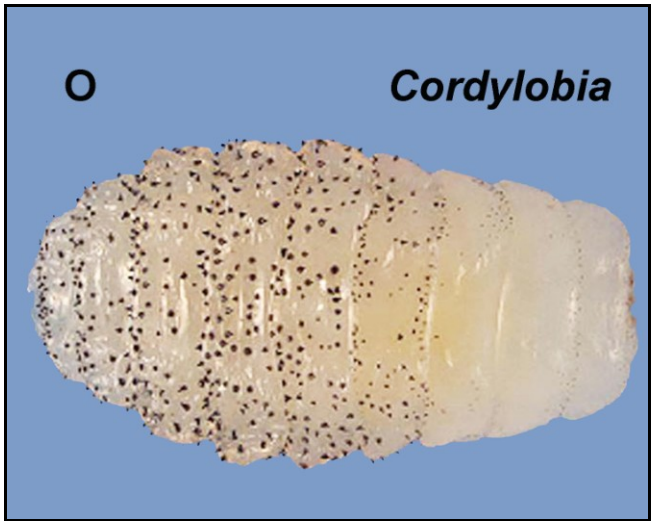


Fig 3. ***Morphological of maggots affecting children.***

In all the imgaes, maggots are oriented with their oral end towards the left side. (Photo credits: Fig. A-J are reproduced with permission from Dr. Lyn Knott, School of Veterinary Sciences, University of Queensland; Fig. K by Acarologiste from Wikimedia Commons under CC-BY-SA-4.0; Fig. L by Daktaridudu from Wikimedia Commons under CC-BY-SA-4.0; Fig. M by James Kalisch, Entomology Department, University of Nebraska at Lincoln from DOI:10.29011/2688-6383.000023 under fairuse; Fig. N from the Western College of Veterinary Medicine, University of Saskatchewan under fairuse; Fig. O by Rebecca Graham, from the Pest and Diseases Image Library (Image ID: 5489545) at Bugwood Center for Invasive Species and Ecosystem Health, University of Georgia under CC-BY-NC 3.0)

ETIOPATHOGENESIS

In 1897, Stecle described the link between myiasis and flies.[27] Myiasis is not one disease, but a group of disorders caused by different species of dipteran larvae. Pathogenesis of obligate and facultative myiasis differs significantly. Obligate larvae essentially require a host to complete their life cycle and they are aggressive in nature. They may painlessly penetrate intact skin and start feeding on even live tissues when necrotic tissues are exhausted. On the other hand, facultative maggots mostly grow in dead decaying matter. They are less aggressive and can grow only on pre-existing necrotic tissues. As they are incapable of causing harm to the live tissues of the host, they are ideal for MT. Two of the primary facultative blowfiy larvae (*Lucilia sericata*, *Lucilia cuprina*) are widely used in MT. *Oestridae* have both facultative and obligate species while *Sarcophagidae* are mostly facultative pests. There are only 3 obligate screwworms[2] (*Cochliomyia hominivorax*, *Chrysomya bezziana* and *Wohlfahrtia magnifica*) which are highly dangerous that they may even kill the host.

Modes of Contracting

Susceptible hosts may contract maggots by many different ways of oviposition: intradermal implantation, contact inoculation, squirt inoculation,

environmental contamination and physical relocation. Raely gravid flies stuck matured larvae to the host skin or hair (larviposition).

Intradermal implantation

Some species of obligate maggots such as the *Dermatobia* oviposit using a phenomenon called *phoresis*.[2] In this, female flies stick the eggs on to the belly of a blood sucking slave insect (e.g. ticks, mosquitoes) without affecting their flying ability. These 'porter insects' inoculate the eggs into the dermis of host during a blood meal. Sudden temperature change inside the host-skin causes the eggs to hatch. Alternatively, the larvae hatched on the surface of porter flies may crawl into the host-dermis at the site of piercing with proboscis. As this transfer happens mostly during sleep, the host is usually unaware of the implantation. In this way, usually a single egg or a couple of eggs can be transmitted.

Contact inoculation

Gravid female flies of facultative species are often attracted by the fetid smell of bacterial metabolic byproducts (e.g. hydrogen sulphide, methane and ethane), alkaline pH of wounds or the smell of flesh and blood. They lay eggs on open wounds when they stroll and feed on the necrotic debris. Hundreds of eggs may be oviposited by a single fly in one sitting. *Hypoderma* oviposit on body hairs.

Squirt inoculation

Body orifices of the host are effectively guarded from insects by robust physiological protective mechanisms such as cerumen (ear) and sneezing reflex (nose). In these circumstances, the female flies oviposit near the orifices so that the hatched maggots may crawl into the lumen. Alternatively, they - with great force - squirt hundreds of eggs directing towards the body orifices while flying at a distance (hence the name 'blowfly').[2] Presence of purulent discharge, as in suppurative otitis media, attracts blowflies.

Environmental contamination

Some species of flies oviposit on green leafs and grass. *Cordylobia* species prefer to lay eggs on soiled linen and undergarments. The eggs are then transmitted to the vulnerable hosts by physical contact. Special adhesive fluid secreted on the surface of the eggs enables their quick transfer by adhesion even if the contact time is very brief.[2] Eggs may also be transferred by contaminated hands into the nose.[27]

Physical relocation

Saprophytic larvae living in contaminated food or drink are ingested by the careless host. Mostly accidental myiasis is acquired in this way. Improper food handling, poor eyesight, mental dysfunctions and attention deficit while eating are the causes of this tragedy.

Pathogenic Behavior

After oviposition, warmth of the host tissue causes hatching of the eggs. The emerging larvae undergo 3 stages of molting, called first, second and third instars respectively. The third instars feed on the live or necrotic tissues and prepare themselves for pupation. The first two stages of instars are highly antigenic, provoking intense host immune reaction. However, third instars learn to downplay thier antigenicity in order to develop symbiosis with the host. The adaptive mechanisms exhibited by third instars include: (1) Alimentary secretions and excretions of maggots (ASEM) contain several chemicals and proteolytic enzymes that control the growth of competitive bacteria in the wound. (*Vide infra*) (2) Maggots, to their survival advantage, develop symbiosis with certain bacteria. For example, presence of *Proteus* in the larval gut is essential for the synthesis of bacteriostatic chemicals in the ASEM which in turn protect the larvae from other pathogenic bacteria. (3) They develop sharp body spines and oral hooklets that prevent accidental slippage from the host before full maturation. (4) They reduce antigenicity of the cuticle to avoid being targeted by the host immune

cells or antibodies. (5) They also secrete immunomodulatory chemicals such as urea to blunt the immune attacks of the host. (6)ASEM also contain several anti-inflammatory modulators that help the maggots from being trapped in the inflammatory fibrosis.

As maggots are highly photophobic they bury their head-ends deep inside the host tissue by burrowing arborizing tunnels. This process is enabled by digestive enzymes present in ASEM and devouring mouth apparatus. When maggots mature into pupae, they either slip out quietly or are ejected out of the host by the pressure of tissue edema.[2] As this happens mostly during night sleep, the host may not be aware of the pupal exit. Pupae get buried into the soil to complete their life cycle before emerging as adult flies. (Fig. 2)

The basic pathogenicity of maggots can be summarized as follows: (1) Damage caused to the host tissue by digestive enzymes and chemicals present in the ASEM; (2) Mechanical damaged caused by the chewing and crawling activities of the maggots; (3) Provocation of immunemediated inflammatory reaction in the host; (4) Systemic absorption of larval excreta and toxin and (5) Promotion of secondary bacterial infections, especially that of *Proteus* and *Escherichia coli*.

RISK FACTORS

Myiasis is known as a disease of uncleanliness and poverty.[39] More than 90% of the affected patients are illiterates and economically poor while only 5% are of middle class.[27,39] It affects those who are unable to maintain personal hygiene or those who live in unhygienic conditions. Unemployment, illiteracy, improper garbage disposal, close proximity of farm animals and unsafe water resources are associated with myiasis. Although it is generally common in rural and slum communities, in an Iranian study 86% of patients were from urban areas.[40] Cultural practices that predispose to myiasis include smearing cowdung on house floors, native treatment of wounds with raw herbal pastes made of contaminated leaves, defecation in open grasslands, sitting or sleeping on mud floor, using dried camel dung to mop-up the last few drops of urine (an Arabian habit), applying honey to wounds as home remedy and bathing in public pools where cattles are also washed.

Those who cannot care for themselves because of physical or mental disabilities are more prone for myiasis. They include infants, mentally retarded (e.g. cerebral palsy), neuromuscular cripples (e.g. paraplegia), visually challenged, orphans without proper care-takers and debilitated patients (e.g. malnutrition and cancers).[41]

Predisposing necrotic tissue that attract facultative myiasis include fungating tumors, noma, pyoderma, leprosy, neuropathic or trophic ulcers (e.g. meningomyelocele, bedsores), necrotizing faciitis (e.g. Fournier's gangrene), contaminated traumatic wounds due to farm or road accidents and purulent discharge from body orifices (e.g. atrophic rhinitis, suppurative otitis media).

A warm humid climate is essential for the hatching of dipteran eggs. Hence, myiasis is typically a disease of tropics and subtropics. It is common in sea-shore areas than at high altitudes.

DEMOGRAPHY

Distribution of myiasis is global with the highest incidence in the tropical countries. Previously, the fly species of a geographic location were thought to be unique and specific. However, with increasing international travels, the geographic differences are slowly fading away.[4] For example, *Cochliomyia* (New World screw-worm) maggots entered Libya in 1988 from America, which is 7000 km away.[42,43] On the other hand, in 2014, *Chrysomya* (Old World screw-worm) larva from Africa threatened the Australian health care system.[44] *Chrysomya bezziana* (20%) and *Lucilia sericata* (30%) are the two most common forms of myiasis in

Asian and African countries.[40] *Oestrus ovis* infestations are rare in children.

Species identification is rarely done. Myiasis is not a notifiable disease. Physicians often consider it a minor problem of wound healing. There are no official data bases of human myiasis. Consequently, authentic demographic data on myiasis is scarce.[3,4] The exact incidence of clinical myiasis is not known.[45] Traumatic wound myiasis has been reported in 5.1% of septic wounds in an Egyptian hospital.[3,46] In 1993, Singh from Rohtak, India reported 94 cases of pediatric myiasis over a span of 6 years.[47] Pediatric infestations form 38% of all human myiasis.[47]

Myiasis is an occupational hazard for shepherds, animal handlers and agricultural workers. It occurs without any age or sex predilection. However, a systematic review of oral myiasis found it to be common in boys.[48] The youngest reported patient is a 1-day-old newborn.[49] There are some evidences that the demographic pattern of myiasis is changing over time. For example, in 1979-80, about 30% of ENT (ear-nose-throat) myiasis occurred in the first decade of age, while it dropped to 15% in 2003-04.[27]

Biological life-cycle of various species of flies differs with seasons. Cases of *Wohlfahrtia* and *Cuterebra* infestation are common in summer months, *Oestrus ovis* in autumn, *Hypoderma* in winter and *Cordylobia* in rainy seasons (when sylvan, the natural host, approaches villages). *Gasterophilus* maggots occurs throughout year without any seasonal fluctuation in incidence.[4] In India 10% of myiasis occurred during January-March, 0% during April-June, 15% during July-September and 75% during October-December.[27]

PATHOLOGY

Pathological changes caused by maggots differ between surface dwelling facultative maggots and aggressively invading obligate maggots. Wound myiasis is usually benign. Chemicals present in the ASEM of facultative larvae suppress inflammatory reaction of the host tissues. On the other hand, obligate maggots may cause considerable damage to host tissue by the strong digestive enzymes present in ASEM. These enzymes are capable of eroding even bones.

Infected soft tissues show progressive liquefaction necrosis and hemorrhage.[2] Marginal acanthosis is often seen on the walls of larval burrows.[50] Maggots may damage host blood vessels by the strong oral hooklets and digestive enzymes. Torrential bleeding from eroded major vessels has been reported.[51,52] The typical response of the host immune system to the presence of invasive maggots is dense infiltration of lymphocytes, eosinophils, histiocytes, mast cells, plasma cells, Langerhans cells and Langhans cells.[2] Local edema, peripheral hyperemia, dilated capillaries and regional lymphadenopathy are also seen.

Cell wall protein of maggots triggers T-helper cells, which in turn stimulate plasma cells and B-lymphocytes to produce antibodies.[2] This results in partial resis-tance against reinfection seen in some species, but this immunity is short lived.[2] Fibroblasts tend to halt the invasion of maggots.[2] Migratory maggots of *Hypoderma* species leave behind a trail of edematous yellow-green gelatinous track with eosinophilic infiltration.[2] *Dermatobia* often provoke cell mediated immunity.[2] After the departure of maggots, wound heals fast within 5-10 days.[2]

Myiasis is frequently associated with secondary bacterial infection. The isolates were *Staphylococcus aureus* in 90%, *Escherichia coli* in 2% and *Klebsiella* in 8% of cases.[27]

CLINICAL FEATURES

It is not known as to what proportion of the clinical symptoms is attributable to the predisposing necrotic wounds and how much can be attri-

butable to the maggots per se.[53] General symptoms caused by the toxins or coexisting bacterial infection include fever, generalized or local itching, lymphadenopathy, poor appetite, weight loss and anemia. Local manifestations of myiasis differ according to the infesting species and the affected anatomical area. *(Vide infra)*

Furuncular Myiasis

Furuncular myiasis, also known as *warble disease* in animals, was first described by Blanchard, and later by Sanchez in 1893.[54,55] It is caused by obligate aggressive larvae such as *Dermatobia, Cordylobia, Cuterebra* and *Wohlfahrtia*.[29] They painlessly penetrate the intact skin within 5-60 min of contact and spend 4-80 days in the host.[4,29] Symptoms develop within 2 days of oviposition; but usually 2-7 weeks elapse between the onset of first symptom and identifying the correct clinical diagnosis.[54]. Young children are more vulnerable because of thin skin and poor immunity.

As the eggs of *Dermatobia* are implanted by blood sucking porter insects, lesions are predominantly distributed in exposed body parts such as the limbs and head-neck. On the other hand, *Cordylobia* ovipositing on soiled linen are common in perineum, trunk, thighs and buttocks. Palms and soles are never affected.[54] Usually only one worm is seen per lesion. Rarely multiple maggots, as many as 28, have been be reported.[4,54] Coalescence of closely located lesions may create an illusion of multiple maggots within one lesion. *Dermatobia* lesions are usually single while that of *Wohlfahrtia* are in crops.

At the site of maggot penetration, itching or occasionally a sharp pain is felt. Children may scratch the entry site and cry unconsolably even before the appearance of a skin lesion. The larvae evoke strong immunological reaction at the entry point that they cause a red and tender furunculoid (boil-like) lesion of 0.2 to 2 cm size. Through a pathognomonic central punctum, the respiratory spiracles at the rear end of maggot are exposed to the atmosphere. Unless examined with a magnifying glass the punctum is easily missed. Exacerbated pain during night sleep, formication (a strange sensation of something crawling underneath the skin), paresthesia, a drop of sticky blood stained discharge through the punctum (excreta of the maggot), foul smell and crusting (dried ASEM) are usually seen. In a 5-year-old girl, pseudo-pulsation due to writhing of worms was mistaken for a vascular lesion.[56] Superadded bacterial infection results in diffuse cellulitis and purulent exudate with tiny gas bubbles. Regional lymphadenopathy and systemic symptoms are rare. In scalp lesions focal alopecia around the punctum is common. Morphological variation of lesion includes bullae, vesicles, abscess, ecchymosis, cellulitis, pustules and ulcers.[4]

When left untreated, furuncular myiasis heals spontaneously in 8-30 days when the matured pupae depart from the host. Lesions usually heal without leaving any trace; however, occasionally pigmentation, hypertrophic scar or keloids may result especially in malnourished children.[4]

Sanguinivorous (Blood Sucking) Maggots

Congo floor maggots typically live in soil, but approach the host during night time just for a blood meal. Their importance lies in transmission of diseases such as the African sleeping sickness.

Wound (Traumatic) Myiasis

Maggot infestation of soft-tissue wounds is caused by both facultative and obligate maggots.[3] *Cochliomyia hominivorax*, *Chrysomya bezziana*, and *Wohlfahrtia magnifica* are the most common species involved. In USA, 87% of wound myiasis (WM) is due to *Lucilia sericata, Phormia regina,* and *Cordylobia*.[8] *Cochliomyia* forms 62% of WM in Brazil. Other involved species are *Dermatobia, Musca domestica, Chrysomya megacephala, Parasarcophaga, Calliphora, Lucilia cuprina* and *Sarcodexia lambens.*

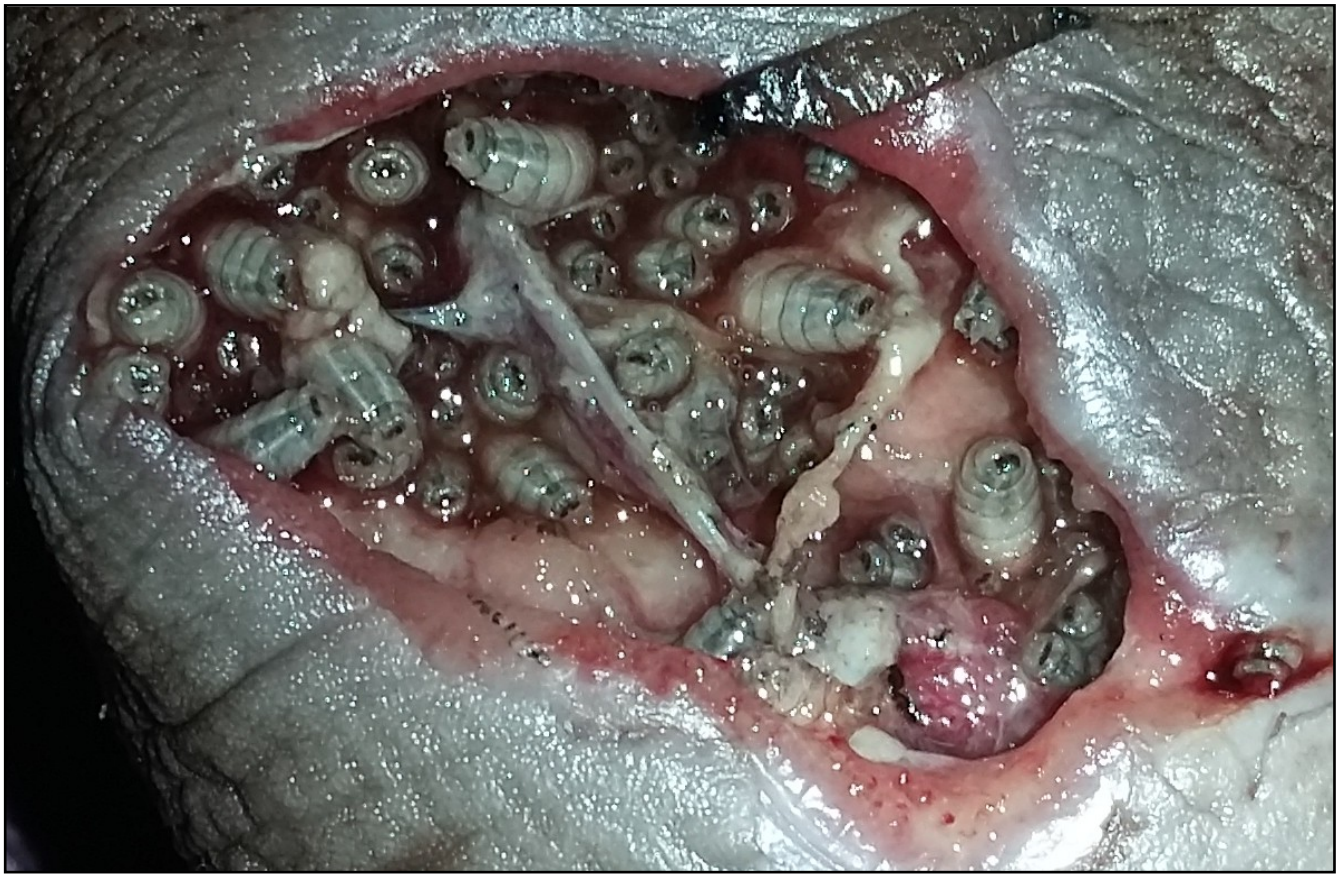


Fig 4. *Traumatic wound myiasis in a 10-year-old boy. The paired block dots are the exposed respiratory spiracles at the posterior end of the maggots. (© Raveenthiran)*

Usually only one species is involved, while in 3% of cases mixed species may be noted.[4,57] Presence of maggots attracts other flies to lay more eggs on to the wound.

Chrysomya bezziana and *Cochliomyia hominivorax* may cause severe pain by scratching with their sharp spines while crawling. There may be as many as 100-500 worms in the deep arborescent burrows of the wounds. (Fig. 4) Scanty amount of foul smelling blood-stained discharge is caused by the excreta of maggots. When necrotic tissues are exhausted, obligate maggots may start feeding on healthy tissue including the blood vessels and the underlying viscera.[52] Regional lymphadenitis is common.

Migratory (Subdermal or Creeping) Myiasis

Sometimes, an enthusiastic maggot starts migrating aimlessly and is doomed to get trapped deep inside the host tissues. Very rarely, the wondering worm may emerge out penetrating the overlying skin. Migratory lesions are of 2 types: the superficial itchy serpentine erythematous linear tunnels caused by *Gasterophilus* and the painful subcutaneous evanescent cysts caused by *Hypoderma*.[41] About 20% of *Cuterebra* and 2% of *Dermatobia* infestations become migratory myiasis. Ontogenic development of the wondering larvae is arrested depending upon the antigenicity of the species and the depth of migration. For example, migratory *Gasterophilus* larvae seldom pass beyond the first instar stage.

Migratory maggot is common in exposed body parts. The early symptoms are similar to that of furuncular myiasis. As the larva starts migrating, it leaves behind a trail of raised palpable serpiginous red lesion with its rear end healing and fading. Migration of *Gasterophilus* larvae is more bizarre and serpiginous than that of *Hypoderma*. Maggots may survive up to one month and may migrate 1-30 cm beneath the skin.[4,29] Exceptionally, *Hypoderma* can migrate as far as 2-30 cm in 24 hrs and as fast as 125-150 cm in 12 hrs.[4] Usually, larvae migrate in superficial planes, but rarely they may invade internal viscera. Migratory maggots may cause ascites, pleuro-pericardial effusion, regional or systemic lymphadenopathy, arthralgia, myalgia, scrotal edema and meningitis.[4] Neural invasion may cause paralysis of limbs, blindness or death.

Left untreated, migratory myiasis may get spontaneously cured, but with a varying degree of fibrosis and calcification of the dead worm.

Fig 5. *Umbilical myiasis (© Raveenthiran)*

Umbilical Myiasis

Umbilical infestation is the commonest form of neonatal myiasis.(Fig.5) Fewer than 35 cases have been reported in the literature, of which 60% are from Nigeria and India.[58-61] The incidence is very high in Nigeria that, 12 out of 55 neonates (22%) examined for omphalitis were having myiasis.[60,61] It occurs between 2-20 days of neonatal life with a median of 7 days.[58,59] *Cochliomyia hominivorax*, *Chrysomya megacephala*, *Sarcophaga vilosa* and *Musca domestica* are the common species. Infestation is facilitated by local cultural practices such as applying cow dung or herbal paste made of oviposited leaves to the umbilical cord stump.[62] Omphalitis and portal vein thrombosis are the potential complications.

Nasal Myiasis

Nasal infestation forms about 11% of all myiasis in pediatric practice.[4,45,47] It is commonly due to *Cochliomyia hominivorax*, *Chrysomya bezziana*, *Oestrus ovis*, *Wohlfahrtia magnifica*, *Lucilia sericata*, *Drosophila* and *Calliphora vicina.* Nasal oviposition is facilitated by purulent rhinorrhea or when the protective sneezing reflex is impaired as in atrophic rhinitis, leprosy, rhinoscleroma and tuberculosis. Local pain, headache, mucopurulent rhinorrhea, epistaxis, blocked nose, mouth breathing and anosmia are the usual symptoms. Volley of sneeze expelling maggots is reported in 20% of the affected children.[4,63] In one case, as many as 388 worms were recovered.[19] Maggots slipping back into the throat, especially during night sleep, may cause nocturnal dry cough, dyspnoea, stridor, laryngospasm and even death.

Maggots may corrode even the bone and cartilage, thus leading to complications such as nasal septal perforation, orbital cellulitis, pharyngeal ulcers, saddle nose deformity, oronasal fistula, cerebrospinal fluid (CSF) rhinorrhea, pneumocephalus and meningitis.[4]

Aural Myiasis (Otomyiasis)

Ear infestation forms about 86% of all myiasis in children.[4,47] *Cochliomyia hominivorax*, *Wohlfahrtia magnifica*, *Chrysomya bezziana*, *Chrysomya megacephala*, *Sarcophaga* and *Parasarcophaga crassipalpis* are the common species of otomyiasis. Purulent otorrhea of suppurative otitis media is a risk factor. It is usually unilateral, involving the external auditory canal or middle ear. Rarely, it may be bilateral. It is common below 10 years of

age. The youngest reported patient was a 1-day-old newborn.[49] Passage of worms (81%), otalgia (41%), otorrhea (44%), ear bleed (50%), formication, itching (33%), foul smelling (83%), tinnitus, vertigo, restlessness, perforation of the tympanic membrane and deafness are common.[64] Affected young infants may bang their head without any obvious reason. It is not known if the tympanic membrane is actively perforated by the maggots or if it is a pre-existing lesion that attracted larvae. Worms lodged in the mastoid air cells are difficult to remove and they usually die and get calcified.

Oro-pharyngeal Myiasis

Since the first description by Lawrence in 1909, fewer than 70 cases of pediatric oral myiasis have been reported.[48] It is a form of WM due to poor oral hygiene and suppurative gingivitis. Sleeping with open mouth is a prerequisite for ovipositing. Thus, the risk factors include uncorrected cleft lip, noma, sleep apnea syndrome, adenoid enlargement, facial trauma and anterior open-bite dentition. Breast feeding infants may acquire invisible eggs from mother's unclean breast.[65] Rarely, oral myiasis is due to consumpsion of contaminated food or drink.

It is usually caused by *Cochliomyia hominivorax, Wohlfahrtia magnifica, Musca domestica, Chrysomya bezziana, Oestrus ovis, Hypoderma bovis, Hypoderma tarandi, Musca nebulo, Gasterophilus intestinalis and Calliphora vicina.* The angle of mouth, lips and anterior gingivae are frequently involved. Number of maggots retrieved vary from 1 to 112.[48] About 50% of the affected children are below 5 years of age.[48] Local pain, swelling, redness, halitosis, fornication, sore throat, retching, vomiting and dry cough are the common symptoms. Once, a fibrosed maggot in the submucosal plane was mistaken for a salivary adenoma.[66] Extensive tissue destruction may result in oro-facial or oronasal fistulae.

Ophthalmomyiasis

Eye involvement may be superficial involving the orbital socket and extraocular apparatus (ophthalmomyiasis externa, extraocular or orbital myiasis) or deep involving the chambers of the eye ball (ophthalmomyiasis interna, intraocular or ocular myiasis, ophthalmomyiasis profonde). They form 2% of all pediatric myiasis.[4] In a review of 27 cases of ophthalmomyiasis, there were 2 children, both males, aged 1.5 and 10 years respectively. In one of them maggots infested the empty socket following enucleation of the eyeball for retinoblastoma.[67]

Orbital myiasis involves conjunctiva and lacrymal apparatus. It is caused commonly by *Oestrus ovis and r*arely by *Rhinoestrus purpureus*, *Dermatobia hominis*, *Chrysomya bezziana*, *Lucilia* and *Cuterebra.* Onset of symptoms is dramatically sudden with a foreign body (gritty) sensation and excessive lacrimation. Other symptoms include epiphora, fornication beneath the eyelids, redness of the conjunctiva, photophobia, edema of the eyelids sub-conjunctival hemorrhage, pseudo-membrane formation and punctate keratopathy. The lesion is usually unilateral. Hard nodular lesion of eyelids caused by furuncular type maggots may mimic hordeolum. Number of maggots vary from 1-20. Orbital myiasis may spread to the nose by larvae creeping through the nasolacrimal duct and vice versa. If untreated, symptoms may last for 7-10 days before spontaneous resolution.

Ocular myiasis is a complication of orbital infestation. It affects the anterior or posterior chambers of the eyeball. Maggots may have penetrated the sclera using their strong oral hooklets; but the point of entry is often not obvious. The vitreous chamber is more often affected than the aqueous chamber. Ocular myiasis is more serious than the orbital form, as it endangers the vision. Although, a single worm is usually found inside the eyeball, cases with as many as three worms and bilateral involvement have been reported in the literature.

Anterior chamber myiasis may mimic the uveitis. Eyeball myiasis causes redness of eye, floaters, ocular pain, blindness and scotoma. *Hypoderma tarandi* is more aggressive in causing blindness. Fundus examination may show typical dead worm floating in the vitreous humor. Maggots crawling beneath the retina may leave behind atrophic retinal pigment-epithelial tracks with a characteristic crisscross pattern. Vitreous hemorrhage, fibrovascular proliferation, exudative retinal detachment and retinal scarring are the serious complications of ocular myiasis.

