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Hon'ble Health Minister Mr. Mansukh Mandaviya, Minister of State for Health & Family Welfare- Prof. S.P. Singh Baghel, Minister of State for Health & Family Welfare- Dr. Bharti Pravin Pawar and Dr. B.K. Rao-Senior Consultant in Institute of Critical Care Medicine, Sir Ganga Ram Hospital, New Delhi, during the online demonstration session of CPR programme conducted by NBEMS, to the nation.



Fig. A



Fig. B

A. Female cloaca case. Preoperative appearance and studies.
B. Postoperative appearance

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EDITORIAL

- Rising trend on cardio-vascular diseases in India after COVID-19**
Minu Bajpai and Abhijat Sheth 157

ORIGINAL ARTICLES

- The Persistent Cloaca**
Jayant Radhakrishnan, Rahul Gupta, Shumyle Alam and Anthony C. Chin 168

- A Study to Evaluate the Complications of Laparoscopic Renal and Adrenal Surgeries Using Modified Clavien-Dindo Classification**
Anil Kumar Nallabothula, Anirudh Suseel Nalumar, Harsha Vardhana Varma Mudunuri, Ashish Kumar Jayant and Anupam Bhargava 192

- Decompressive Craniectomy Outcomes in Cerebral Venous Sinus Thrombosis: A Comprehensive Analysis from a Tertiary Neurosurgical Center**
Rahul Sharma, Anand Katkar, Ashok Bhanage and Premkumar Reddy G 205

- Evaluating Anemia Counseling: A Study among Pregnant Women and Adolescent Girls in Bhanvad Taluka, DevBhoomi Dwarka, Gujarat**
Mithun Sanghavi, Swati Misra, Eshwar Kumar Gupta and Dipesh Parmar 219

- Endoscopic Pilonidal Sinus Treatment {Epsit} vs Limberg Flap for Pilonidal Sinus: A Single Center Experience**
Sudarsan Srikanth, Jameel Akhter, Surabhi Sreekumar and Poojitha 233

- Oral health situation analysis and integration of Massive Open Online Course Modules on Oral Health Promotion in WHO SEAR Countries: A Comprehensive Report**
Ritu Duggal, Vijay Mathur, Harsh Priya, Bharathi M Purohit, Shalini Gupta, Deepika Mishra, Manali Deb Barma, Sneha Malhotra, Upendra S Bhadauria and Deepali Agarwal 244

- A Comprehensive Review of Polatuzumab vedotin: Mechanisms, Clinical Applications, and Future Prospects**
Divya Shanthi and Sathiya Vinotha 256

CASE REPORTS

- Isolated Polycystic Disease of the Pancreas**
Vaibhavi P Patel, Parikh Tirth Vinaykumar, Siddharth S Parmar and Rajveersingh Chavda 264

- Cryptorchidism with Duplicate Vas Deferens and Epididymis: A Rare Case with Literary Review**
Anil Kumar Nallabothula, Anirudh Suseel Nalumar, Anupam Bhargava, Ashish Kumar Jayant and Harsha Vardhana Varma Mudunuri 270



National Board of Examinations - Journal of Medical Sciences
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EDITORIAL

Rising trend on cardio-vascular diseases in India after COVID-19

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"Post-acute COVID-19," "persistent COVID-19," "post-COVID syndrome," "long haulers," and "long COVID-19." These terms reflect the ongoing health issues experienced by individuals weeks or even months after their initial COVID-19 infection. The potential mechanisms contributing to these long-term symptoms include tissue and cell damage affecting vascular flow, increased blood clotting, the persistence of viral fragments or proteins in tissues, and alterations in the immune system. Furthermore, there are frequent reports of chronic cardiovascular effects of COVID-19, even among individuals with no prior cardiovascular disease (CVD), underscoring the significant impact of the virus on the cardiovascular system. Patients with pre-existing heart conditions are

particularly noted to have a worse prognosis during the acute phase of the SARS-CoV-2 infection [1].

There are several ways in which COVID-19 can affect the heart, resulting in temporary or lasting damage to heart tissue, such as lack of oxygen, myocarditis, blood vessel involvement, stress cardiomyopathy, etc. However, there is a lack of specific long-term data regarding the outcomes and impact on individuals with underlying heart diseases who have survived COVID-19 hospitalization. These mechanisms highlight the complex ways in which COVID-19 can impact the cardiovascular system, emphasizing the importance of monitoring and managing cardiac complications in individuals affected by the virus.

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Rising trend on cardio-vascular diseases in India after COVID 19

Non-communicable diseases, which are thought to be responsible for over 60% of all fatalities, include diabetes, chronic respiratory conditions, malignancies, CVD, and more. Cardiovascular diseases (CVDs), including ischemic heart disease and cerebrovascular accidents (strokes), are the primary cause of 17.7 million deaths worldwide. The World Health Organization estimates that India is responsible for one-fifth of all global deaths, particularly among younger people. According to the Global Burden of Disease report, India has an age-standardized CVD death rate of 272 per 100,000 people, which is significantly higher than the 235 global average [2,3].

Indians were affected by CVDs ten years earlier than people in the West. The high death rate, quick progression, and early age of initiation of CVD are very concerning to us Indians. The rates of coronary artery disease (CAD) are highest among Indians, and traditional risk factors are unable to account for this elevated risk. Regarding the cardiac mortality and morbidity for the Indian subcontinent, there are no systematic data collection methods, and the majority of deaths occur at home with the reason of death unknown. Data on CV morbidity and mortality from hospitals could not accurately reflect the whole burden of CV disease. Compared to 15.2% and 6.9%, respectively, in 2015, CVDs accounted for 28.1% of all deaths and 14.1% of all disability-adjusted life years (DALYs) in India [4].

Additionally, elevated blood pressure and cholesterol are most common in these states. Acute coronary syndrome and ST-elevation myocardial infarction (MI) are currently the most common in India. In addition to other CVDs,

hypertensive heart disease is a major issue in India, accounting for 261,694 fatalities in 2013 (a 138% rise from 1990). In India, rheumatic heart disease is still considered to be an epidemic, with 1.5–2 cases per 1000 people [5].

The frequency of CAD is three times higher in migrant Asian Indians than in the native population. Indians are liable to get hospitalized 2–4 times more frequently for complications of CAD, in comparison with other ethnic groups, and admission rates are 5–10 times higher for populations younger than 40 years. For Indians living in India, the prevalence of CAD is 11% for those without diabetes and 21.4% for those with diabetes. The prevalence of CAD in rural parts of the country is nearly half that in urban population [5].

Cardiovascular complications are associated with COVID-19, emphasizing that these sequelae can occur not only in symptomatic patients but also in asymptomatic individuals. It notes that up to 20% to 30% of patients hospitalized with severe COVID-19 show evidence of myocardial involvement, which can manifest as elevated troponin levels, venous thromboembolism, heart failure, and arrhythmias. Elevated troponin levels in acutely symptomatic patients have been linked to poor outcomes and higher in-hospital mortality rates. The passage highlights various proposed mechanisms for cardiovascular complications of COVID-19, including direct damage to cardiomyocytes, hypoxia-related damage, microvascular dysfunction, thrombosis, and cytokine storm.

Furthermore, myocardial involvement may initiate an inflammatory process leading to fibrosis, which can be detected through cardiac magnetic

resonance imaging (MRI). Long-term sequelae of COVID-19-related myocardial involvement may include increased cardio-metabolic demands, myocardial fibrosis or

scarring, persistent left ventricular dysfunction, heart failure, arrhythmias, inappropriate sinus tachycardia, and autonomic dysfunctions (Figure 1).

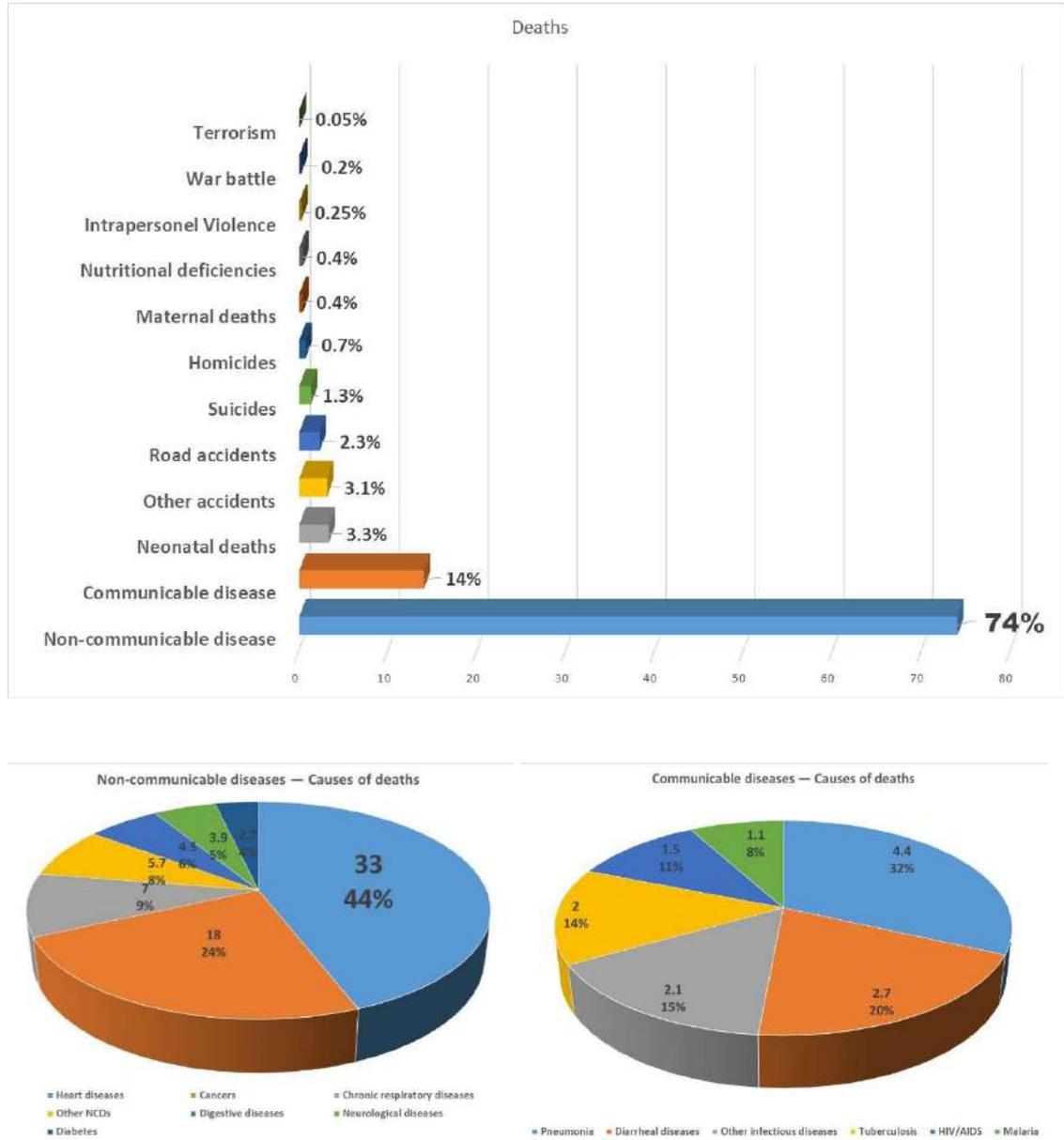


Figure 1. Global burden of disease
Source: IMHE Global Burden of Diseases and Global Terrorism Database

OurWorldData.org – Research Data to Make Progress Against the World’s Largest Problems

Global trend on cardio-vascular diseases after COVID-19

Cardio-vascular diseases (CVD) are responsible for nearly one-third of all deaths globally, with over 17 million deaths attributed to them in 2017 alone. The number of deaths from CVD has been steadily increasing, and it is projected to be the cause of more than 23 million deaths worldwide by 2030 [6].

While age-standardized mortality rates for CVD have declined over the last decades, the actual number of deaths has increased significantly. This trend underscores the growing burden of CVD on global health. Although developing countries have historically experienced higher rates of death from CVD, the disease is increasingly prevalent in developed countries as well. Over three-quarters of CVD deaths occur in low- and middle-income countries, indicating a growing epidemic problem in recent years. Several factors contribute to the high burden of CVD, including diabetes, obesity, a lack of physical activity, hypertension, an unhealthy diet, and excessive alcohol consumption. These risk factors highlight the importance of preventive measures and lifestyle interventions in reducing the incidence and impact of CVD. The CVD imposes significant costs on healthcare systems worldwide, affecting quality of life, life expectancy, and healthcare expenditures in various countries. To counter the increasing trend of CVD, it requires comprehensive strategies aimed at prevention, management, and treatment to improve public health outcomes and reduce healthcare costs.

Hon'ble Health Minister Mr. Mansukh Mandaviya's statement on the finding from the Indian Council of Medical Research (ICMR) highlights a concerning trend of rising heart attacks among young individuals who have recovered from severe COVID-19 infection

The ICMR conducted a detailed study that revealed a rise in heart attacks among young individuals who had recovered from severe COVID-19 infections. The study recommended that individuals who had severe COVID-19 should avoid extra labor and strenuous activities such as laborious running and exercise for a specified period, typically one to two years, to prevent heart attacks. There has been an increasing number of incidents of young individuals in their twenties and thirties experiencing fatal heart attacks, with some attributing these fatalities to COVID-19 or its treatment. This trend is concerning and underscores the need for further research and awareness about the cardiovascular implications of COVID-19. Recent reports have documented heart attack deaths among young individuals during cultural events such as garba nights in Gujarat. Despite medical teams being on standby at venues, there have been instances of fatal heart attacks during these events. The reported heart attack deaths during garba events include individuals as young as 17 years old, highlighting the severity of the issue and the impact on the younger population [7] (Figure 2).



Figure 2. Hon'ble Health Minister Mr. Mansukh Mandaviya, Minister of State for Health & Family Welfare- Dr. Bharti Pravin Pawar & Minister of State for Health & Family Welfare- Prof. S.P. Singh Baghel and Dr. Abhijat Sheth, President- National Board of Examinations in Medical Sciences

Cardiovascular diseases (CVDs), the world's leading cause of death, claim as many as 17.9 million lives each year, globally. According to a study by the Indian Council of Medical Research and the Registrar General of India, India accounts for approximately 60 percent of the world's heart disease burden [8].

The Hon'ble Prime Minister's flagship Ayushman Bharat programme guarantees impoverished and deprived classes free medical care up to Rs 5 lakh

The National Board of Examination in Medical Sciences has geared up for providing expert care in the public sector, thus impeding the development of the rural cardiology system's infrastructure.

NBEMS - Joint Accreditation scheme

To utilize the available resources of hospitals and to provide them with an opportunity to provide PG training, joint accreditation of the hospitals has been approved by the Governing Body, NBEMS, in its meeting dated 25.11.2022 [9].

The aim is to deploy cardiologists and thoracic and cardiovascular surgeons at the grassroots to attend to sudden emergencies. Most of the casualties occur due to a lack of skills to deal with in golden hours, which could be easily handled by using non-conventional cardiac skills.

Community Health Centres/ Primary Health Centres across UP and even in some other parts of the country would receive a support system to deal with growing cases of CVD [10].



Figure 3. Hon'ble Health Minister Mr. Mansukh Mandaviya, Minister of State for Health & Family Welfare- Prof. S.P. Singh Baghel, Minister of State for Health & Family Welfare- Dr. Bharti Pravin Pawar and Dr. B.K. Rao-Senior Consultant in Institute of Critical Care Medicine, Sir Ganga Ram Hospital, New Delhi, during the online demonstration session of CPR programme conducted by NBEMS, to the nation.

A team of experts led by Parul Naib from Delhi University, Pulkit Kumar from Tata Institute of Social Sciences, Sudha Chandrashekar from the London School of Hygiene and Tropical Medicine at the University of London, and Owen Smith from Harvard University in Cambridge, Massachusetts, USA, examined **“Trends in Cardiac Care Utilization under Ayushman Bharat.”**

The team examined every cardiac claim submitted under the Pradhan Mantri Jan Arogya Yojana (PM-JAY) for a span of 17 months, starting in September 2018 and ending in February 2020. It was discovered by 26%, suggesting that a substantial amount of the program was used to give people from the lowest socioeconomic groups free cardiac treatment [11].

According to a study that was published in the medical journal "The

Lancet Global Health," the number of fatalities in India in 2015 that were attributable to cardiovascular disease was rising, accounting for more than 25% of all deaths in the nation. According to the survey, young people and those living in rural areas are the groups most affected. According to the data, between 2000 and 2015, the death rate from ischemic heart disease rose significantly among Indians aged 30 to 69 living in rural areas, surpassing that of urban areas [10].

This has virtually compelled a process of brain drain to other countries as most of the trained non-conventional clinical cardiologists left the country and started practicing in countries like Europe, USA and UAE—where they are much in demand. The private sector is also engaging them, like Medanta, Narayana Hrudayalaya Bangalore, Escorts Heart Institute, U.N.

Mehta Heart Hospital in Gujarat, Asian Heart Institute Mumbai and many more. These MBBS doctors learned clinical cardiology under the best experts and are capable of saving lives during the initial moments after a heart attack.

Government steps to strengthen the cardio-vascular services in the country

The government of India has taken several steps to tackle the shortage of cardiologists and improve cardiovascular healthcare in the country, such as increasing medical education, specialized training programs, encouraging public-private partnerships, telemedicine and telecardiology services, capacity building, public awareness and prevention programs, regulatory measures and incentives for rural services.

These steps demonstrate the government's efforts to address the shortage of cardiologists and improve cardiovascular healthcare in India, with a focus on increasing access to specialized care, promoting preventive measures, and strengthening the healthcare infrastructure.

Role of NBEMS to increase the workforce of cardiologists in the country

The National Board of Examinations in Medical Sciences (NBEMS) plays a crucial role in generating and strengthening cardiologists in India through various initiatives related to medical education, training, and certification. By accrediting training programs in cardiology, NBE ensures that these programs meet the required standards of education and training, thereby producing competent cardiologists. The NBE conducts various postgraduate medical examinations, including Diplomate of National Board (DNB) examinations in

various specialties, including cardiology. These examinations provide a pathway for medical graduates to specialize in cardiology and obtain certification as cardiologists. The NBE sets standards for medical education and training in cardiology, including curriculum development, assessment methods, and training guidelines. By establishing and maintaining high standards, NBE contributes to the quality of cardiology education and training in India.

The NBE also promotes continuous professional development among cardiologists by offering certification and recertification programs, continuing medical education (CME) activities, and other initiatives to update their knowledge and skills. NBE supports research and innovation in cardiology by encouraging research activities among trainees and providing platforms for presenting and publishing research findings. This contributes to the advancement of knowledge and practices in the field of cardiology. The NBE ensures quality assurance in cardiology training programs and examinations through rigorous evaluation processes, including regular inspections, assessments, and feedback mechanisms. This ensures that cardiologists trained through NBE-accredited programs meet the required standards of competence and professionalism.

Role of Wellness centres in the Hon'ble Prime Minister's flagship Ayushman Bharat programme: Preventive care and frequent health checkups

Preventive care and frequent health checkups can play a significant role in controlling the prevalence of CVDs in India, as they can help identify and manage

risk factors early, prevent the onset of CVD, and reduce the burden of the disease. Regular health checkups can help identify risk factors for CVD, such as high blood pressure, high cholesterol, diabetes, obesity, and smoking. Early detection of these risk factors allows for timely interventions to manage and control them. Preventive care visits provide an opportunity for healthcare providers to educate individuals about healthy lifestyle choices, including a balanced diet, regular exercise, smoking cessation, and stress management. These lifestyle modifications can help reduce the risk of developing CVD. Health checkups may include screenings for CVD, such as electrocardiograms (ECGs), echocardiograms, and stress tests. These screenings can help detect early signs of heart disease or abnormalities in heart function, allowing for timely interventions and treatment.

Regular health checkups and preventive care can help prevent complications associated with CVD, such as heart attacks, strokes, and heart failure. By managing risk factors and addressing early signs of CVD, individuals can reduce their risk of developing these complications. Implementing preventive care measures and promoting frequent health checkups at the population level can have a broader impact on reducing the prevalence of CVD in India. By improving access to preventive care services and promoting health awareness, the burden of CVD can be reduced across the population.

Emergency, critical care services and cardiopulmonary resuscitation (CPR)

Strengthening emergency medicine and critical care services is indeed crucial for effectively addressing the burden of

CVDs in India. Emergency medicine and critical care services are essential for providing timely and effective management of acute cardiovascular events such as heart attacks (acute myocardial infarction) and strokes. Prompt initiation of treatments like thrombolytic therapy, percutaneous coronary intervention (PCI), and clot-busting medications can significantly improve outcomes and reduce mortality rates. Strengthening emergency medicine services can help reduce the time from arrival at the hospital to the administration of life-saving interventions like thrombolytic therapy or PCI (known as door-to-needle and door-to-balloon times, respectively). This requires well-equipped emergency departments, trained staff, and efficient systems for triage and patient management. Improving pre-hospital emergency care services, including ambulance systems and emergency medical services (EMS), is critical for ensuring the timely transport of patients with acute cardiovascular emergencies to hospitals equipped to provide appropriate care. This includes training paramedics and emergency medical technicians (EMTs) in the recognition and initial management of cardiac emergencies.

Increasing the capacity and capabilities of critical care units, including coronary care units (CCUs) and intensive care units (ICUs), is essential for managing critically ill patients with complex cardiovascular conditions. This involves ensuring the availability of specialized equipment, such as ventilators, cardiac monitors, and defibrillators, as well as a skilled multidisciplinary team of healthcare providers. Providing specialized training and education programs for healthcare professionals in emergency medicine, critical care, and cardiology is essential for

building a skilled workforce capable of managing acute cardiovascular emergencies. This includes continuous medical education (CME) programs, workshops, and simulation-based training sessions.

Leveraging telemedicine technologies can help extend the reach of emergency medicine and critical care services to underserved areas, enabling remote consultation, triage, and decision-making support for healthcare providers managing cardiovascular emergencies. Investing in research and quality improvement initiatives focused on emergency cardiovascular care can help identify best practices, optimize protocols and pathways, and continuously improve the quality of care delivered to patients with CVD in emergency settings.

NBEMS CPR workshop

The National Board of Examinations in Medical Sciences (NBEMS) has planned a nation-wide programme to raise awareness and train the public in cardiopulmonary resuscitation (CPR). The first workshop was conducted on December 06, 2023 & Union Health Minister Dr. Mansukh Mandaviya launched this public awareness campaign on Cardiopulmonary Resuscitation (CPR) Awareness by NBEMS. "It is imperative that patient receive treatment immediately after suffering a cardiac arrest; hence, awareness and adequate training in CPR are paramount, stated the Hon'ble Minister, Dr. Mansukh Mandaviya. More than 20 lakh people participated in the CPR training across the nation. CPR plays a crucial role in spreading awareness about cardiopulmonary resuscitation (CPR) during emergency situations. CPR is a life-saving skill that can significantly improve

the chances of survival for individuals experiencing cardiac arrest or other medical emergencies. The NBEMS CPR workshop educates participants on the proper techniques and procedures for performing CPR effectively, empowering them to act swiftly and confidently in emergency situations. During a cardiac arrest or similar emergency, every second counts. The NBEMS CPR workshop teaches participants how to recognize the signs of cardiac arrest, initiate CPR promptly, and continue life-saving measures until professional medical help arrives. This immediate response can make a crucial difference in saving lives.

Impact of vaccination after COVID-19 infection

The study by Zubair Akhtar et al. acknowledged the potential for reporting bias, particularly concerning the availability of mRNA vaccines in developed countries. This bias may influence the observed differences in cardiac events between vaccine types [12].

Despite the observed cardiac risks associated with COVID-19 vaccination, the study emphasizes that the personal and public health benefits of COVID-19 immunization far outweigh the modest cardiac risks. Furthermore, any cardiac events following vaccination were generally transient and resolved within a few days or weeks.

Population-based studies have indicated that the risk of cardiac complications, including myocarditis and pericarditis, is significantly higher after SARS-CoV-2 infection compared to mRNA COVID-19 vaccination, particularly among adolescents aged 12–17 years [13].

The outcome of CVD in COVID-19 vaccinated individuals compared to non-vaccinated or uninfected populations can vary based on several factors, including the severity of the COVID-19 infection, pre-existing cardiovascular health status, and individual risk factors.

While myocarditis and pericarditis are known complications of mRNA COVID-19 vaccines, especially in certain demographic groups, the incidence is rare, and the benefits of vaccination in preventing COVID-19 outweigh the risks associated with these cardiac complications. It's essential for healthcare providers and individuals to weigh the risks and benefits of vaccination based on individual health status and vaccination eligibility.

Conclusion

The COVID-19 infection is identified as an independent risk factor for cardiovascular disease. Patients with COVID-19 may experience various cardiovascular complications, including myocardial injury, pericarditis, coagulopathy, myocardial infarction, heart failure, arrhythmias, and persistent post-acute adverse cardiovascular outcomes. While the COVID-19 vaccination is generally cardioprotective, it can lead to myocarditis or pericarditis in some cases, particularly among young males. However, the incidence of myopericarditis following vaccination is lower compared to the risk associated with SARS-CoV-2 infection.

Increased awareness among primary care physicians regarding potential cardiovascular causes of non-specific post-COVID-19 symptoms is crucial. This includes recognizing the signs of myocarditis, pericarditis, and other cardiac complications in younger adults and

promptly referring them for further evaluation and management. Additionally, optimal management of cardiovascular risk factors and clear diagnostic, referral, and management pathways for patients with non-specific symptoms are essential to rule out cardiac complications.

The current scenario emphasizes the importance of comprehensive strategies to address the cardiovascular implications of COVID-19 infection and vaccination, including early detection, prompt management, and preventive measures to mitigate the risk of cardiovascular complications in affected individuals.

Statements and Declarations

Conflicts of interest

The authors declares that they do not have conflict of interest.

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ORIGINAL ARTICLE

The Persistent Cloaca

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Abstract

Normally, at birth, females have separate perineal orifices for the urinary, reproductive and digestive tracts, while males have individual openings for the urogenital and digestive tracts. However, during fetal development, both sexes pass through a stage when all the systems empty into a common chamber, the cloaca. The cloaca opens to the exterior through a solitary opening until it divides into the various systems. Occasionally, the tracts do not separate and the baby is born with a persistent cloaca. It is a complex anomaly that is often associated with abnormalities in other systems that have to be taken into consideration to manage the patient appropriately.

Keywords: Persistent cloaca, anorectal anomalies, cloacal anomalies

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Graphical Abstract

THE PERSISTENT CLOACA

Jayant Radhakrishnan, Rahul Gupta, Shumyle Alam and Anthony C. Chin

Abstract
Normally, at birth, females have separate perineal orifices for the urinary, reproductive and digestive tracts while males have individual openings for the urogenital and digestive tracts. However, during fetal development, both sexes pass through a stage when all the systems empty into a common chamber, the cloaca. The cloaca opens to the exterior through a solitary opening until it divides into the various systems. Occasionally the tracts do not separate and the baby is born with a persistent cloaca. It is a complex anomaly that is often associated with abnormalities in other systems that have to be taken into consideration to manage the patient appropriately.

Male cloaca

Results:
The objectives of treatment are not only for the patient to attain socially acceptable fecal and urinary continence but to permit girls to function sexually, to have the capacity to get pregnant and carry babies to term. In males also, fecal and urinary control and the potential for sexual activity are the goals of management.



National Board of Examinations
Journal of Medical Sciences

Conclusion: Persistent cloaca is a complicated problem that must be approached after careful evaluation at a center with experience in the management. With proper care excellent results are achievable. Therefore, it seems reasonable that, if not all patients with a persistent cloaca, at least long-channel patients should be transferred as soon as possible to a center with the expertise to treat them. Improper intervention prior to referral makes subsequent correction difficult and results in poor outcomes.

Introduction

The overwhelming majority of placental mammals have separate orifices from which they evacuate urinary, genital and intestinal contents. In contrast, amphibians, birds, and reptiles have a single cavity, the cloaca (from Latin: sewer). Urine, stool and products of reproduction are stored in the cloaca and expelled through its solitary orifice [1].