Tracheal (Tracheostomy) Myiasis

Maggots may complicate tracheostomy wounds especially in mentally retarded children.[68] It is commonly due to *Chrysomya bezziana, Cochliomyia hominivorax, Lucilia sericata* and *Musca domestica*.[68]

Urogenital Myiasis

Vaginal (female genital) myiasis

Vaginal infestation is predisposed by not wearing undergarments. Dysuria, genital itching and foul smelling leucorrhea are common. Rarely, vaginal maggots may crawl into uterine cavity; but more commonly the prolapsed uterus in meningomyelocoele is colonized.[69,70]

Preputial, penile or scrotal (male genital) myiasis

Fewer than 6 cases of genital myiasis have been reported in male children[71-73] and the youngest was 7-months old.[73] *Cordylobia anthropophaga* and *Dermatobia hominis* are the commonly isolated species.[73] Although the preputial sac of phimotic boys may get infested, more often furuncular lesions occur on the penile shaft and the scrotum. An 8-year-old boy with scrotal myiasis was mistaken for testicular torsion.[74] A 3-year-old boy with preputial myiasis was mistakenly treated with antibiotics for balanitis.[75]

Urethral (internal urogenital) myiasis

Migratory maggots may rarely reach the urethra or bladder and get mummified. *Megaselia scalaris*, *Psychoda albipennis*, *Eristalis tenax*, *Fannia canicularis, Piophila, Fannia scalaris*, and *Muscina stabulans* are the commonly retrieved species from the bladder. Urinary symptoms include dysuria, lumbago, hematuria, sterile pyuria, albuminuria, uroliths and acute retention of urine. Cystoscopy is not only diagnostic but also therapeutic.[76]

Bone and Joint (Pin-site) Myiasis

Interestingly, maggots that were historically used to treat chronic osteomyelitis, are also considered aa a disease when they occur spontaneously in the exposed bones. 'Pin-site myiasis', a form of WM, occurring at the site of external fixator pins is a well known to complicate Ilizarov procedure in adults and children.[77] Fungating osteosarcomas with myiasis are fortunately rare nowadays.[78] Creeping maggots may detach mucoperiosteum from the bone cortex thereby causing excruciating pain and new bone formation.

Cerebrospinal Myiasis

Since the first description in 1939 by Frumin and Katsnelson[79] fewer than 20 cases of cerebral myiasis have been reported in the world literature, of which 30% are in children.[80,81] It is often a complication of orbital, aural or nasal infestation.[79,82,83] Nasal maggots invade the frontal lobe (38%), while aural larvae invade the temporal lobe (14%).[80] In 19% of cases, they occurred following head injuries.[Ramon] Frequently, the trailing end of the migratory tunnel of maggots heals completely that the route of entry into the brain remains undetectable in 33% of cases.[80,82] The youngest reported patient was a 5-month-old infant, in whom *Dermatobia* maggots from a scalp wound penetrated the anterior fontanelle to cause fatal cerebral myiasis.[84] The commonly involved species are *Hypoderma bovis* (14%), *Lucilia sericata* (5%), *Dermatobia hominis (14%)* and *Hypoderma lineatum* (5%), the duration of symptoms

ranges from 10 days to 2 years.[81] Non-specific chronic headache, seizures, altered sensorium, motor paralysis, intracranial hypertension and extra-pyramidal symptoms are the common symptoms. Interestingly, associated meningitis is rare due to the bacteriostatic property of ASEM.[80]

Gastro-intestinal Myiasis

The first adult patient of intestinal myiasis was reported by Herms and Gilbert in 1930.[85] It is a form of pseudomyiasis due to accidental ingestion of tiny maggots in contaminated food or drink. Oropharyngeal maggots may also get swallowed. The youngest patient was 1-year old.[85] The cuticle of maggots can withstand the corrosive action of gastric acid. In fact, 2 children with gastric infestation have been described.[85] *Sacrophaga haemorrhoidalis* is the commonest gastro-intestinal maggot in the Indian subcontinent.[85] Other reported species are *Fannia canicularis*, *Hermetia illucens*, *Piophila casei*, *Muscina stabulans*, *Megaselia scalaris*, *Eristalis tenax*, *Musca domestica*, *Phormia regina*, *Lucilia cuprina*, *Tubifera* tenax, *Sarcophaga crassipalpis, Sarcophaga peregrina* and *Stomoxys calcitrans.* Concomitant helminthic infestations are common in intestinal myiasis. Asymptomatic passage of maggots in stool or vomitus is not rare. But it should carefully be differentiated from post-defecation contamination from the environment. Vague abdominal colic, flatulence, dyspepsia, rectal bleed, nausea, hematemesis, vomiting and perianal itching are the frequent symptoms.[85,86] Duration of symptoms may range from 2 weeks to 5 years.[85]

Anorectal Myiasis

In 1972, Aspock reported the first and youngest case of rectal myiasis in a 4-month-old boy due to *Fannia canicularis*.[87] Since fewer than 10 cases of rectal myiasis have been reported in the pediatric literature. Usually eggs are laid in the perineum and the hatched maggots crawl into the rectum. A 12-month-old infant got infested by consuming contaminated over-ripe banana.[88] A 4-year-old Indian girl developed *Chrysomya bezziana* larvae on the surface of prolapsed rectum.[89] Rectal myiasis causes bleeding which may be just blood streaks in stool, dripping at the end of defecation or frank bloody diarrhea.[90,91] A 8-month-old with *Sacrophaga* infestation passed hundreds of worms in bloody diarrhea.[91] In a 2-year-old girl, rectal myiasis due to *Parasarcophaga crassipalpis* was associated with Salmonella food poisoning.[92] Recto-perineal fistula has been reported in association with anorectal myiasis.[93]

Pulmonary Myiasis

Lung infestation is a form of pseudomyiasis due to *Cuterebra, Alouattamyia baeri*, *Megaselia spicularis*, and *Gasterophilus.* Maggots reach the lungs either by aspiration from oro-pharyngeal lesions or through pleural space from chest-wall wounds. Brassy cough, blood tinged sputum and wheezing are the common symptoms. Chest x-ray or CT scan may show coin-shaped opacity or calcification.[94]

Nosocomial and Epidemic Myiasis

In 1980, Mielke and Schlote first reported hospital acquired myasis.[4,95] It is rare in rich countries and is under-reported from poor countries. As a result its exact incidence is not known. Although it can be easily dismissed as a deficiency of medical services, true hospital and community outbreaks have been reported, especially from intensive care units and preterm nurseries.[95-99] It is commonly due to *Lucilia sericata*, *Megaselia scalaris*, *Sarcophaga, Cochliomyia hominivorax* and *Musca domestica*. Nasal and ostomy infestations are common.

DIFFERENTIAL DIAGNOSIS

The diagnosis of myiasis is straightforward when the worms are clinically visible. On the other hand, when they are hidden in deep burrows, the lesion may be mistaken for a variety of diseases. (Table 6) Particularly, pseudomyiasis poses considerable diagnostic challenge. Detailed travel history and careful clinical examination are the key for correct diagnosis.

Table 6. Differential diagnosis of pediatric myiasis

Clinical Type	Differential Diagnosis
Aural	Otitis externa, Suppurative otitis media, Eustachian catarrh, Foreign body
Furuncular	Delusional parasitosis, Furuncle (Boils), Cellulitis, Insect bite allergy, Infected sebaceous cyst, Tungiasis, Pyoderma, Herpes, Hemangioma, Arteriovenous malformation, Prickly heat (Miliaria)
Migratory	Cutaneous larva migrans, Gnathostomiasis, Sparganosis, Hyper-eosinophilic Syndrome
Nasal	Sinusitis, Upper respiratory tract infection, Allergic rhinitis, Foreign bodies of nose, Atrophic rhinitis, Rhinoscleroma, Leprosy, Tuberculosis
Ocular	Retinal Detachment, Uveitis, Cavernous sinus thrombosis, Chorio-retinitis, Endophthalmitis
Orbital	Foreign body, Conjunctivitis, Keratitis, Peri-orbital cellulitis, Uveitis, Chalazion, Benign floaters
Oropharyngeal	Pharyngitis, Salivary adenoma, Diphtheria, Peritonsillar abscess, Agranulocytosis, Dental abscess
Penile	Balanitis, Urinary Tract Infection, Urethritis
Pulmonary	Branchial Asthma, Eosinophilia Syndrome, Pulmonary larva migrans
Rectal	Fissure-in-Ano, Rectal Polyp, Intussusception, Enterobiasis
Scrotal	Torsion of testis, Idiopathic scrotal edema
Umbilical	Omphalitis, Patent vitello-intestinal duct
Vaginal	Foreign body, Vulvo-vagnitis

INVESTIGATION

Maggots in deep burrows can be easily seen using a magnifying lens or dermascope. Bright light should be avoided during physical examination. Subcutaneous maggots can be made to wriggle and their movement can be observed by smearing the affected area with liquid paraffin. The mineral oil not only blocks the ventilatory punctum and causes the larvae to writhe in suffocation, but also makes observation of their movements easier by reflecting the light.

Endoscopes such as bronchoscope, nasopharyngoscope, otoscope, sigmoidoscope and cystoscope are useful not only in diagnosis but also in therapeutic removal of maggots. Ophthalmomyiasis necessitates slit lamp examination. However, it is essential to paralyze the maggots with topical anesthetics (lignocaine or cocaine), lest the photophobic worms migrate deeper inside the tissues on examination with a bright light.

Maggots hiding in deeper tissues can be visualized by ultrasonography using a high frequency (10-18 MHz) probe.[100] The larvae are seen as echogenic spindle-shaped structures within hypoechoic fluid filled burrow and pulsatile blood vessels in the periphery (the Bouer's criteria).[100] The thick cuticle and spines of maggots cast distal acoustic shadow.[100] In longitudinal sections of the worms, undulations of the body segmentation may be appreciated. Doppler ultrasonography is 100% sensitive in demonstrating the wriggling movements of maggots.[54,100] Computed tomographic (CT) scan and magnetic reesonance imaging (MRI) are not more useful than Doppler ultrasound in cutaneous myiasis. However, they together with plain x-rays are useful in pulmonary, mastoid, cerebral or bone myiasis.

Hematological investigations are suggestive but not specific of myiasis. For example, *Dermatobiasis* which mimics an abscess will cause eosinophilia rather than neutrophilic leukocytosis.[2]

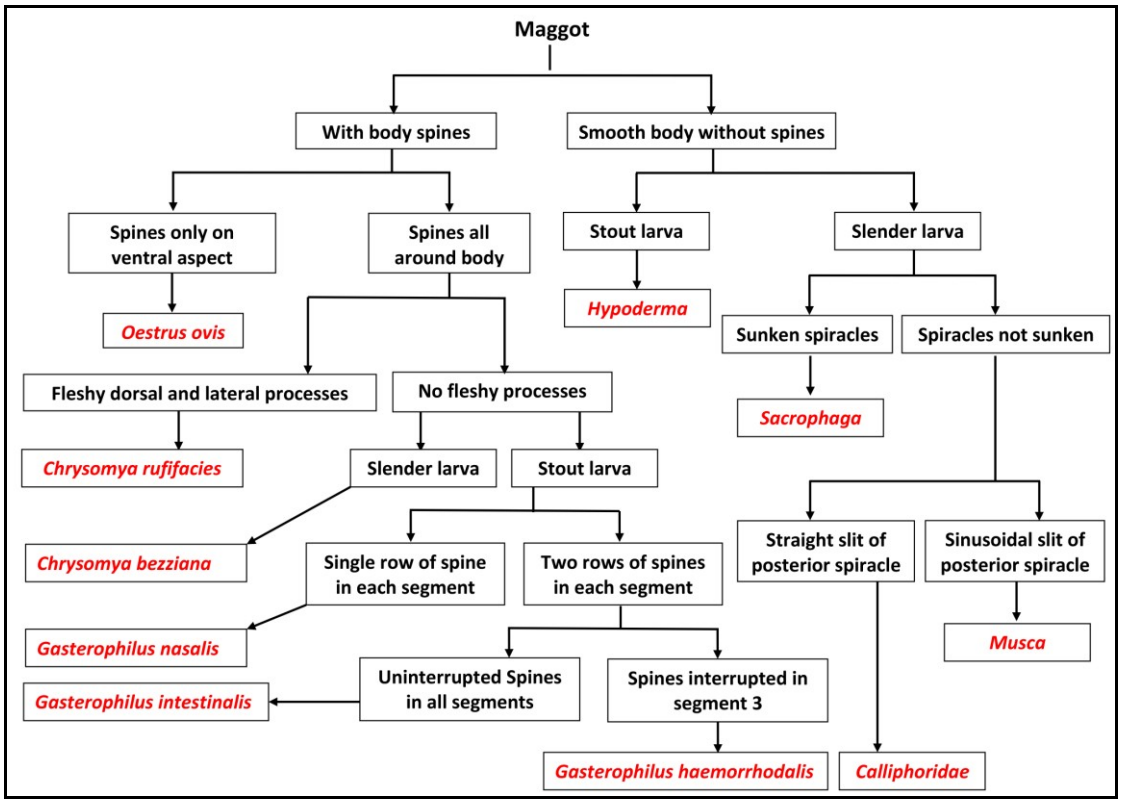


Fig 6. *Dichotomous, maggot identification key. (Based on the data from the School of Veterinary Sciences, University of Queensland) https://shire.science.uq.edu.au/bb/parasitology/maggots/mag got.html*

High levels of IgE, elevated C-reactive protein and increased erythrocyte sedimentation rate may be present.

Attempted development of serodiagnosis has not been successful because of antigenic cross reactivity between various species.[2] However, the exception is *Hypoderma*. It periodically releases hypodermin C, an antigen which can be diagnosed by enzyme-linked-immunosorbent serologic assay (ELISA) technique.[2] Polymerase chain reaction (PCR) aided testing of cytochrome oxidase-I (COI) of mitochondrial DNA is also used in the diagnosis of *Hypoderma*. Indirect haemagglutination test and double immuno-fluorescence test with crude antigenic extracts can detect the first and second instar larvae. But these techniques are less sensitive for third instar larvae. An intradermal injection test is used to diagnose *Gasterophilus* infestations in animals.[2,3]

Fine-needle aspiration cytology (FNAC) and tissue biopsy are usually not indicated. However, Pogany demonstrated that an unspecified species of larva could be aspirated from a neck mass using a 23G needle.[101] Obviously maggots with sharp body spines, such as the *Dermatobia,* can not be aspirated. Needle puncturing of maggots may leave them killed in deep burrows making their removal difficult and may cause leaking of toxic body fluids provoking anaphylaxis. Hence, FNAC cannot be recommended for routine clinical use.

Exact species identification of maggots extracted from lesions is not essential for the clinical management of myiasis. Further, required entomological expertise is not readily available in many resource limited centers. For these reasons, most of the reports on myiasis do not mention the infesting species. However, academically it is desirable to establish the taxonomic identity of the

parasite. Maggots are identified by their external morphology. (Fig. 3) Third instars are easier to identify than the first two instars which lack the characteristic morphological features. Hence, preserving the retrieved maggots without any structural alteration is essential. Formalin is not an ideal preservative as it causes shrinkage and hardening artifacts. The extracted worms are transported to entomology lab after being killed in hot (but not boiling) water and preserved in 70% alcohol. When the services of a trained entomologist is not available, the dichotomous Maggot Identification Key available online at the website of the University of Queensland[102] can be of great help to practicing pediatric surgeons. (Fig. 6)

As identification of adult flies is easier than that of the larval forms, some entomologists recommend maggot culturing on raw meat or Robertson cooked meat medium allowing them to mature in the laboratory.[85] More recently, molecular diagnostics are applied for accurate identification of the maggot species.[3]

TREATMENT

When left untreated, myiasis may spontaneously resolve when the larvae mature into pupae and leave the host. Further, maggots are also therapeutically used in healing chronic ulcers (*vide infra*). Therefore, one may wonder if it is essential to treat naturally occurring WM as a disease. It is to be emphasized that all naturally occurring maggots should be removed from wounds because of the uncertainty of the species involved and because of the risk of superadded bacterial infection and tissue damage.[103] It is to be remembered that not all maggots are beneficial.[104] Some of the obligate parasites may cause extensive tissue damage, hemorrhage and death of the host.[84,51,52] Wild maggots may also transmit infections like tetanus, gas gangrene and synergistic necrotizing fasciitis.[104] Therefore, all clinical myiasis should be treated as a disease. [103]

Manual (Mechanical) Removal

The best method of removing maggots is gentle manual picking with forceps.[105,106] Alternatively gargling, syringing, rinsing, sternutatories, purgatives (polyethylene glycol) and mouth washes can be appropriately used in removing surface larvae. Removal of worms from the deep dermis is often aided by squeezing with a wooden spatula. Optical magnification (e.g. handheld lens, dermascope or operating microscope) is helpful, especially in cavity myiasis. But, photophobic larvae frequently hide in deep burrows and are not easily accessible. Usually multiple session are required for complete removal of all the worms. Maggots may also resist removal by firmly anchoring to the host tissues by using oral hooklets and body spines. Forced extraction results in rupturing of maggots and incomplete removal. Left over dead maggots may cause anaphylatic shock or foreign body reaction. *Dermatobia* is very resistant to manual extraction due to innumerable stiff body spines.[107] Their attempted removal is not only painful but also unsuccessful with the risk of rupturing the larva. Tiny cruciate incisions are often used to remove them furuncle.

Several chemical agents have been used either to to facilitate manual removal of maggots or to kill them. They include asphyxiants, vermifuges, baits, larvicides and paralyzers.

Asphyxiants

Maggots are highly photophobic that burry their head-end deep inside the tissues. However, they keep their rear-end spiracles exposed for breathing. Blocking their atmospheric access make them suffocated and cause them move out of burrows seeking fresh air.[2] This property of maggots is exploited in treating them. Liquid paraffin is the ideal agent as it not only asphyxiates the worms but also lubricates their removal from burrows. Alternatively, covering the area with an adhesive tape or ointments (e.g. Polymyxin B) can be used. Occlusion of the punctum for 24 hr may be

required to achieve the desired effect. It is important to avoid tight occlusion which may kill the worms instead of making them just uncomfortable. A dead worm can never be extracted from arbores-cent burrows. Intense inflammatory reaction evoked by a dead worm may actually be more troublesome than the presence of live worms.

Vermifuges

Mild chemical irritants are ideal as they cause maggots to move out of burrows. Turpentine oil is the oldest known vermifuge which is still being used in modern medicine. It was first recommended in 1870 by Center who experimented with 20 different chemicals.[18] Calomel is said to be more effective and less painful than turpentine as they neutralize ASEM in addition to causing worm irritation.[19] Natives of Bengal use extract of the plant *Ocimum sanctum* (Holy Basil or Tulasi).[19]

Paralyzers

Drugs like lignocaine, 15% chloroform in olive oil and ether temporarily paralyze the maggots and facilitate their manual extraction.[107] Lignocaine has the added advantage of reducing pain to the host. Although these agents do not kill the worms, the stupefied maggots may remain in deep tunnels thus become inaccessible for manual picking. Liquid nitrogen may stiffen the larvae facilitating their easy extraction.

Laser Destruction

Although manual removal of wild maggots is always desirable, the only exception is ocular myiasis. Subretinal and vitreous maggots are often killed with laser and left in situ without any active efforts to remove them. This is supplemented with drugs to suppress inflammation (topical steroids and mydriatics). Only those maggots floating in the visual axis require surgical removal. Rarely affected eye may need to be enucleated to prevent sympathetic ophthalmia and intracranial extension of the infestation.

Larvicides

Larvicidal chemicals are generally not preferred for the following reasons: (1) They leave behind dead maggots in deep burrows provoking strong inflammatory reaction; (2) Sudden release of large amount of toxins from the killed maggots may precipitate anaphylactic shock; (3) The chemical agents, when absorbed into the host circulation, may cause systemic toxicity; (4) They pollute the environment; (5) Insects are known to develop resistance to pesticides. However, larvicides are occasionally used in aggressive maggots such as *Dermatobia*, *Wohlfahrtia, Cochliomyia*, *Chrysomya* and *Oestrus ovis*. They are also indicated when the maggots are wandering inaccessibly in deeper tissues or when their removal is deemed to cause more damage to the host tissues.

Several pesticides including organo-phosphorus compound have been successfully used in animals and human adults. (Table 7) However, their safety and efficacy in children are not yet well stablished. Among the slow-acting larvicides, ivermectin has been extensively studied.[2,108-111] It causes release of gamma-amino-butyric acid which is toxic to maggots. Ivermectin is used as topical paste (1% solution in propylene glycol applied for 2 hr), oral tablet (15 μg/kg per day; maximum 400μg/kg) or as subcutaneous local injection around the lesion. Topical ivermectin has the risk of converting WM into migratory myiasis by repelling the maggots into deeper tissues. Oral ivermectin is ideal for migratory myiasis although they have the disadvantage of leaving behind the dead larvae in the host tissues. Rarely, spontaneous external emigration of worms with oral ivermectin has been reported. At the oral dosage of 200 μg/kg (single dose) ivermectin kills 99% of WM.[2] Topical trichlorphon kills larvae within 28-52 hr of application while ivermectin does so at 50-64 hr.[2] Mesalazine, albendazole, mebendazole, and levamizole are other alternatives to oral ivermectin. They are specifically used in intestinal myiasis. In a case of rectal myiasis due to *Fannia canicularis* levamizole

Table 7. Drugs and chemical used in the treatment of maggots

Rapid Larvicides (mostly of topical use)[2]
- Chlorfenvinphos*
- Chlorpyrifos*
- Closantel * (oral, topical, IM)[2]
- Coumaphos*
- Crotoxyphos *
- Crufomate *
- Cyromazine*
- DDT *
- Decamethrine *
- Diazinon*
- Dichlofenthion*
- Dichlorvas*
- Fenchlorphos*
- Fenthion *
- gamma-BHC*
- iodofenphos*
- Lufenuron *
- Milbemycin*
- Moxidectin *
- Phenylbutazone
- Phosalone*
- Propoxur*
- Rafoxanide * (oral or IV Use) [2]
- Temephos *
- Tetrachloroethylene (oral)[85]
- Thymol (enema)[85]
- Toxaphene (Camphechlor)*
- Trichlorphon * (topical)

Slow larvicides (systemic or topical use)
- Abamectin* (subcutaneous)
- Albendazole †
- Avermectin*
- Clindamycin †
- Decamethrin*
- Doramectin* (subcutaneous)
- Ivermectin † (oral, topical, subcutaneous, IV)
- Levamizole
- Milbemycin*
- Moxidectin*

(Continued)

Vermifuges (Topical use)
- Carbolic acid[18]
- Hydrogen peroxide
- Mercurials [18]
- Phenol
- Potassium permanganate
- Povidone iodine
- Sodium hypochlorite
- Tobacco juice [18]
- Turpentine oil [18]

Paralyzing Agents (Topical use)
- Chloroform[31]
- Ether
- Ethyl chloride
- Lignocaine
- Liquid nitrogen

Worm asphyxiants (Topical use)
- Butter
- Chewing gum[54]
- Dipping affected part in water
- Hydrocolloid dressing[54]
- Nail polish[54]
- Olive oil
- Paraffin wax or oil
- Pine oil
- Resins or glues[54]
- Sealing with cello tapes
- Vaseline (petrolatum jelly)

Baits
- Bacon
- Lard[54]

Agents of Unknown mechanism
- Calomel
- Mesalazine[90]
- Nitrofurazone 0.2% (Topical)
- Polymyxin B (Topical)
- Warm water immersion[54]

Compiled from: Hall,[2] Cedello,[54] Center[18]
** These agents are extrapolated from veterinary and adult practices. Pediatric experience with them is almost non-existent. Their safety in children is yet to be studied.*
† The 3 are given together as triple therapy
BHC - Benzene hexachloride, DDT - Dichloro-diphenyl-trichloro-ethane

was useful when albendazole was ineffective.[90] A triple therapy of oral ivermectin (15 mg/kg/d for 3 days) followed by clindamycin (300 mg t.i.d for 5 days) and albendazole (400 mg b.i.d for 3 days) has been used successfully.[48,111] However, it must be emphasized that pediatric experience with these larvicidal agents is very limited.

Baits

Bacon therapy, a form of baiting, is known to ancient Indians (*vide supra*). In 1993, Brewer et al from Massachusetts General Hospital 'rediscovered' it [14,17] without any reference to Sushruta who described exactly the same technique 3000 years ago. Within 3 hr of applying raw bacon fat the worms attracted by the smell will come out which then can be manually removed.[112] On careful analysis of the published descriptions, the modern bacon therapy appears to act more like an asphyxiant rather than as a bait.

Surgical debridement

Flies are principally attracted towards necrotic wounds. Hence, it is logical to do complete surgical debridement both as therapy and prophylaxis. However, this is inappropriate if vital organs are involved.

Antibiotics

Bacterial infections not only attract gravid flies but also can occur as a complication of myiasis. Therefore, concomitant administration of broad spectrum antibiotics and tetanus prophylaxis are indicated in all patients with myiasis.[113]

COMPLICATION

It is not clear as to whether all the complications of myiasis are attributable to the larvae or to the predisposing necrotic wounds.[53] For example, tetanus is a well known complication of myiasis; but it may just be due to the original trauma. Complications of myiasis may be general or site and species specific. There are some evidences to say that complications are becoming infrequent with modern treatment. For example, palatal perforation in nasal myiasis fell from 12% in 1980 to 2.5% in 2004.[27] Site specific complications are already described above in respective subsections.

Maggots transmit several helminthic, viral and bacterial diseases. Eggs of *Ascaris, Enterobius* and *Trichuris* remain undigested in the gut of *Lucilia sericata*.[114] Diseases like typhoid, pasturella, kala azar, shigella, anthrax and polio are known to be transmitted by maggots.[114]

Prions are infectious protein particles that cause serious neurodegenerative diseases like spongiform encephalopathy, Creutzfeldt–Jakob disease and Kuru. It is suspected that maggots may be vectors and reservoirs for horizontal transmission of prions.[115] In fact, prion has been isolated from *Sarcophaga carnaria* maggots and adult *Drosophila melanogaster*.[115] Prion proteins replicate inside the maggots' nervous system using the larval DNA. It remains to be confirmed if children with myiasis develop neurodegenerative diseases in adult life.

PREVENTION

Prophylaxis of wound myiasis includes eradication of flies and preventing their access to wounds. Spraying insecticides and proper garbage disposal will largely control breeding of flies. Pine oil repels gravid flies.[2] Biological larvicides such as *Tolypocladium niveum* (a fungus), *Bacillus thuringiensis* (a bacteria) and *Macrocheles muscaedomestica* (a mite) are used to kill maggots in garbage.[2] Genetic control by the laboratory bred sterile male flies is shown to be effective in reducing fly-population.

Children should be prohibited from sleeping out door and playing nude without undergarments. Contact with farm animals should be restricted and monitored. Drying clothes in bright sunlight and ironing them with hot press are effective against *Cordylobia*.[75]

Anti-maggot vaccines were tried with great enthusiasm. Maggots are too large to be affected by the host phagocytes. But, host antibodies can kill them by binding the peritrophic membrane in the larval gut, when they feed on the host serum.[2] However, the attempts of vaccine development were not successful due to antigenic cross reactivity.[2] The third stage instars also develop defense mechanism against the host immune cells and antibodies. Recently, Hypodermin A&B the digestive enzymes of *Hypoderma* are found to be suitable target antigens for vaccine production.

Livingston topically applied a jelly made of ground maggots to prevent myiasis. Its effectiveness is attributed to the ASEM present in the paste. But, this was soon discarded as the paste evoked more allergic reactions and inflammation.

PROGNOSIS

Recurrent infestation is reported in 15% of nasal myiasis and it increases to 80% when associated with suppurative rhinitis.[19] About 1% of nasal myiasis,[4] 8% of otomyiasis,[64] and 5% of ophthal momyiasis[68] cause death by meningitis. Cerebral myiasis has the highest mortality of 50%.[80] Death may not be exclusively attributed to the presence of maggots.[53] Rather, the original suppurative condition that predisposes to myiasis and secondary bacterial infection should also be accounted. Interestingly, maggot infested wounds are less often associated with systemic sepsis, a phenomenon attributed to bacteriostatic properties of the ASEM.

MAGGOT THERAPY

Maggot therapy (MT, also known as larval therapy or biosurgery) is a form of iatrogenic WM.[38,116-118] Among the several species of maggots (Table 8), *Lucilia sericata* has been extensively studied as a therapeutic agent.[24] Interestingly, it is a serious pest causing fatality in farm animals.[2] Maggots are said to promote the healing of chronic wounds by 3 different mechanisms: (1) Enhancing wound debridement, (2) Microbial disinfection and (3) Promoting cellular proliferation. Each of these goals is achieved by several physical and chemical properties of maggots.