Embryology

In the human fetus, the cloaca is a temporary widening of the hindgut at 5 weeks of gestation. It is lined with endoderm and it divides into the posterior hindgut and the anterior urogenital section by the 7th week. The original hypothesis was that a wedge of mesenchyme, known as the urorectal septum (URS), develops between

the allantois and the hindgut and it grows in a craniocaudal direction until it fuses with the cloacal membrane, thereby dividing the cloaca into the urogenital sinus (UGS) anteriorly and the rectum posteriorly. It was further believed that the failure of the URS to extend caudally and to fuse with lateral infoldings of the cloaca, Rathke's folds, resulted in anorectal anomalies, including the persistent cloaca. This theory was upended in 1986 when van der Putte demonstrated in the pig embryo that the shift of the dorsal part of the cloaca and the adjacent gut to the body surface of the tail groove is the predominant event in the development of the anorectum. In other words, the caudal extension of the URS is a secondary, passive process that results from differential growth in the cloacal region which, in turn, causes the caudal body axis

of the embryo to unfold. The dorsal part of the cloacal membrane must regress for this step to transpire. The genesis of this part of the cloacal membrane prevents the normal shift of the anorectum to the surface of the body, resulting in the development of anorectal malformations. According to van der Putte the size and nature of this defect dictate the type of resultant anorectal anomaly. Van der Putte further postulated that deformities of the cloacal membrane could be the cause of cloaca-derived malformations [2]. In 1995, Kluth et al. compared the cloacal region of normal rats to that of semi-dominant gene mice (SD mice), as the latter often have abnormal cloacae. They found that in the embryos of abnormal mice, the cloacal membrane is too short and the site of the future anal opening is not identifiable, while in normal rats, this site can be identified soon after the cloacal membrane forms. They found a variety of cloacal malformations in SD mice that conform to the types of anorectal malformations found in humans. Their findings also confirmed that a normal URS forms passively when cloacal development is normal and that the URS is not the primary agent in normal cloacal development [3]. In 2018, passive growth of the URS was demonstrated in human embryos between 4 and 10 weeks of development. The authors found that the ventral and central areas of the cloaca grew rapidly, while there was almost no growth in the cranial and dorsal areas. This differential growth resulted in separate urogenital and anorectal compartments. Later, differential growth straightened the curved caudal body axis of the embryo and in the process, the

URS advanced caudally. Dysregulation of dorsal cloacal growth could be the cause of anorectal malformations [4].

Although we now believe that we understand cloacal development, we still do not know why normal differentiation of the cloaca is thwarted in some instances [5].

Incidence

Persistent cloaca is estimated to occur in 1:20,000–50,000 births. It is reported almost exclusively in females with a 46 XX chromosomal structure. However, it has been identified in human [6,7] and canine [8] males on rare occasions. Whether it is truly rare in males or has been misdiagnosed as an anorectal malformation with a fistula to the urethra is an unanswered question.

Pathological anatomy

In Females, The basic abnormality in female patients is a confluence between the bladder-urethral complex, the Müllerian structures and the hindgut. All the structures empty into one cavity, which opens to the exterior through a solitary perineal orifice under a hooded clitoris. However, there is considerable internal variation amongst the components that must be delineated carefully in each patient before settling on a plan for surgical correction [9,10].

To manage these girls appropriately, the following issues must be defined clearly:

1. Whether the components fuse at the same location or at different sites. The site of communication between the components must be precisely localized.

2. The length of the common channel helps determine the surgical approach and the likelihood of eventual urinary continence. It has generally been accepted that a common channel shorter than 3cm. can be repaired entirely from a sacral approach, while in those with a common channel longer than 3 cm an abdominal exposure is also required [11-13].
 3. The length of the urethra also matters. A urethral length of 1.5 cm with a short common channel requires a simpler perineal operation and results in better continence. On the other hand, if the urethra is short and the common channel is long, not only is abdominal exposure required, but ultimately, continence is not as satisfactory [14].
 4. An anatomically normal bladder could be distended if the urethra is obstructed, or it may fail to empty if associated spinal lesions render it neurogenic. It could also be distended with mucus if the Müllerian structures and/or the hindgut drain directly into it.
 5. The greatest variations occur in the Müllerian structures and they must be carefully identified. Any of the following variations are possible: a normal uterus and vagina opening at a common confluence with the bladder and hindgut: a normal uterus perched atop a vagina filled with mucus and/or urine: a didelphic uterus in which one or both sides are distended. Furthermore, the septum between the halves may be partial or complete and the vagina and uteri may be symmetric or asymmetric if one side is atretic. Finally, the two Müllerian ducts may enter the bladder separately [13].
 6. The hindgut typically enters the confluence, but rarely, it may end blindly without any communication with the other structures. In addition, when the two Müllerian ducts do not fuse and enter the bladder individually, the hindgut may also drain between the two of them directly into the bladder.
 7. It is also essential to determine whether the hindgut has descended far enough towards the perineum to be brought down to its desired location using only a perineal approach or whether the abdomen would have to be entered to free up an adequate length of the rectum.
- Cloacal anomalies may be classified as follows [9]:
1. Persistent cloaca. It is the most common lesion and has a solitary perineal opening.
 2. Cloaca variant.
 3. Posterior cloaca.
 4. Posterior cloaca variant.
 5. Urogenital sinus (UGS).
 6. Cloacal dysgenesis. It has no perineal opening and is the least common and most severe of all these anomalies.
- In Males.** In males with a persistent cloaca, the urinary tract and the rectum drain into a short common channel, which constitutes the only external outlet for both structures. On occasion, atrophic Müllerian

structures may also communicate with the common channel [6,7].

Associated abnormalities

To facilitate diagnosis and therapy, other abnormalities seen in these patients could be divided into two broad groups: associated developmental lesions and coexistent complications.

Associated developmental lesions.

Persistent cloaca is the most severe form of anorectal anomaly; hence, vertebral, cardiac, tracheo-esophageal, renal and limb (VACTERL) abnormalities are to be expected. It is particularly important to look for spinal and renal/urinary abnormalities [15].

In patients with persistent cloaca, vertebral anomalies affect the lower vertebrae and the sacrum. Some vertebrae may be missing or deformed, the sacrum could be deformed or absent and a tethered spinal cord may create a neurogenic bladder and also cause problems with fecal control and evacuation after reconstruction.

Renal and urinary abnormalities are also common. Renal agenesis, horseshoe kidney, hydronephrosis, ureteric duplication, vesicoureteric reflux and obstructive uropathy have all been reported. It is important to note that sometimes these problems may not be identified initially.

Numerous limb abnormalities of varying severities may also coexist. Examples are polydactyly, syndactyly, radial aplasia and hypoplasia or displacement of the thumb.

Coexistent complications.

Coexistent complications involving the

bladder, Müllerian structures and hindgut mentioned previously are much more of a problem and require careful evaluation and management either before or when the cloaca is being corrected.

Antenatal diagnosis

Antenatal ultrasonography is the obvious screening test since it is noninvasive, easy to carry out, avoids radiation and is inexpensive. Suspicious findings consist of cystic lesions in the pelvis of a female fetus, a highly placed rectum, fetal ascites, renal abnormalities, an irregular appearance of the bladder, oligohydramnios and ambiguous genitalia. However, all these findings could be due to other pathologies. It is claimed that a fluid-filled, dilated colon with intraluminal enteroliths and meconium in the form of debris in the urinary tract constitutes the most specific combination of findings to diagnose a persistent cloaca [9]. Additionally, the anal sphincter can also be evaluated by ultrasonography.

Magnetic Resonance Imaging (MRI) of the fetus is the definitive test to confirm the diagnosis and also to evaluate each entity in detail. MRIs are carried out under carefully defined criteria to help differentiate the six variants of cloacal abnormalities mentioned above [9] (Figures 1 to 5).

1A.



1C

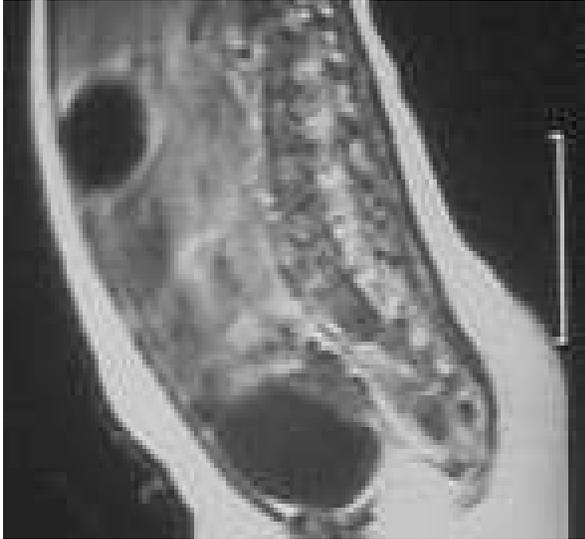


1B.



1D.

1E.



1F.

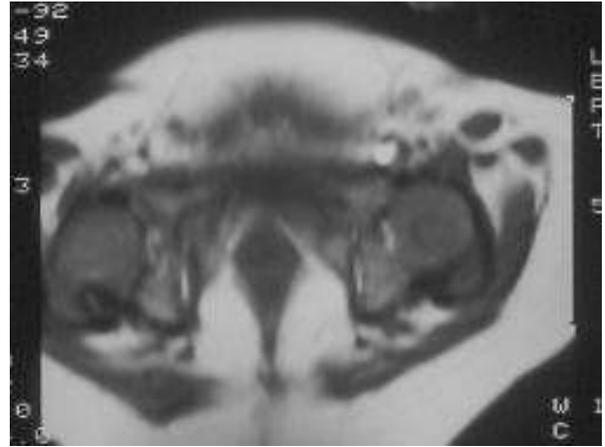


Figure 1. Female persistent cloaca case #1. This patient was referred for evaluation as her abdomen was distended, and no perineal orifices were visible to the examiner.

1A. Patient is supine. She has mild clitoral hypertrophy and excess of preputial skin. The solitary orifice at the base of the clitoris is hidden by the skin fold. The midline raphe is well-defined and she had a well-developed anal sphincter.

1B. Plain anteroposterior abdominal radiograph at birth demonstrating massive abdominal distension with a ground glass appearance and a few loops of intestine displaced upwards by a very large pelvic mass, the hydrocolpos. The hydrocolpos was catheterized and drained through the urogenital sinus (UGS) and a diverting colostomy was created in the descending colon.

The patient was studied in detail once she was thriving.

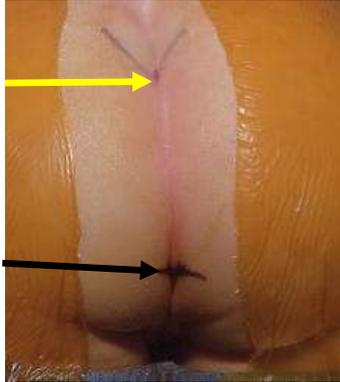
1C. Lateral radiograph of sinogram. The yellow arrow points to the bladder, and the white arrow indicates the UGS and vagina. The red arrow demonstrates the hindgut.

1D. Lateral radiograph of the distal colostogram through the colostomy. There is an adequate length of the distal colon for a pull-through procedure.

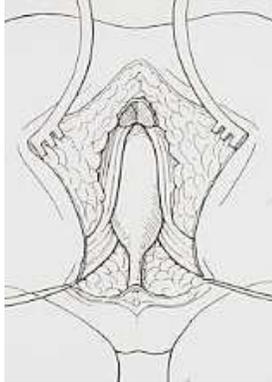
1E. MRI: Normal spinal cord with no tethering.

1F. MRI: Normal anal sphincter mechanism on transverse section through the pelvis.

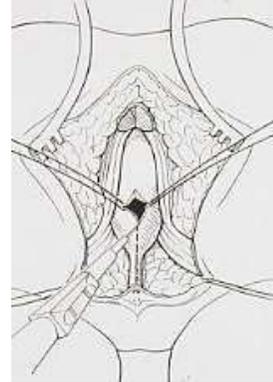
1G.



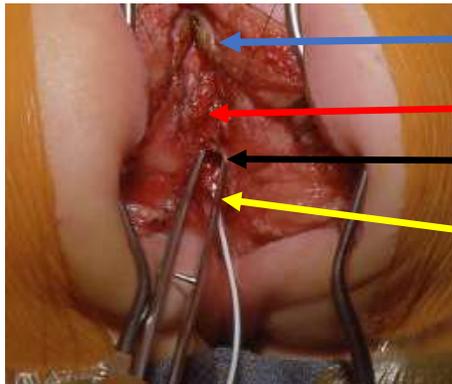
1H.



1J.

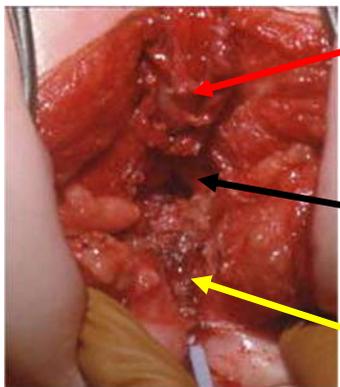


1K.



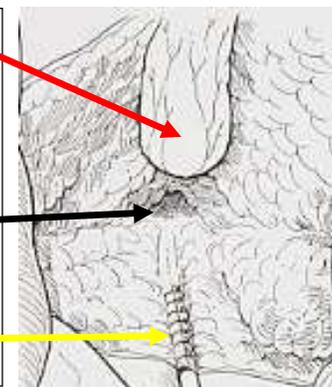
- Divided coccyx
- Open hindgut
- Vaginal opening (forceps within)
- Open UGS (with vessel loop)

1L.

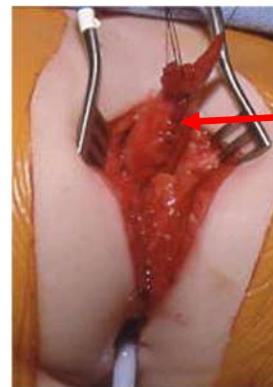


- Hind gut
- Vagina
- Neourethra

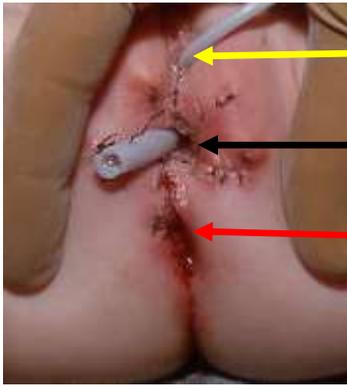
1M.



1N.



1O.



1P.



Case #1 cont'd. Steps of surgical reconstruction by posterior sagittal ano-recto-vagino-urethroplasty (PSARVUP)

1G. With the patient in the knee-chest position the tip of the coccyx (yellow arrow) and the center of the anal sphincter (black arrow) delineate the extent of the incision.

1H. The midline incision is deepened until the posterior wall of the cloaca and urogenital sinus are clearly visualized.

1J. The posterior wall of the cloaca and UGS are incised down to the perineal opening.

1K. Photograph of the open cloaca demonstrating the open hind gut (red arrow), the vaginal opening (black arrow) and the open UGS (yellow arrow).

1L and 1M. Photograph and illustration of the structures after dissection is complete. Hind gut (red arrow), vaginal orifice (black arrow) and urethra reconstructed from the UGS (yellow arrow).

1N. The urethra has been reconstructed and the vagina mobilized and sutured to the perineum. The hind gut has been mobilized and tapered in preparation for it to be sutured at the perineum (red arrow).

1O. Appearance of the perineum 1 week after surgery with the patient supine. The foley catheter (yellow arrow) is in the neourethra, the rubber tube (black arrow) is in the vagina and the neoanus is visible posteriorly (red arrow).

1P. Antero-posterior radiograph of the postoperative voiding cystourethrogram (VCUG) demonstrating good capacity, shape and retaining ability of the bladder. She was able to void spontaneously.

2A.



2B.



Figure 2. Female persistent cloaca case #2. This patient was referred to us at 4 months old as she was not thriving. Her birth weight was 7 lbs. and she had not gained any weight in the 4 months. She was also septic. At birth a diverting ileostomy had been carried out elsewhere. Later they also carried out a

vesicostomy believing that her recurrent hydrocolpos was due to retrograde drainage of urine into the vaginae. The vesicostomy did not correct the problem.

2A. Photograph demonstrating a loop ileostomy (red arrow) and vesicostomy (yellow arrow).

2B. Close up of the vesicostomy showing severe candidiasis.

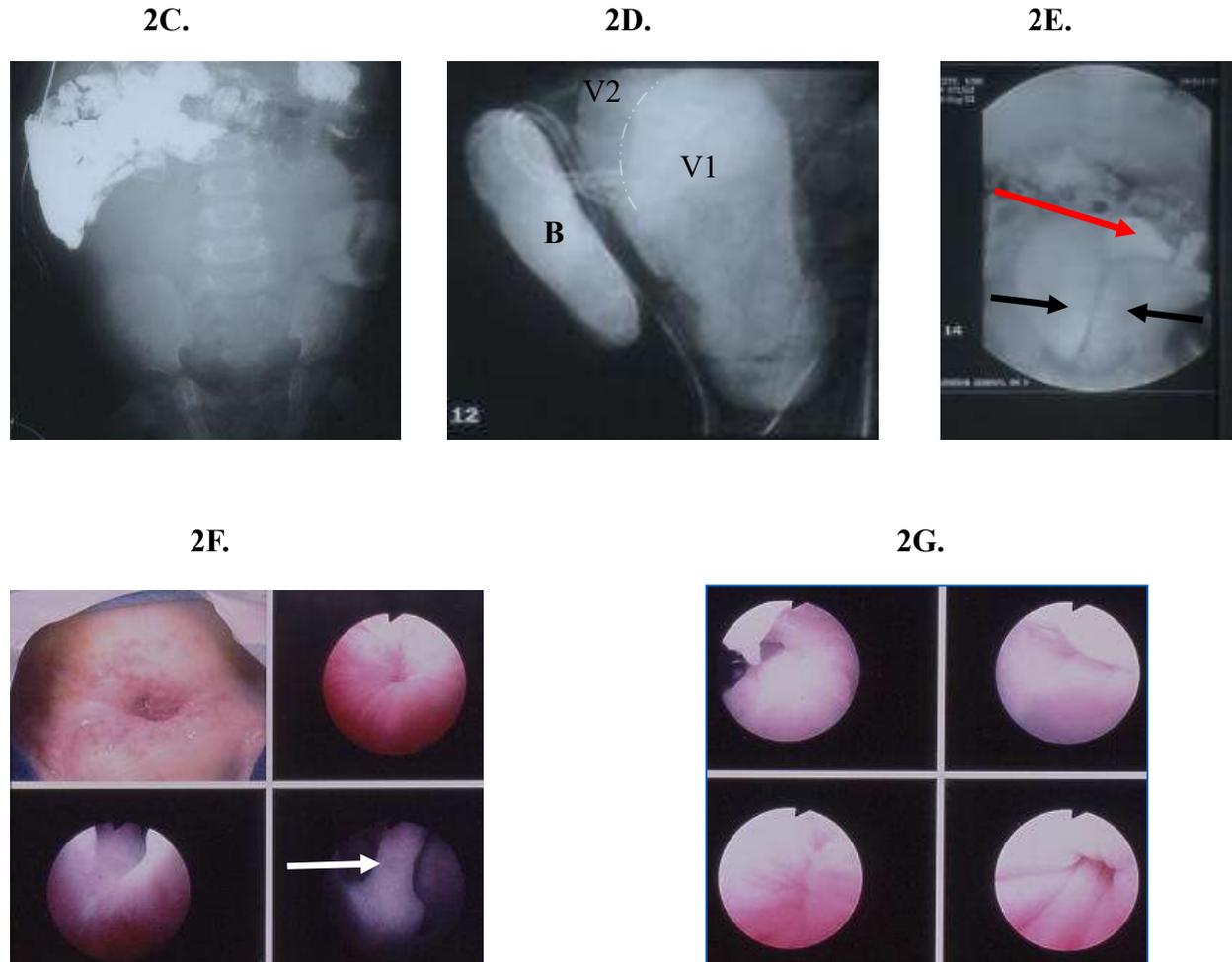


Figure 2. Female cloaca case #2 (cont'd.). Preoperative studies

2C. Upper gastrointestinal contrast study revealing upward displacement of intestines by a large mass (hydrocolpos) arising from the pelvis.

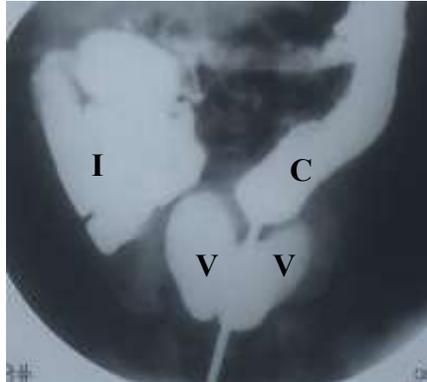
2D. Sinogram carried out through the UGS after plugging the vesicostomy. The bladder (B) is seen to the left and the large double vagina to the right. The two halves of the vagina are V1 and V2. The dashed white line in an arc demarcates the upper extent of the left vagina with the right vagina visible above it.

2E. Antero-posterior view of the sinogram after emptying the bladder. The two hemivaginae (black arrows) are visualized and contrast also enters the colon (red arrow).

2F. Cystoscopy through the vesicostomy. Top left: Vesicostomy. Top right: Bladder neck visualized from within the bladder. Bottom left: Septum between the hemivaginae visualized upon entering the UGS from the bladder. Bottom right: Orifice visible on the septum between the vaginae (white arrow).

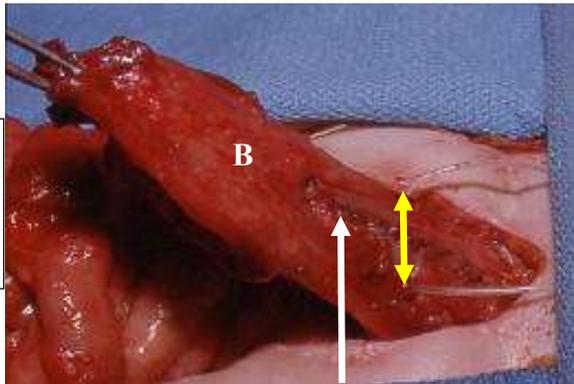
2G. Top left: Cystoscope passed from the perineum through the UGS into the orifice on the intervaginal septum. Top right, Bottom left and Bottom right: Normal colonic mucosa.

2H.



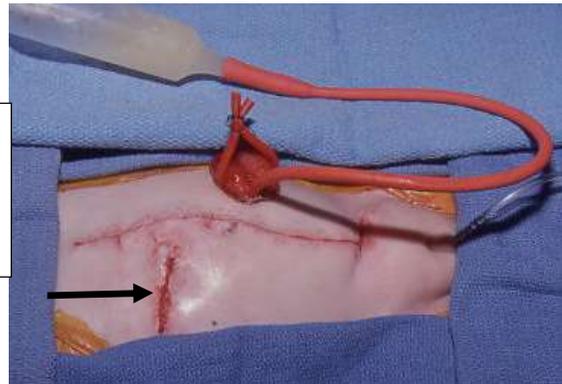
2H. Radiographic contrast instilled through the cystoscope demonstrating the two vaginae (V), the cystoscope entering the colon through the septum and contrast filling the colon (C) and distal ileum (I).

2J.



H
E
A
D

2K.



H
E
A
D

Female cloaca case #2 (cont'd.) First operation

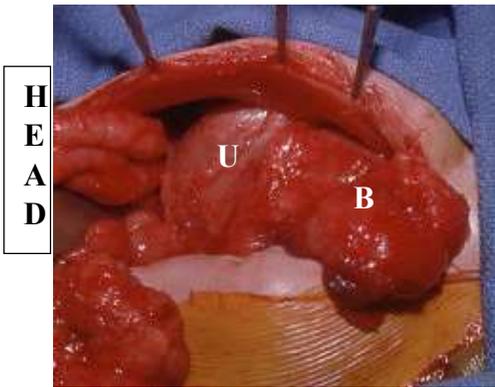
Case #2 cont'd. First operation to correct the problems created by the previous management.

Both photographs were taken from the patient's right side. Her head is to the viewer's left in the photographs.

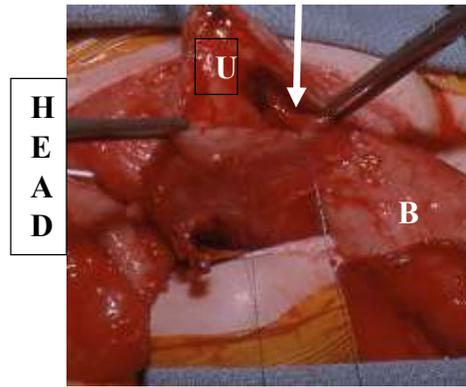
2J. An extremely large bladder (B) is visible. The vesicostomy has been closed (white arrow) after cannulating both ureters (yellow double headed arrow).

2K. After closure of the ileostomy (black arrow) a left colon diverting colostomy has been created. The red rubber catheter is introduced into the proximal limb of the colostomy and a copious amount of mucous has been aspirated from the colon and distal ileum. The recurrent hydrocolpos was due to intestinal mucus filling the vaginae.

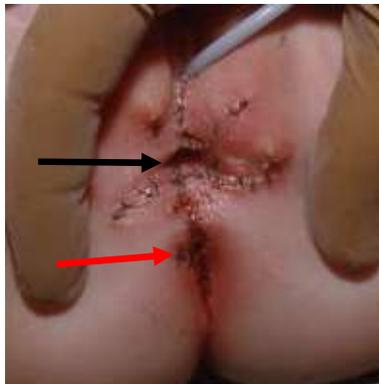
2L.



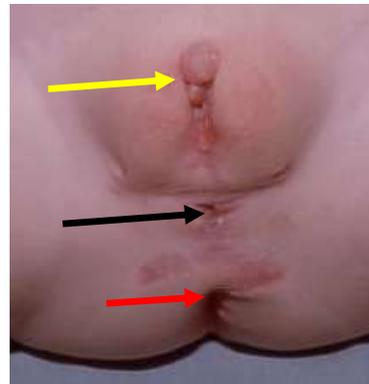
2M.



2N.



2O.



2P.



2Q.



Case #2 cont'd. Second and definitive operation.

Both photographs were taken from the patient's right side. Her head is to the viewer's left in the photographs.

2L. A thick-walled uterus and vagina (U) and an empty bladder (B) are seen.

2M. The uterus and vagina are opened (U). The white arrow points to the septum between the vaginae which is being held up with an Allis clamp. The septum was resected. The bladder (B) has been displaced out of the pelvis.

2N. Appearance of the perineum 1 week post-operatively. The patient is supine with a Foley catheter in the urethra. The black arrow points to the vagina and the red arrow to the neoanus. A Z-plasty in the

perineal skin and subcutaneous tissues was required to move the anus backwards into the center of the sphincter mechanism.

2O. Perineum of the patient 6 months later with excessive skin at the clitoris hiding the urethra (yellow arrow). The black arrow points to the vagina while the red arrow demonstrates the anus.

2P. Skin incisions used in the operation to improve the appearance of the perineum. The urethra was moved posteriorly to the point between the two arrows marked with ink in the anterior midline and the two labio-scrotal folds were displaced posteriorly along the two inked arrows on the sides. The inked dotted lines point to the previous incision scars which were not crossed to avoid damaging the blood supply of the skin flaps.

2Q. Post operative appearance of the perineum after 6 months with the patient supine.

3A.



3B.



3C.



Figure 3. Perineum after Total Urogenital Mobilization.

Perineum of another patient in the supine position.

3A. The entire urogenital sinus is freed and brought down to the perineum.

3B. The posterior wall of the urethra is being constructed (yellow arrow).

3C. The completed procedure with a Foley catheter in the urethra. The vagina is demonstrated by the black arrow.

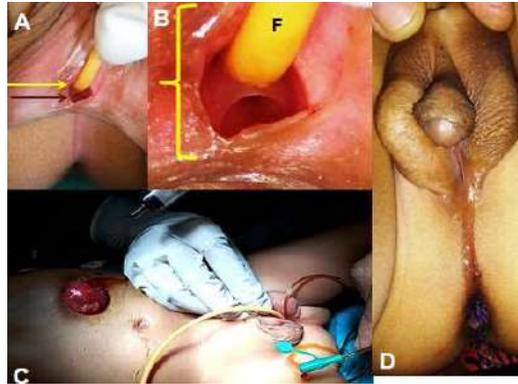


Figure 4. Male persistent cloaca.