Table 8. Commonly used medicinal maggots

Calliphora vicina
Chrysomya rufifacies
Lucilia caesar
Lucilia cuprina
Lucilia illustris
Lucilia sericata *
Musca domestica
Phormia regina
Protophormia terraenovae
Wohlfahrtia nuba †

** Most commonly used species*
† When necrotic tissues are exhausted, they notoriously start feeding on healthy tissue

Wound Debridement

Medicinal maggots (MM) with body spines, while crawling, scrub the wound that loosens necrotic debris and breaks biofilm (a layer of coagulum under which bacteria thrive protected from the attacks of immune cells and antibiotics). By breaking biofilms, MM improve antibiotic penetration. ASEM contain proteolytic enzymes such as the matrix metallo-proteinase(MMP), trypsin-like and chymotrypsin-like serine protease, leucin-amino-peptidase, carboxy-peptidase and collagenase that are resistant to human wound protease inhibitors. These digestive enzymes liquefy necrotic tissue and biofilms which are subsequently consumed by the MM. Each maggot is estimated to eat up 25-30 mg of necrotic debris in 24 hr.[24, 38] Being a facultative maggot *Lucilia* does not cause any damage to the viable host-tissue. MM do not affect the functioning of host immune cells or inflammatory repairs.

Infection Control

The natural habitat of maggots is decaying matters and feces. Hence, they are endowed with natural protection against bacteria. This is attributed to the antibacterial chemicals in ASEM such as the urea, calcium carbonate, ammonium bicarbonate, lucifensin, diptericin, allantoin, calcium picrate, picric acid, phenyl-acetaldehyde and phenyl-acetic acid. Deoxy-ribonuclease (DNAse) present in ASEM not only facilitates breaking of biofilms, but also inhibits bacterial proliferating. Aelloferons in ASEM are capable of inhibiting even viruses and protozoa like *Leishmania*. ASEM also alkalinizes the wound pH and make it hostile for bacteria. By all these mechanisms, MM significantly reduce the microbial population of the wound.

Epithelial Proliferation

Physical crawling of larvae stimulates the electrical potential of the repairing cells and releases epithelial growth factors. Chemicals in ASEM such as the cysteine, glutathione, sulphydryl radicals and haemolymph are shown to stimulate the growth of fibroblasts, endothelial cells, angiogenic cells and neural cells. They also promote fibroblast migration, vascular perfusion, tissue oxygenation and phagocyte maturation. ASEM inhibits generation of superoxide and release of myeloperoxidase from activated neutrophils, thereby down-regulates inflammation. This is further enhanced by increased synthesis of cyclic AMP in neutrophils. Although MM subdue host inflammation, they do not affect phagocytosis or apoptosis. Cytokines like IFNγ and IL-10 in ASEM promote healing. MMP in the ASEM has positive influence over hemostasis, thrombosis, keratinocyte migration, collagen degradation and tissue remodeling. They also increase epithelial growth factor and interleukin-6-stimulated fibroblasts. ASEM causes macrophages and neutrophils to switch from their original pro-inflammatory role to pro-angiogenic function. ASEM inhibits complement activation and breaks down C3 and C4 proteins.[38]

Techniques of Maggot Application

The number of MM required for therapy varies between 5 and 600 according to the size of the wound.[117,118] There are two different methods of applying MM to the wound: In *free-range therapy* they are directly applied to crawl on the wound. However, this is not well received as children are terrified by creeping worms. In *biobag method* MM are not directly applied to the wound. Rather they are kept in nylon bags with tiny pores of 100-400 micron size. These biobags, when applied to the wound, allow seepage of ASEM, thus providing the benefits of chemical action.[119] Several studies have shown that biobag method is less effective than free-range therapy as it deprives the benefits of physical action of maggots.

Demerits of Maggot Therapy

Anxiety and repulsion of patients, foul smell of the ASEM and aesthetic unacceptability are the major demerits of MT. Further, the duration of treatment is very prolonged, that two cycles of 48-72 hr therapy sessions per week for 3-4 weeks is needed for complete wound debridement. The beneficial effects of maggots are short lived, that they need to be applied as 'maintenance therapy' even after achieving complete debridement until the wound heals completely. Some of the randomized controlled trials have questioned the very benefits of MT.[120]

Although MT inhibit *Pseudomonas* and Gram positive organisms (*Staphylococcus aureus* and Group A&B *Streptococcus*), they actually increase the population of gram negative bacteria (*Escherichia coli* and *Proteus*). This is probably due to the reduced competition from Gram positive bacteria. MM in fact develop symbiosis with *Proteus* in their gut to facilitate synthesis of bacteriostatic chemicals in ASEM.

MT is contraindicated near body cavities, orifices and fistulae where physical removal of maggots at the completion of therapy will be difficult. Other

contraindications of MT include life endangering or limb threatening infections, acute gangrene, and wounds close to great vessels.[38] Ammonia present in ASEM may get absorbed and cause toxicity especially in children with liver impairment.

Pediatric Experience with Maggot Therapy

Although extensively studied in adults with leg ulcers, MT is very rarely used in children.[121-123] In 1931, William Baer used wild maggots to treat more than 100 children with osteomyelitis and soft tissue wounds.[12,23] Subsequently the enthusiasm with MT was dampened by the availability of broad-spectrum antibiotics and by the entomophobia of children. Recently, few case reports have appeared in the pediatric literature.[121-123]

EPILOGUE

Although there are several randomized controlled trials (RCT) on 'maggot therapy' there are none on the 'therapy of maggots'. It is difficult to design an RCT because of the heterogeneity of the involved larval species. Most of our understanding about myiasis is derived from veterinary practice. Global warming, climate change and international travels are expected to change the clinical pattern and incidence of myiasis. Growing resistance of flies to insecticides may, in future, significantly affect the treatment and its outcome.[3]

REFERENCES

[1] Zumpt F. Myiasis in man and animals in the Old World. Butterworths, London, 1965.

[2] Hall M, Wall R. Myiasis of humans and domestic animals. Adv Parasitol 1995; 35: 257-334.

[3] Hall MJ, Wall RL, Stevens JR. Traumatic Myiasis: A neglected disease in a changing world. Annu Rev Entomol. 2016; 61: 159-76.

[4] Francesconi F, Lupi O. Myiasis. Clin Microbiol Rev. 2012 Jan; 25(1): 79-105.

[5] Caumes E, Carrière J, Guermonprez G, Bricaire F, Danis M, Gentilini M. Dermatoses associated with travel to tropical countries: a prospective study of the diagnosis and management of 269 patients presenting to a tropical disease unit. Clin Infect Dis. 1995 Mar; 20(3): 542-8.

[6] Diaz JH. The global epidemiology, public health outcomes, management, and prevention of reemerging ectoparasitic diseases. *Trop Med Health 2008;* 36(1): 1-10

[7] Pechter EA, Sherman RA. Maggot therapy: the surgical metamorphosis. Plast Reconstr Surg. 1983 Oct; 72(4): 567-70.

[8] Sherman RA, Hall MJ, Thomas S. Medicinal maggots: an ancient remedy for some contemporary afflictions. Annu Rev Entomol. 2000; 45: 55-81.

[9] Collier R. Medicinal maggots cross border at a crawl. CMAJ. 2010 Feb 9; 182(2): E123-4.

[10] Hall M, Donovan S. Forensic entomology: what can maggots tell us about murders? Biologist (London). 2001 Dec; 48(6): 249-53.

[11] Greenberg B. Flies through history. In: Greenberg B (ed). Flies and Disease - Vol 2. Princeton University Press, New Jersy, 1973. pp 2-18.

[12] Whitaker IS, Twine C, Whitaker MJ, Welck M, Brown CS, Shandall A. Larval therapy from antiquity to the present day: Mechanisms of action, clinical applications and future potential. Postgrad Med J. 2007 Jun; 83(980): 409-13.

[13] Parke EC. Flies from meat and wasps from trees: Re-evaluating Francesco Redi's spontaneous generation experiments. Stud Hist Philos Biol Biomed Sci. 2014 Mar; 45: 34-42.

[14] Bhishagratna KKL. An English translation of the Sushruta Samhita. Vol-3 - Uttara-Tantra. S.L.Bhaduri, Calcutta. 1916. pp 108, 115, 132, 137.

[15] Aswani KV, Abdul Shukkoor MM, Rajasree G. Extraction of *Krimi* (maggots) in *Dushtavrana* (non-healing ulcer) by *Susrutha's* concept of *Mamsaachadana:* A rare case report. Int J Ayush CaRe. 2021 Oct-Dec; 5(4):247-252.

[16] Deb D, Das K. Maggot therapy or *Krimi Utpattikara Chikitsa*: A review of history and clinical applications. Int Ayurvedic Med J (online). March 2023. DOI: 10.46607/iamj09p7032023

[17] Brewer TF, Wilson ME, Gonzalez E, Felsenstein D. Bacon therapy and furuncular myiasis. JAMA. 1993 Nov 3; 270 (17): 2087-8.

[18] Center W. A case of Peenash. Ind Med Gaz. 1870 Feb 1; 5(2):38-39.

[19] Bosmia AN, Zimmermann TM, Griessenauer CJ, Shane Tubbs R, Rosenthal EL. Nasal myiasis in Hinduism and contemporary otorhinolaryngology. J Relig Health. 2017 Aug; 56(4): 1263-1281.

[20] Mazzarello P. Achilles and the maggots. Nature.1999 Nov 18; 402(6759): 237.

[21] Agnelli S, King RB. Aural myiasis in ancient Rome: Celsus and the ear maggots. J Laryngol Otol. 2023 Dec; 137(12): 1345-1348.

[22] Manring MM, Calhoun JH. Biographical sketch: William S. Baer (1872-1931). Clin Orthop Relat Res. 2011 Apr; 469(4): 917-9.

[23] Baer WS. The treatment of chronic osteomyelitis with the maggot (larva of the blow fly). J Bone Joint Surg. 1931 July; 13(3): 438-475. {Reprinted: Clin Orthop Relat Res. 2011 Apr; 469(4): 920-44.}

[24] Sherman RA. Mechanisms of maggot-induced wound healing: what do we know, and where do we go from here? Evid Based Complement Alternat Med. 2014; 2014: 592419.

[25] Shamloul G, Khachemoune A. Reappraisal and updated review of maggot debridement therapy in chronic lower extremity ulcers. Int J Dermatol. 2023 Jul; 62(7): 962-968.

[26] Syam K, Joiya SA, Khan S, Unnikrishnan PN. Maggot debridement therapy for chronic leg and foot ulcers: A review of randomized controlled trials. Adv Skin Wound Care. 2021 Nov 1; 34(11): 603-607.

[27] Arora S, Sharma JK, Pippal SK, Sethi Y, Yadav A. Clinical etiology of myiasis in ENT: a reterograde period-interval study. Braz J Otorhinolaryngol. 2009 May-Jun; 75(3): 356-61.

[28] Hope FW. On insects and their larvae occasionally found in the human body. Trans Entomolo Soc London 1837; 2: 256-271.

[29] McGraw TA, Turiansky GW. Cutaneous myiasis. J Am Acad Dermatol. 2008 Jun; 58(6): 907-29.

[30] Sherman R. Putting wild maggots on your head is not "maggot therapy," but it does suggest pre-existing pathology. Plast Reconstr Surg. 2007 Nov; 120(6):1737-1738.

[31] Doss B. Maggots in nose or so-called Vermes Nasi. Ind Med Gaz. 1874 Apr 1; 9(4): 96.

[32] Hunter JM. Bot-fly maggot infestation in Latin America. Geogr Rev. 1990 Oct; 80(4): 382-398.

[33] James MT. The flies that cause myiasis in man. US Dept of Agriculture. Pub.no. 631. Washington. 1947. pp 1–175.

[34] Bishopp FC. Flies which cause myiasis in man and animals: Some aspects of the problem. J Economic Entomol 1915 June; 8(3), 317-329.

[35] Patton WS. Notes on the myiasis-producing Diptera of man and animals. *Bull Entomol Res*. 1921; 12(3): 239-261.

[36] Chan JC, Lee JS, Dai DL, Woo J. Unusual cases of human myiasis due to Old World screw-worm fly acquired indoors in Hong Kong. Trans R Soc Trop Med Hyg. 2005 Dec; 99(12):914-8.

[37] Sarwar M. Typical flies: Natural history, lifestyle and diversity of Diptera. In: Sarwar M (ed.) Life cycle and development of Diptera. Intech Open 2020. DOI: 10.5772/intechopen.91391 {accessed on 10 Sep 2024}

[38] Wollina U, Karte K, Herold C, Looks A. Biosurgery in wound healing - the renaissance of maggot therapy. J Eur Acad Dermatol Venereol. 2000 Jul; 14(4): 285-9.

[39] Rodrigues FT, D'Acri AM, Lessa CSS, Aguiar VM. Profile of pediatric patients with myiasis treated at a tertiary hospital in Rio de Janeiro. An Bras Dermatol. 2021 May-Jun; 96(3):369-372.

[40] Azarmi S, Akbarzadeh K, Ekrami A, Sheikh Z, Dehghan O. Scalp myiasis associated with soft tissue sarcoma lesion: a case report and review of relevant literature. BMC Infect Dis. 2024 Jan 5; 24(1): 51.

[41] Bapat SS. Neonatal myiasis. Pediatrics. 2000 Jul; 106(1): E6.

[42] Lindquist DA, Abusowa M, Hall MJ. The New World screw worm fly in Libya: a review of its introduction and eradication. Med Vet Entomol. 1992 Jan; 6(1):2-8.

[43] Gabaj MM, Beesley WN. American screwworm fly in Libya. Vet Rec. 1989 Feb 11; 124(6):152.

[44] Beckett SD, Spradbery JP, Green PE, Urech R and James P. Old World screw-worm fly: Risk of entry into Australia and surveillance requirements. A report for Animal Health Australia, Canberra (November, 2014)

[45] Singh A, Singh Z. Incidence of myiasis among humans-a review. Parasitol Res. 2015 Sep;114(9):3183-99.

[46] Seif AI, Kholeif HA, Koura EA, El-Alfi NM. Experience with traumatic myiasis among patients with septic wounds. J Trop Med. 1992 Dec; 2 (1): 115–19.

[47] Singh I, Gathwala G, Yadav SP, Wig U, Jakhar KK. Myiasis in children: the Indian perspective. Int J Pediatr Otorhino laryngol. 1993 Jan; 25(1-3): 127-31.

[48] Jain A, Taneja S. Oral myiasis affecting pediatric patients: a systematic review. J Stomatol Oral Maxillofac Surg. 2022 Jun; 123(3): e32-e36.

[49] Jain S, Audhya A; Madhupriya; Nagpure PS. Aural myiasis in a 1-day-old neonate. Indian J Med Sci. 2008 Apr; 62 (4): 164-6.

[50] Goldman L. Pyodermic myiasis in children: observations with special reference to torsalo. Am J Dis Child. 1945 May; 69(5): 280-282.

[51] Bozorgmehr R, Rezaei MM, Jafarabadi K, Alibeik N, Rahi mian N, Ahmadinejad M. A rare case of carotid artery injury during maggot debridement therapy for locally advanced parotid gland carcinoma. Int J Surg Case Rep. 2023 Oct; 111: 108844.

[52] Steenvoorde P, Oskam J. Bleeding complications in patients treated with maggot debridement therapy. Int J Low Extrem Wounds. 2005 Mar; 4(1): 57-8.

[53] Sherman RA, Bjork R. Association does not mean causation, especially when it concerns maggots. Wounds. 2018 Dec; 30(12):382.

[54] Cedillo MRQ, Leon-Urena H, Contreras-Ruiz J, Arenas R. The value of Doppler ultrasound in diagnosis in 25 cases of furunculoid myiasis. Int J Dermatol. 2005 Jan; 44(1): 34-7.

[55] Al-Juaid A, Al-Zahrani W. Furuncular Myiasis in a Child: A Case Report and Literature Review. Saudi J Med Med Sci. 2017 Jan-Apr;5(1):77-79.

[56] Boruk M, Rosenfeld RM, Alexis R. Human botfly infestation presenting as peri-auricular mass. Int J Pediatr Oto rhinolaryngol. 2006 Feb; 70(2): 335-8.
[57] Fernandes LF, Pimenta FC, Fernandes FF. First report of human myiasis in GoiaS state, Brazil: frequency of different types of myiasis, their various etiological agents, and associated factors. J Parasitol. 2009 Feb; 95 (1): 32-8.
[58] Sharma SB, Gupta R. Newborn umbilical cord stump myiasis: Report of an earliest presentation. J Indian Assoc Pediatr Surg. 2023 Jan-Feb; 28(1): 86-87.
[59] Barolia DK, Singh AP, Tanger R, Gupta AK. Umbilical myiasis in a human neonate treated with turpentine oil. J NTR Univ Health Sci 2020 Apr-June; 9 (2):143-5.
[60] Ogbalu OK, Eze CN, Manuelrb B. A new trend of omphalitis complicated with myiasis in neonates of the Niger Delta, Nigeria. Epidemiology (Sunnyvale) 2016; 6(2): 231. doi:10.4172/2161-1165.1000231
[61] Ogbalu OK, Achufusi TD, Orlu EE, Bawo DS, Adibe CH, Kumbe L, Azuonwu O, Amadi E. Human myiasis in neonates and children of the Niger Delta wetlands and South East Nigeria. J Cosmetic Dermatol Sci Appl. 2011 Dec; 1: 171-176. {DOI:10.4236/jcdsa.2011.14026}
[62] Ruiz-Zapata JD, Figueroa-Gutiérrez LM, Mesa-Franco JA, Moreno-Gutierrez PA. Umbilical myiasis by *Cochliomyia hominivorax* in an infant in Colombia. Front Med. 2020 Jan 22; 6: 292.
[63] Sharma H, Dayal D, Agrawal SP. Nasal myiasis: review of 10 years experience. J Laryngol Otol. 1989 May; 103(5): 489-91.
[64] Yuca K, Caksen H, Sakin YF, Yuca SA, Kiriş M, Yilmaz H, Cankaya H. Aural myiasis in children and literature review. Tohoku J Exp Med. 2005 Jun; 206(2): 125-30.
[65] Ogbalu OK, Achufusi TG, Adibe C. Incidence of multiple myiases in breasts of rural women and oral infection in infants from the human warble fly larvae in the humid Tropic-Niger Delta. Int J Dermatol. 2006 Sep; 45(9): 1069-70.
[66] Kamboj M, Mahajan S, Boaz K. Oral myiasis misinterpreted as salivary gland adenoma. J Clin Pathol. 2007 Jul; 60(7): 848.
[67] Huang YL, Liu L, Liang H, He J, Chen J, Liang QW, Jiang ZY, He JF, Huang ML, Du Y. Orbital myiasis: A case report and literature review. Medicine (Baltimore). 2020 Jan; 99(4): e18879.
[68] Blejter J. Tracheostomy wound myiasis in a child: case report and review of the literature. Case Rep Pediatr. 2012; 2012: 317862.
[69] Demir AD, Iraz M, Ipek DNS. Urogenital myiasis caused by *Psychoda albipennis* in a child. Turk Ped Arch 2015; 50: 65-8
[70] Kataria U, Siwach S, Gupta S. Myiasis in female external genitalia. Indian J Sex Transm Dis AIDS. 2013 Jul; 34(2): 129-31.
[71] Singh A, Kaur J. Occurrence of human urogenital myiasis due to neglected personal hygiene: a review. Trans R Soc Trop Med Hyg. 2019 Jan 1; 113(1): 4-10.
[72] Jesuyajolu DA, Jesuyajolu P. Furuncular myiasis affecting the glans penis of a young boy caused by the larvae of *Cordylobia anthropophaga* (the tumbu fly): a case report. Pan Afr Med J. 2022 May 27; 42: 75.
[73] Passos MR, Barreto NA, Varella RQ, Rodrigues GH, Lewis DA. Penile myiasis: a case report. Sex Transm Infect. 2004 Jun; 80(3): 183-4.
[74] Wakid MH, Sharafeldein YS, Almakki AA, Alidrisi DA, Bashinim AA. Scrotal Myiasis in a Child Due to Cordylobia anthropophaga. Cureus. 2024Apr30;16(4): e59417.
[75] Egbuchulem KI, Ogundipe HD, Olulana DI, Ojediran TO. A rare finding of distal penile furuncular myiasis in a child of a Nigerian health care worker. Ann Ib Postgrad Med. 2023 Aug; 21(2): 103-105.
[76] Rath BK, Das PK. Urogenital myiasis. Indian Pediatr. 2000 Jul; 37(7): 797-8.
[77] Lageju N, Neupane D, Jaiswal LS, Phuyal U. Pin-tract myiasis after external bone fixation: A case report and review of literature. Int J Surg Case Rep. 2022 Jun; 95: 107247.
[78] Saluja HK, Jadhav S. Diaphyseal tibial osteosarcoma with myiasis. Cureus. 2022 Dec 20; 14(12): e32718.
[79] Kalelioglu M, Akturk G, Akturk F, Komsuoglu SS, Kuzeyli K, Tigin Y, Karaer Z, Bingol R. Intracerebral myiasis from Hypoderma bovis larva in a child. Case report. J Neuro surg. 1989 Dec; 71(6): 929-31.
[80] Ramon-Cuellar JF, Mejía-Cordovez JA, Quinones-Ossa GA, Leal-Isaza JP, Vargas-Osorio MP, Ramirez-Munoz JD, Ordonez-Rubiano EG, Ramirez AP, Amarillo DG, Hakim-Daccach F. Cerebral myiasis, an unexpected intraoperative finding - A case report and systematic review. World Neurosurg. 2024 Jun; 186: 138-144.
[81] Curzi C, Bartoletti V, Canova G, Giordan E. A severe case of brain myiasis: treatment rationale and review of literature. Asian J Neurosurg. 2021 Sep14; 16(3): 582-586.
[82] Pouillaude JM, Dupont J, Gilly R, Lapras C. Intracerebral myiasis in a child. Pediatr Radiol. 1980 Nov; 10(2):121-3.
[83] Labbe A, Desvignes V, Meyer M, Campagne D, Cohen F, Dechelotte P. Meningite a Hypoderma bovis. A propos d'un nouveau cas pediatrique [Hypoderma bovis meningitis. Apropos of new pediatric case]. Ann Pediatr (Paris). 1983 Apr; 30(4): 277-80.
[84] Rossi MA, Zucoloto S. Fatal cerebral myiasis caused by the tropical warble fly, Dermatobia hominis. Am J Trop Med Hyg. 1973 Mar; 22(2): 267-9.
[85] Udgaonkar US, Dharamsi R, Kulkarni SA, Shah SR, Patil SS, Bhosale AL, Gadgil SA, Mohite RS. Intestinal myiasis. Indian J Med Microbiol. 2012 Jul-Sep; 30(3): 332-7.
[86] Chen XQ, Zhang KL, Shan QW. Bloody diarrhea caused by intestinal myiasis in an infant: A case report and review

of pediatric literature. J Trop Pediatr. 2021 Jul 2; 67(3): fmaa037.
[87] Aspock H. Rektale Myiasis durch Fannia canicularis (Linne) und *Muscina stabulans* (Fallen) [Rectal myiasis due to Fannia canicularis (Linne) and *Muscina stabulans* (Fallen)]. Dtsch Med Wochenschr. 1972 Aug 11; 97(32): 1174-5.
[88] North DE, Matteson KL, Helgerson SD, Richards F Jr, Stewart JM, Baum L, Catts EP. Intestinal myiasis in a baby attending a public health clinic. Nurse Pract. 1987 May; 12(5): 60-3.
[89] Rathi S, Pednekar K, Pathak A, Singh P. Screw-worm myiasis of prolapsed rectum. Indian Pediatr. 2014 Jan; 51 (1): 53-4.
[90] Karabiber H, Oguzkurt DG, Dogan DG, Aktas M, Selimoglu MA. An unusual cause of rectal bleeding: intestinal myiasis. J Pediatr Gastroenterol Nutr. 2010 Oct; 51(4): 530-1.
[91] Hasegawa S, Miwata H, Masuda S, Naruse H, Ozaki T. An infantile case of intestinal myiasis. Acta Paediatr Jpn. 1992 Feb; 34(1): 87-9.
[92] Nagakura K, Isozaki M, Shigeta M, Shimamura T, Tachi bana H, Kaneda Y, Kano R. A case report of intestinal myiasis. Tokai J Exp Clin Med. 1984 Dec; 9(5-6): 345-8.
[93] Zardi EM, Iori A, Picardi A, Costantino S, Petrarca V. Myiasis of a perineal fistula. Parassitologia. 2002 Dec; 44 (3-4): 201-2.
[94] Ahmed MJ, Miller A. Pulmonary coin lesion containing a horse bot, Gasterophilus. Report of a case of myiasis. Am J Clin Pathol. 1969 Oct; 52(4): 414-9.
[95] Mielke U. Nosocomial myiasis. J Hosp Infect. 1997 Sep; 37(1): 1-5.
[96] Espinoza-Gomez F, Rojas-Larios F, Cruz-Sanchez S, Rodri guez-Hernandez A, Delgado-Enciso I. Outbreak of nosocomial myiasis by Cochliomyia macellaria (Diptera, Calliphoridae) in a hospital in Colima, Mexico. Am J Trop Med Hyg. 2023 Sep 11; 109(4):748-751.
[97] Maleki-Ravasan N, Shayeghi M, Najibi B, Oshaghi MA. Infantile nosocomial myiasis in Iran. J Arthropod Borne Dis. 2012 Dec; 6(2): 156-63.
[98] Martinez-Rojano H, Huerta H, Hernandez-Triana LM, Ruiz Perez EF, Sámano R. Nosocomial myiasis caused by *Lucilia sericata* (Diptera: Calliphoridae) in a pediatric patient in Mexico. Case Rep Infect Dis. 2020 Jan 29; 2020: 1285459.
[99] Clyti E, Deligny C, Nacher M, Del Giudice P, Sainte-Marie D, Pradinaud R, Couppie P. An urban epidemic of human myiasis caused by *Dermatobia hominis* in French Guiana. Am J Trop Med Hyg. 2008 Nov; 79(5): 797-8.
[100] Bouer M, Rodriguez-Bandera AI, Albizuri-Prado F, Lobos A, Gubeling W, Wortsman X. Real-time high-frequency colour Doppler ultrasound detection of cutaneous *Derma tobia hominis* myiasis. J Eur Acad Dermatol Venereol. 2016 Dec; 30(12): e180-e181.
[101] Pogany P, Szucs E, Lichtenberger G, Vass L. Diagnosis of myiasis by fine needle aspiration cytology: a case report. Acta Cytol. 2008 Mar-Apr; 52(2): 228-30.
[102] https://shire.science.uq.edu.au/parasites/insects/maggots/maggots-identification.php (Accessed on 16 August 2024)
[103] Sherman RA, Jesus KD. Removing unwanted maggots. J Emerg Med. 2015 Feb; 48(2): 213.
[104] Sherman RA, Nguyen A, Dastpak V, Fonseca-Munoz A, Parizad N, Siavash M, Yusuf MA. Not all maggots are created equal; not all maggots are therapeutic. Clin Case Rep. 2024 Jul 17; 12(7): e9163.
[105] Elder JW, Grover CA. Wound debridement: lessons learned of when and how to remove "wild" maggots. J Emerg Med. 2013 Oct; 45(4): 585-7.
[106] Nayak SP, Reddy YL, Patil PH, Harugop AS, Narasimman DB, Neema K. Importance of an ENT surgeon in maggot removal, improper attempt leads to deadly complications: A case report. Indian J Otolaryngol Head Neck Surg. 2023 Jun; 75(2): 1204-1206.
[107] Nunzi E, Rongioletti F, Rebora A. Removal of *Dermatobia hominis* larvae. Arch Dermatol. 1986 Feb; 122(2): 140.
[108] Pereyra-Rodríguez JJ, Bernabeu-Wittel J, Conejo-Mir MD, Ruiz-Perez de Pipaon M, Conejo-Mir J. Treatment of cutaneous myiasis associated with scalp psoriasis in a 13-year-old girl with oral ivermectin. J Am Acad Dermatol. 2010 Nov; 63(5): 908-9.
[109] Duque FL, Ardila CM. Oral myiasis caused by the screwworm Cochliomyia hominivorax treated with subcutaneous ivermectin and creolin: report of six cases after trauma. Dent Traumatol. 2011 Oct; 27(5): 404-7.
[110] Clyti E, Nacher M, Merrien L, El Guedj M, Roussel M, Sainte-Marie D, Couppie P. Myiasis owing to *Dermatobia hominis* in a HIV-infected subject: Treatment by topical ivermectin. Int J Dermatol. 2007 Jan; 46(1): 52-4.
[111] Patel BC, Ostwal S, Sanghavi PR, Joshi G, Singh R. Management of malignant wound myiasis with ivermectin, albendazole and clindamycin (triple therapy) in advanced head-and-neck cancer patients: A prospective observational study. Indian J Palliat Care. 2018 Oct-Dec; 24(4): 459-464.
[112] Bernhard JD. Bringing on the bacon for myiasis. Lancet. 1993 Dec 4; 342(8884): 1377-8.
[113] Fox H, Seeger FH. Tetanus in a patient with a maggot-colonised head tumor. Anaesth Intensive Care. 2013 Mar; 41(2): 272-3.
[114] Greenberg B. Flies and Disease. Vol 2: Biology and disease transmission. Princeton University Press, New Jersey. 1973.
[115] Lupi O. Myiasis as a risk factor for prion diseases in humans. J Eur Acad Dermatol Venereol. 2006 Oct; 20(9): 1037-45.
[116] Morris D, Flores M, Harris L, Gammon J, Nigam Y. Larval therapy and larval excretions/secretions: A potential

treatment for biofilm in chronic wounds? A systematic review. Microorganisms. 2023 Feb 11; 11(2): 457.
[117] Fleischmann W, Grassberger M, Sherman R. Maggot therapy - A handbook of maggot-assisted wound healing. Georg Thieme Verlag, New York 2004.
[118] Stadler F. A Complete Guide to Maggot Therapy. Open Book Publishers, Cambridge 2022
[119] Grassberger M, Fleischmann W. The biobag - a new device for the application of medicinal maggots. Derma tology. 2002; 204(4): 306.
[120] Opletalova K, Blaizot X, Mourgeon B, Chêne Y, Creveuil C, Combemale P, Laplaud AL, Sohyer-Lebreuilly I, Domp martin A. Maggot therapy for wound debridement: a randomized multicenter trial. Arch Dermatol. 2012 Apr; 148(4): 432-8.
[121] Birdsong M, McIltrot KH, Ascenzi J. Pediatric maggot debridement therapy: A case study. J Pediatr Surg Nurs. 2014 April; 3(2): 60-64. DOI:10.1097/JPS.0000000000000021
[122] Orkiszewski M, Madej J, Kilian T. The use of maggot therapy in paediatric surgery: a case report. World Wide Wounds. Mar 2006. {Availabe from: http://www.worldwidewounds.com/2006/march/Orkiszewski/Use-Maggot-Therapy-Paediatric-Surgery.html} (Accessed on 15 September 2024)
[123] Perez-Acevedo G, Bosch-Alcaraz A, Torra-Bou JE. Larval therapy for treatment of chronic wounds colonized by multiresistant pathogens in a pediatric patient: A Case study. J Wound Ostomy Continence Nurs. 2022 Jul-Aug 01; 49(4): 373-378.