Photographs showing single midline perineal opening at the proximal part of the scrotum with small cavity underneath it (A); two smaller openings, one anterior (with Foley's catheter *in situ* shown by yellow arrow) and other posteriorly placed (black arrow), are seen in the posterior wall of the cavity (B); infant feeding tube placed in posteriorly placed rectal opening and irrigation of the distal colonic loop done to confirm the rectal opening in the common channel (C); bilaterally descended testes, penoscrotal transposition, severe chordee, and perineal hypospadias along with neanus are seen (D).

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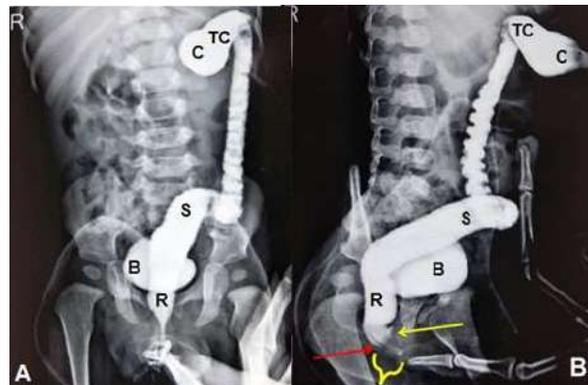


Figure 5. Male persistent cloaca (case cont'd.)

Nonfluoroscopic well-tempered pressure-augmented distal colostogram: Anteroposterior view (A) showing rectum tapering before entering into the cavity along with complete bladder filling; lateral view (B) showing rectum descending below the "T" line; rectum (red arrow) and urethra (yellow arrow) communicating with a common channel (yellow bracket). TC: Transverse colon, C: Colostomy, S: Sigmoid colon, R: Rectum, B: Bladder

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The primary purpose of antenatal evaluation is to alert the family and physicians that the baby has a major congenital anomaly that will require planned delivery at a center where the baby could be cared for appropriately. Early termination of pregnancy is not indicated.

Clinical presentation in females

Externally, there is an enlarged clitoris with an excess of preputial skin, even though they do not have disorders of sex development. Variations in the numbers and locations of perineal openings depend upon the type of anomaly. A persistent cloaca has a solitary opening close to the base of the clitoris, while the solitary opening of the posterior cloaca is where a normal anus would be located. The two openings in a cloaca variant are for the UGS, where the urethra would normally be located, and an anteriorly displaced anus. The posterior cloaca variant also has two openings, which are the anal opening in its normal position and a posteriorly displaced UGS. Patients with cloacal dysgenesis do not have an external opening [9].

A lower abdominal mass due to hydrocolpos is found in about 30% of patients [13]. Since the common channel appears to be open in them, it is assumed that a one-way valve mechanism results in the vagina or vaginae being distended with fluid.

On occasion, outflow from the bladder is obstructed and this, in turn, leads to obstructive megaureters and hydronephrosis.

Clinical presentation in males

They usually have a penoscrotal hypospadias with a bifid scrotum and possibly a penoscrotal transposition. The urinary and digestive systems open parallel to each other at the cranial end of a short and wide common channel within the pelvis, with a single orifice to the exterior. The lesion has to be differentiated from penoscrotal hypospadias and bifid scrotum in a patient with an anorectal anomaly and a rectobulbar or rectoperineal fistula. The latter lesions do not have a common channel and the fistula meets the urethra at an acute (Y-shaped) or right angle (T-shaped) [6].

Evaluation

The initial diagnosis of a persistent cloaca is relatively straightforward as the patient has a solitary orifice. At this time, it is worthwhile to also evaluate the appearance of the perineum. If it is “characterless” or flat, it indicates a high lesion with a poor or nonexistent anal sphincter mechanism. On the other hand, a well-defined anal dimple and midline raphe suggest the presence of an underlying sphincter mechanism and one should expect a good functional result after reconstruction.

The next priority is to identify any associated problems that may be incompatible with life. After a careful local and general physical examination, a series of studies are indicated. The first test should be an ultrasonographic examination of the pelvis, abdomen and spine to evaluate the pelvic organs, kidneys and spinal cord. Next, antero-posterior and lateral radiographs of the pelvis, abdomen, chest, spine and sacrum are indicated. Further

studies to determine the cardiac and renal status may be required and it is essential to rule out esophageal atresia by passing a nasogastric tube. The above studies should suffice to prepare the patient for the required immediate interventions.

After urgent conditions have been dealt with, definitive studies are obtained in a controlled environment. The anatomy must be defined precisely by using all available modalities, such as cystography and sinography (instilling radiographic contrast into every perineal orifice), under fluoroscopic control and with still radiographs. This is followed by panendoscopy (endoscopic evaluation through every orifice) with photographic documentation to permit reevaluation. Colon contrast studies are used to define the level of anorectal obstruction. A cloacogram in 3-D is particularly valuable. Definitive information is obtained by MRI of the perineal area, spine, sacrum and upper abdominal organs. All other systems must also be evaluated to determine the general status and suitability of the child for major surgery.

Management

Management can be divided into initial urgent care and elective definitive treatment.

Initial care for females. Diversion of the hindgut and drainage of the hydrocolpos are typically required urgently. This is best carried out with a completely diverting high sigmoid or descending colon colostomy with a separate mucus fistula. It is essential to leave an adequate length of distal colon *in situ* for a bowel pull-through

and, if necessary, for the construction of the vagina. Until the cloaca is definitively corrected, the hindgut must stay connected to it to function as an outflow channel for vaginal and bladder collections if they do not drain through the UGS. It is not recommended to divert the bowel at the level of the right transverse colon or proximal to it because this results in a large colonic reservoir for urine and mucus, which flows back and forth, causing urinary and vaginal infections. Hyperchloremic metabolic acidosis could develop if enough urine is absorbed from the colon.

The hydrocolpos must be drained immediately, preferably through the UGS, not only to prevent it from compressing the bladder outlet but also to keep it from getting infected. On occasion, a self-retaining catheter may have to be placed to drain the vagina or vaginae to one of the lower abdominal quadrants. If both segments of a duplicated Müllerian system are distended, a window created in the intervening septum will permit a single catheter to drain both sides. Occasionally, a vaginostomy may be necessary to decompress the vagina.

Next, the bladder may require catheterization. Rarely, catheterization is not feasible and a vesicostomy may be needed to alleviate a persistent urinary tract obstruction.

Definitive treatment for females. Treatment of these patients has to be individualized; therefore, no blanket rule is applicable to all of them. However, the following fundamental principles are worth bearing in mind:

1. Definitive treatment should be delayed, within limits, until the evaluation is complete and the patient is thriving.
2. The surgeon(s) must be familiar with all surgical options and all aspects of management.
3. When necessary, other specialists must be consulted prior to the repair and they should be available when the repair is carried out.
4. The team must be willing to modify operative plans as indicated by findings at surgery.
5. The above fact must be conveyed to the family prior to scheduling the child for surgery.
6. Management of the family's expectations is critical and they must be aware that multiple operations and procedures may be required over the patient's lifetime to correct the anomaly and deal with issues that arise in the future.
7. It is best to carry out the entire reconstruction in one operation, as it is difficult to reoperate in a pelvis that has been scarred by previous surgery.
8. It is essential to not perform a procedure that would make future operations more difficult or impossible. It is especially important not to excise any organs or structures without careful thought, as they cannot be replaced.
9. The entire body, from the nipples down to the knees, must be prepared circumferentially for surgery to allow for a change in the approach if so dictated by the findings.

10. The team must be prepared to operate through the posterior or anterior sagittal route, and the abdomen.

Surgical principles in females. In a rare condition such as persistent cloaca, it is essential to be familiar with the experience of two surgeons with the greatest experience in treating these patients and to be prepared for any eventuality. In 1959, Gough brought attention to a series of cloaca patients from the Great Ormond Street Hospital (GOSH) for children in London, UK [16]. However, Hendren was the first to carry out concerted, extensive and successful efforts to correct these problems [17-20]. Subsequently, Peña has collected a huge series of patients and he has made the most important advances in their care [11-13].

The following principles are now clear:

1. The operation should start along the posterior sagittal route.
2. If the hindgut reaches the level of the third sacral vertebra, it can be brought down to the perineum without an abdominal exploration and if the common channel is <3 cm, the entire procedure can be carried out through the posterior sagittal approach [10].
3. In 1997, Peña publicized a new surgical technique, entitled total urogenital mobilization, to deal with any UGS <3cm. in length [21]. It has numerous advantages over separating the vagina from the UGS. Total urogenital mobilization reduces operative time and complications greatly and the cosmetic result is better. The first part of the operation is unchanged, but once the cloaca is identified, the hindgut is

separated from the UGS. The suspensory ligament of the urethra is then divided and the entire UGS complex is mobilized and brought down to the perineum. The urethra is constructed at the perineum. Excess tissue of the UGS is sutured to the sides of the perineum to create the labia.

4. Patients with a common channel >3cm require the addition of an abdominal component to the procedure. Additional urologic procedures should be carried out at the same time. In these patients, special techniques, described below, may be necessary to make the vagina reach the perineum. In some patients, extended transabdominal urogenital mobilization is required [13].
5. Patients with an extra-long common channel of >5cm. may not be reparable even with the added abdominal approach for total urogenital mobilization. In this group, the posterior aspect of the pubis may be carved out to shorten the distance that the urethra and vagina traverse to reach the perineum. In patients with an ultralong channel, after carefully identifying and protecting the ureters with catheters, the bladder and urethra may have to be separated from the genitalia. The UGS forms the neourethra and is used for clean intermittent catheterization (CIC), which is required by the majority of these patients [13]. If a vesicostomy is carried out, it should be closed by 5–6 years of age and ureteric reimplantation can be carried out at the time. It is worthwhile to use anticholinergics to try to avoid bladder augmentation.

6. About half of these patients have Müllerian ductal anomalies that have to be addressed. As mentioned above, if the ducts have not fused, the septum between the two has to be resected partially or completely to permit adequate drainage of the system.

The first choice for repairing the vagina is a vaginal pull-through, which is possible in the majority of patients. If it is not possible, a variety of procedures are available, depending on the findings. In the case of a dilated vagina, a wide, inferiorly based vaginal flap can be created and rotated down on itself. The flap and native vagina are then tubularized to form a thinner vagina that extends to the perineum [11]. When the vagina ends just short of the perineal surface, vascularized, laterally based perineal skin flaps can be laid into the opening to augment the introitus of the vagina. However, one has to be careful to avoid being too aggressive in developing perineal skin flaps, as they might alter the shape of the perineal body and affect the urethral opening. If there are two large hemivaginae and hemiuteri, the septum and one hemiuterus could be excised, both hemivaginae tubularized, and the upper end of the hemivagina on the side from which the uterus was excised turned down to the perineum. Peña termed this procedure the vaginal switch [12]. If none of these options are available, the rectum, colon, or even ileum could be utilized to bridge the gap to the perineum or to create an entire vagina [11,22]. If a very dilated rectum is available, the layers of its mesentery could be separated and the bowel divided

longitudinally to develop a vascularized neovagina [12,23].

Initial care in males. It is best to initially divert the hindgut completely from the cloaca. No other procedure is urgently necessary.

Definitive treatment for males

1. Once the hindgut has been diverted and the patient is thriving, he is studied for associated anomalies.
2. The first step in definitive management is to correct the anorectal anomaly and reconstitute the cloacal channel to form the proximal urethra.
3. The final step in repair is the correction of the hypospadias, penile and scrotal anomalies.
4. If Müllerian remnants are found, they are best excised at the time of the rectal surgery.

Surgical principles in males. Management of the lesion in males is simpler, but there are a few important principles.

1. Regardless of the distance between the rectal pouch and the proposed site of the neoanus, the procedure can be carried out without an abdominal exploration [6,7].
2. The length of the cloacal channel is inconsequential since it does not have to be mobilized to reach the perineum.
3. After it is repaired, the cloacal channel must conform in circumference to the proximal and distal urethra to avoid developing a diverticulum or a stricture.

4. Genital reconstruction should be delayed until the cloaca and the anorectum are well healed and do not require further instrumentation or manipulation.
5. It is probably best to close the colostomy after all the entities are well healed.

Management of the tethered cord

A tethered cord has to be released surgically, but the operation can be carried out after 1 year of age. A renal ultrasound and voiding cystourethrogram (VCUG) may be carried out 3 months later to determine the status of the kidneys and the bladder. Urodynamic studies are only reliable after the child is a year old. To follow-up, yearly ultrasound examinations are adequate unless abnormalities are found on ultrasonography.

Results

The objectives of treatment are not only for the patient to attain socially acceptable fecal and urinary continence but also to permit girls to function sexually and to have the capacity to get pregnant and carry babies to term. In males as well, fecal and urinary control and the potential for sexual activity are the goals of management.

If female patients are treated correctly by surgeons conversant with the various modalities necessary to manage them appropriately, they can have productive lives. Peña [13] has reported excellent results in a series of 490 patients, most of whom were treated through the posterior sagittal approach, with only 38% requiring a laparotomy. Furthermore, in 63% of patients, the genital reconstruction

consisted of a vaginal pullthrough. Fifty-three percent of his patients have spontaneous daily bowel movements, while the rest stay clean with a bowel management regime consisting of daily enemas.

Just over half of Peña's patients were naturally continent of urine after correction, while another quarter remained dry with intermittent catheterization *via* the native urethra. Twenty percent of patients required catheterizable urinary diversion. Almost 80% of patients with a common channel >3cm. required intermittent catheterization, while less than 30% of those with a common channel <3cm. did so. However, it is essential to remember that urinary control or continence does not imply that the urinary tract is normal. Furthermore, even in the absence of structural abnormalities, there is a risk of progressive renal injury in these patients. Therefore, their urinary tracts must be evaluated for life.

Complications are to be expected in any major, intricate reconstructive procedure and additional procedures and operations are inevitable over time. The more common complications pertaining to the urinary and genital structures are persistent UGS, vaginal strictures or atresia, urethrovaginal fistulae and urethral atresia. Complications affecting the hindgut are rectal prolapse, stricture, retraction, dehiscence, and atresia. In addition, on occasion, the rectum may be mislocated [13].

Information on urinary and fecal control and sexual activities in male patients is scant and there is no overview of the condition to date [6,7].

Future considerations

Future studies must standardize techniques and results. Some issues worth considering are discussed below.

First, based upon his extensive experience, Peña has created a valuable algorithm for the management of cloacae based upon the length of the common channel [13]. It would be easier to plan reconstructions, compare results and decide on management strategies if this algorithm were kept in mind.