Address for communication: *Dr. V. Raveenthiran, Email: vrthiran@gmail.com*

Received 20 Sep 2024; Accepted 30 Sep 2024

Acknowledgements: None
Conflicts of Interest: None declared by the author
Source of Funding : None
Ethical concerns : None (Review of literature)

Citation: *Raveenthiran V. Maggot infestation (Myiasis) in children. Pediatr Surg Trop 2024; 1(4): 216-248.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 249-252
DOI: 10.5281/zenodo.13893376

Case Report

Pediatric Sigmoid Volvulus

Cristina Fernandez, Shannon Yoo, Sathyaprasad Burjonrappa

Department of Pediatric Surgery, Rutgers Robert Wood Johnson Medical School, New Brunswick, New Jersey, NJ08901, USA.

Keywords
Acute abdomen
Colectomy
Colonic obstruction
Colonoscopy
Laparoscopy
Sigmoid colon Volvulus

Abbreviations
SV - Sigmoid volvulus

Abstract

Sigmoid volvulus is a rare cause of colonic obstruction in children. In this age group it has higher morbidity and mortality than that of adults. There is a paucity of literature on pediatric sigmoid volvulus, and thus treatment strategies are largely based on adult data. Regardless of the patient age, prompt intervention is imperative in all cases. Emergent colectomy is typically reserved for cases complicated by gangrene, perforation and peritonitis; while non-operative emergency decompression followed by elective surgical resection is preferred in uncomplicated cases. In this report, we describe a 17-year-old female with sigmoid volvulus, who successfully underwent flexible sigmoidoscopic detorsion, followed by rectal tube decompression for 2 days and then elective laparoscopic sigmoid colectomy with primary anastomosis.

INTRODUCTION

Sigmoid volvulus is a rare, yet potentially life-threatening cause of large bowel obstruction in children.[1] It occurs when a loop of sigmoid colon twists upon its mesentery. Depending on the degree and severity of torsion, the volvulus may progressively cause bowel obstruction, ischemic infarction, gangrene, perforation and potentially septic shock.[1] Thus, immediate intervention is critical for survival.

The current approach to pediatric SV closely follows the management algorithm in adults. It is based on the initial presentation of the patient. Emergent surgery on admission is required for those who present with complications such as ischemic necrosis, perforation, or peritonitis. Non-surgical emergency endoscopic detorsion followed by elective laparoscopic sigmoidectomy, is the favored approach in uncomplicated SV. In this report, we present the successful management of uncomplicated SV in an adolescent patient. We emphasize the importance of tailored management of SV based on the initial presentation and complications.

CASE REPORT

A 17-year-old female presented to the Emergency Department with a 3-day history of intermittent abdominal pain, multiple episodes of emesis and nausea. Two days prior to the onset of pain, she had had fever of 100.1^{0}F, followed by non-bloody

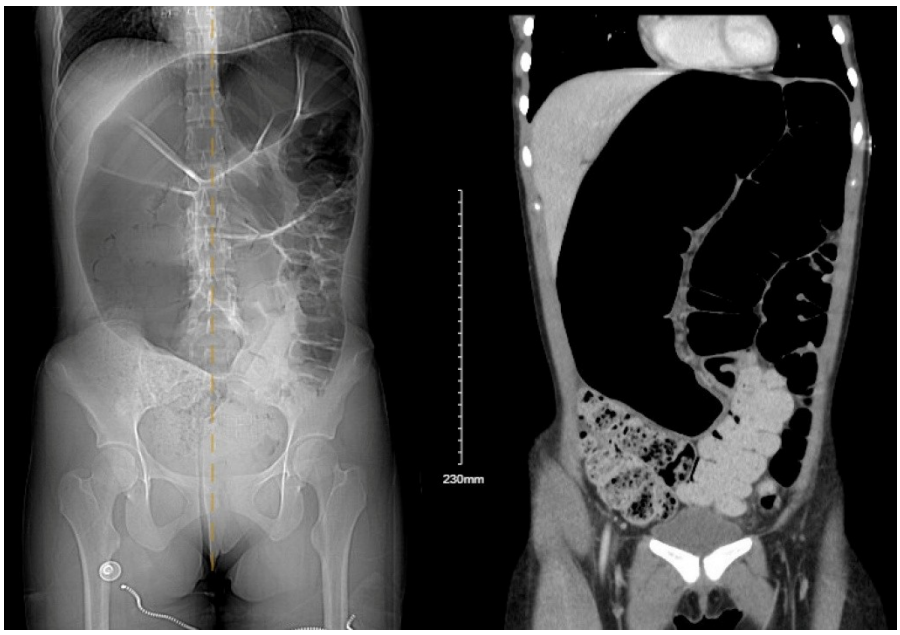


Fig 1. *Abdominal X-Ray (left panel) and coronal section of computed tomographic (CT) scan (right panel) showing the classic 'coffee-bean sign' of sigmoid volvulus.*

diarrhea on the next day. She denied passing any flatus. Previously she was healthy without any major illness.

On physical examination, she had tachycardia (116 beats per min) and hypertension (143 / 77 mmHg). She was afebrile and stable with a respiratory rate of 18 per min and oxygen saturating of 98% on room air. She was having distension with diffuse tenderness throughout the abdomen.

Her leukocyte count was elevated to 24,500/mm^3. Plain x-ray of abdomen showed coffee-bean sign that is typical of SV. (Fig. 1) Contrast enhanced CT scan of abdomen confirmed SV as evidenced by a markedly dilated air-filled and tortuous loop of sigmoid colon. (Fig. 1) There was also considerable amount of stool retained in the ascending and the transverse colon. No abscess or free air was observed.

She underwent flexible sigmoidoscopy, followed by rectal tube decompression for 2 days. Five days after the initial presentation, she underwent laparoscopic sigmoid colectomy with primary anastomosis after preoperative bowel preparation and intravenous cefoxitin. The resected colon was thin and dilated measuring approximately 51 cm in length. The patient recovered well by tolerating oral intake, achieving adequate pain control, and ambulating safely during her hospitalization. She was discharged on the fourth post-operative day without any complications.

DISCUSSION

SV is a rare cause of colonic obstruction in children. Risk factors of SV include redundant sigmoid colon, anatomically a long mesocolon with narrow base and colonic dysmotility. Among adults, these predisposing risk factors occur with old age and chronic constipation. In pediatrics, a systematic review of 256 cases of SV demonstrated correlation with Hirschsprung disease (10%), neurological or developmental disorders (10.9%), and constipation (10.2%).[2] However, 57.4% of patients in

that systematic review did not have any known predisposing risk factors.

Volvulus can be potentially life threatening. The isolated twisted bowel segment may become ischemic, leading to gangrene, perforation, and septic shock.[1] Hence, immediate intervention is critical to avoid fatality.

Endoscopic detorsion is non-invasive and highly effective treatment, with a success rate as high as 75-95% among adults.[2,3] However, successful endoscopic decompression is followed by a recurrence rate as high as 84%, rendering it a less effective and less reliable long-term solution.[4,5] The time interval between endoscopic decompres-sion and recurrence varies from a few days to several months, with a median of 2 months as noted in a retrospective study of 168 patients.[4] Rectal tube decompression or barium enema under fluoroscopic control are other non-surgical interventions that are favored in pediatric age group. The later methods have similar success rate, yet have lesser risk of iatrogenic perforation than sigmoidoscopy.[1,2] Similar to endoscopic intervention, rectal tube and fluoroscopic enema are not definitive procedures as they are asso-ciated with a recurrence rate as high as 38-57% in children.[2] With every subsequent episodes of SV, recurrence and mortality rates increase, underscoring the critical need for a definitive treatment after the first presentation.[4]

Surgery is the most definitive treatment, which typically involves resection of the redundant colon (sigmoid colectomy). Colonic resection may be followed by temporizing colostomy or primary colorectal anastomosis. Surgical options that do not involve resection, such as operative detorsion, sigmoidopexy, and mesosigmoidoplasty have been shown to be inferior to sigmoid resection in recurrence prevention among adults.[6] There are no reported cases of recurrent SV after sigmoidectomy.[3] The high-risk of recurrence after endoscopic detorsion, coupled with the high morbidity and mortality associated with recurrent volvulus, makes definitive sigmoid colectomy the best course of action during the initial admission or promptly afterward.[3,5,7]

The most opportune time for sigmoid colectomy is few days after a successful non-surgical detorsion, rather than immediately upon the initial presentation. Elective surgery following successful non-surgical initial decompression has lower mortality rate than emergency surgery.[8] Emergent surgery has been shown to have higher rates of postoperative complications, open surgeries and stoma creation that require secondary take-down surgeries as compared to post-decompression elective laparoscopic surgery.[1,8] For this reason, emergency surgery is typically reserved for complicated cases and failed attempts of non-surgical detorsion.[2,5,9]

The best course of treatment for SV in both children and adults is dependent on the nature of initial presentation. Those who present with complicated volvulus, including perforation and ischemic necrosis of the bowel, will always require emergent surgery. Those who present with an uncomplicated volvulus should undergo non-surgical decompression, followed by elective resection after proper bowel preparation. The case reported herein demonstrates a successful treatment of non-complicated pediatric SV using colonoscopy, followed by elective surgical sigmoidectomy with primary anastomosis.

REFERENCES

[1] Salas S, Angel CA, Salas N, Murillo C, Swischuk L. Sigmoid volvulus in children and adolescents. J Am Coll Surg. 2000 Jun; 190(6): 717-23.

[2] Hencke J, Loff S. Recurrent Sigmoid Volvulus in Children-Our Experience and Systematic Review of the Current Literature. Children (Basel). 2023 Aug 24; 10(9): 1441.

[3] Atamanalp SS. Treatment of sigmoid volvulus: a single-center experience of 952 patients over 46.5 years. Tech Coloproctol. 2013 Oct; 17(5): 561-9.

[4] Johansson N, Rosemar A, Angenete E. Risk of recurrence of sigmoid volvulus: a single-centre cohort study. Colorectal Dis. 2018 Jun; 20(6): 529-535.
[5] Parolini F, Orizio P, Bulotta AL, Garcia Magne M, Boroni G, Cengia G, Torri F, Alberti D. Endoscopic management of sigmoid volvulus in children. World J Gastrointest Endosc. 2016 Jun 25; 8(12): 439-43.
[6] Vogel JD, Feingold DL, Stewart DB, Turner JS, Boutros M, Chun J, Steele SR. Clinical Practice Guidelines for Colon Volvulus and Acute Colonic Pseudo-Obstruction. Dis Colon Rectum. 2016 Jul; 59(7): 589-600.
[7] Ismail A. Recurrent colonic volvulus in children. J Pediatr Surg. 1997 Dec; 32(12): 1739-42.
[8] Lee K, Oh HK, Cho JR, Kim M, Kim DW, Kang SB, Kim HJ, Park HC, Shin R, Heo SC, Ryoo SB, Park KJ; Seoul Colorectal Research Group (SECOG). Surgical Management of Sigmoid Volvulus: A Multicenter Observational Study. Ann Coloproctol. 2020 Dec; 36 (6): 403-408.
[9] Lou Z, Yu ED, Zhang W, Meng RG, Hao LQ, Fu CG. Appropriate treatment of acute sigmoid volvulus in the emergency setting. World J Gastroenterol. 2013 Aug 14; 19 (30): 4979-83.

Address for communication: *Dr. Sathyaprasad Burjonrappa, Email: Sb2058@rutgers.edu*

Received: 12 May 2024; Accepted: 3 July 2024

Acknowledgements: None
Conflicts of Interest: None declared by the author
Source of Funding : None
Ethical concerns : None (Report of routine clinical care)

Citation: *Fernandez C, Yoo S, Burjonrappa S. Pediatric Sigmoid Volvulus. Pediatr Surg Trop 2024; 1(4): 249-252.*

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 253-257
DOI: 10.5281/zenodo.13893511

Case Report

Midgut Volvulus - A Rare Cause of Recurrent Acute Kidney Injury in Adolescence

Febin Abraham[1], Georgie Mathew[1], Deepthi Raranveettil[1], Swati Kiran Shiri[1], Lakshmi Devi Naorem[1], Naresh Shanmugam[1], Anurega Selvaraj[1], Aiswarya Manoharan[2], John Kuttichirayil Thomas[3], Indira Agarwal[1]

Departments of Pediatric Nephrology [1], Radiology [2] and Pediatric Surgery [3], Christian Medical College, Vellore, Tamil Nadu, India.

Keywords
Acute kidney injury
Midgut volvulus
Malrotation of midgut
Adolescent Pediatrics
Pseudo-Bartter syndrome

Abbreviations
AKI - Acute kidney injury

Abstract

A15-year-old boy presented with recurrent episodes of non-bilious vomiting for the past 9 years. During these episodes he also had dehydration and oliguria. He required hospitalization during each episode that was marked by features of acute kidney injury. A plain radiograph showing ground glass opacity raised suspicion. Imaging studies confirmed intestinal malrotation with intermittent midgut volvulus. Following correction of malrotation by Ladd's procedure, he became asymptomatic. Delayed presentation of malrotation beyond infancy is rare. Recurrent acute kidney injury is an unusual complication of late manifesting malrotation in adolescence.

INTRODUCTION

Midgut volvulus due to intestinal malrotation usually presents in the early infancy. Delayed presentation leads to diagnostic dilemmas and hence increased morbidity and mortality. In this report, we describe a case of midgut malrotation with an extraordinary diagnostic delay leading to significant complications.

CASE REPORT

A 15-year-old boy, the second-born child of non-consanguineous parents, was admitted with recurrent bouts of vomiting for the past 9 years. Antenatal and perinatal periods were uneventful. He had appropriate development for the age. After his mother's death 10 years ago (cause unknown), he was living with his relatives and there was no reported child abuse during this period.

He presented with non-bilious, non-projectile, and non-hemorrhagic vomiting that occurred typically after food. Symptoms occurred semiannually, starting at the age of 6 years. The frequency of symptoms worsened progressively after the age of 12 years, with monthly episodes in the last 2 years and fortnightly episodes in the last few months. With the passage of time, the quantity of vomitus (currently 2-3 large cups) and its intensity (each episode lasting for 5-6 days at a stretch) have also

Table 1: Renal parameters at the previous and current inpatient admissions

Parameters	Mar 2023		April 2023		May 2023		Oct 2023	
	Adm	Dis	Adm	Dis	Adm	Dis	Adm	Dis
Blood Urea (mg/dl)	110	-	66	-	109	18	29	-
Serum Creatinine (mg/dl)	2.3	0.8	1.6	0.8	6.2	0.7	3.8	0.7
e-GFR (ml/min per 1.73 m^2)	22	63	32	63	8	72	13	72
Serum Sodium (mEq/l)	124	134	134	136	131	134	142	139
Serum potassium (mEq/l)	2.7	4.3	4.7	3.6	2.5	3.7	2.9	4.9
pH	7.59	7.35	7.39	7.31	7.56	7.39	7.47	-
Bicarbonate (mEq/L)	57	23	28	21	46	23	27	21
Ionized Calcium (mEq/l)	0.76	1.02	1.48	-	0.98	1.22	1.09	-

e-GFR- Estimated glomerular filtration rate, Adm - Day of admission, Dis- Day of Discharge

worsened. Few episodes of bilious vomiting, were observed recently, which were preceded by abdominal fullness and discomfort. He did not tolerate solid food, and preferred liquids instead.

For the last 4 months, fortnightly episodes of large quantity, bilious vomiting were associated with severe dehydration, reduction in the urine output and constipation. Three inpatient admissions each lasting for 8-10 days were required for correction of severe dehydration, metabolic alkalosis, hyponatremia, severe hypokalemia, and stage-3 acute kidney injury (AKI) (Table 1).

He self reported symptomatic relief during nil per oral therapy with nasogastric drainage. At a peripheral centre potassium chloride and indomethacin were given to correct recurrent hypokalemic metabolic alkalosis, although there was no documented polyuria. Diarrhea, jaundice, bulky or oily stools, headache, aura, seizures or altered sensorium were absent during the preceding 9 years. He denied using any traditional or over-the-counter medications. Poor growth was recorded during the recent past visits.

On presentation to our centre, he was severely dehydrated and hypotensive, but with a normal sensorium. Initial pulse rate was 110/min with low volume, respiratory rate was 22/min and blood pressure was 80/50 mm Hg. He was afebrile and was not dyspnoeic, maintaining SpO_2 99% on room air. Severe wasting and stunting were observed with a height of 139 cm (-3.1 standard deviation score - SDS), weight of 30.2 kg (-2.8 SDS), and body mass index of 15.6 kg/m^2 (-1.3 SDS).

The abdomen was soft with mild epigastric fullness, but without any visible peristalsis. Bowel sounds were diminished in the lower quadrants. The liver and spleen were not palpable. Hypovolemic shock corrected with bolus infusion of 20 ml/kg of isotonic fluids and it was followed by correction of dehydration.

An erect radiograph showed a gasless abdomen with a small fundal gas bubble. Barium swallow study demonstrated a grossly distended stomach with abrupt narrowing of the bowel at the proximal duodenum. (Fig. 1) A negligible amount of contrast was seen in the distal bowel.

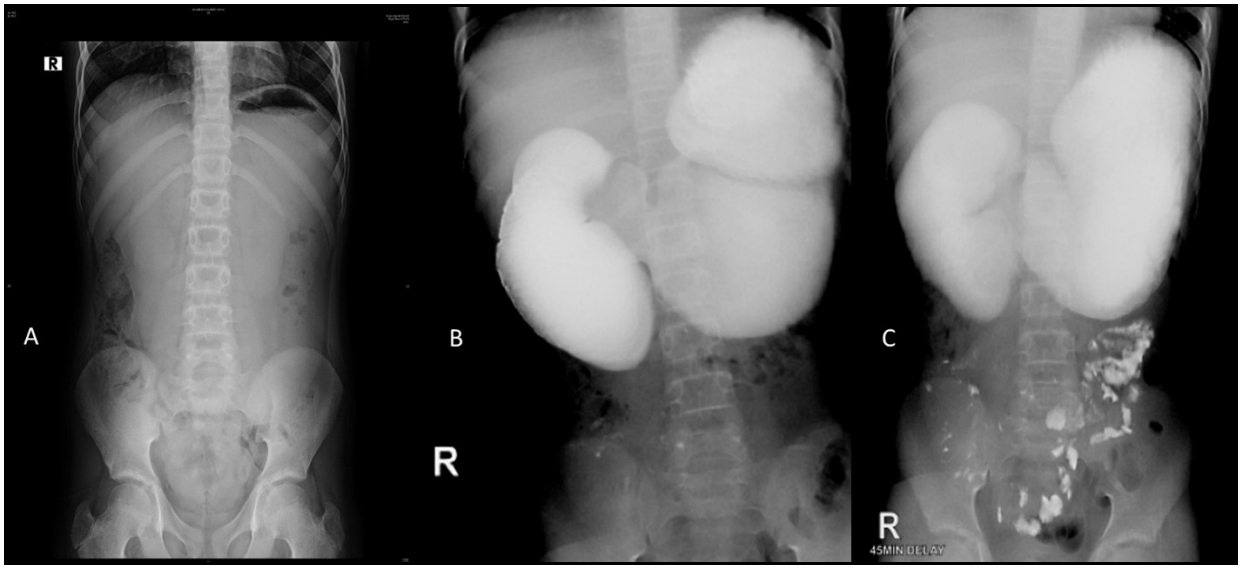


Fig 1: *Contrast radiography (A) Erect plain radiograph showing gasless abdomen; (B) Barium meal follow through showing massive dilatation of the stomach and the duodenum with abrupt cutoff at the third part of the duodenum. (C) Delayed picture showing minimal passage of contrast into the distal small bowel.*

Near-complete obstruction at the distal duodenum was recognized. MRI of the abdomen showed features of midgut malrotation with volvulus. (Fig. 2) Ultrasonography showed near-normal sized kidneys (right kidney length 74 mm; left kidney length 74 mm; -0.36 and -0.97 SDS respectively) with increased echogenicity, but without any hydroureteronephrosis.

He was finally diagnosed with recurrent metabolic alkalosis secondary to severe gastric losses, malrotation of the intestine with midgut volvulus, recurrent AKI in the context of acute-on-chronic kidney disease and chronic malnutrition.

After initial stabilization, a laparotomy was done. Small bowel volvulus with Ladd's band was found. After counterclockwise derotation, Ladd's band was divided, the mesenteric base was broadened and the small intestine was repositioned after straightening the duodenum. Postoperatively he recovered uneventfully and was discharged after a few days, with complete normalization of the metabolic parameters.

DISCUSSION

In this report, we present a patient who presented with multiple metabolic crises and recurrent reversible AKI due to midgut malrotation with volvulus. The absence of bilious vomiting and the episodic nature of the symptoms precluded an early diagnosis in this patient. It appears that the patient had had recurrent episodes of volvulus with transient derotation, leading to the episodic nature of symptoms.

Intestinal malrotation affects roughly 1 in 500 newborns, and volvulus occurs in approximately 1 in 2500 live births.[1] Normally, between 6 and 12 weeks of gestation, the intestine rotates 270^{o} with the superior mesenteric artery (SMA) as the axis. The duodeno-jejunal loop shifts to the left side of the SMA, while the ceco-colic loop moves to the right side, resulting in the final anatomic arrangement.[2] In cases of partial rotation (90^{o} - 180^{o}), the cecum remains in the mid-upper abdomen and becomes fixed to the right lateral wall by the Ladd's bands. These bands may compress the duodenum, leading to obstruction.

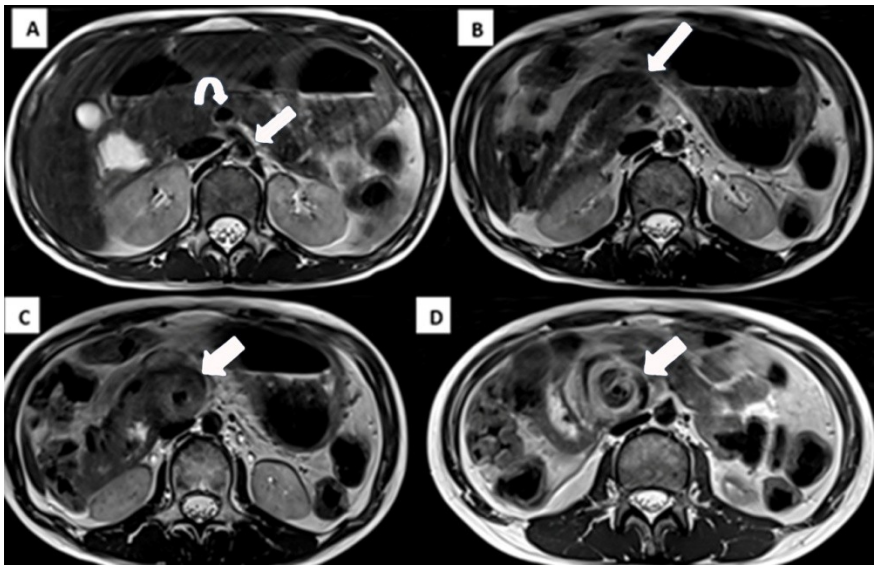


Fig 2: *Magnetic rersonance imaging (T2 high-resolution axial sections through the upper abdomen) showing the superior mesenteric artery (white arrow) coursing to the right of the superior mesenteric vein (bent arrow) (Panel A). The third part of the duodenum (arrow) does not cross the midline (Panel B & C) and is seen spiralling with the mesentery forming a "cork-screw" appearance (arrows in the panels C & D). These features are typical of midgut volvulus due to intestinal malrotation.*

The classical clinical presentation of midgut malrotation is that of bilious vomiting in the neo-natal period or infancy. An untreated malrotation may be complicated by midgut volvulus which in turn leads to hypovolemia, shock, perforation, peritonitis, bowel necrosis and systemic complications. Rarely, spontaneous derotation may cause intermittent relief of symptoms. The diagnosis is eventually reached by acute crisis, and rarely mortality occurs.

Some patients experience poor weight gain and have difficulty in tolerating solid food. This subtle clue is very important for early diagnosis and intervention.[4] Recurrent metabolic alkalosis with AKI is a rare presentation of midgut malrotation that can mimic some of the salt-wasting tubulopathies such as Bartter syndrome and Gitelman syndrome. Ultrasound is the first-line diagnostic imaging of choice in midgut volvulus. Its sensitivity is 94% and specificity is 100%. In cases that go undetected upper GI contrast studies, CT or MRI may be useful. [5]

The management of midgut volvulus involves several key steps. Preoperatively, brief, effective fluid resuscitation is needed to address dehydration or shock. Undue delay in surgical correction of the volvulus can lead to bowel gangrene. Nasogastric tube is necessary for decompression of the gastrointestinal tract. Broad-spectrum antibiotics should be administered to cover the bowel flora. The primary treatment choice involves an emergency laparotomy and Ladd's procedure, during which the volvulus is derotated and the mesentery is widened to prevent recurrence.[6] It is imperative to exclude other associated gastro-intestinal anomalies (e.g. duodenal web, intestinal pseudo-obstruction, ciliary dyskinesias). Extra-intestinal anomalies such as genitourinary, cardiovascular and craniofacial abnormalities may rarely coexist with midgut malrotation.[2]

REFERENCES

[1] Svetanoff WJ, Srivatsa S, Diefenbach K, Nwomeh BC. Diagnosis and management of intestinal rotational abnormalities with or without volvulus in the pediatric population. Semin Pediatr Surg. 2022 Feb;31(1):151141.

[2] Martin V, Shaw-Smith C. Review of genetic factors in intestinal malrotation. Pediatr Surg Int. 2010 Aug; 26(8): 769-81.

[3] Shah MR, Levin TL, Blumer SL, Berdon WE, Jan DM, Yousefzadeh DK. Volvulus of the entire small bowel with normal bowel fixation simulating malrotation and midgut volvulus. Pediatr Radiol. 2015 Dec; 45(13): 1953-1956.

[4] Karlslätt KS, Husberg B, Ullberg U, Nordenskjöld A, Wester T. Intestinal malrotation in children: Clinical presentation and outcomes. Eur J Pediatr Surg. 2024 Jun; 34 (3): 228-235.
[5] Nguyen HN, Kulkarni M, Jose J, Sisson A, Brandt ML, Sammer MBK, Pammi M. Ultrasound for the diagnosis of malrotation and volvulus in children and adolescents: a systematic review and meta-analysis. Arch Dis Child. 2021 Dec; 106(12): 1171-1178.
[6] Langer JC. Intestinal Rotation Abnormalities and Midgut Volvulus. Surg Clin North Am. 2017 Feb; 97(1): 147-159.

Address for communication: *Dr Georgie Mathew, Email: georgie.mathew@cmcvellore.ac.in*

Received: 30 July 2024; Accepted: 4 August 2024

Acknowledgements: None
Conflicts of Interest: None declared by the author
Source of Funding : None
Ethical concerns : None (Report of routine clinical care)

Citation: *Abraham F, Mathew G, Deepthi R, Shiri SK, Naorem LD, Shanmugam N, Selvaraj A, Manoharan A, Thomas JK, Agarwal I. Midgut volvulus - A rare cause of recurrent acute kidney injury in adolescence. Pediatr Surg Trop 2024; 1(4): 253-257.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 258-261
DOI: 10.5281/zenodo.13893572

Case Report

Pancreatic Heterotopia

Sindhu Anirudhan Adarsh[1], Pampa Ch Toi[2], Krishna Kumar Govindarajan[1]

Departments of Pediatric Surgery [1] and Pathology [2], Jawaharlal Institute of Postgraduate Medical Education and Research (JIPMER), Pondicherry 605006, India.

Keywords
Ectopic Pancreas
Heterotopia
Intestinal obstruction
Intussusception
Pain abdomen

Abbreviations
GI - Gastro intestinal
PH - Pancreatic heterotopia
USG - Ultrasonography

Abstract

Pancreatic heterotopia (PH) is a rare congenital anomaly that is also known as ectopic pancreas or pancreatic choristoma. It is usually asymptomatic but rarely may cause abdominal pain, lower gastrointestinal bleeding or intussusception. This report describes two cases of PH: a 9-month-old boy presented with intussusception and a 6-year-old girl presented with a jejuno-colic inflammatory mass. Surgical excision of PH cured both of them.

INTRODUCTION

Pancreatic heterotopia (PH) is a rare congenital anomaly that mostly remains silent for several years. It is also known as ectopic pancreas or pancreatic choristoma. Rarely PH may become symptomatic, presenting with abdominal pain. The common age of presentation is usually the third decade. In the pediatric age group, it forms the pathological lead point of intussusception.(1)

The pathogenesis of PH is explained by the misplacement theory. During the embryonic rotation and fusion of the ventral and dorsal pancreatic buds, some of the pancreatic tissue may get separated from the main gland, and gets attached to the adjoining structures such as the stomach, small bowel, colon and Meckel's diverticulum. This ectopic tissue has its own independent vascularity and ductal com-ponents. The degree of maturation of the ectopic tissue is variable.(2,3) In PH, the pancreatic tissue is noted within the submucosa of the gastrointestinal (GI) tract, making its endoscopic diagnosis difficult. Gastrointestinal stromal tumor (GIST), leiomyoma and submucosal lymphoid hyperplasia or tumors are the closest differential diagnosis of PH.(2,3) Endoscopy or endoscopic ultra sound (EUS) combined with fine-needle aspiration cytology may be helpful in the diagnosis. Surgical excision is the only effective treatment of PH. In this report we share our experience with 2 such cases of PH.

CASE REPORTS

Case 1

A 9-month-old male child presented with non-bilious vomiting and excessive crying. On clinical examination, the abdomen was soft and non-tender without any palpable mass. Plain x-ray of

the abdomen was non-contributory. An ultrasonography (USG) revealed ileo-colic intussusception. After a failed attempt of USG-guided hydrostatic reduction, it was manually reduced at laparotomy (Fig. 1A), where upon a palpable nodule of 2 x 1.5 cm was noted at its apex. The lead-point was located at the distal ileum at about 10 cm proximal to the ileo-cecal junction. (Fig. 1B) The involved segment of the ileum was resected and primary end-to-end bowel anastomosis was done. Post-operative recovery was uneventful.

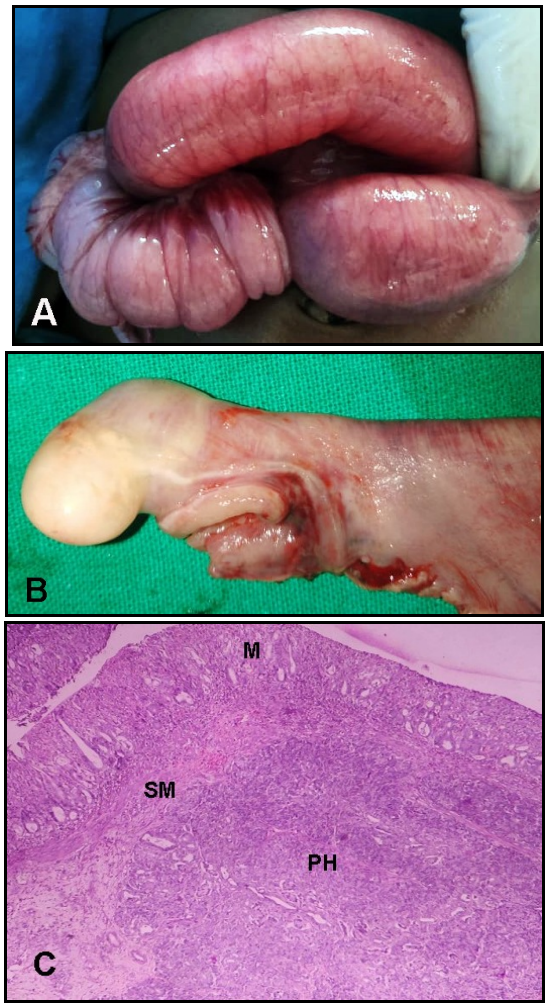


Fig 1. *Pancreatic heterotopia (case 1): (A) Intra-operative photograph showing ileo-colic intussusception, (B) pathological lead point at the apex of the reduced intussusception, (C) Histopathology of the apical nodule showing pancreatic heterotopia (PH) deeper to the mucosa (M) and submucosa (SM) Magnification 20X, Hematoxylin-Eosin staining*

Histopathology of the resected nodule (Fig. 1C) revealed presence of heterotopic pancreatic tissue in the submucosa along with focal ulceration of the overlying mucosa. The child was well on follow-up at 8 months.

Case 2

A 6-year-old girl presented with insidious onset of abdominal pain and vomiting. On clinical examination, a vague mass was felt in the upper abdomen. Diagnostic USG was not helpful due to interference of bowel gas. Plain radiograph was also non-contri butory. Persistent pain and the palpable mass prompted an exploratory laparotomy.

Intraoperatively, the proximal jejunum was found to be adherent with the transverse colon and the omentum. (Fig 2A) Further dissection revealed a nodule of 2 x 1.5 cm in the ante-mesenteric border of the jejunum. The nodule was excised by wedge resection of bowel and the rent was repaired. Histology confirmed the nodule as PH. (Fig 1C) Retrospectively, a diagnosis of ectopic pancreas causing inflammatory mass was made. The child was doing well on follow-up at 6 months.

DISCUSSION

PH is usually present in the submucosa of the upper GI tract, including the stomach, duodenum and less commonly in the jejunum, ileum and Meckel's diverticulum. It is rarely found in the esophagus, liver, gallbladder, biliary tree, spleen, omentum, lungs, mediastinum, Fallopian tubes and the umbilicus.[4] Comparatively PH is rarer in the lower GI tract than the upper tract. PH may also be complicated by inflammation and stricture. Ductal adenocarcinoma arising from rectal PH has been described.[1] Incidental diagnosis of PH during laparotomy is well known. In our cases, PH was identified in the ileum and the jejunum in one case each. These are less common sites according to the literature.

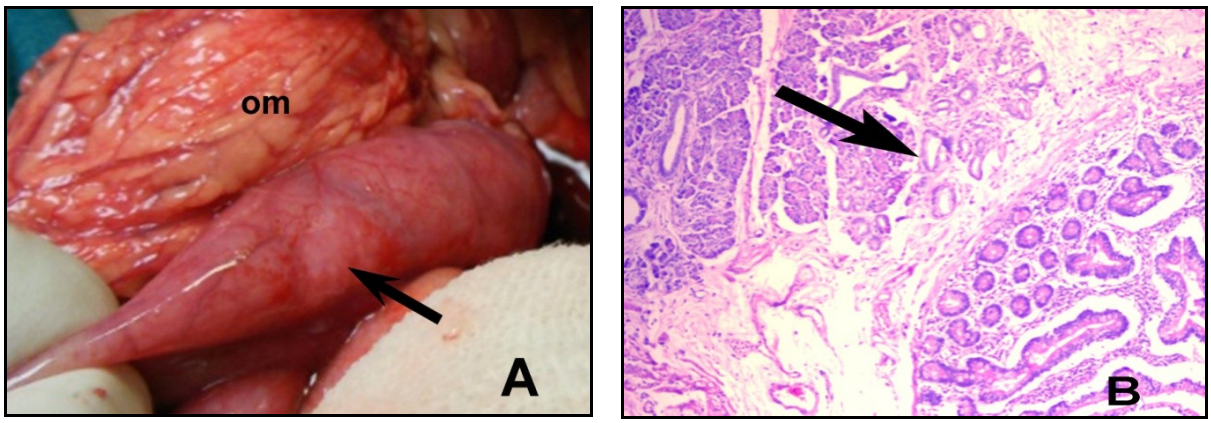


Fig 2. *Pancreatic heterotopia (case 2): (A) Intra-operative photograph showing the jejunal nodule (arrow) and the adherent omentum (om); (B) Histopathology of the nodule showing typical pancreatic acini (arrow) in the submucosa of the Jejunum (Magnification 100X, Hematoxylin-Eosin staining)*

PE may cause symptoms related to mechanical complications (e.g. gastric outlet obstruction, intus susception), bleeding from the ulcerated mucosa or metaplasia leading to malignancy in the late adulthood. Even though exceedingly rare, neoplasia arising from PH have been described even in children.[1] In our patients, one presented with intussusception and another with inflammatory bowel obstruction.

It is not possible to differentiate between normal and ectopic pancreatic tissue in histology as PH may have all the normal components (acini, ducts, islet cells) of pancreas. PH is also prone for common diseases of the orthotopic pancreas such as acute pancreatitis and pseudocysts.[5]

Persano et al. reported 14 pediatric patients with PH, of whom 50% were asymptomatic. Older children with PH were more often symptomatic than infants and toddlers. The most common presenting symptoms were melena and recurrent abdominal pain. PH was more frequently recognized during emergency surgery than with elective operations.[6]

Preoperative diagnosis of PH is often difficult. If identified before surgical management, it will facilitate planning of operative procedures including minimal access or endoscopic procedures. When located in the stomach or duodenum, PH can be suspected based on the typical findings of upper GI series. In contrast imaging PH is seen as a mass with broad base and smooth surface that is characteristic of extra-mucosal intramural tumors. Umbilication (visualization of a contrast-filled pit at the center of the lesion) is thought to represent the ductal remnant of PH. In magnetic resonance imaging (MRI) PH is seen as hyper- or iso-intense lesion in comparison with the orthotopic pancreas in unenhanced T1-weighted images and is seen as iso- or hypo-intense lesions in the T2-weighted images. On dynamic MRI, PH appears as an iso-intense lesion in comparison to the orthotopic pancreas in the arterial phase image. Radiologic findings of PH distal to the duodeno-jejunal junction are non-specific.[7]

In children with dyspepsia, upper GI endoscopy may reveal PH. As the lesion is located in the submucosal layer, endoscopy can be useful in the diagnosis as well as therapy (endoscopic resection whenever feasible).[8]

Another rare presentation of PH is an antenatally detected intra-abdominal cyst, postnatally complicated with unexplained of neonatal hypoglycemia. Hypoglycemia unresponsive to medical treatment

will improve on excision of the cyst containing PH.[9] It could be a differential diagnosis of acute cholangitis or obstructive jaundice when the ectopic tissue is located at the ampulla of Vater.[10]

Heinrich's histological classification of PH has no direct implications in the clinical management. It is very difficult to diagnose PH purely based on the macroscopic appearance during surgery, and hence histological confirmation is mandatory.

Table 1: Heinrich's classification of Pancreatic Heterotopia*

Type	Histological Description
1 (Complete)	Presence of typical pancreatic tissue comprising of acini, ducts, and islet cells
2 (Canalicular)	Presence of only ducts
3 (Exocrine)	Presence of only acinar tissue
4 (Endocrine)	Presence of only islet cells

* *Ref. Heinrich[11]*

CONCLUSION

PH is an uncommon entity, especially as a symptomatic lesion causing pediatric acute abdomen. It should be considered in the differential diagnosis of recurrent abdominal pain, lower GI bleed or intussusception. Diagnosis of PH would require histopathological confirmation after excision.

REFERENCES

[1] Yang ZH, Gao JB, Yue SW, Yang XH, Guo H. Synchronous ectopic pancreatoblastoma in a child: a case report. J Korean Med Sci. 2011 Jun; 26(6): 832-5.

[2] Chin NH, Wu JM, Chen KC, Lee TH, Lin CK, Chung CS. Pancreatic heterotopia in the small bowel: a case report and literature review. Pancreas. 2022 Jul 1; 51(6):700-4.

[3] Nguyen TL, Kapur S, Haerer SC, Gurda GT, Folkers ME. Unusual tissue - Unusual issue: Pancreatic heterotopia presenting as gastric outlet obstruction. Case Rep Gastro enterol. 2021 Mar 11; 15(1): 338-343.

[4] Zhou Y, Zhou S, Shi Y, Zheng S, Liu B. Endoscopic submucosal dissection for gastric ectopic pancreas: a single-center experience. World J Surg Oncol. 2019 Apr 16; 17 (1): 69.

[5] Tang XB, Liao MY, Wang WL, Bai YZ. Mesenteric heterotopic pancreas in a pediatric patient: A case report and review of literature. World J Clin Cases. 2018 Nov 26; 6 (14): 847-853.

[6] Persano G, Cantone N, Pani E, Ciardini E, Noccioli B. Heterotopic pancreas in the gastrointestinal tract in children: a single-center experience and a review of the literature. Ital J Pediatr. 2019 Nov 9; 45(1): 142.

[7] Kung JW, Brown A, Kruskal JB, Goldsmith JD, Pedrosa I. Heterotopic pancreas: typical and atypical imaging findings. Clin Radiol. 2010 May; 65(5): 403-7.

[8] Zhong YS, Shi Q, Yao LQ, Zhou PH, Xu MD, Wang P. Endoscopic mucosal resection/endoscopic submucosal dissection for gastric heterotopic pancreas. Turk J Gastro enterol. 2013; 24(4): 322-9.

[9] Seymore NM, Zoghbi B, Sotelo-Avila C, Farmakis SG. Pancreatic heterotopia in a neonatal abdomino-pelvic cyst. Pediatr Radiol. 2019 Mar; 49(3): 415-418.

[10] Christodoulidis G, Zacharoulis D, Barbanis S, Katsogridakis E, Hatzitheofilou K. Heterotopic pancreas in the stomach: a case report and literature review. World J Gastroenterol. 2007 Dec 7; 13(45): 6098-100.

[11] Heinrich H. Ein Beitrag zur Histologie des sogenannten akzessorischen Pankreas. Virchows Arch A Pathol Anat Histopathol. 1909; 198: 392–440.

Address for communication: *Dr Krishnakumar Govindarajan, Email: kkpeds@gmail.com*

Received 5 June 2024; Accepted 4 August 2024

Acknowledgements : None
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : None (Report of routine clinical care)

Citation: *Adarsh SA, Toi PC, Govindarajan KK. Pancreatic heterotopia. Pediatr Surg Trop 2024; 1(4): 258-261.*

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 262-268
DOI: 10.5281/zenodo.13893578

Clinical Study

Surgical Management of Thyroid Illnesses in Children

Edwin Alvarez Torres, Humberto Lugo Vicente

Section of Pediatric Surgery, Department of Surgery, University of Puerto Rico School of Medicine, San Juan, Puerto Rico.

Keywords
Calcium supplements
Endocrine surgery
Hashimoto thyroiditis
Hemostatic sealant
Hyperthyroidism
Hypothyroidism
Neuromonitoring
Papillary carcinoma
Thyroid disorders
Thyroidectomy

Abbreviations
CLND - Central lymph node dissection
FNAC - Fine needle aspiration cytology
INM - Intra-operative neuromonitoring
MNG - Multi-nodular goiter
PC - Papillary carcinoma
RLN - Recurrent laryngeal nerve
SLN - Superior laryngeal nerve
TE - Thyroidectomy
TSH - Thyroid stimulating hormone

Abstract

Background: *Thyroid disorders represent a significant proportion of pediatric endocrine problems. Surgical intervention is often indicated in thyroid cancers, hyperthyroidism and solitary non-toxic nodules.*

Methods: *This study outlines the surgical experience with 116 pediatric patients who underwent a total of 124 thyroidectomies from 2001 to 2018 at the University Pediatric Hospital in San Juan, Puerto Rico. Analyzed data include patient age, gender, race, weight, medical history, type of thyroid disorder, nodule size, pre-operative ultrasound and fine-needle aspiration cytology (FNAC) findings, type of thyroid resection, operative duration, novel surgical techniques, recovery period, and complications.*

Results: *The diagnoses included well-differentiated thyroid carcinoma (39%), Hashimoto disease (16%), follicular adenoma (14%), and Graves' disease (12%). The most frequent procedures were total thyroidectomy with central lymph node removal (44%), hemithyroidectomy with isthmusectomy (35%), and total thyroidectomy alone (10%). Complications occurred in 7 patients. The common complications include recurrent laryngeal nerve injury (2%) and neck hematoma (2%).*

Conclusions: *Pediatric patients with thyroid disorders can benefit from thyroidectomy based on the specific condition and its stage. Accurate diagnosis, appropriate surgical treatment, and the application of innovative techniques resulted in 124 successful thyroidectomies with only seven complications. The use of intra-operative nerve monitoring and vascular hemostatic sealants reduced the risk of recurrent laryngeal nerve injury and shortened operative times. Postoperative oral calcium and calcitriol therapy effectively mitigated the risk of postoperative hypocalcemia following total gland removal.*

INTRODUCTION

Thyroid disorders forms a significant proportion of pediatric endocrine problems, ranking globally as the second most prevalent pathology. These disorders affect 37 out of 1,000 school-aged children in the United States, leading to metabolic abnormalities that impact growth and development. The clinical outcomes of thyroid illness vary depending on the child's age and the nature of the disorder.[1] Surgical management is common in thyroid disorders such as goiter, solitary non-toxic nodule, malignancy and auto-immune diseases (e.g. Graves disease). Surgical options range from removing the entire gland (total thyroidectomy) to removing a single lobe (hemithyroidectomy) or the isthmus (isthmusectomy). The extent of resection is often determined by the nature of the illness. Pediatric surgeons in the USA usually do 2-3 thyroidectomies per year. While thyroidectomies are generally safe and effective, they do carry specific risks such as hypocalcemia, recurrent laryngeal nerve (RLN) injury, cervical hematoma, wound infection and hypertrophied scars.

Over the past decade, several new techniques have emerged to reduce the complications of thyroid operations. One such technique is intraoperative neuromonitoring (INM) of the RLN that has been shown to improve the surgical outcomes.[2] This method enables real-time assessment of the nerve function before and after gland removal. It enables complete removal of gland with reduced incidence of RLN injury.[2] Another innovative technique is the hemostatic vascular sealants, which aim to minimize postoperative bleeding and expedite the surgical process.[3] Despite its widespread use, literature indicates that their effectiveness may be limited.[3]

This study is aimed to document the surgical experience of the authors in performing thyroid operations and to draw educative inferences from that. Additionally, the study sought to identify the surgical complications encountered during and after thyroid operations. The application of novel surgical techniques to improve the outcomes and to reduce the complications was also examined.

PATIENTS AND METHODS

This retrospective study involves 116 consecutive children who underwent a total of 124 thyroid operations between 2001 and 2018, under the care of the senior author (HLV) at the University Pediatric Hospital of Puerto Rico. Collected data include patient age, gender, race, weight, medical history, nature of the thyroid disorder, lesion size, pre-operative imaging details, fine-needle aspiration cytology (FNAC) results, type of resection, surgical duration, novel techniques used, hospital stay and complications.

All the patients were Hispanics. Among them, 90 (79%) were female, and 26 (21%) were male. The mean age at operation was 15 years (range 3-21 years) and the mean weight was 60 kg (range 16-150 kg).

RESULTS

The list of pathologies, nature of surgical operations done and complications are summarized in Tables 1 to 3.

Table 1. Frequency of thyroid disorders

Pathology	n (%)
Papillary Carcinoma	54 (39%)
Hashimoto Disease	23 (16%)
Follicular Adenoma	20 (14%)
Graves Disease	17 (12%)
Nodular Hyperplasia	11 (8%)
Diffuse Nodular Toxic Goiter	5 (4%)
Oncocytic Adenoma	4 (3%)
Adenomatoid Nodule	3 (2%)
Medullary Carcinoma	1(1%)
C-cell Hyperplasia	1(1%)
Multi-nodular Goiter	1(1%)

Table 2. Surgical intervention of thyroid disorders in children

Nature of Operation	n (%)	Mean OR time (min)	Mean Hospital stay (days)
Total TE + CLND	55 (44%)	123	1
Hemi-TE	44 (35%)	97	1
Total TE	13 (10%)	150	2
Completion TE†	7 (6%)	98	1
Lymphadenectomy	3 (2%)	-	-
Central TE*	2 (2%)	60	1
Total operations	124		

*TE - Thyroidectomy, CLND - Central lymph node dissection, OR - Operating Room. * Also known as isthumectomy, †Done within 7 days of initial operation*

Table 3. Complications of thyroid surgeries

Complications	n (%)
Injury to the recurrent laryngeal nerve*	2 (1.6%)
Hematoma	2 (1.6%)
Hypocalcemia	1 (0.8%)
Pulmonary edema	1 (0.8%)
Stitch abscess / silk granuloma	1 (0.8%)
Total	7

** Intraoperative neuromonitoring was done in 74cases*

All the patients with Graves disease were treated with total thyroidectomy. In 7 children, reversible complications occurred. There were 2 cases of recurrent laryngeal nerve (RLN) injury (1.6%). In the first case, INM detected nerve injury during total thyroidectomy for Graves disease with a 4kg goiter. The divided nerve was immediately repaired using an operating microscope, and the patient recovered completely 5 months later. The second case developed post-operative hoarseness due to RLN palsy, which resolved after 3 months. There were also two cases of hematoma, both requiring neck re-exploration within 24 hours of the original operation. They underwent hematoma drainage and fibrin glue application. One patient developed symptomatic hypocalcemia. Patients who underwent total thyroidectomy were post-operatively treated with 2.4g of oral calcium carbonate and 0.5 µg of oral vitamin-D supplement for 1 month, with the dosage being reduced gradually by every week.

INM of the RLN was used in 74 resections, and hemostatic vascular sealants in all the 124 cases. The mean operating time for various operations is shown in Table 2. Total thyroidectomy done for Graves disease took the longest operating time with a mean of 150 minutes. These patients also required the longest post-surgical hospital stay, averaging 2 days, primarily to manage temporary hypocalcemia.

DISCUSSION

The overall risk of post-operative complications after thyroidectomy is approximately 2%. The common complications include operative damage to the RLN, hypocalcemia and neck hematoma. Permanent hypoparathyroidism is a serious but rare complication of total thyroidectomy. It often requires lifelong supplementation of vitamin-D.[4] Transient hypoparathyroidism is more common due to perioperative reversible ischemia of the parathyroid glands. Fortunately, only 1-4% of the parathyroid deficiencies occurring after thyroidectomy are permanent.[4] In our series, all those who underwent total thyroidectomy received oral supplementation of 2.4g of calcium carbonate and 0.5µg of vitamin-D, resulting in a low incidence of postoperative hypocalcemia.

Thyroid surgery may cause temporary or permanent damage to the RLN and superior laryngeal nerve (SLN), resulting in hoarseness of voice, dysphonia, dysphagia, pulmonary aspiration and dyspnoea.[5,6]. Bilateral RLN damage can be life-threatening due to airway obstruction.[5,7] The RLN is particularly susceptible to operative injury during the separation of the thyroid gland at the

ligament of Berry. RLN injury occurs in 3-11% of cases. Factors influencing nerve damage include the nature of pathology (benign vs. malignant), the extent of thyroid resection (lobectomy vs. total thyroidectomy), the number of attempt of resection (primary vs. reoperation) and the experience of the surgeon.[8] Complications are common in patients with malignant tumors and lymph node involvement, or in those undergoing re-operation with neck dissection. Proper surgical exposure and identification of both nerves during surgery is crucial to avoid damage.[5,9] Over the past decade, INM has evolved as the technique of choice. In this technique, electrodes are placed through the endo tracheal tube to monitor the nerve function during the surgical dissections.[6] INM allows not only intra-operative identification of nerves but also enable recording their function, thus providing legal protection in case of damage.[9] INM can also locate the site of injury and determine if the injury is repairable, as demonstrated in one of our cases. INM also makes thyroid surgery safer for the inexperienced young surgeons and trainees.[10] It helps in identifying anatomical variations present in less than 5% of patients.[5] INM has become an asset in complex thyroid dissections, such as those involving sub-sternal goiters, re-do operations, and total thyroidectomy for Graves disease.[8,11,12] Electric nerve testing at the end of thyroidectomy can predict postoperative nerve function and prevent bilateral vocal cord paralysis.[5,10] Despite the lack of class-1 evidence from randomized controlled clinical trials, INM may be considered as the standard-of-care in pediatric thyroid operations. Currently, it is the only method available to verify the functional intactness of the RLN and SLN during the procedure.[6] Although rare, hypocalcemia and vocal cord paralysis can significantly impact the quality of life. RLN injury is a major cause of medico-legal litigation in thyroid and parathyroid surgeries, underscoring the need for INM.[2,13,14]

Hemostatic sealants are important in preventing post-operative bleeding, which can be life-threatening due to acute airway obstruction. It occurs in 1.5-4% of thyroidectomies.[15,16] Causes of post-operative bleeding include ligature slippage, re-opening of the cauterized veins due to retching or Valsalva maneuver during recovery, increased blood pressure, continued capillary oozing from the thyroid gland or inadequate hemostasis.[15,16] Neck hematoma may cause airway obstruction from tracheal compression, requiring urgent surgical intervention or sometimes immediate bedside decompression.[17,18] Hematomas typically develop within 24 hours of surgery, though 20% may occur as late as 3 days.[19] Patients with post-operative neck hematoma present with dyspnea, neck pain, dysphagia or local swelling.[17,19] Early recognition and prompt intervention, including intubation or tracheotomy, are essential to save life.[17-19] Many of the hematomas are caused by arterial bleeding from the upper pole vessels. The frequency of hematoma does not reduce with the usage of neck drains.[20] The risk of postoperative hemorrhage is a deterrent for outpatient thyroid surgery or early discharge from hospital.[15,20] Risk factors of neck hematoma include age, sex, race, obesity, geographic region, co-morbidities, Graves disease, bleeding disorders, previous neck operations and the nature of current surgical procedure. Total thyroidectomy, sub-sternal thyroidectomy and radical neck dissection are associated with increased frequency of hematoma.[16,19,21] Hospital characteristics such as the type of hospital (teaching vs. non-teaching), location and patient volume do not correlate with the risk of hematoma.[16,19,21] Parathyroidectomy has a lower incidence of neck hematoma as compared to thyroidectomy.

Thyroid nodules in children are four times more likely to be cancerous than in adults. They warrant estimation of serum levels of thyroid-stimulating hormone (TSH) and calcitonin, ultrasonography and FNAC. Elevated calcitonin level is suggestive of medullary thyroid cancer. Micro-calcification,

indistinct margins of the nodules and variable echo-texture are the sonographic features of malignant nodules.[22] FNAC is the most accurate method of diagnosing thyroid malignancy, but it is often limited by the nodule size.[23] The risk of malignancy increases in iodine-deficient regions or with autoimmune thyroiditis.

Papillary carcinoma (PC) is the most common thyroid malignancy in children. It has excellent overall prognosis with prompt surgery and radio-iodine therapy.[24-26]. It often presents with multi-focal nodules with neck metastasis.[26,27] Distant metastasis is more common in children than in adults.[24,25] PC spreads sequentially from the thyroid to the central lymph node and then to ipsi-lateral nodes. Routine central lymph node dissection (CLND) during total thyroidectomy for PC reveals impalpable histological metastasis in 50-60% of cases.[26-28] Cervical lymph node involvement in PC does not affect the overall survival, because the residual nodal disease is easily treated with radio-active iodine.[27,28] CLND enables accurate staging and improves the efficacy of radio-iodine treatment.[24-27] Routine prophylactic lateral neck dissection is not recommended in children with PC.[26,27] Rather, routine CLND is recommended in children with palpable nodes to increase disease-free survival.[24,25] A lymph node ratio > 0.45 correlates with a higher risk of loco-regional recurrences.[26,28] CLND decreases tumor burden and improves survival in children with positive nodal metastasis.[26,27] Total thyroidectomy with CLND enables accurate tumor staging and effective radioiodine therapy.[24,27]

Graves disease is the most common cause of hyper thyroidism. Total thyroidectomy is indicated when thyrotoxicosis is refractory to medical treatments such as the anti-thyroid drugs and radio-active iodine.[30,31] In the past 20 years, total thyroidectomy is increasingly preferred over medical treatment due to its near zero recurrence rate, predictable postoperative hypothyroidism and low complication rate.[30,31]

Hashimoto thyroiditis is the most common auto-immune disorder of the endocrine system. It is often complicated by malignancies, especially the PC.[32] Primary treatment of Hashimoto thyroiditis is conservative, although thyroidectomy is often done for malignancy or symptomatic goiters.[32,33]

Non-toxic multinodular goiter (MNG) refers to diffuse thyroid enlargement with multiple non-functioning nodules. It primarily affects adolescents.[34,35] About 8% of MNG may undergo malignant transformation, especially in familial cases or in those with previous cervical irradiation.[34] In MNG, total thyroidectomy has lower morbidity than subtotal thyroidectomy.[13,36]

Follicular adenomas are common benign thyroid neoplasms that present as solitary nodules. Differentiating them from follicular carcinoma is often difficult, yet important.[37] Hemithyroidectomy is recommended for follicular adenoma diagnosed by FNAC. However, if histology of the excised specimen shows malignancy, completion (total) thyroidectomy is essential. Nodular thyroid hyperplasia involves non-cancerous growth affecting the thyroid gland, potentially causing enlargement of one or both lobes.