Common channel <1 cm. A posterior sagittal anorecto-vaginoplasty in which the rectum is separated from the vagina. The UGS is left untouched. The lateral and posterior walls of the vagina are mobilized to suture to the future labia. The minimal female hypospadias is inconsequential since the urethra is clearly visible. These patients are continent of urine and stool.

Common channel 1-3 cm. The majority of patients fall into this category. They are operated through the posterior sagittal approach and do not require additional abdominal exposure. Through this approach, the rectum is separated from the vaginal or Müllerian structures. After total urogenital mobilization, approximately 2 cm. of length is obtained to bring the UGS down to the perineum, where it is split in the sagittal plane and the urethra and vagina are sutured to the neolabia.

Common channel 3-5 cm. In these patients, also the operation starts from the posterior sagittal approach and after the internal anatomy is defined, total urogenital

mobilization is carried out. If enough length is not obtained, the patient is placed supine and the abdomen is entered through an infraumbilical incision. The bladder dome is pulled upward and the lateral attachments of the bladder are divided. The UGS is brought up in the space between the bladder and the pubis and all avascular attachments of the bladder and the urethra are divided. If this maneuver gives the necessary length the UGS is brought back down and the previously described repair is carried out.

If the UGS can still not be anastomosed properly to the perineum, the posterior half of the pubic cartilage is resected to create a more direct route for the UGS to reach the perineum.

In cases where, in spite of all the above maneuvers, the UGS cannot be anastomosed at the perineum, the urinary tract must be separated from the vagina/vaginae. To do, this the bladder is opened and both ureters are catheterized as they pass out in the wall between the vagina and the bladder. Reconstruction is carried out after the complete separation of the two structures. The choices are a vaginal switch, a vaginal flap, or the use of a bowel segment to bridge the gap or for the entire vagina.

Common channel >5 cm. These patients are best approached from the abdomen since the rectum and the vagina/vaginae open into the trigone of the bladder, where the ureters also open. Upon separation, the bladder neck may be damaged or even destroyed. If the bladder neck is competent, the UGS can be left as the channel for CIC. In some situations, the bladder neck can be closed and a vesicostomy fashioned. Heroic efforts

to reconstruct the urethra or to pull through a short urethra are not justified since most, if not all, of these patients will have neurogenic bladders and the extensive efforts to mobilize structures can negatively impact the vaginal or rectal pullthrough.

Occasionally, an extremely high rectum cannot be reached from the posterior sagittal approach. In this situation, the rectum has to be dissected from the abdomen.

Secondly, since the length of the common channel is vital to the ultimate results, it may be worthwhile to measure it with relation to a fixed bony point rather than leave it to endoscopic measurements that are subject to operator error and can vary depending upon the age and size of the child when the measurements are made. In all other forms of anorectal malformations, we use fixed bony points as landmarks. It has been suggested that the pubic symphysis would make the ideal bony landmark for cloacal lesions. The common channel is short if the confluence is below the pubic symphysis, long when the confluence is at or above its upper border and intermediate when the components fuse behind the symphysis [24].

Thirdly, there are no true long-term follow-up studies yet. While the immediate and mid-term results of surgical repair seem to be acceptable in females, we must know what happens to these patients throughout their lives. It is also essential that detailed follow-up information be obtained for males.

Fourth, another modality that may be valuable for the abdominal part of the operation could be laparoscopy instead of

laparotomy. In the past, laparoscopy has only been used for repair of the rectum in patients with a high rectal pouch and a low UGS. Now, it is also being used to mobilize the vagina and there is some resurgence of interest in laparoscopic urogenital separation over total urogenital mobilization. With this modality too, studies are few and there are no long-term results [25].

Finally, one wonders if there is any place for the use of tissue expanders to obtain adequate perineal skin and tissue for the repair. If the vagina cannot be brought to the perineum, we have used broad-based, vascularized perineal skin flaps laid into the introitus to bridge the gap. The use of tissue expanders on either side of the perineal orifice would increase the amount of skin available and the flaps could be placed further into the introitus to complete the vaginal reconstruction. The skin may also be helpful in the repair of the urethra. Liu et al. used tissue expansion in persistent, cloaca but they placed the expanders within the UGS [26]. While they succeeded in widening the UGS to create a separate urethra and vagina, their technique is problematic because all their patients required a vesicostomy and hospitalization for the 3–4 weeks it took for sufficient tissue expansion. Thus, their technique is cumbersome, results in prolonged hospitalization, increases hospital costs tremendously, is uncomfortable for patients and is inconvenient for families. Tissue expanders placed on the perineum on either side may obviate these problems.

Conclusion

Persistent cloaca is a complicated problem that must be approached after careful evaluation at a center with experience in management. With proper care, excellent results are achievable. Therefore, it seems reasonable that, if not all patients with a persistent cloaca, at least long-channel patients should be transferred as soon as possible to a center with the expertise to treat them. Improper intervention prior to referral makes subsequent correction difficult and results in poor outcomes.

Statements and Declarations

Competing interests

None of the authors have any competing interests.

Conflict of interest

None of the authors have any conflict of interest.

Author contributions

All four authors reviewed the literature and contributed to the manuscript.

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ORIGINAL ARTICLE

A Study to Evaluate the Complications of Laparoscopic Renal and Adrenal Surgeries Using Modified Clavien-Dindo Classification

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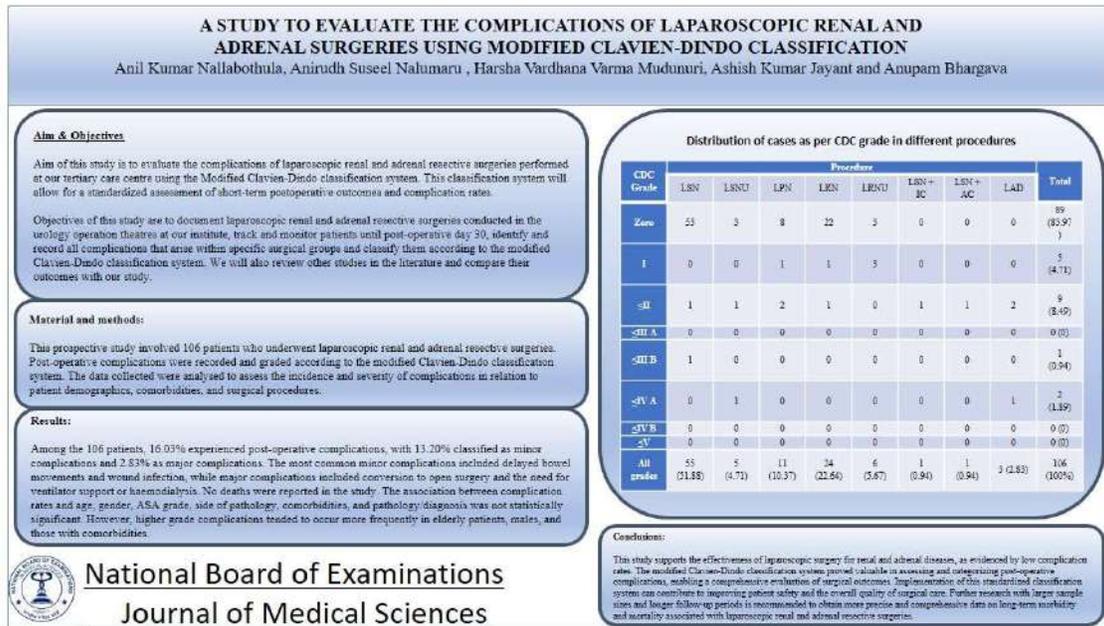
Abstract

Background: Laparoscopic surgery has become a widely accepted approach for the treatment of renal and adrenal diseases. However, there is a need to assess the post-operative complications associated with these procedures using standardized classification systems. The modified Clavien-Dindo classification system offers a comprehensive framework for evaluating surgical outcomes and determining the severity of complications. **Methods:** This prospective study involved 106 patients who underwent laparoscopic renal and adrenal resective surgeries. Post-operative complications were recorded and graded according to the modified Clavien-Dindo classification system. The data collected were analysed to assess the incidence and severity of complications in relation to patient demographics, comorbidities, and surgical procedures. **Results:** Among the 106 patients, 16.03% experienced post-operative complications, with 13.20% classified as minor complications and 2.83% as major complications. The most common minor complications included delayed bowel movements and wound infection, while major complications included conversion to open surgery and the need for ventilator support or haemodialysis. No deaths were reported in the study. The association between complication rates and age, gender, ASA grade, side of pathology, comorbidities, and pathology/diagnosis was not statistically significant. However, higher grade complications tended to occur more frequently in elderly patients, males, and those with comorbidities. **Conclusions:** This study supports the effectiveness of laparoscopic surgery for renal and adrenal diseases, as evidenced by low complication rates. The modified Clavien-Dindo classification system proved valuable in assessing and categorizing post-operative complications, enabling a comprehensive evaluation of surgical outcomes. Implementation of this standardized classification system can contribute to improving patient safety and the overall quality of surgical care. Further research with larger sample sizes and longer follow-up periods is recommended to obtain more precise and comprehensive data on long-term morbidity and mortality associated with laparoscopic renal and adrenal resective surgeries.

Keywords: Clavien-Dindo classification, Risk factor, complication, Postoperative complications, Adrenal, Kidney, Laparoscopy, Nephrectomy, Adrenalectomy, Transperitoneal laparoscopic surgery

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Graphical Abstract



Background

Laparoscopic technology has revolutionized urological surgeries by providing several advantages over traditional open surgery, including reduced postoperative pain, shorter hospital stays, and faster recovery [1-3]. However, despite these benefits, laparoscopic procedures come with unique challenges and potential complications. Introduced over three decades ago, laparoscopy initially gained significant enthusiasm in urology, with the technique being widely adopted for various procedures such as pelvic lymphadenectomy, nephrectomy, and varicocelectomy [4]. Its popularity grew rapidly in both academic and private centres, leading to its widespread use in routine urology practice.

However, this initial enthusiasm was eventually followed by disappointment due to several factors [5]. First, laparoscopy proved to be more difficult than many had anticipated. During the early years of urological laparoscopy, researchers were

still trying to identify appropriate indications, and many were not fully aware of the limitations of the technique. Additionally, most reports at the time reflected the challenges encountered during the learning phase, rather than focusing on the outcomes and complications of advanced laparoscopy. Some of the indications initially identified did not meet expectations.

Of the initial indications of urologic laparoscopy, only nephrectomy has stood the test of time. Although pelvic lymphadenectomy was associated with less morbidity than its open surgical counterpart, it has become less common as a result of intense screening for prostate cancer and the fact that a retro-pubic prostatectomy typically includes lymphadenectomy anyway. Some groups have even abandoned laparoscopy for this indication altogether.

Laparoscopic varicocelectomy has been considered an ideal operation for those inexperienced with laparoscopy. However,

surgical efficiency cannot compete with antegrade or retrograde sclerotherapy, and the recurrence rate of microsurgery is lower. The long-term efficacy of varicocelectomy is difficult to assess, so cost effectiveness remains the primary argument in favour of this procedure [4].

In contrast to general surgery, where laparoscopy has become the norm for procedures like cholecystectomy, urology has been slower to identify good and frequent indications for the technique. However, as more experience was gained, the accumulating results demonstrated that, for certain indications like Adrenalectomy [6], laparoscopy proved superior to open surgery in every aspect. Several other good indications soon followed including laparoscopy for the impalpable testis, nephrectomy for benign and malignant disease, adrenalectomy, pyeloplasty, and radical prostatectomy [7,8].

As the number and complexity of laparoscopic procedures increase, so do the potential complications, which can result in longer hospital stays and increased costs [9-11]. The recognition and classification of complications associated with urological laparoscopic procedures are becoming increasingly important. Although various studies have published the rates of complications encountered in urological laparoscopic procedures [9,10], there is currently no widely accepted classification system for reporting these complications. To enable the comparison of complication rates across different surgical centres and techniques, it is necessary to use a standardized complication classification system [12].

Centuries ago, Celsius introduced the assessment of postoperative complications, which he referred to as "professional mishap," and was followed

indirectly by Hammurabi's code. The code specified that if a surgeon used a bronze lancet on a high-status patient leading to their death, the surgeon's hands would be amputated as punishment [9].

In 1992, Pierre-Alain Clavien and colleagues introduced a new system of classification that transformed the assessment of postoperative complications for general surgical practice, it was updated by Dindo in 2004 and was subsequently validated for urology [9,11,13]. It differentiates complications, sequelae, and failures and provides a standardized approach for grading and reporting postoperative complications in urology.

Now, the Clavien-Dindo classification system provides a standardized metric for evaluating surgical complications, offering an objective and convenient way to quantify and grade complications. It has been widely adopted in various surgical specialties [14].

While previous studies have compared complications in laparoscopic renal and adrenal surgeries, there is a limited number of prospective studies that have utilized the Modified Clavien-Dindo classification system to assess these procedures [15].

A study, by Çetin et al. (2018), analysed complications of laparoscopic urological surgeries using the Clavien classification system¹⁶. The overall postoperative complication rate was 18.9%, with laparoscopic radical nephrectomy and laparoscopic simple nephrectomy having the lowest complication rates. Laparoscopic partial nephrectomy had the highest complication rate.

A prospective cohort study, by Agrawal et al. (2017), evaluated complications in patients undergoing laparoscopic nephrectomy using the

Clavien-Dindo scale [17]. The study found a complication rate of 46%, with 34 low-grade and 11 high-grade complications. There were no mortalities, and the complication rates were similar for inflammatory and non-inflammatory causes.

A study, by Balci (2018), examined complications of laparoscopic procedures, including radical nephrectomy, pyeloplasty, and adrenalectomy, using the Clavien-Dindo classification [18]. Laparoscopic surgery was found to be safe and effective, with low complication rates in selected patients with renal and adrenal diseases.

By employing the Modified Clavien-Dindo classification system in a prospective manner, this study will provide valuable insights into the quality and safety of laparoscopic urological procedures. The findings will contribute to the existing literature on complications in laparoscopic surgery and help improve patient outcomes.

Therefore, the aim of this study is to evaluate the complications of laparoscopic renal and adrenal resective surgeries performed at our tertiary care centre using the Modified Clavien-Dindo classification system. This classification system will allow for a standardized assessment of short-term postoperative outcomes and complication rates.