CONCLUSION

Children with thyroid disorders are significantly benefited from thyroidectomy. Among the 124 thyroidectomies there were only 7 complications. This improved outcome is attributable to INM, hemostatic sealants and oral supplementation of calcium plus calcitriol which were instrumental in reducing the incidence of RLN injury, neck hematoma and hypocalcemia, respectively.

REFERENCES

[1] Oyenusi EE, Ajayi EO, Akeredolu FD, Oduwole AO. Pattern of thyroid disorders in children and adolescents

seen at the Lagos University Teaching Hospital, Nigeria, over a 10-year period. Niger Med J. 2017 May-Jun; 58 (3): 101-106.

[2] Barczyński M, Konturek A, Stopa M, Hubalewska-Dyde jczyk A, Richter P, Nowak W. Clinical value of intraoperative neuromonitoring of the recurrent laryngeal nerves in improving outcomes of surgery for well-differentiated thyroid cancer. Pol Przegl Chir. 2011Apr;83(4):196-203.

[3] Scaroni M, von Holzen U, Nebiker CA. Effectiveness of hemostatic agents in thyroid surgery for the prevention of postoperative bleeding. Sci Rep. 2020 Feb 4; 10(1): 1753.

[4] McLeod M, Doherty GM. Thyroid Surgery. In: Evans SRT. (ed) Surgical pitfalls - prevention and management. W.B. Saunders, New York. 2009. pp 397-405.

[5] Deniwar A, Kandil E, Randolph G. Electrophysiological neural monitoring of the laryngeal nerves in thyroid surgery: review of the current literature. Gland Surg. 2015 Oct; 4(5): 368-75.

[6] Cirocchi R, Arezzo A, D'Andrea V, Abraha I, Popivanov GI, Avenia N, Gerardi C, Henry BM, Randolph J, Barczyński M. Intraoperative neuromonitoring versus visual nerve identification for prevention of recurrent laryngeal nerve injury in adults undergoing thyroid surgery. Cochrane Database Syst Rev. 2019 Jan 19; 1(1): CD012483.

[7] Henry BM, Graves MJ, Vikse J, Sanna B, Pękala PA, Walo cha JA, Barczyński M, Tomaszewski KA. The current state of intermittent intraoperative neural monitoring for prevention of recurrent laryngeal nerve injury during thyroidectomy: a PRISMA-compliant systematic review of overlapping meta-analyses. Langenbecks Arch Surg. 2017 Jun; 402(4): 663-673.

[8] Wong KP, Mak KL, Wong CK, Lang BH. Systematic review and meta-analysis on intra-operative neuro-monitoring in high-risk thyroidectomy. Int J Surg. 2017 Feb; 38: 21-30.

[9] Zhang D, Pino A, Caruso E, Dionigi G, Sun H. Neural monitoring in thyroid surgery is here to stay. Gland Surg. 2020 Jan; 9(Suppl 1): S43-S46.

[10] Zheng S, Xu Z, Wei Y, Zeng M, He J. Effect of intraoperative neuromonitoring on recurrent laryngeal nerve palsy rates after thyroid surgery - A meta-analysis. J Formos Med Assoc. 2013 Aug; 112(8): 463-72.

[11] Barczyński M, Konturek A, Pragacz K, Papier A, Stopa M, Nowak W. Intraoperative nerve monitoring can reduce prevalence of recurrent laryngeal nerve injury in thyroid reoperations: results of a retrospective cohort study. World J Surg. 2014 Mar; 38(3): 599-606.

[12] Wojtczak B, Sutkowski K, Kaliszewski K, Barczyński M, Bolanowski M. Thyroid reoperation using intraoperative neuromonitoring. Endocrine. 2017 Dec; 58(3): 458-466.

[13] Makay O. Less than total thyroidectomy for goiter: when and how? Gland Surg. 2017 Dec; 6(Suppl 1): S49-S58.

[14] Barczyński M, Konturek A, Pragacz K, Papier A, Stopa M, Nowak W. Intraoperative nerve monitoring can reduce prevalence of recurrent laryngeal nerve injury in thyroid reoperations: results of a retrospective cohort study. World J Surg. 2014 Mar; 38(3): 599-606.

[15] Lee HS, Lee BJ, Kim SW, Cha YW, Choi YS, Park YH, Lee KD. Patterns of Post-thyroidectomy Hemorrhage. Clin Exp Otorhinolaryngol. 2009 Jun; 2(2): 72-7.

[16] Suzuki S, Yasunaga H, Matsui H, Fushimi K, Saito Y, Yamasoba T. Factors associated with neck hematoma after thyroidectomy: a retrospective analysis using a Japanese inpatient database. Medicine (Baltimore). 2016 Feb; 95 (7): e2812.

[17] Adigbli G, King J. Airway management of a life-threatening post-thyroidectomy haematoma. BMJ Case Rep. 2015 Dec 15; 2015: bcr2015213578.

[18] Materazzi G, Ambrosini CE, Fregoli L, De Napoli L, Frus taci G, Matteucci V, Papini P, Bakkar S, Miccoli P. Prevention and management of bleeding in thyroid surgery. Gland Surg. 2017 Oct; 6(5): 510-515.

[19] Dehal A, Abbas A, Hussain F, Johna S. Risk factors for neck hematoma after thyroid or parathyroid surgery: ten-year analysis of the nationwide inpatient sample database. Perm J. 2015 Winter; 19(1):22-8.

[20] Fan C, Zhou X, Su G, Zhou Y, Su J, Luo M, Li H. Risk factors for neck hematoma requiring surgical re-intervention after thyroidectomy: a systematic review and meta-analysis. BMC Surg. 2019 Jul 24;19(1):98.

[21] Salem FA, Bergenfelz A, Nordenström E, Dahlberg J, Hessman O, Lundgren CI, Almquist M. Evaluating risk factors for re-exploration due to postoperative neck hematoma after thyroid surgery: a nested case-control study. Langenbecks Arch Surg. 2019 Nov; 404 (7): 815-823.

[22] Giovanella L, Imperiali M, Ferrari A, Palumbo A, Furlani L, Graziani MS, Castello R. Serum thyroglobulin reference values according to NACB criteria in healthy subjects with normal thyroid ultrasound. Clin Chem Lab Med. 2012 Jan 26; 50(5): 891-3.

[23] Alvarado-Santiago M, Alvarez-Valentin D, Bermudez RO, Gonzalez-Sepulveda L, Allende-Vigo M, Rodriguez SE, Tumanyan SR. Fine-needle thyroid aspiration biopsy: clinical experience at the endocrinology clinics of the University Hospital of Puerto Rico. PR Health Sci J. 2017 Mar; 36(1): 5-10.

[24] Zong Y, Li K, Dong K, Yao W, Liu G, Xiao X. The surgical choice for unilateral thyroid carcinoma in pediatrics: Lobectomy or total thyroidectomy? J Pediatr Surg. 2018 Dec; 53(12): 2449-2453.

[25] Parisi MT, Eslamy H, Mankoff D. Management of differentiated thyroid cancer in children: Focus on the American Thyroid Association Pediatric Guidelines. Semin Nucl Med. 2016 Mar; 46(2): 147-64.

[26] Machens A, Elwerr M, Thanh PN, Lorenz K, Schneider R, Dralle H. Impact of central node dissection on post-operative morbidity in pediatric patients with suspected or proven thyroid cancer. Surgery. 2016 Aug; 160(2): 484-92.
[27] Sakorafas GH, Sampanis D, Safioleas M. Cervical lymph node dissection in papillary thyroid cancer: current trends, persisting controversies, and unclarified uncertainties. Surg Oncol. 2010 Jun; 19(2): e57-70.
[28] Rubinstein JC, Dinauer C, Herrick-Reynolds K, Morotti R, Callender GG, Christison-Lagay ER. Lymph node ratio predicts recurrence in pediatric papillary thyroid cancer. J Pediatr Surg. 2019 Jan; 54(1): 129-132.
[29] Fridman M, Krasko O, Branovan DI, Dabryian S, Pisa renko A, Lo CY, Lam AK. Factors affecting the approaches and complications of surgery in childhood papillary thyroid carcinomas. Eur J Surg Oncol. 2019 Nov; 45(11): 2078-2085.
[30] Barakate MS, Agarwal G, Reeve TS, Barraclough B, Robin son B, Delbridge LW. Total thyroidectomy is now the preferred option for the surgical management of Graves' disease. ANZ J Surg. 2002 May; 72(5): 321-4.
[31] Bobanga ID, McHenry CR. Treatment of patients with Graves disease and the appropriate extent of thyroidectomy. Best Pract Res Clin Endocrinol Metab. 2019 Aug; 33(4): 101319.
[32] Caturegli P, De Remigis A, Chuang K, Dembele M, Iwama A, Iwama S. Hashimoto's thyroiditis: celebrating the centennial through the lens of the Johns Hopkins hospital surgical pathology records. Thyroid. 2013 Feb; 23(2): 142-50.
[33] Gyory F, Lukacs G, Juhász F, Mezösi E, Szakall S, Vegh T, Máth J, Balazs G. Surgically treated Hashimoto's thyroiditis. Acta Chir Hung. 1999; 38(3-4): 243-7..
[34] Ríos A, Rodríguez JM, Canteras M, Galindo PJ, Balsalobre MD, Parrilla P. Risk factors for malignancy in multinodular goitres. Eur J Surg Oncol. 2004 Feb;30(1):58-62.
[35] Al-Fifi S, Rodd C. Multinodular goiter in children. J Pediatr Endocrinol Metab. 2001 Jun; 14(6): 749-56.
[36] Yoldas T, Makay O, Icoz G, Kose T, Gezer G, Kismali E, Tamsel S, Ozbek S, Yılmaz M, Akyildiz M. Should subtotal thyroidectomy be abandoned in multinodular goiter patients from endemic regions requiring surgery? Int Surg. 2015 Jan; 100(1): 9-14.
[37] Norris JJ, Farci F. Follicular Adenoma. [Updated 2023 Apr 23]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 Jan. {Available from: https://www.ncbi.nlm.nih.gov/books/NBK559320/}

Address for communication: *Dr. Humberto Lugo Vicente, Email: humberto.lugo@upr.edu*

Received: 12 July 2024; Accepted: 20 September 2024

Acknowledgements : None
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : None (Retrospective observational study describing routine clinical care)

Citation: *Torres EGA, Vicente HL. Surgical management of thyroid illnesses in children. Pediatr Surg Trop 2024; 1(4): 262-268.*

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 269-277
DOI: 10.5281/zenodo.13893592

Review Article

Conservative Management of Omphalocele Using Escharotic Agents Available in Resource-Constrained Settings: A Scoping Review

Florent Tshibwid A Zeng[1], Nathalie Dinganga Kapessa[1], Alagie Baldeh[2], Luc-Beau Ihaku Kombe[3], Gracia Mitonga Kamwangen[4], Trésor Kibangula Kasanga[1], Willy Arung Kalau[1]

[1]*Department of Surgery, Université de Lubumbashi, Lubumbashi, Democratic Republic of the Congo.*
[2]*Department of Surgery, Edward Francis Small Teaching Hospital, Banjul, Gambia.*
[3]*Medical Center Lumière, Lubumbashi, Democratic Republic of the Congo.*
[4]*Department of Surgery, Mwangeji Provincial General Hospital, Kolwezi, Democratic Republic of the Congo.*

Keywords
Non-operative management
Escharotic agent
Exomphalos
Omphalocele
Resource-limited Setting

Abbreviations
DE - disodium eosin
GV - gentian violet
HIC - High-income country
LMIC - *Low-middle-income country*
NA - *Acacia nilotica*
PI - Povidone-iodine
SSD - Silver sulfadiazine

Abstract

Background: *Conservative management of omphalocele is an accessible method in resource-constrained settings. This review summarizes the literature on the usage of escharotic agents in omphalocele.*

Methods: *Articles published between January 2000 and August 2023, were screened following the PRISMA guidelines.*

Results: *Twenty studies met the inclusion criteria. Five of them used disodium eosin (DE), in 249 patients, five used povidone-iodine (PI) in 70, four used silver sulfadiazine (SSD) in 38, four used honey in 55, one used gentian violet (GV) in 27, and two used Acacia nilotica (AN) paste in 47. The length of hospital-stay ranged from 5 to 239 days, and the time to full enteral feeding ranged from 2 days to 45 weeks. The overall complication rate (n=128 patients) was 26.4% for all escharotic agents. Sac related complications occurred in 80 patients, gastrointestinal complications in 42, and drug-related toxicity (especially iodine and silver) in 4 cases. Sac infection was common with DE and SSD, but rare with GV. Mortality appears to be increased with protective dressing (e.g. DE, PI, and honey) as compared to open dressing (e.g. GV and AN paste).*

Conclusion: *Escharotic agents easily available in resource-constrained settings are valuable therapeutic aids in the non-operative management of omphalocele. However, meaningful comparison of various agents is precluded by the lack of standardized treatment and uniform reporting.*

INTRODUCTION

Omphalocele (exomphalos) is the most common congenital defect of the abdominal wall, with an incidence of 1 in 5,000 live births.[1] It occurs due to defective fusion of primitive embryonic folds between the sixth and the tenth week of gestation. During this time, several organs are in a critical phase of their formation[2] Disorders affecting the closure of the abdominal wall can also affect these organs, resulting in associated malformations in 30 to 80% of patients with omphalocele.[1]

The management of omphalocele in high-income countries (HIC) has evolved into primary surgical closure in minor omphalocele and staged repair in major omphalocele. Conservative management (delayed closure or non-operative management) as an alternative, is reserved for those cases with life-threatening associated congenital malformations.[3,4]. With improvements in prenatal diagnosis, neonatal intensive care, and neonatal anesthesia, the prognosis of this condition has improved, considerably and mortality is now mainly due to associated congenital malformations, especially the cardiac anomalies.[5] On the contrary, in low- and middle-income countries (LMIC), the management of omphalocele is still a challenge.[6] This is due to the scarcity of accurate prenatal diagnosis. Many patients are born far from dedicated tertiary centers, leading to subsequent rupture of the sac during transport, infection, dehydration, hypothermia and hypoglycemia.[6] Even when the affected patients attend specialist neonatal-care facilities, operative management is still challenging due to the lack of pediatric anesthetists and pediatric surgeons, leading to an increased mortality.[7] In a Senegalese cohort, rupture of sac and primary closure were identified as the risk factors for mortality in omphalocele, no matter if it were major or minor type.[8] This led to the adaptation of conservative management as the gold standard of omphalocele management in some African centers, regardless of the size of the abdominal wall defect or associated anomalies.[9]

From the historical usage of alcohol and mercurochrome as escharotic agents, techniques of conservative management are now more innovative and safe. Modern conservative therapies include nanocrystalline-silver applications, hydrogel dressings and negative pressure therapy.[4] However, some of the newer artifices are not easily available in LIMC. With this background, we reviewed the published evidence on the outcomes of treatment with commonly available topical agents in LMIC.

METHODS AND MATERIALS

Search Strategy

Two authors searched PubMed and Google Scholar for publications on the conservative management of omphalocele using the strategy detailed in the appendix-I.

Eligibility Criteria

We considered studies on conservative management of omphalocele published between January 2000 and August 2023 that used topical agents easily available in LIMC. Chosen topical agents included *Acacia nilotica* (AN) paste, di-sodium eosin (DE), gentian violet (GV), honey, povidone-iodine (PI) and silver sulfadiazine (SSD). Excluded were theoretical articles, study using any other topical agents, reports using negative pressure, and publications that are not in English or French.

Study Selection

Two authors independently selected articles using Rayyan® online software following the PRISMA guidelines for scoping review.[10] (Fig. 1) Duplicate publications were manually removed.

Data Extraction and Analysis

Two authors extracted data on a Microsoft Excel sheet using OfficeTM-2019. Extracted variables include number of patients, escharotic agent used, frequency of topical applications, use of a supportive dressing, time to full enteral feeding, length of hospital stay, discharge criteria, time to epithelia-

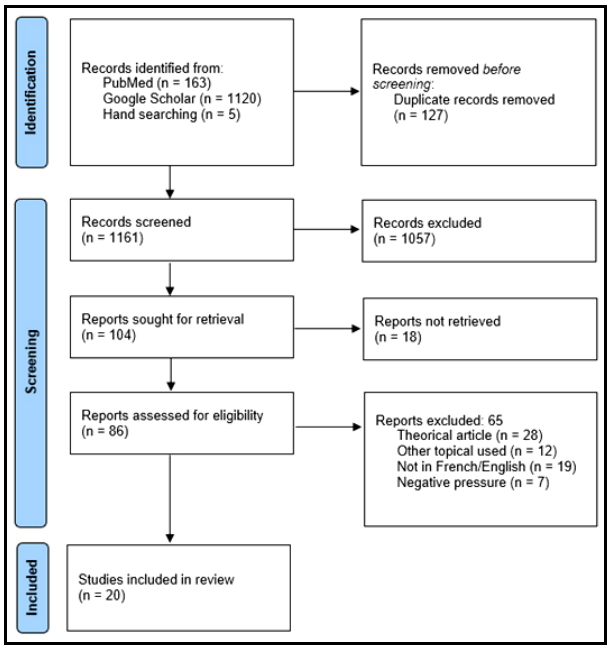


Fig 1. *PRISMA flowchart*

lization, toxicity of the therapeutic agents, complications, mortality and its causes. Results were grouped on the basis of escharotic agents.

RESULTS

This review included 20 studies including 485 patients.[9,11-29] Table 1 summarizes the treatment details of various agents. Discharge criteria were not stated in 9 studies.[9,14,16,22,23,25-28] When stated, discharge criteria were based on patient stability (absence of life-threatening associated malformations or need of mechanical ventilatory assistance),[13,15,17,18,20,21,24,29] nutritional independence (full enteral feeding with weight gain),[15,17–21,24,29] and assurance of good home-care of the sac (ability of parents to do dressing or evidence of sac epithelialization).[11,12,20,21,24] Details on the time to full oral feeds was not stated in 11 publications.[9,11-14,16,22,23,26,28,29]. For the remaining papers, it varied according to the presence of life-threatening comorbidities. It ranged from 2 days to 45 weeks.[15,17-21,24,25,27]

The overall complication (128 patients) rate was 26.4% for all escharotic agents.[9,11-29] Sac related complications (bacterial colonization, infection and rupture) occurred in 80 patients, gastro-intestinal complications (ileus and gastro-jejunal perforations) in 42, and drug-related toxicity (especially iodine and silver) in 4 cases.(Table 2) Mortality was reported in 114 patients (23.5%) (Table 3)

DISCUSSION

The sac of omphalocele constituted of the outer amnion, the intermediate Wharton's jelly and the inner peritoneum. Conservative management with topical agents aims to transform the sac into a ventral hernia through progressive epithelialization of the amnion layer.[4] This procedure has evolved ever since its first description in 1899 by Ahlfeld.[30] Nowadays, several escharotic agents have been proven to be safe. However, some may not be easily available or affordable in resource-constrained areas, where omphalocele still has high mortality.[6]

Compared to surgical repairs, conservative treatment of giant omphalocele has the advantages of early full enteral feeding, lower need for ventilatory support, and shorter hospital stay.[31,32] This perfectly suits the resource-limited settings where parenteral nutrition and artificial respirators are often not available.[6] Furthermore, non-operative management has lower morbidity and mortality than surgical repairs.[31,32]

Concerning the preparation of different topical agents, there is considerable scope for improvement by standardizing their concentration. Many authors[10,12] used DE, PI and AN paste without dilution, while others[11] diluted it. Considerable variations were also noted in the frequency of application. Standardization of the concentration and frequency of application of a given topical agent would allow future multicentric studies, thus contributing to better understanding of its therapeutic role.

Table 1. Use of topical escharotic agents in omphalocele

Escharotic agent	Year	n	Preparation Strength	Application frequency	Protective Dressing of the sac	Hospital stay (days)	Time to full feeds (weeks)
Disodium eosin							
Ngom[9]	2004	50	2% aq.sol	BID	NR	NR	NR
Andriamanarivo[14]	2012	10	2% aq.sol	BID	GD	7 - 18	11 - 14
Kouame[12]	2014	173	2% aq.sol	BID	NR	21 ± 6	10 ± 1
Habou[13]	2017	13	2% aq.sol	QAD	GD+EB	4 - 17	6 - 11
Kasanga[11]	2021	3	2% aq.sol in 500ml NS	TID	NR	30 - 33	8 - 13
Povidone-iodine							
Whitehouse[15]	2010	6	1–2.5% aq.sol	OD / QAD	GD + Brace	7 - 111	12 - 24
Pandey[17]	2014	24	10% aq.sol	BID	Suspension+ Sand bags	14 ± 3	NR
Eltayeb[18]	2015	12	5% aq.sol	BID	None	19 ± 13	7 ± 6
Malhotra[16]	2016	1	5% aq.sol	OD	GD	LFU	LFU
Rattan[19]	2020	27	NR	OD / QAD	GD	18 ± 3	10 ± 2
Silver sulfadiazine							
Lee[24]	2006	15	1% cream	OD	GD + EB	5 - 239	NR
Lewis[26]	2010	2	1% cream	OD	GD + EB	LFU	LFU
Ein[25]	2012	20	1% cream	OD	NR	NR	20
Echendu[27]	2021	1	1% cream	OD	GD + EB	14	16
Honey							
Nicoara[21]	2014	5	MHG	QAD / QW	GD + EB	21 - 121	7 - 17
Ekot[23]	2017	1	Natural honey	OD	GD+ EB	14	6
Bode[20]	2018	25	Natural honey	OD	Insect net	NR	4 - 6
Lawrence[22]	2021	24	MHG	QAD / BI7D	GD + EB	7 - 66	4 - 28
Gentian violet							
Mitul[28]	2012	27	1% aq.sol	BID	None	21 - 28	4 - 6
Acacia nilotica paste							
Moustafa[29]	2005	35	25mg AN in 15ml GV	BID	None	5 - 15	6 - 8
Eltayeb[18]	2015	12	1mg AN in 1 ml GV	BID	None	16 ± 8	8 ± 5

AN - Acacia nilotica; aq.sol - Aqueous solution; BI7D - Bis in 7 die (twice weekly); BID - Bis in die (twice a day); EB - Elastic bandage; GD - Gauze dressing; GV - Gentian violet; LFU - Lost to follow-up; MHG - Medical grade honey gel; NR - Not reported; NS - Normal saline; OD - Omne in die (Once daily); QAD - Quaque altera die (every other day); QW - Quaque week (Once weekly); TID - Ter in die (thrice a day)

Table 2. Complications of non-operative management of omphalocele

Topical Agent	Ref.	n	Complications n (%)	Mortality n (%)
Disodium eosin	(9,11–14)	249	Ileus 40 (16 %) Sac infection 38 (15 %)	64 (26 %)
Povidone iodine	(15–19)	70	Iodine intoxication 2 (2.8%) Sac infection/sepsis 3 (4%) Sac rupture 2 (2.8%)	19 (27%)
Silver sulfadiazine	(24–27)	38	Blood silver elevation 2 (5%) Sac infection/sepsis 6 (16%) Sac rupture 5 (13%) Multiple* 1 (2.6%)	7 (18 %)
Honey	(20–23)	55	Sac bacterial colonization 15 (27%) Sac infection/sepsis 3 (5.4%) Sac rupture 1 (1.8%)	12 (22 %)
Gentian violet	(28)	27	Sac rupture 3 (11%)	4 (15 %)
***Acacia nilotica* paste**	(18,29)	47	Sac infection 2 (4%) Sac rupture 2 (4%)	8 (17%)

** Includes gastric necrosis, jejunal perforation, acute kidney injury and aspiration pneumonia*

Table 3. Mortality of omphalocele treated with various topical agents

	DE (n=249)	PI (n=70)	SSD (n=38)	Honey (n=55)	GV (n=27)	AN paste (n=47)
Mortality (n)	64	19	7	12	4	8
Causes of death						
Sepsis	47 (73%)	5 (26%)	1 (14%)	2 (17%)	-	3 (38%)
Unspecified associated malformation	5 (8%)	-	-	-	-	5 (63%)
Cardiovascular failure	2 (3%)	2 (3%)	-	5 (42%)	4 (100%)	-
Respiratory failure	1 (2%)	5 (26%)	6 (86%)	2 (17%)	-	-
Unspecified complications	9 (14%)	7 (37%)	-	-	-	-
Others*	-	-	-	3 (25%)	-	-

** Includes sac rupture, multi-organ failure and care withdrawal*
AN: Acacia nilotica, DE: Disodium eosin, GV: Gentian violet, PI: Povidone iodine, SSD: Silver sulfadiazine

Several studies have mixed-up the therapeutic agents, making analytical interpretation difficult. For example, AN paste has been shown to be a useful escharotic agent with same efficiency as that of topical honey application.[18] However, AN powder had been dissolved in GV, which by itself is a well-established topical agent.[28]. Therefore, it would not be possible to tell if the therapeutic effect of the AN paste is due to GV, AN, or both. Indepen-dent pharmacological role of AN can be

evaluated only if it is dissolved in normal saline rather than GV. After detailed analysis of the literature, we propose a scheme of therapeutic standardization for each topical agent. If this is followed in future reporting, a meaningful meta-analysis is possible.

The application of supporting elastic bandage varied indifferent studies. Dressing with elastic bandage would prevent rupturing of the sac which is associated with poor prognosis.[8] Therefore, its application from the commencement of treatment would reduce the risks of sac rupture. This application should be soft and not attempting to reduce the omphalocele contents. Such forced bandage of may lead to either rupture of the sac or abdominal compartmental syndrome. With epithelialization, sac rupture is unlikely to occur. Therefore, the objective of the elastic bandage is to achieve partial reduction of the omphalocele content and restrain enlargement of the ventral hernia by providing counter-resistance to intra-abdominal pressure. Elastic bandage will facilitate increasing intra-abdominal capacity and will reduce the need for mesh while repairing the resultant ventral hernia.[33] We believe that application of elastic bandage should be continued until the surgical closure of the ventral hernia.

The use of escharotic agents has the potential risk of toxicity. Some of the agents are well proven to be safe. These include DE, honey, GV and AN paste. DE is non-absorbable and hence of low toxicity potential.[14,34] Although increased risks of hepatocellular carcinoma and thyroid tumors have been reported in mice fed with GV[35], it is generally considered safe in humans. There are no reports of GV-linked cancer in human beings.[35] Transient hypothyroidism due to the Wolff-Chaikoff effect is the most feared toxicity of PI. Therefore, a regular estimation of thyroxin levels is mandatory while using PI in omphalocele. Thyroid function tests done prior to the starting of PI treatment will differentiate drug-induced thyroid deficiency from congenital hypothyroidism. The frequency of routine thyroxin estimation suggested by different authors varies from 7 to 14 days.[15,17,18] In transient hypothyroidism, PI treatment may be continued with periodic monitoring of thyroxin.[15] Malhotra[16] reported symptomatic thyrotoxicosis after 3 days of using 10% PI in a premature newborn. Based on the study of Pandey,[17] serum thyroxin estimation once in 10 days seems to be appropriate, considering its affordability in LMIC. However, in symptomatic patients, thyroxin estimation should be done daily.[16] Extreme caution should be exercised when using PI in preterm neonates, because of immature Wolff-Chaikoff effect (auto-regulatory iodine-induced inhibition of thyroxin synthesis and release), easy absorption of iodine through the neonatal skin, immaturity of the thyroid gland and impaired renal clearance of iodine.[36]. As a result of this, thyrotoxicosis may occur with PI treatment in pre-term babies.[16] The major complication of all silver-containing topical agents is due to systemic absorption of the silver. Although SSD is extensively used in the clinical practice, blood silver levels are seldom estimated. Lewis recorded elevated blood silver levels on day 21 and 22 respectively in 2 patients; however, they were clinically asympto-matic.[26] This raises a question of safety on the usage of SSD in the conservative management of omphalocele. Well designed large sample studies are required to evaluate the safety of SSD.

Sac infection was common when a dressing was used to cover the sac as it is with DE and SSD. Absolute asepsis should be observed in performing these dressings. The use of GV was not associated with infectious complications; this may be due to the dark colour of GV masking subtle infection. It is also possible that GV has excellent antibacterial property as compared to other substances. Rupture of the sac was the second most common complication. In patients with protective dressing, the rupture may be secondary to an infection that digests the epithelialized sac. In the

absence of protective dressings, rupture is often due to a mechanical injury.

Mortality appears to be increased with protective dressing (e.g. DE, PI, and honey) as compared to open dressing (e.g. GV and AN paste). It is possible that the large studies had more patients with co-existing malformations, which is a well-known cause of mortality in omphalocele.