Objectives of this study are to document laparoscopic renal and adrenal resective surgeries conducted in the urology operation theatres at our institute, track and monitor patients until post-operative day 30, identify and record all complications that arise within specific surgical groups and classify them according to the modified Clavien-Dindo classification system. We will also review other studies in the

literature and compare their outcomes with our study.

Material and Methods

This prospective observational study was conducted in the department of Urology at Sri Venkateswara Institute of Medical Sciences (SVIMS), Tirupati, Andhra Pradesh. The study included all consecutive patients who underwent laparoscopic renal and adrenal resective surgeries from August 2021 to January 2023. Ethical approvals were obtained, and written informed consent was obtained from all participants. Data collection involved recording patients' demographics, clinical information, and postoperative complications based on the modified Clavien-Dindo classification scale. Follow-up data were collected through outpatient visits or telephonic interviews until discharge and up to 30 days postoperatively.

Inclusion criteria encompassed both male and female patients who underwent laparoscopic trans-peritoneal renal and adrenal resective procedures, including simple nephrectomy, partial nephrectomy, radical nephrectomy, radical nephro-ureterectomy, and adrenalectomy, in the Urology OT at SVIMS. For cases of malignancy, organ-confined malignancy evaluated preoperatively with imaging was included. Elective procedures were performed after thorough preoperative evaluation, optimization of comorbidities, and stabilization in functional tumour cases. Exclusion criteria consisted of patients refusing to participate in the study and those undergoing palliative or cytoreductive surgeries.

Patient evaluation involved a comprehensive assessment of eligible patients. A detailed patient history was

obtained, including renal and adrenal complaints, comorbidities, substance abuse, and prior abdominal surgical history. Physical examinations, including BMI, vital data, general physical examination, and abdominal examination, were performed.

Laboratory investigations included routine surgical profile tests such as complete blood count, serum creatinine, serum electrolytes, coagulation profile, random blood sugar, serological tests (HBV, HCV, and HIV), and urine culture. Trans-abdominal ultrasound with a 5 Hz probe was conducted to evaluate the genitourinary system. Relevant imaging and nuclear scans, such as contrast CT KUB or MRI for diagnosis confirmation and tumour staging, were performed. DTPA renogram was used to assess renal function in cases of suspected PUJO or planned nephrectomy to document kidney function. For suspected functional adrenal tumours, blood and urine meta/nor-metanephrine assessments were conducted, and consultations with an endocrinologist were sought for pre-operative stabilization. Pre-anaesthetic evaluation, ASA grading, and additional investigations as recommended by the anaesthetist were carried out. Routine ECG was performed, and patients with abnormal ECG or prior cardiac issues underwent 2D ECHO for cardiac evaluation.

Preoperative preparation included bowel cleansing with proctoclysis enema administered twice the night before and early morning on the day of surgery. For deep vein thrombosis prophylaxis, knee varicose stockings were provided, and low molecular weight heparin was subcutaneously administered 12 hours before the operation and continued every 24 hours postoperatively until the patient was fully mobilized. Antibiotic prophylaxis consisted of using a third-generation cephalosporin.

All the surgical procedures were performed by a single chief surgeon with assistants. Trans-peritoneal simple/radical nephrectomy/radical nephro-ureterectomy involved 3-4 trocar accesses with the patient in the lateral decubitus position (Figure 1). Hasson's technique was used to generate pneumoperitoneum. The colon was mobilized medially, and the ureter was dissected cranially to define the hilar structures of the kidney. Renal artery and vein were released, doubly clipped, and divided. Posterior and superior dissection was performed, and the adrenal gland was preserved or removed based on intra-operative assessment. The ureter was divided, and the kidney was removed intact through a separate incision.



Figure 1. Port placement for procedures - **A**: Right side, **B**: Left side

For laparoscopic partial nephrectomy, after releasing the renal artery and vein, Gerota's fascia was opened, and the renal mass was identified. The boundary between the mass and normal renal parenchyma was determined, and the renal artery was clamped to achieve warm ischemia. The mass was removed using cold scissors, and the collecting system and bleeding areas were sutured. Surgicell cushions were placed, and the renal parenchyma was closed. The specimen was removed through a separate incision.

Laparoscopic adrenalectomy involved mobilizing the colon medially and dissecting between Gerota's fascia and the mesocolon. The vasculature was identified and confirmed before isolating and clipping the adrenal vein. Dissection was carried out, and care was taken to avoid injury to surrounding structures. The adrenal gland was freed within the abdomen and extracted intact. Trocar sites were closed, and a nasogastric tube and urethral catheter were inserted in all patients.

The modified Clavien-Dindo classification is used to categorize complications that occur after surgery [14]. The grades are as follows:

Grade I: Any deviation from the normal postoperative course that does not require pharmacological or invasive interventions.

Examples include prolonged drainage, delayed bowel movements, hernia treated conservatively, and urinary retention requiring catheterization.

Grade II: Complications requiring pharmacological treatment with drugs other than those allowed for Grade I. Blood transfusions and total parenteral nutrition are also included. Examples include high fever requiring antibiotic therapy, wound site infection, blood loss requiring transfusion, and respiratory infections.

Grade IIIa: Complications requiring surgical, endoscopic, or radiologic interventions under local anaesthesia. Examples include abscess requiring drainage and angio-embolization of a bleeding vessel.

Grade IIIb: Complications requiring surgical, endoscopic, or radiologic interventions under general anaesthesia. Examples include conversion to open surgery for reasons other than failure to progress and injury to major blood vessels.

Grade IVa: Life-threatening complications requiring ICU management and involving single organ dysfunction. Examples include splenectomy, respiratory failure requiring ventilator support, and renal failure needing dialysis.

Grade IVb: Life-threatening complications requiring ICU management and involving multi-organ dysfunction.

Grade V: Death of the patient.

If a patient experiences multiple complications, all are recorded, and the most severe complication is graded.

The data was recorded prospectively in an electronic database and analysed using Excel and IBM SPSS software. The sample size was calculated to be 40 based on a confidence level of 95% and a margin of error of 5%. Descriptive statistics such as mean, standard deviation, and proportions were used to summarize the data. Tests such as the Chi-square test and t-test were performed for comparisons between variables, with a significance level of $p < 0.05$ considered statistically significant.

Observations and Results

A total of 106 patients underwent laparoscopic renal and adrenal resective surgeries at Sri Venkateswara Institute of Medical Sciences (SVIMS) between August 2021 and January 2023 (Table 1). Among them, 103 patients had renal and ureteric pathologies, while 3 patients had adrenal pathologies. The age ranged from 6 to 81 years with a mean age of 48.85 years. There were 47 men and 59 women in the study. Most patients (64) were classified as ASA grade I, followed by grade II (24) and grade III (18). The distribution of pathologies showed 62 left-sided and 44 right-sided cases. A total of 42 patients had comorbidities, with the most common

being diabetes mellitus (21) and hypertension (17). The diagnoses included various benign conditions (66 cases) and malignancies (40 cases) (Table 2). The various laparoscopic surgical procedures performed included nephrectomy, partial nephrectomy, nephro-ureterectomy, adrenalectomy and other combined surgeries (Table 3).

In the study, out of 106 patients, complications were observed in 16.03% of patients, with 13.20% experiencing minor complications and 2.83% experiencing major complications. The most common minor complications were delayed bowel movements and wound infection, while major complications included conversion to open surgery and the need for ventilator support or haemodialysis. There were no deaths in the study.

The analysis of factors associated with complications showed that the rates did not significantly differ based on age ($p=0.446$), gender ($p=0.189$), ASA grade ($p=0.294$), side of pathology ($p=0.113$), comorbidities ($p=0.220$), or pathology/diagnosis ($p=0.749$). However, higher grade complications tended to occur more frequently in elderly patients, males, and those with comorbidities.

Though there were trends observed, the p-values for these associations were not statistically significant. Probably the observed differences may have occurred by chance rather than being truly indicative of an association.

Table 1: Patient data

S. No.	Age	Sex	Comorbidities	ASA Grade	Diagnosis	Side	Surgery	Complications	CDC Grade
1	69	M	DM, CKD, HTN	III	STONE	R	LSN	Wound infection, Delayed bowel movements	II
2	35	M		I	GUTB	L	LSN + AC	Wound infection, blood transfusion, atelectasis	II
3	67	M	CVA	III	RCC	R	LRN		
4	73	F		I	RCC	R	LRN		
5	25	F		I	RCC	L	LRN		
6	64	M	HTN	II	TCC	L	LRNU	Delayed bowel movements	I
7	27	F		I	PUJO	L	LSN		
8	45	F	CKD, HTN	III	STONE	R	LSN		
9	52	M	DM, CVA	III	PUJO	R	LSN		
10	43	F	PTB	II	GUTB	R	LSN		
11	70	F	HTN, BA	II	RCC	L	LRN		
12	45	F		I	RCC	R	LRN		
13	50	F		I	STONE	R	LSNU	Wound infection	II
14	60	M	CAD	III	TCC	R	LRNU		
15	43	F		I	RCC	R	LRN		
16	36	M		I	RCC	L	LRN		
17	34	F		I	PUJO	L	LSN		
18	50	M		I	DONOR	L	LSN		
19	68	M	CKD, CVA	III	RCC	L	LRN	Delayed bowel movements	I
20	64	M	CKD, HTN	III	STONE	L	LSNU	Dialysis requirement	IV A
21	35	M	HTN, CVA	III	PUJO	L	LSN		
22	54	F	HYPOTHYROID	II	RCC	L	LRN		
23	38	F		I	PUJO	R	LSN		
24	51	M	DM, COPD	III	TCC	R	LRNU	Abundant drainage through drain	I
25	58	M		I	STONE	L	LSN		
26	43	M		I	RCC	R	LRN		
27	51	F		I	PUJO	L	LSN		
28	47	M		I	RCC	L	LRN		
29	55	M		I	STONE	L	LSN		
30	53	F	DM, HTN	II	STONE	L	LSN		
31	51	M		I	STONE	L	LSN		
32	64	M		I	RCC	L	LRN		
33	60	M		I	RCC	R	LRN		
34	67	M	HTN	II	RCC	R	LRN		
35	62	M	DM	II	PUJO	L	LSN		
36	55	F		I	STONE	R	LSN		
37	58	F		I	RCC	R	LRN		
38	60	F		I	STONE	L	LSN		
39	21	F		I	AML	R	LPN	Delayed bowel movements	I
40	53	M	DM	II	GUTB	L	LSN		
41	71	F	DM, CVA	III	RCC	L	LRN		
42	32	F	HYPOTHYROID	II	STONE	L	LSN		
43	48	F		I	RCC	R	LPN		
44	25	F		I	RCC	R	LRN		
45	58	M	COPD	III	STONE	L	LSN		
46	46	F		I	STONE	L	LSN		
47	53	F	PTB	II	GUTB	R	LSN		
48	65	M		I	STONE	R	LSN		
49	43	F		I	DONOR	L	LSN		
50	66	F		I	RCC	L	LRN		
51	59	F		I	STONE	L	LSN		
52	61	M	DM, CVA	III	TCC	L	LRNU		
53	42	M	HTN	II	PUJO	L	LSN		
54	44	F		I	RCC	L	LPN		
55	43	F		I	GUTB	R	LSN + IC	Wound infection, blood transfusion, Abundant drainage through drain	II
56	50	F		I	RCC	L	LRN	Blood transfusion	II
57	61	F		I	STONE	R	LSN		
58	48	M	DM	II	RCC	L	LPN		
59	56	M	DM	II	AML	R	LSN		
60	51	M		I	STONE	L	LSN		
61	27	F		I	XGPN	R	LSN	Delayed bowel movements, Conversion to open (for reason other than failure to progress)	III B
62	18	M		I	PUJO	L	LSN		
63	34	F	PTB	II	GUTB	L	LSN		
64	56	F	DM	II	RCC	L	LRN		
65	33	M		I	RCC	R	LRN		

66	38	F		I	STONE	L	LSN		
67	46	M	DM	II	GUTB	L	LSN		
68	56	F	DM	II	STONE	R	LSN		
69	61	F		I	STONE	L	LSN		
70	76	M	HTN	II	Adrenal Mass	L	LAD	Conversion to open (for reason other than failure to progress), Ventilatory support	IV A
71	31	F		I	PUJO	R	LSN		
72	26	F		I	RCC	R	LPN		
73	66	F	CKD, HTN	III	STONE	L	LSN		
74	48	M		I	RCC	L	LRN		
75	25	F		I	GUTB	R	LSN		
76	37	F	DM, BA	II	PUJO	R	LSN		
77	29	F		I	PUJO	L	LSN		
78	46	M		I	RCC	R	LPN	Atelectasis	II
79	61	F		I	RCC	L	LPN		
80	53	F	CAD	III	STONE	R	LSN		
81	33	M		I	PUJO	L	LSN		
82	41	F		I	STONE	L	LSN		
83	56	F	DM, HTN, COPD	III	VUJO Ca Cx	L	LSN		
84	54	M	DM, HTN	II	Adrenal Mass	R	LAD	Intercostal pain	II
85	12	M	HTN	II	Renal Artery Stenosis	L	LSN		
86	51	M		I	STONE	L	LSN		
87	40	F		I	STONE	L	LSN		
88	46	M		I	STONE	R	LSN		
89	7	F		I	PUJO	L	LSN		
90	30	F		I	Adrenal Mass	R	LAD	Atelectasis	II
91	81	M	DM, HTN, CVA	III	TCC	R	LRNU	Delayed bowel movements	I
92	53	M		I	STONE	L	LSN		
93	51	F		I	STONE	L	LSN		
94	64	F	DM, CAD	III	RCC	R	LPN		
95	45	F		I	STONE	L	LSN		
96	42	F	DM, HTN	II	STONE	L	LSN		
97	6	M		I	VUR + Ureterostomy	L	LSN		
98	64	M		I	RCC	R	LPN		
99	56	F	DM	II	STONE	R	LSN		
100	70	F	DM, HTN	II	RCC	L	LPN	Wound infection	II
101	67	F		I	RCC	L	LRN		
102	76	F		I	RCC	R	LRN		
103	61	M		I	STONE	R	LSNU		
104	62	F	CAD	III	TCC	L	LRNU		
105	46	M		I	PUJO	R	LSN		
106	19	M		I	Duplex UP VUJO	L	LPN		

Table 2. Diagnosis (Pathology) wise distribution of patients

S. No	Diagnosis	Number of patients
1.	Pheochromocytoma	3
2.	AML	2
3.	Donor	2
4.	Duplex moiety with UP NFK	1
5.	GUTB	8
6.	PUJO	15
7.	RCC	33
8.	Renal artery stenosis with NFK	1
9.	Stone disease with PFK/NFK	32
10.	TCC	6
11.	Ca Cervix with VUJO with NFK	1
12.	VUR (S/P ureterostomy) with NFK	1
13.	XGPN	1
	Total	106

Table 3. Procedure wise distribution of patients

S. No	Procedure	Number of patients
1.	LSN	55
2.	LRN	24
3.	LPN	11
4.	LRNU	6
5.	LSNU	5
6.	LSN + IC	1
7.	LSN + AC	1
8.	LAD	3
	Total	106

Discussion

In this prospective study, 106 patients with renal and adrenal pathologies underwent laparoscopic resective procedures. The post-operative complications were classified according to the modified Clavien-Dindo classification system, which provided an objective and simple grading system. The overall rate of complications in this study was 16.03%, with 13.20% experiencing minor complications and 2.83% experiencing major complications.