Table 4. Suggested standardization of topical therapy of omphalocele

Topical agent	Suggested strength of preparation	Suggested frequency of application
Disodium eosin[12]	2% aq.sol	Twice daily
Povidone iodine[30]*	5% aq.sol	Twice daily
Silver sulfadiazine[14]	1% cream	Daily
Honey[16]	Natural honey	Daily
Gentian violet[22]	1% aq.sol	Twice daily
Acacia nilotica [24]	25g of AN in 15 ml of Gentian violet	Twice daily

** Should be used with caution in preterm*

It would be interesting to study the comparative efficacy of different escharotic agents in the management of omphalocele. To facilitate future comparison, usage of each topical agent should be standardized. Reporting should also be uniform. Crucial data are often missing from some of the published reports. To allow future comparison of different escharotics, we suggest that each report must include the following data: (a) a description of associated malformations, (b) full information of the topical agent including its nature, concentration and frequency of application, (c) the use of an elastic bandage or another supportive dressing, (d) information about enteral feeding including starting time, tolerance and time to full feeding, (e) discharge criteria and length of hospital stay, (f) estimation of blood levels of the topical agents used (especially mandatory for PI and SSD), (g) time to epithelialization, (h) complications and their management, (i) results of swab and blood culture to diagnose infection, (j) age at mortality, (k) cause of death, and (l) timing of surgical repair of the ventral hernia.

During this review, some topical agents were not considered due to their questionable efficiency (e.g. normal saline) or due to a higher toxicity rate (e.g. ethanol). Recently, drowsiness which is a sign of alcohol intoxication has been reported in all neonates treated with topical ethanol.[37] As it may affect the brain development and pose a risk of apnea, we do not consider it safe. Animal studies have also shown abnormal brain development in mice with neonatal ethanol intoxication.[38,39]

CONCLUSION

Several escharotic agents are used in resource-constrained settings and they are valuable therapeutic arsenal in the conservative management of omphalocele. But a meaningful comparison of various agents is precluded by lack of standardized treatment and uniform reporting.

REFERENCES

[1] Verla MA, Style CC, Olutoye OO. Prenatal diagnosis and management of omphalocele. Semin Pediatr Surg. 2019 Apr; 28(2): 84-8.

[2] Khan FA, Hashmi A, Islam S. Insights into embryology and development of omphalocele. Semin Pediatr Surg. 2019 Apr; 28(2): 80-3.

[3] Skarsgard ED. Immediate versus staged repair of omphaloceles. Semin Pediatr Surg. 2019 Apr; 28(2): 89-94.

[4] Wagner JP, Cusick RA. Paint and wait management of giant omphaloceles. Semin Pediatr Surg. 2019 Apr; 28 (2): 95-100.

[5] Ayub SS, Taylor JA. Cardiac anomalies associated with omphalocele. Semin Pediatr Surg. 2019Apr;28(2):111- 4.

[6] Global PaedSurg Research Collaboration. Mortality from gastrointestinal congenital anomalies at 264 hospitals in 74 low-income, middle-income, and high-income countries: a multicentre, international, prospective cohort study. Lancet. 2021 Jul 24; 398(10297): 325-339.

[7] Ekenze SO, Ajuzieogu OV, Nwomeh BC. Challenges of management and outcome of neonatal surgery in Africa: a systematic review. Pediatr Surg Int. 2016 Mar; 32 (3): 291-9.

[8] Ndour O, Fall AF, Alumeti D, Fall M, Diouf C, Ndoye A, Ngom G, Ndoye MM. Etude des facteurs pronostiques de l'omphalocèle au service de Chirurgie Pediatrique du CHU Aristide Le Dantec de Dakar : A propos de 95 cas. Rév CAMES-Série A. 2009; 08: 103-6.

[9] Ngom G, Fall I, Sankale AA, Konate I, Dieng M, Sanou A, Ndiaye L, Ndoye M. Evaluation de la prise en charge de l'omphalocèle a Dakar [Evaluation of the management of omphalocele at Dakar]. Dakar Med. 2004; 49(3): 203-6.

[10] Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, Moher D, Peters MDJ, Horsley T, Weeks L, Hem pel S, Akl EA, Chang C, McGowan J, Stewart L, Hartling L, Aldcroft A, Wilson MG, Garritty C, Lewin S, Godfrey CM, Macdonald MT, Langlois EV, Soares-Weiser K, Moriarty J, Clifford T, Tunçalp O, Straus SE. PRISMA Extension for Scoping Reviews (PRISMA-ScR): Checklist and Explanation. Ann Intern Med. 2018 Oct 2; 169(7): 467-473.

[11] Kasanga TK, Bilond TM, Zeng FTA, Mujinga HMW, Mukakala AK, Kapessa ND, Musapudi EM, Mwamba FK, Katambwa PM, Nafatalewa DK, Badypwyla IT, Mukangala SI, Ngoie CN, Cabala VPK, Banza MI, Musanzayi SM. Traitement conservateur des omphaloceles geantes par l'eosine aqueuse disodique 2%: une serie des cas [Conservative treatment of giant omphaloceles with dissodic 2% aqueous eosin: a case serie]. Pan Afr Med J. 2021 May 21; 39: 63.

[12] Kouame BD, Odehouri Koudou TH, Yaokreh JB, Sounkere M, Tembely S, Yapo KG, Boka R, Koffi M, Dieth AG, Ouattara O, da Silva A, Dick R. Outcomes of conservative treatment of giant omphaloceles with dissodic 2% aqueous eosin: 15 years' experience. Afr J Paediatr Surg. 2014 Apr-Jun; 11(2): 170-3.

[13] Habou O, Adamou H, Magagi IA, Ali OM, Magagi A, Moustapha H, Sabo R, Abarchi H. The conservative treatment of giant omphalocele by tanning with povidone iodine and aqueous 2% eosin solutions. Ann Pediatr Surg. 2017; 13(3): 125-8.

[14] Andriamanarivo M, Hunald F, Rajaonarivony M, Raherison A, Rakotovao M. Treatment of giant omphalocele in Madagascar [Le traitement des omphaloceles geantes a Madagascar]. e-Mem Acad Natl Chir. 2012; 11(1): 48-51.

[15] Whitehouse JS, Gourlay DM, Masonbrink AR, Aiken JJ, Calkins CM, Sato TT, Arca MJ. Conservative management of giant omphalocele with topical povidone-iodine and its effect on thyroid function. J Pediatr Surg. 2010 Jun; 45 (6): 1192-7.

[16] Malhotra S, Kumta S, Bhutada A, Jacobson-Dickman E, Motaghedi R. Topical iodine–induced thyrotoxicosis in a newborn with a giant omphalocele. Am J Perinatol Rep. 2016 Apr; 6(2): e243-5.

[17] Pandey V, Gangopadhyay AN, Gupta DK, Sharma SP, Kumar V. Non-operative management of giant omphalocele with topical povidone-iodine and powdered antibiotic combination: early experience from a tertiary centre. Pediatr Surg Int. 2014 Apr; 30(4): 407-11.

[18] Eltayeb A, Mostafa M. Topical treatment of major omphalocoele: Acacia nilotica versus povidone-iodine: A randomised controlled study. Afr J Paediatr Surg. 2015; 12(4): 241-6.

[19] Rattan KN, Singh J, Dalal P, Rohilla R. Conservative management of giant omphalocele: 20-year experience from a tertiary care center in North India. J Pediatr Neo natal Individ Med. 2020; 9(1): e090105.

[20] Bode C, Ademuyiwa A, Elebute O. Formal saline versus honey as escharotic in the conservative management of major omphaloceles. Niger Postgrad Med J. 2018; 25(1): 48-51.

[21] Nicoara CD, Singh M, Jester I, Reda B, Parikh DH. Medicated Manuka honey in conservative management of exomphalos major. Pediatr Surg Int. 2014 May; 30(5): 515-20.

[22] Lawrence L, Gavens E, Reda B, Hill T, Jester I, Lander A, Soccorso G, Pachl M, Gee O, Singh M, Arul GS. Exomphalos major: Conservative management using Manuka honey dressings and an outreach surgical nursing team. J Pediatr Surg. 2021 Aug; 56(8): 1389-1394.

[23] Ekot EA, Emordi VC, Osifo DO. Does omphalocele major undergo spontaneous closure? J Surg Case Rep. 2017 Aug 21; 2017(8): rjx156.

[24] Lee SL, Beyer TD, Kim SS, Waldhausen JHT, Healey PJ, Sawin RS, et al. Initial non-operative management and delayed closure for treatment of giant omphaloceles. J Pediatr Surg. 2006 Nov; 41(11): 1846-9.

[25] Ein SH, Langer JC. Delayed management of giant omphalocele using silver sulfadiazine cream: an 18-year experience. J Pediatr Surg. 2012 Mar; 47(3): 494-500.

[26] Lewis N, Kolimarala V, Lander A. Conservative management of exomphalos major with silver dressings: are they safe? J Pediatr Surg. 2010 Dec; 45(12): 2438-9.

[27] Echendu ST, Okpala BC, Ikechebelu JI, Nwajiaku LA, Eleje GU, Nwachukwu CE, Ofojebe CJ, Ezeigwe CO, Okpala AN. Giant omphalocele with successful conservative management in a low income setting: A Case Report. J Med Case Rep Rev. 2021 June 16; 4(6): 903-6.

[28] Mitul AR, Ferdous KMN. Initial conservative management of exomphalos major with gentian violet. J Neonatal Surg. 2012 Oct 1; 1(4): 51.

[29] Moustafa MM, Osman MA, Ismael K. A new method for non-operative treatment of giant omphalocele. Al-Azhar Assiut Med J. 2005; 3(2): 1-11.

[30] Ahlfeld H. Alkohol bei der Behandlung inoperabler Bauchbruche. Monatsschr Geb Gynakol. 1899; 10: 124.

[31] Bauman B, Stephens D, Gershone H, Bongiorno C, Oster holm E, Acton R, Hess D, Saltzman D, Segura B. Management of giant omphaloceles: A systematic review of

methods of staged surgical vs. non-operative delayed closure. J Pediatr Surg. 2016 Oct; 51(10): 1725-30.
[32] Binet A, Scalabre A, Amar S, Alzahrani K, Boureau C, Bastard F, Lefebvre F, Koffi M, Moufidath S, Nasser D, Ouattara O, Kouame BD, Lardy H. Operative versus conservative treatment for giant omphalocele: Study of French and Ivorian management. Ann Chir Plast Esthet. 2020 Apr; 65(2): 147-153.
[33] Evbuomwan I, Durell J, Lakhoo K, Ali AE. Exomphalos and Gastroschisis. In: Ameh EA, Bickler SW, Lakhoo K, Nwomeh BC, Poenaru D. (ed) Pediatric Surgery: A comprehensive textbook for Africa. 2nd edn. Cham: Springer Nature. pp 597-604. {DOI: 10.1007/978-3-030-41724-6_56}
[34] Rueda-Fernandez M, Melguizo-Rodriguez L, Costela-Ruiz VJ, de Luna-Bertos E, Ruiz C, Ramos-Torrecillas J, Illescas-Montes R. Effect of the most common wound antiseptics on human skin fibroblasts. Clin Exp Dermatol. 2022 Aug; 47(8): 1543-1549.
[35] Maley AM, Arbiser JL. Gentian Violet: a 19th century drug re-emerges in the 21st century. Exp Dermatol. 2013 Dec; 22 (12): 775-80.
[36] Aitken J, Williams FL. A systematic review of thyroid dysfunction in preterm neonates exposed to topical iodine. Arch Dis Child Fetal Neonatal Ed. 2014 Jan; 99(1): F21-8.
[37] Talabi A, Sowande O, Adejuyigbe O. Challenges in the management of omphalocele in Ile-Ife, Nigeria. J Clin Neo natol. 2020; 9(4): 280-5.
[38] Goodlett CR, Thomas JD, West JR. Long-term deficits in cerebellar growth and rota-rod performance of rats following 'binge-like' alcohol exposure during the neonatal brain growth spurt. Neurotoxicol Teratol. 1991 Jan; 13(1): 69-74.
[39] Hamilton GF, Murawski NJ, St Cyr SA, Jablonski SA, Schiffino FL, Stanton ME, Klintsova AY. Neonatal alcohol exposure disrupts hippocampal neurogenesis and contextual fear conditioning in adult rats. Brain Res. 2011 Sep 15; 1412: 88-101.

APPENDIX

Search strategy for PubMed

Using MeSH and the following syntax
((Omphalocele OR Exomphalos) AND
(Conservative OR Delayed) AND Management))

Search strategy for Google Scholar

'Advanced' option was used, with the following syntax:

- in "with all the words": Omphalocele
- in "with at least one of the words": Omphalocele, Exomphalos, Delayed; Conservative, Management
- in "where my words occur"
- in the title of the article

Address for communication: *Dr. Florent Tshibwid A Zeng, Email: tshibwidflo@gmail.com*

Received 18 June 2024; Accepted 27 August 2024

Acknowledgements: The author FTAZ is grateful to BEBUC Excellence Scholarship which funds his specialization in pediatric surgery
Conflicts of Interest: None declared by the author
Source of Funding : None
Ethical concerns : None (Literature Review)
Data Availability : Available from the corresponding author upon reasonable request

Citation: *Zeng FTA, Kapessa ND, Baldeh A, Kombe LBI, Kamwangen GM, Kasanga TK, Kalau WA. Conservative management of omphalocele using escharotic agents available in resource-constrained settings: A scoping review. Pediatr Surg Trop 2024; 1(4): 269-277.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 278-283
DOI: 10.5281/zenodo.13893756

Clinical Study

Bedside Cystometry as a Simple Alternative to Urodynamic Studies in Resource-Limited Settings in Diagnosing Pediatric Bladder Dysfunctions: A Single-Blinded Prospective Comparative Study

Sandip Kumar Rahul, Yogesh Kumar Sarin

Department of Pediatric Surgery, Maulana Azad Medical College and Lok Nayak Hospital, New Delhi-110002, India.

Keywords
Bedside cystometry
Urinary bladder dysfunction
Urodynamic study

Abbreviations
BC - Bedside cystometry
CVP - Central venous pressure
EBC - Expected bladder capacity
OBC - Observed Bladder Capacity
PUV - Posterior Urethral Valve
UDS - Urodynamic study

Abstract

Background: *Proper urodynamic evaluation of children is often limited by the cost, technical expertise and availability of the involved equipments. Bedside cystometry (BC) could be a useful alternative in the absence of facility for urodynamic study (UDS). In this study, the predictive value of BC is compared with that of the UDS in diagnosing functional abnormalities of the bladder in children.*

Methods*: This single-blinded prospective comparative study involved 30 children of age 4-18 years who were diagnosed with various urinary anomalies (e.g. posterior urethral valves, neurogenic bladder, bladder exstrophy and dysfunctional voiding). All of them underwent both BC and UDS. To mimic the resource-limited settings, BC was done using saline manometer and without rectal pressure monitoring.*

Results*: The overall sensitivity of BC was 92%, specificity was 100%, positive predictive value was 100% and negative predictive value was 95%. False-negative results were found in 8% of patients. Detrusor instability was detected by UDS and BC in 12 and 11 patients respectively. The observed differences between the two groups were not statistically significant. A high kappa value (k = 0.93) confirms a near-perfect agreement between the two diagnostic procedures.*

Conclusion*: BC could measure bladder volume and pressure as accurately as UDS. Therefore, it could be a valid, reliable, acceptable and cheap alternative to UDS in resource-limited settings where UDS facilities are not available.*

INTRODUCTION

Urodynamic study (UDS) is an essential tool in the diagnosis of urinary bladder dysfunctions. UDS evaluates both the static and dynamic elements of the bladder function. It is a sophisticated multi-channel study that consists of cystometry (filling

phase behavior of the urinary bladder), uroflometry (pressure-volume dynamics of voiding) and electromyography (study of the sphincter contractions). As it yields a wide range of information, it is considered to be the gold standard. However UDS requires complex and costly equipments that are not easily available in many resource-limited centers.

Although bedside cystometry (BC) is no match to UDS, it could be a simple yet valuable screening tool for the assessment of filling-phase behavior of the urinary bladder. Unlike UDS, it is done at bedside by a single-channel technique using simple and easily available materials in any hospital.[1] Most of the previous studies comparing BC and UDS have been done in adults, and there is a paucity of pediatric literature. The present study is intended to compare the predictive value of BC with that of standard UDS.

MATERIALS AND METHODS

Study Cohort

This single-blinded prospective comparative study was conducted in the department of pediatric surgery at a tertiary care center over a period of one year. Study cohort consists of 30 co-operative children aged 4-18 years who were diagnosed with posterior urethral valves (PUV), neurogenic bladder, vesical exstrophy or voiding dysfunction. All of them required assessment of bladder function as part of clinical management. Patients with urinary tract infections and those who could not be catheterized were excluded. Informed parental consent and child assent were obtained.

Study Design

All the study subjects underwent both BC and UDS in the same sitting. UDS was taken as the gold standard, against which BC was compared. BC was done in all but one before the UDS. Additional urological assessments such as urological imaging, blood biochemistry and urine culture were done as indicated.

Study Parameters

Demographic details, clinical history and physical examination findings were recorded for each of them. Subjective and objective variables were studied. Subjective variables included the filling volume and pressure at the first sensation of bladder filling, at the first desire to void and at the strong desire to void. Objective variables were pressure recordings at specified bladder volumes (e.g. 25%, 50%, 75% and 100% of the expected bladder capacity), bladder compliance and the presence or absence of detrusor instability. The bladder volume and pressure were also recorded at the time of any significant event such as cough.

Technical Standardization

Prior to the study, the rectum was emptied by administering bisacodyl suppository or sodium phosphate enema. At the outset, children were asked to empty the bladder on their own. Post-void residual volume was noted on catheterization. Privacy was ensured during the procedures. Koff's formula [30 x (age in years + 2) ml] was used to calculate the expected bladder capacity (EBC) for each patient.[2,3] Pubic symphysis was the taken as the reference point of manometer level and atmospheric pressure as the reference pressure in both UDS and BC.

Compliance of bladder was assessed by measuring intra-vesical volume and pressure at two standard points as per the recommendations of the International Continence Society.[4,5] Bladder compliance was expressed as the ratio of change in bladder volume against the corresponding change in bladder pressure till the time of strong desire to void.

Protocol of Urodynamic Study

An independent resident trainee, who was blinded to the findings of BC, performed UDS as per the prescribed standard methodology.[4,5] Laborie™ urodynamic machine - version 6 (Laborie Medical Technologies Corp, New Hampshire, USA) was

used. Software incorporated in this multi-channel equipment automatically calculated the detrusor pressure from intra-abdominal and intra-vesical pressures and continuously displayed these 3 values in real-time.

Before starting the study, intactness of all the tube connections were checked using cough response test. Air bubbles were removed from all the tubes. Transducers were calibrated by zero balancing at the level of the pubic symphysis.

Endpoints of filling were defined as follows: (a) Strong urge to void and inability to hold urine further (b) Uncomfortable bladder sensation (c) Uncontrolled voiding during the study (d) Persisting bladder pressure >40 cmH2O (e) Infused volume > 150 % of EBC (f) Rate of leakage more than the rate of infusion.

Protocol of Bedside Cystometry

BC was performed by the principal investigator (the first author). Children were asked to void at the outset and the force of stream (qualitative) and volume of urine (quantitative) were recorded. With aseptic precautions, two sterile 5 Fr infant-feeding tubes were simultaneously inserted into the bladder through the urethra or through any other conduit such as the Mitrofanoff channel. Care was taken that the catheters were not too much inside the bladder, as this may irritate the detrusor or trigone. The amount of residual urine, if any, in the bladder was recorded. Both the tubes were fixed to the lower abdominal wall with an adhesive tape. One of the tubes was used to fill the bladder; the other served as monitoring channel connected to a manometer. Pediatric burette sets were used to fill the bladder with measured quantities of normal saline at an appropriate rate. Central venous pressure (CVP) manometer that is based on the level of water column was used to measure the pressure in cm H_2O. The same parameters as that of UDS were studied. A note was made of any sudden wide fluctuation in pressure that is suggestive of bladder instability.

Monitoring of the rectal pressure was deliberately omitted in BC group in order to mimic the ground reality of resource-limited settings. (For further justification *vide infra*).

Statistical Analysis

Paired t-test or Mann-Whitney test was used for analyzing the quantitative data and chi-square test for the qualitative data. Agreement between BC and UDS was analyzed using Cohen's kappa statistics. P-value <0.05 was considered significant to disprove null hypothesis. Sensitivity, specificity and predictive value were also calculated for UDS and BC.

RESULTS

The mean age of patients (25 boys, 5 girls) was 9.4 years (SD 3.48; median 8.5 years). Among them 87% were aged 5-15 years. The mean EBC was 331 ml (range was wide due to the differences in the age of the patients). Table 1 summarizes the comparison of urodynamic values of UDS and BC. Detrusor instability was detected by UDS and BC in 12 and 11 patients respectively. The observed differences between the two groups were not statistically significant.

The overall sensitivity of BC was 92%, specificity was 100%, positive predictive value was 100% and negative predictive value was 95%. False-negative results were found in 8% of patients. The diagnostic accuracy of BC, taking UDS as the gold standard, was 97%. A high kappa value ($k = 0.93$) confirms a near-perfect agreement between the two diagnostic procedures.

DISCUSSION

Bladder dysfunction causes significant morbidity in many patients with PUV, neurogenic bladder, vesical exstrophy and dysfunctional voiding. It is

therefore important to have a periodic monitoring of the bladder condition in these patients. The pressure and volume characteristics of bladder are a measure of its functional status and overall health. Often, these parameters guide us in specific management or intervention. Urodynamic study is now considered indispensable in the management of bladder malformations and dysfunctions.

Table 1. Comparison of urodynamic parameters with bedside cystometry

Study Parameter	UDS*	BC*	*P - value*
Subjective Parameters			
OBC (ml)	254 ± 194	265 ± 200	0.32
EBC/OBC (%)	80 $\pm$ 55	82 $\pm$ 59	0.73
Volume at first bladder sensation (ml)	22 ± 42	27 ± 53	0.18
Pressure at first bladder sensation (cm H_2O)	2 ± 4	2 ± 5	0.45
Volume at first desire to void (ml)	105 ± 90	111 ± 125	0.35
Pressure at first desire to void (cm H_2O)	8 ± 19	7 ± 15	0.32
Volume at strong desire to void (ml)	203 ± 165	214 ± 175	0.32
Pressure at strong desire to void (cm H_2O)	14 ± 28	16 ± 22	0.31
Objective Parameters			
Pressure at 25% EBC (cm H_2O)	4 ± 9	4 ± 6	0.23
Pressure at 50% EBC (cm H_2O)	8 ± 18	8 ± 13	0.48
Pressure at 75% EBC (cm H_2O)	12 ± 20	13 ± 20	0.32
Pressure at 100% EBC (cm H_2O)	19 ± 20	19 ± 20	0.50
Bladder Compliance	18.7 ± 30.1	16.5 ± 24.5	0.27

** Values as Mean $\pm$ 2 standard deviations. BC - Bedside cystometry, EBC - Expected bladder capacity, OBC - Observed bladder capacity, SD - Standard deviation, UDS - Urodynamic study*

The cystometric techniques have evolved ever since 1882, when Mosso and Pellacani first used a water manometer to measure bladder pressure and recorded it on a smoked drum.[6] Initially, simple methods were used; but with passage of time, accuracy and consistency of recordings improved with advancements in digital recording systems and transducers. Neale described the use of a CVP manometer as a simple tool of measuring bladder pressure.[7]

The modern-day UDS requires an elaborate, cumbersome and costly setup that is available only in a few tertiary-care hospitals. Single-use UDS catheters are costlier than the two infant feeding tubes, pediatric burette set and CVP manometer used in BC. However, such low-cost alternatives are acceptable only if the diagnostic accuracy is not compromised. Previously published studies were mostly pertinent to adult females and geriatric patients.[8-12] We could find only a single pediatric study of BC in spina bifida patients.[13] There is a paucity of literature regarding this in the pediatric age group, although UDS has been widely used in them.[14,15] Several authors have acknowledged the usefulness of single-channel urodynamics in the evaluation and treatment of urinary incontinence.[16-23]

Bates, in 1970, showed the importance of differentiating between the gross intra-vesical pressure, the contribution of intra-abdominal pressure and the true detrusor pressure in judging the real pathophysiology.[24] It was after this publication, multi-channel cystometry became popular and developed to reach its present complexity.[24] Recently, Cheriyan et al found that the gross vesical pressure and detrusor pressure (vesical pressure minus intra-abdominal pressure) are comparably identical in children. They also noted several artifacts with the usage of additional rectal catheter in an already anxious child.[25,26]

Subjective variables measured in this study need accurate active feedbacks communications from the patients regarding their subjective perceptions (e.g. sensation of filling, desire to void). The younger the child, the more difficult it will be to have this interaction. For this reason children younger than 4 years were not included in this study. The mean age of patients in this study was 9.4 years with a standard deviation of 3.5 years. Previously published studies involve mainly adults or geriatric patients.

Ouslander reported a sensitivity of 75%, a specificity of 78% and a positive predictive value of 85% for simple cystometry.[8] Other workers have also confirmed similar outcomes with simple cystometry.[9,12] Wheeler et al[10] who compared simple water manometer cystometry with 4-channel electronic cystometry in females with voiding dysfunction found that the water manometer had an accuracy of 93% in detecting detrusor instability and had a good correlation of bladder volume with the 4-channel manometry.

Unlike multi-channel UDS machines, BC does not have software for automatic recording and storing of data. As they have to be done manually, the observer must be vigilant while performing BC. With practice this skill is acquired easily and the learning curve is not prolonged.

CONCLUSIONS

BC could measure the changes in bladder volume and pressure as accurately as UDS and could detect bladder instability with high sensitivity, specificity and accuracy. It therefore is a valid, reliable, acceptable, easy-to-perform and cost-effective alternative to UDS in resource-challenged settings without UDS facilities.

REFERENCES

[1] Cole EE, Dmochowski RR. Office urodynamics. Urol Clin North Am. 2005 Aug; 32(3): 353-70.

[2] Koff SA. Estimating bladder capacity in children. Urology. 1983 Mar; 21(3): 248.

[3] Berger RM, Maizels M, Moran GC, Conway JJ, Firlit CF. 'Bladder capacity (ounces) equals age (years) plus 2' predicts normal bladder capacity and aids in diagnosis of abnormal voiding patterns. J Urol. 1983 Feb; 129 (2): 347-9.

[4] Abrams P, Cardozo L, Fall M, Griffiths D, Rosier P, Ulmsten U, van Kerrebroeck P, Victor A, Wein A; Standardisation Sub-committee of the International Continence Society. The standardisation of terminology of lower urinary tract function: report from the Standardisation Sub-committee of the International Continence Society. Neurourol Urodyn. 2002; 21(2): 167-78.

[5] Abrams P, Cardozo L, Fall M, Griffiths D, Rosier P, Ulms ten U, Van Kerrebroeck P, Victor A, Wein A; Standardisation Sub-Committee of the International Continence Society. The standardisation of terminology in lower urinary tract function: report from the standardisation sub-committee of the International Continence Society. Urology. 2003 Jan; 61(1): 37-49.

[6] Mosso A, Pellacani P. Sur les fonctions de la vessie. Arch Ital Biol. 1882; 1:97-128.

[7] Neale R. Simple cystometry. Lancet. 1977 Oct 29; 2(8044): 927.

[8] Ouslander J, Leach G, Abelson S, Staskin D, Blaustein J, Raz S. Simple versus multichannel cystometry in the evaluation of bladder function in an incontinent geriatric population. J Urol. 1988 Dec; 140(6): 1482-6.

[9] Sutherst JR, Brown MC. Comparison of single and multichannel cystometry in diagnosing bladder instability. Br Med J. 1984 Jun 9; 288 (6432): 1720-2.

[10] Wheeler JS, Niecestro RM, Fredian C, Walter JS. Comparison of a simple cystometer with a multichannel cystometer in females with voiding dysfunction. Int Urogynecol J. 1991 June; 2:90-3.

[11] Johnson TM, Muirhead L, Busby-Whitehead J. Urinary incontinence. In: Busby-Whitehead J, Durso SC, Arenson C, Elon R, Palmer MH, Reichel W. (ed). Reichel's Care of

the Elderly: Clinical Aspects of Aging. Cambridge Univ Press; 2022. pp. 350-63.
[12] Fonda D, Brimage PJ, D'Astoli M. Simple screening for urinary incontinence in the elderly: a comparison of simple and multichannel cystometry. Urology. 1993 Nov; 42 (5): 536-40.
[13] Bangar A, Karmarkar S. Simple clinical and bedside urodynamic evaluation is sufficient for successful management decisions in spina bifida patients with urinary incontinence and the machine urodynamics are not necessary. Fluids Barriers CNS. 2007; 4 (Suppl 1), S21 (Conference abstract, 51st Annual Meeting of the Society for Research into Hydrocephalus and Spina Bifida, Heidelberg, Germany, 27–30 June 2007). {DOI: 10.1186/1743-8454-4-S1-S21}
[14] Karmarkar SJ, Muthal PB, Kulkarni BK. Urodynamic patterns in spina bifida: Classification, comparison and the need for international consensus. J Indian Assoc Pediatr Surg. 1998; 3: 10-4.
[15] Agarwal M, Rath B, Kaza RC, Talukdar B, Puri RK. Urodynamic study of children with voiding problems. Indian Pediatr. 1995 Mar;32(3):307-11.
[16] Hodgkinson CP, Ayers MA, Drukker BH. Dyssynergic detrusor dysfunction in the apparently normal female. Am J Obstet Gynecol. 1963 Nov 15; 87: 717-30.
[17] Willington FL. Therapeutic distension for bladder instability in the elderly. In: Proceedings of the VIII International Continence Society Meeting. Manchester: Pergamon Press; 1978. pp 13-7.
[18] Eastwood HD. Incontinence: who can benefit from simple investigation? Modern Geriatrics .1978; 8: 39-43.
[19] Stanton SL. What is the place of urodynamic investigations in a district general hospital? Br J Obstet Gynaecol. 1983 Feb; 90(2): 97-9.
[20] Jeffcoate TN, Francis WJ. Urgency incontinence in the female. Am J Obstet Gynecol. 1966 Mar 1; 94(5): 604-18.
[21] Zamli AH, Ratnalingam K, Yusmido YA, Ong KG. Diagnostic accuracy of single channel cystometry for neurogenic bladder diagnosis following spinal cord injury: a pilot study. Spinal Cord Ser Cases. 2017 May 4; 3: 16044.
[22] Wyndaele JJ, THi HV, Pham BC, Kovindha A, Huong VT, Weerts E. The use of one-channel water cystometry in patients with a spinal cord lesion: practicalities, clinical value and limitations for the diagnosis of neurogenic bladder dysfunction. Spinal Cord. 2009 Jul;47(7):526-30.
[23] Ricci Arriola P, Solá Dalenz V, Pardo Schanz J. Estudio de la incontinencia de orina femenina mediante urodinamia monocanal: comparación con los síntomas de ingreso. Análisis de 590 mujeres [Study of female urinary incontinence with single channel urodynamics: comparison of the symptoms on admission. Analysis of 590 females]. Arch Esp Urol. 2009 Mar;62(2):115-23.
[24] Bates CP, Whiteside CG, Turner-Warwick R. Synchronous cine-pressure-flow-cysto-urethrography with special reference to stress and urge incontinence. Br J Urol. 1970 Dec; 42(6): 714-23.
[25] Cheriyan A, George AJP, Devasia A, Chandrasingh J. Can rectal catheters be avoided during paediatric urodynamic studies? Arab J Urol. 2019 Sep 25; 18(1): 41-46.
[26] Farag F. Re: Can rectal catheters be avoided during paediatric urodynamic studies? Arab J Urol. 2019 Dec 4; 18 (1): 47.

Address for communication: *Dr. Yogesh Kumar Sarin, Email: yksarin@gmail.com*

Received 22 June 2024; Accepted 16 August 2024

Acknowledgements : None
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : Authors declare securing IRB approval

Citation: *Rahul SK, Sarin YK. Predictive value of bedside cystometry versus urodynamic study in diagnosing bladder functional abnormalities in children: A single-blinded prospective comparative study. Pediatr Surg Trop 2024; 1(4): 278-283.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 284-285
DOI: 10.5281/zenodo.13893805

Brief Communication

Buried Penis: Parental Perceptions and Surgical Options

Rajah Shunmugam, Vinodh Suppiah

Division of Paediatric Surgery, Gleneagles Hospital, Kota Kinabalu, Sabah, Malaysia.

Keywords
Buried penis
Penile anomaly
Foreskin
Buck's fascia
Penopubic angle
Penoscrotal angle
Z-plasty
Genital reconstruction

Abstract

Buried penis is a rare congenital anomaly due to dysplasia of the dartos fascia and lack of penile skin fixation to the Buck's fascia. This report describes an experience with 8 boys with buried penis. In young children, the parental concerns were invisible penis, dribbling of urine and smelly urine. Unfurling of inner prepuce appears to be suitable for severe variety of buried penis, while Z- Plasty is suitable for mild or moderate varieties.

We reviewed the medical records of 8 boys who were operated for buried penis between 2017 and 2021. Their age range was 3 months to 15 years. The parental concerns were invisible penis, dribbling of urine and smelly urine in 2 infant boys while small size of penis and undue shyness in 6 older boys. Using the classification of Chin et.al.[1] they were categorized into severe, moderate or mild deformity. Surgical technique was chosen according to the availability of outer shaft skin and the degree of penopubic and penoscrotal angles. Unfurling of inner prepuce was done in 3 children with severe buried penis (Fig. 1). Z-plasty using a part of scrotal skin was done to gain good penopubic and penoscrotal scrotal angles in the remaining 5 boys with mild or moderate anomaly.

Persistent post-operative edema was noted in 3 children who had undergone unfurling of inner prepuce. (Fig. 2) Preputial edema resolved in 1 to 6 months. Slight bleeding and wound infection in 2 patients required repeated dressing and antibiotics. The remaining 3 boys had an uneventful post-operative period. Follow-up ranged between 2 months and 4 years. All the children and parents were satisfied with the appearance and the length of the penis.

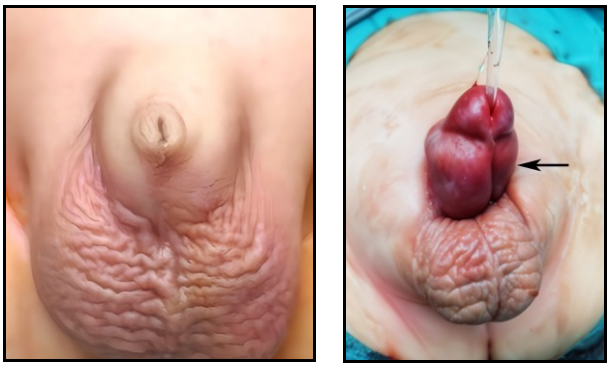


Fig 1. *Burried penis: Preoperative (left panel) and post-operative (right panel) appearance. Arrow indicates the unfurled inner prepuce*

Buried penis is a rare congenital anomaly due to dysplasia of the dartos fascia, lack of penile skin fixation to the Buck's fascia and deficiency of shaft

skin.[1-4] Chin[1] classified buried penis into three groups based on the deficiency of the penile skin. They proposed a ratio of the length of the preputial skin (S) and that of the penile shaft (P). The length of the foreskin was measured by gentle stretching and that of the penile shaft by pressing the pubic fat. Buried penis is categorized as severe if the S/P ratio (S/Px100) is < 30%, moderate if it is 30-70% and mild if it is >70%. In our series 3 children had severe buried penis.

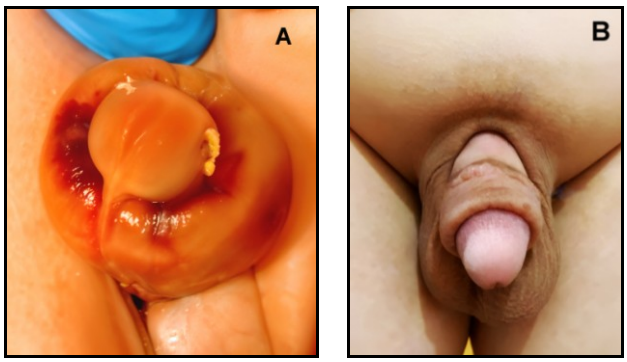


Fig 2. *Post-operative results of burried penis repaired by unfurling the inner prepuce: (A) Immediate post-operative appearance showing significant edema of the inner prepuce, (B) The final outcome*

A 3-month-old infant with severe anomaly had thick fibrous tissue causing severe chordee, glanular hypospadias and long inner prepuce (LIP) as reported by Hadidi.[3] There was severe deficiency of the shaft skin, and it was fixed to the pubis to re-define the penopubic angle. The entire inner prepuce was used to cover the shaft in this patient while only a part of inner prepuce was used in the remaining 2 patients with severe buried penis. In mild or moderate anomalies, complete excision of the abnormal tissue and fat at the base of the penis, raising the scrotal skin flap to cover the base of the penis combined with Z-Plasty helped in creating a good penoscrotal angle and increasing the length of the penile shaft.

Our limited experience suggests that unfurling of the inner prepuce is suitable to treat severe degree of buried penis while Z-plasty is suitable for mild or moderate anomalies.

REFERENCES

[1] Chin TW, Tsai HL, Liu CS. Modified prepuce unfurling for buried penis: a report of 12 years of experience. Asian J Surg. 2015 Apr; 38(2): 74-8.

[2] Redman JF. Buried penis: congenital syndrome of a short penile shaft and a paucity of penile shaft skin. J Urol. 2005 May; 173(5): 1714-7.

[3] Hadidi AT. Buried penis: classification surgical approach. J Pediatr Surg. 2014 Feb; 49(2): 374-9.

[4] De Jesus LE, Dekermacher S, Anderson KM. Severe forms of concealed penis without hypospadias: Surgical strategies. Indian J Urol. 2015 Oct-Dec; 31(4): 344-8.

Address for communication: *Dr. Rajah Shunmugam, Email: srajah1@hotmail.com*

Received 23 April 2024; Accepted 16 August 2024

Acknowledgements : None
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : Authors declare securing IRB approval

Citation: *Rajah S, Vinodh S. Buried penis: Parental perceptions and surgical options. Pediatr Surg Trop 2024; 1(4): 284-285.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Pages 286-290
DOI: 10.5281/zenodo.13893821

Case Report

Pediatric Cranioencephalic Trauma of Ballistic Origin

Malangu Mhacks, Feruzi Marius, Yogolelo Rosy, Mutomb Sarah, Tshishiku DJonny, Arung Willy

Neurosurgery Unit, Department of Surgery, Faculty of Medicine, University of Lubumbashi, Lubumbashi 2008, Democratic Republic of the Congo.

Keywords
Head injury
Ballistic trauma
Firearm wounds
Brain injury
Central nervous system

Abbreviations
GCS - Glasgow coma scale
GOSE - Glasgow Outcome Scale-Extended

Abstract

We report 2 cases of stray bullet injury to brain in children of age 3 months and 7 years respectively. One of them had an occipital penetrating wound (with no exit) and the other had trans-temporal wound (with both entry and exit points). The delay in doing a CT scan was 1 and 8 days respectively. The average duration of surgery was 80 min. The average time to complete control imaging was 18.5 days. Follow-up imaging showed cerebral swelling and re-bleeding in one patient while it was unremarkable in the other child. Pediatric head injuries due to firearms are very rare, but with high morbidity and mortality. Their management in limited resource settings is challenging.

INTRODUCTION

Head injuries caused by firearms are often fatal but fortunately rare in the pediatric population. However, their incidence in civilian population has increased in recent years due to a surge in armed conflicts, urban banditry and terrorism.[1-3] About 5 to 13% of injuries in France are due to weapons and 1% of all victims had gunshot wounds. They are common in the head, neck and trunk.[2] In this report, we present our experience in managing two children with ballistic head injury.

CASE PRESENTATION

Case 1

An 3-month-old male infant was referred in an unconscious state from Likasi to Lubumbashi (126 km distance; 3hr-10min travel). He developed sudden cry and bleeding from the temporal scalp during sleep.(Fig. 1) The history is suggestive of injury due to stray bullet (which would have entered the bedroom through the ceiling). Initial resuscitation had been done at a near-by hospital. Blood transfusion, intravenous fluids, paracetamol, phenobarbitone and wound dressing had been given. Onset of unconsciousness prompted referral after 24hr.

On arrival at our center, his pediatric Glasgow Coma Score (GCS) was 7/15. He had hypotonia, bilateral miosis, flat anterior fontanel and a head-circumference of 41cm. There was a trans-temporal wound (having both entry and exit points) with extrusion of brain matter. His injury was classified as Matson grade IV-D. The s100 beta

protein was < 0.05µg/l. Pulsatility index was 3 and diastolic velocity was 7. On the second day of hospitalization a CT scan revealed multiple areas of brain edema and hemorrhagic contusion of both parietal lobes, subarachnoid hemorrhage, pneumocephalus and comminuted fracture of the left temporal bone. (Fig.2) Multidisciplinary management involving a neurosurgeon, neurologist, anesthesiologist and critical care specialist was started. Mannitol, normal saline, antibiotics (gentamicin, ceftriaxone, metronidazole), proton pump inhibitor, phenobarbitone, midazolam and oxygen by oro-tracheal tube were given.

Fig 1. *Photograph (case 1) showing temporal wound*

A surgical operation was performed on the second day of admission. Debridement and closure of the scalp wound was done. Post-operative period was marked by tachycardia, anemia (Hemoglobin 8.8 g/dl), hypoalbuminemia (2 g/dl), hypocalcemia (7.9 mg/dl), wound infection, cerebral ischemia, re-bleed and meningitis. Two episodes of cardiac arrest were successfully revived. Eventually, he recovered. On clinical evolution 2 months after the injury, he was well with a Glasgow Outcome Scale-Extended (GOSE) score of 3.

Case 2

A 7-year-old male child, with no particular history of injury, was referred in unconscious state from Fungurume to Lubumbashi (200 km distance; 3hr-20min travel by road). Ten hour before admission he suddenly developed scalp bleeding while playing with his friends and it was followed by a brief period of loss of consciousness. He also vomited several times and developed seizures. He appeared to have been injured by a stray bullet. Skull radiograph done at another hospital showed a metal foreign body (bullet) inside the brain.(Fig.3)

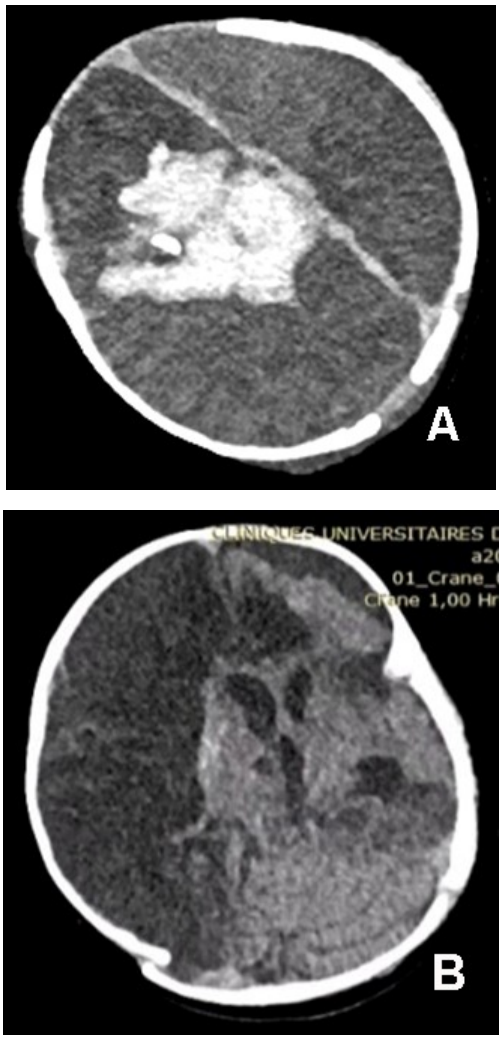


Fig 2. *CT scan (Case 1) showing intra-cerebral bleed (A) and fronto-temporal brain edema (B)*

On arrival, his GCS was 4/15. The pupils were equal and reacting. There was a contused wound of 5 cm diameter (entry wound without an exit point) in the right parietal area. A CT scan of the brain on the day-8 of injury revealed a bullet in the left temporal lobe. The right posterior parietal cortex was contused and edematous with associated temporal bone fracture. Diastolic velocity was 7 and pulsatility index was 3. The s100 beta-protein was < 0.05µg/l. A multidisciplinary treat-

ment was started. Normal saline, mannitol, paracetamol, tramadol, ceftriaxone, metronidazole, sodium valproate and vitamin C were given. On the post-traumatic day-25, the bullet was surgically extracted by trans-cortical approach. Post-operative recovery was uneventful. (Fig. 4) He stayed in the hospital for 40 days.

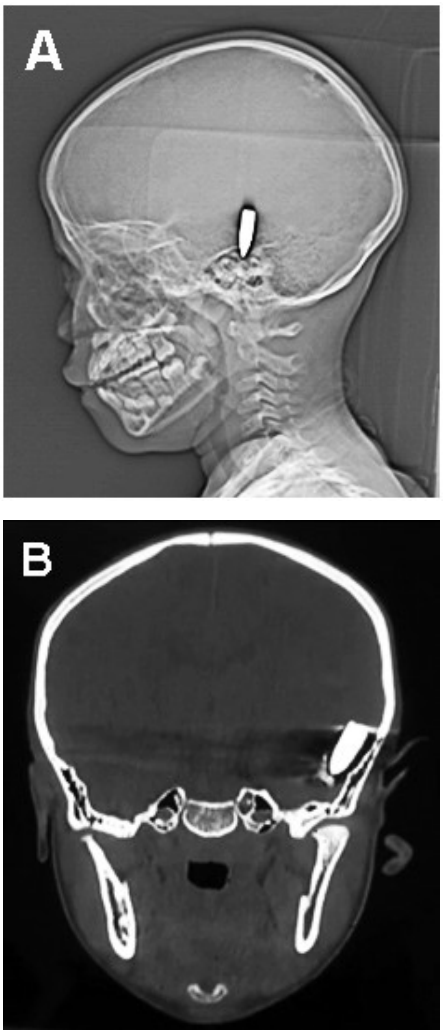


Fig 3. *Plain skull x-ray (A) and CT reconstruction of coronal section (B) of the case 2 showing a bullet in the left temporal brain*

At follow-up 2-months later, his Blantyre score was 5 with a slight ptosis of the left eyelid. He could move all 4 limbs spontaneously; but there was hypertonia of central origin. By then, the surgical wound had healed well and the GOSE score was 3.

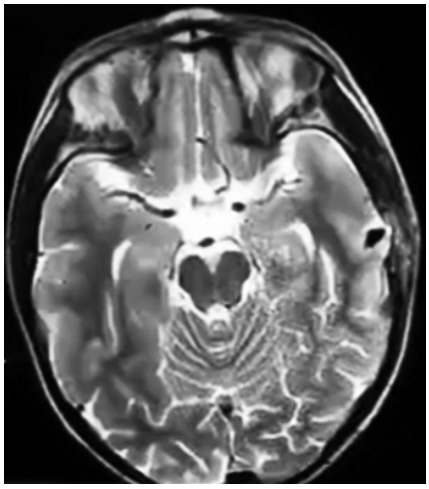


Fig 4. *Follow-up MRI (T2 sequence, axial section) of the case 2, done after 15 days of treatment, showing residual hypodense lesion in the left basi-temporal zone.*

DISCUSSION

Cranioencephalic ballistic injuries are medico-surgical emergencies with a high morbidity and mortality, especially in resource-limited settings. Plasticity of the pediatric brain enables its quick recovery from serious injuries, unlike that of the adults. Therefore, the approach to ballistic head injuries is distinctly different in children.[1-3]

Pediatric head injuries due to firearms are rarely reported in the literature.[1-7,9-11] The mean age of victims is 8.9 years (range 1.6 to 15 years).[1,9] The mean delay in hospital admission is generally prolonged in resource-constrained settings (mean 23hr-43min; range 1 - 65 hours). This is often due to the long distance between the scene of the incident and the specialist surgical center, lack of emergency medical evacuation services and the poor condition of roads. In our hospital, we see more and more of young victims with an average age of 3.6 years (range 3 months - 7 years). The circumstances of firearms injury are exceptional in civil practice (stray bullet). The average delay in specialist consultation is 17 hours (range 10 - 24 hours). The mode of transport of our patients was variable (ambulance or private car) contrary to the literature where patients benefited from the

services of the SAMU (Service d'Aide Médicale Urgente) or the fire brigades.[1,9-11]

Head injuries from firearms are serious with an average Glasgow Coma Score of 7.5 (range 6-9) in the literature.[1,9,13] It was 7 and 14 in our patients respectively. The circumstances of bullet injury in our patients were mysterious. It must have been stray bullets.

As with any head injury, a CT scan is the imaging of choice for the initial assessment. In addition to its availability and low cost, it provides a good demonstration of bone fractures and intracranial hemorrhages.[5] The two main limitations of this technique are artifacts caused by metallic bullets and patient movement. Multi-detector scanners have made it possible to considerably reduce the acquisition time and therefore the movement artifacts. In parallel, the adaptation of acquisition parameters and improvements in reconstruction algorithms have reduced metal artifacts.[3,4,6-9,12] In our patients, the CT scan had been done with a delay of 1 and 8 days respectively.

Management of cranioencephalic ballistic injuries is multidisciplinary involving critical care resuscitators, neurosurgeons, neurologists, physiotherapists and psychologists. The treatment is medico-surgical according to the symptomatology of the patient. The surgical management is controversial as well as challenging.[6,8,12] Some authors prefer minimal local debridement maintaining as much brain tissue as possible, while the others tend to avoid surgery altogether.[8,9] surgical operation may include decompression craniotomy,[11] craniectomy with removal of bone chips, conservative surgical debridement, extraction of the bullet, a water-tight dural closure and providing a skin coverage.[1]

Alvis-Miranda et al[12] summarize the treatment of ballistic head injuries in 4 steps: (1) Immediate saving of life through the control of bleeding and cerebral decompression, (2) Prevention of infection, by extensive debridement of all the contaminated, macerated or ischemic tissue, (3) Prevention of meningo-cerebral scars, (4) Restoration of dural and skin coverings. In our patients, surgical debridement and bullet extraction were indicated. It was done by craniotomy trans-cortical approach with the patients in supine position under general anesthesia and endotracheal intubation. The duration of surgery ranged between 90-130 min.

The severity of ballistic trauma depends on its clinical consequences, the site of injury and the lesion mechanism. Thus, unstable hemodynamic status, injuries involving the neck, trunk (esp. the heart) or groin, injuries by high-velocity bullets or hunting weapons, short-range firing injuries are indications of immediate surgical exploration.[10] The prognosis is variable depending on the expertise of the surgeon, occurrence of infection, surgical accessibility of the lesion site, the type of weapon used, the shooting distance, the age of the patient, the clinical status (e.g. GCS), the ballistic trajectory and the brain scan findings.[6] The additional prognostic factors in our setting are random shooting, low socio-economic level, delay in surgical consultation and insufficient technical platform.

Complications of ballistic head injury may be primary or secondary. Primary complications occurring at the time of original injury include bone fractures, intracranial hemorrhages, cerebral edema, cortical damage, pneumocephalus and pneumo-ventriculia.[6] Secondary complications include associated trauma of other organs, infections (e.g. extracranial soft tissue infection, skull osteomyelitis, meningoencephalitis and ventriculitis), post-operative vascular events (intracranial re-bleed, cortical ischemia due to vessel damage), false aneurysms due to arterial wall laceration, secondary displacement of bullet, hydrocephalus, epilepsy and encephalomalacia.[6,8,12]

CONCLUSION

Ballistic head injuries in children are very rare and serious, with a high morbidity and mortality, especially in resource-limited settings. In our center considerable delay in specialist consultation and inadequacy of technical platform are additional prognostic factors that need to be addressed.

REFERENCES

[1] Hamma OI, Assoumane I, Hissene TM, Hama SMI. Serious head trauma from firearm in children: two cases. Pan Afr Med J Clin Med 2023 Jun 21; 12: 18(9)

[2] Daban JL, Peigne V, Boddaert G, Ondo RO, Paul S, Debien B. Traumatisme Penetrant et Balistique. Le Congrès Medecins, Societe Francaise des Infirmieres Anesthesistes (SOFIA); 2012; 1-16. {Available from https://sofia.medicalistes.fr/spip/IMG/pdf/Traumatisme_penetrant_et_balistique.pdf} Accessed on 10 August 2024

[3] Gervaise A, Foscolo S, Rivierre A, Derelle A, Schmitt E, Braun M, Anxionnat R, Bracard S. Imagerie des traumatismes craniens par arme a feu [Imaging of cranial gun shot traumas]. J Radiol. 2010 Nov; 91(11 Pt 1): 1113-20. French.

[4] Stefanopoulos PK, Hadjigeorgiou GF, Filippakis K, Gyftokostas D. Gunshot wounds: A review of ballistics related to penetrating trauma. J Acute Dis 2014; 3: 178-185. DOI: 10.1016/S2221-6189(14)60041-X

[5] Li Y, Adanty K, Vakiel P, Ouellet S, Vette AH, Raboud D, Dennison CR. Review of mechanisms and research methods for blunt ballistic head injury. J Biomech Eng. 2023 Jan 1; 145(1): 010801.

[6] Alvis-Miranda HR, M Rubiano A, Agrawal A, Rojas A, Mos cote-Salazar LR, Satyarthee GD, Calderon-Miranda WG, Hernandez NE, Zabaleta-Churio N. Craniocerebral gunshot injuries; A review of the current literature. Bull Emerg Trauma. 2016 Apr; 4(2): 65-74.

[7] Tunthanathip T, Duangsuwan J, Wattanakitrungroj N, Tongman S, Phuenpathom N. Clinical nomogram predicting intracranial injury in pediatric traumatic brain injury. J Pediatr Neurosci. 2020 Oct-Dec; 15(4): 409-415.

[8] de Souza RB, Todeschini AB, Veiga JC, Saade N, de Aguiar GB. Traumatic brain injury by a firearm projectile: a 16 years experience of the neurosurgery service of Santa Casa de São Paulo. Rev Col Bras Cir. 2013 Jul-Aug; 40(4): 300-4.

[9] Pollion P, Senamaud-Dabadie K, Schnedecker B, Gonidec S, Musson T, Gromb S. A ten years old child with ballistic type of cranial trauma. Rev Med Legal 2014 Dec; 5 (4): 165-169.

[10] Rouvier B, Lenoir B, Rigal S. Les traumatismes balistique. SFAR - Société Française d'Anesthésie et de Réanimation 1997. {Available from https://urgences-serveur.fr/IMG/pdf/trauma1_balistique_sfar97.pdf} Accessed on 10 August 2024

[11] Moguel REA, Salas CC, Monterrubio RAC, Ramírez MMV, Márquez GM. Bilateral decompressive craniectomy in a patient with head injuries due to gun projectile: a case report. Arch Neurocienci, 2023 Feb;28(3): 44-48.

[12] Alvis-Miranda HR, Adie Villafañe R, Rojas A, Alcala-Cerra G, Moscote-Salazar LR. Management of Craniocerebral Gunshot Injuries: A Review. Korean J Neurotrauma. 2015 Oct;11(2):35-43.

[13] Marroquín AR. Gunshot Wound to the Head: Surgical Management and Cases Report in a Tertiary Care Center in Florencia, Colombia. Open J Modern Neurosurg 2015 July; 5(3): 84-87.

Address for communication: *Dr. Malangu Mhacks, Email : malanguntambwemax@gmail.com*

Received 14 July 2024; Accepted 2 September 2024

Acknowledgements : None
Conflicts of Interest : None declared by the author
Source of Funding : None
Ethical concerns : None (Reporting of routine clinical care)

Citation: *Malangu M, Feruzi M, Yogolelo R, Mutomb S, Tshishiku DJ, Arung W. Pediatric cranioencephalic trauma of ballistic origin. Pediatr Surg Trop 2024; 1(4): 286-290.*

PST PEDIATRIC SURGERY IN TROPICS

Pediatric Surgery in Tropics
2024 (Oct-Dec); Volume 1, Issue 4: Page 291

Book Review

Pediatric Thoracic Surgery

Vivek Gharpure

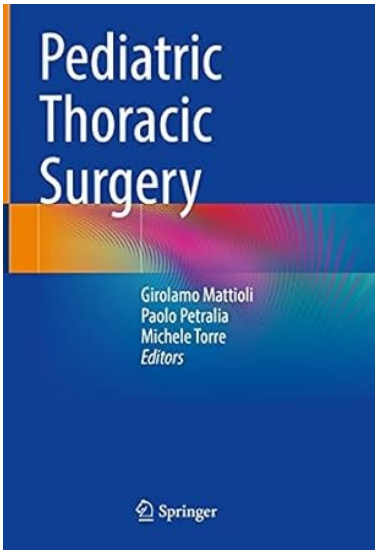
Chief Editor, Pediatric Surgery in Tropics

'**P**ediatric Thoracic Surgery', edited by Mattioli, Petralia and Torre, is written by surgeons working at Giannia Gaslini Institute, Genoa. This book emphasizes that thoracic surgery in children ought to be performed by pediatric surgeons. Considering the limited experience many pediatric surgeons may have in thoracic surgery, authors describe in a simple and direct way, common congenital and acquired thoracic anomalies, and their management, emphasizing the physical, functional and psychological development of the pediatric patient.

The chapters are comprehensive and cover almost all aspects of pediatric thoracic surgery. They include prenatal diagnosis, imaging, anesthesia, ECMO and various specific conditions. Thoracoscopic management is covered in considerable detail. Different tumors of thorax also receive comprehensive discussion.

However, certain conditions of the chest, such as the empyema and other infective conditions are not mentioned at all. Probably such illnesses no longer occur in the authors' practice. We continue to see such patients, and would have appreciated some discussion on those topics.

This book will prove useful to all pediatric surgeons across the world and be a welcome addition to the surgical bookshelf.



Bibliography

Title: Pediatric Thoracic Surgery
Editors: Girolamo Mattioli, Paolo Petralia, Michaele Torre
Publisher: Springer (16 July 2021)
https://doi.org/10.1007/978-3-030-74668-1.
ISBN 978-3-030-74667-4
ASIN: B099PDWVTY
Language : English
File size : 44847 KB
Text-to-Speech : Enabled
Screen Reader : Supported
Enhanced typesetting : Enabled
Print length : 488 pages

Pediatric Surgery in Tropics

Official journal of the Association of Pediatric Surgeons in Tropics

Author Index 2024 (Volume 1)

Pediatric Surgery in Tropics
Official journal of the Association of Pediatric Surgeons in Tropics

Subject Index 2024