The most common complications observed were delayed bowel movements, abundant drainage, wound infection, basal atelectasis of the lung, intercostal pain, blood product transfusion, conversion to open surgery, haemodialysis requirement, and the need for reintubation and mechanical ventilator support. No deaths occurred during the study.

When analysing the factors associated with complications, it was observed that elderly patients, males, and those with comorbidities tended to have a higher occurrence of higher grade complications. However, these associations were not statistically significant.

The correlation between age and complications showed an increasing trend with age, but it was not significant.

Similarly, there was no statistically significant correlation between ASA grades, side of pathology, or comorbidities and the occurrence of complications.

Regarding the type of pathology, higher grade complications were seen more frequently in patients with benign pathologies, while patients with malignancies experienced mostly low grade complications. However, no statistically significant difference was found between the type of pathology and complication occurrence (Table 4).

In terms of the procedures performed, simple resective surgeries had a higher occurrence of complications, likely due to the complexity of the procedure in the presence of pre-existing inflammatory or infective pathology. Donor nephroureterectomy had no complications, possibly due to the healthy status of the kidney and careful patient selection (Table 5).

The majority of complications fell into grades I and II, requiring only pharmacological treatment, while a smaller portion required ICU management or interventional treatment. The Clavien-Dindo classification system was found to be a useful tool in assessing the severity of complications.

Comparing the study with previous research, it was found that the complication rates in this study were comparable or lower than other studies (Table 6). This may be

attributed to the expertise of the main surgeon, a well-trained team, and a streamlined peri-operative management protocol.

Table 4. Distribution of cases as per CDC grade in groups of different pathologies

CDC Grade	Pathology		Total
	Benign	Malignant	
Zero	56	33	89 (83.97)
I	1	4	5 (4.71)
≤II	6	3	9 (8.49)
≤III A	0	0	0 (0)
≤III B	1	0	1 (0.94)
≤IV A	2	0	2 (1.89)
≤IV B	0	0	0 (0)
≤V	0	0	0 (0)
All grades	66 (62.26)	40 (37.74)	106 (100%)

Table 5. Distribution of cases as per CDC grade in different procedures

CDC Grade	Procedure								Total
	LSN	LSNU	LPN	LRN	LRNU	LSN + IC	LSN + AC	LAD	
Zero	53	3	8	22	3	0	0	0	89 (83.97)
I	0	0	1	1	3	0	0	0	5 (4.71)
≤II	1	1	2	1	0	1	1	2	9 (8.49)
≤III A	0	0	0	0	0	0	0	0	0 (0)
≤III B	1	0	0	0	0	0	0	0	1 (0.94)
≤IV A	0	1	0	0	0	0	0	1	2 (1.89)
≤IV B	0	0	0	0	0	0	0	0	0 (0)
≤V	0	0	0	0	0	0	0	0	0 (0)
All grades	55 (51.88)	5 (4.71)	11 (10.37)	24 (22.64)	6 (5.67)	1 (0.94)	1 (0.94)	3 (2.83)	106 (100%)

Table 6. Comparison of present study with previous studies

Study	Type	Year	Number of patients	Complications	MCD Grade	
					Low	High
Hua Xu et. al ²⁶	Retrospective	2014	88	17(19.3%)	18.2%	1.1%
Balcı M. et. al ¹⁸	Prospective	2016	208	13(6.3%)	5.8%	0.5%
Agrawal T. et.al ¹⁷	Prospective	2017	97	45(46%)	35%	11%
Ali S.G. et.al ²⁸	Retrospective	2017	330	65(19.7%)	12.7%	7%
Çetin D et.al ¹⁶	Retrospective	2018	396	75(18.9%)	17.7%	2.2%
Present Study	Prospective	2023	106	11 (16.03%)	13.2%	2.8%

In conclusion, this study provides valuable insights into the post-operative complications associated with laparoscopic renal and adrenal resective surgeries. The modified Clavien-Dindo classification system proved useful in grading the severity of complications. Although certain trends were observed in relation to age, gender, comorbidities, and pathology, they did not reach statistical significance. Further research with larger sample sizes is necessary to validate these findings and identify potential risk factors for complications.

Limitations of this study

This study has several limitations that should be taken into consideration. Firstly, the sample size was relatively small, and the study was conducted over a limited period of time, which may have resulted in limited precision and generalizability of the findings. The short follow-up period also limits the ability to assess long-term morbidity and mortality associated with the procedures. Additionally, as the study was conducted in a single tertiary-level hospital in a specific country, the results may not be representative of other populations or healthcare settings. Despite these limitations, the study provides valuable insights into the post-operative

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complications of elective laparoscopic renal and adrenal resective surgeries.

Conclusion

In conclusion, this study demonstrates the effectiveness of laparoscopic surgery in treating renal and adrenal diseases, with low complication rates observed. The modified Clavien-Dindo classification system proved to be a valuable tool for assessing and categorizing post-operative complications, enabling a clear distinction between different grades of severity. The findings highlight the significance of this standardized classification system in evaluating surgical outcomes and improving patient safety. It is recommended that the modified Clavien-Dindo classification system be widely implemented and incorporated into routine reporting of surgical results. By doing so, the quality of surgical care can be enhanced, leading to better patient outcomes.

Conflicts of interest

The authors declares that they do not have conflict of interest.

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ORIGINAL ARTICLE

Decompressive Craniectomy Outcomes in Cerebral Venous Sinus Thrombosis: A Comprehensive Analysis from a Tertiary Neurosurgical Center

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Abstract

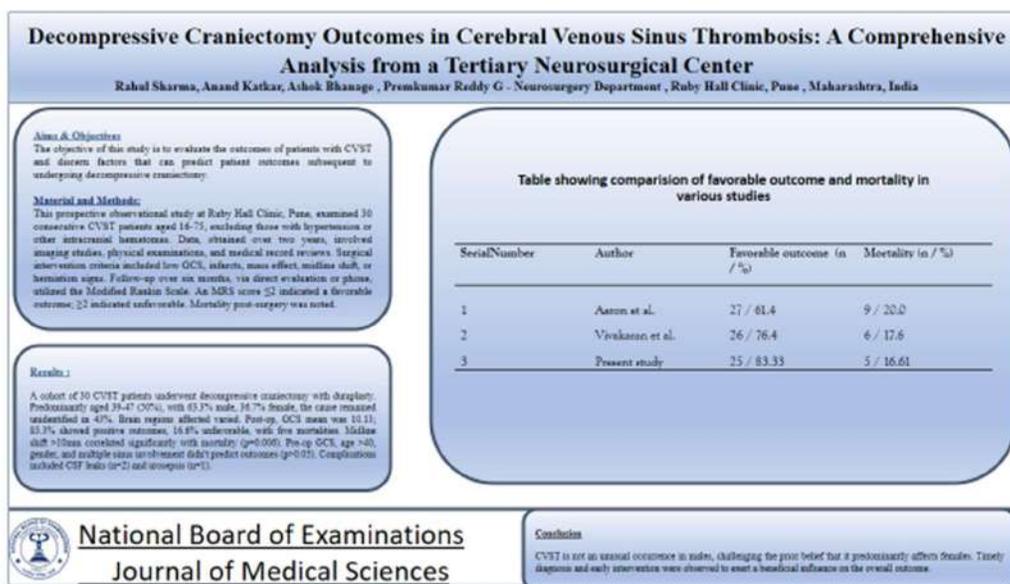
Background: Cerebral venous sinus thrombosis (CVST) is a rare cerebrovascular condition characterized by the formation of blood clots in the cerebral venous sinuses, which are responsible for draining blood and cerebrospinal fluid from the brain. CVST represents a significant cause of stroke in young individuals, with a mortality rate ranging from 6% to 15%. **Methods:** Common clinical presentations include symptoms like headaches, seizures, altered mental state, and focal neurological deficits. In India, a noteworthy occurrence of CVST is observed among postpartum women, while alcoholism poses a significant risk factor for males. **Results:** This study identifies headaches as the most prevalent initial symptom of CVST, followed by seizures and focal neurological deficits. The superior sagittal sinus is the most frequently affected in these patients. Notably, 83.3% of patients in this study achieved a favorable outcome. However, a midline shift exceeding 10mm was identified as a predictive factor for an unfavorable outcome in this series. **Conclusions:** Contrary to previous perceptions, CVST is not uncommon in males. The early diagnosis and prompt intervention have a positive impact on overall patient outcomes. This research sheds light on the importance of recognizing CVST in a broader demographic, its common symptoms, and the critical role of timely intervention for improved patient prognosis.

Keywords: CVST, Cerebral Venous Sinus Thrombosis, Clinical Presentation, Outcome Predictors, Timely Intervention.

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Graphical Abstract



Introduction

Cerebral venous sinus thrombosis (CVST) is a rare cerebrovascular disorder initially described by a 19th-century French physician. CVST primarily involves the blockage of dural sinuses in the brain, often coupled with cortical vein thrombosis. CVST restricts blood and cerebrospinal fluid outflow, resulting in venous infarcts in about 50% of cases [3].

CVST is a significant cause of stroke in young individuals, with mortality rates ranging from 6% to 15%. The International Study on Cerebral Vein and Dural Sinus Thrombosis (ISCVT) reported that most deaths occur within the first 30 days [1].

Patients with CVST typically exhibit symptoms such as headaches, seizures, papilledema, altered consciousness, and focal neurological deficits. These arise from the thrombosis

of intracranial veins and sinuses, leading to hemorrhagic infarctions and increased intracranial pressure. In India, CVST forms a distinct subgroup of cerebrovascular diseases and is a major cause of mortality among women of reproductive age. Postpartum women are particularly susceptible, while alcoholism presents a significant risk factor in males.

Heparin represents the standard treatment for CVST, and most patients respond well to this therapy. However, approximately 20% of patients continue to experience disability or face a fatal outcome [13]. The main cause of death is transtentorial herniation resulting from extensive hemorrhagic infarctions [17].

In cases where patients exhibit a mass effect and midline shift and their condition worsens despite anticoagulation, emergency decompressive surgery has been

proposed as a life-saving measure [1]. Nonetheless, unlike in arterial strokes, decompressive craniectomy is not widely adopted for venous stroke patients, and its role in CVST remains uncertain.

This study aims to assess the outcomes of CVST patients and identify factors that predict patient outcomes following decompressive craniectomy.

Methods and Materials

I) Inclusion criteria

- Patients diagnosed by the imaging studies (CT Brain, MR Brain with Venogram) with CVST
- Patients of age group (16-75) presenting with Cerebral venous thrombosis.

II) Exclusion criteria

- Patient with
- Hypertension
 - Other intra cranial hematoma such as cavernoma etc

Study Area:

This study was conducted in the Department of Neurosurgery, Ruby Hall Clinic, Pune, after obtaining ethical clearance from the ethical committee.

Study population:

Data was obtained from 30 consecutive patients, out of which 19 were male and 11 were female admitted for surgeries at Ruby Hall Clinic, Pune.

Time frame:

Data was collected for a period of 2 years

Study design:

Prospective Observational Study.

Indications for surgery

Indications for surgery include: a low GCS at admission with large infarct on the CT/MRI scan; significant mass effect and midline shift on CT/MRI scan; clinical and radiological signs of transtentorial herniation; deterioration in the sensorium despite anti-edema measures.

Sample size:

Sample size was 30 patients.

Data collection technique and tools:

Data was collected by interviewing the relatives and examining the patients as well as patient's medical records i.e. CT Scan, MRI Scan, perioperatively. All the patients were followed post-operatively for assessment for a period of 6 months. Follow-up data were obtained either through direct clinical evaluation or telephone conversations. The MRS were used to assess the outcome of patients in the follow-up period and was statistically analyzed. Patients with mRS score ≤ 2 at follow-up were considered to have favourable outcome; whereas patients with mRS score ≥ 2 were considered to have poor outcome. Details about death

following the surgical procedure were recorded.

Results

Patient Population

There were 30 patients in the study. All the patients underwent surgical intervention. The mean age was 45.23 years, with a range from 18 to 75 years. There were 19 men (63.3%) and 11 women (36.7%). The mean age of the

patients in the present study was 45.23%. Majority of them were in the age group 39-47 years contributing to 50%. The youngest age being 21 and highest is 73 years. There were 19 men (63.3%) and 11 women (36.7%).

In spite of all the investigations, no cause was found in 43% patients. The brain regions involved are mentioned in table 1. Distribution of thrombosed sinus in Figure 1.

Table 1 provides information about the brain regions involved.

	No. of Cases	Percentage
Brain Regions		
B/L Frontal Lobe	2	6.7%
B/L Frontoparietal Lobe	1	3.3%
LT F-P Lobe	4	13.2%
LT Frontal	1	3.3%
LT Frontotemporal Lobe	2	6.7%
LT FTP Lobe	1	3.3%
LT Parietal Lobe	1	3.3%
LT Parieto-Occipital Lobe	1	3.3%
LT Temporal-Parietal Lobe	4	13.2%
LT Temporo occipital Lobe	1	3.3%
LT Temporal Lobe	5	16.7%
RT Frontoparietal Lobe	3	9.9%
RT FTP Lobe	2	6.7%
RT Parieto-Occipital Lobe	1	3.3%
RT Temporoparietal Lobe	1	3.3%
Total	30	100.0%

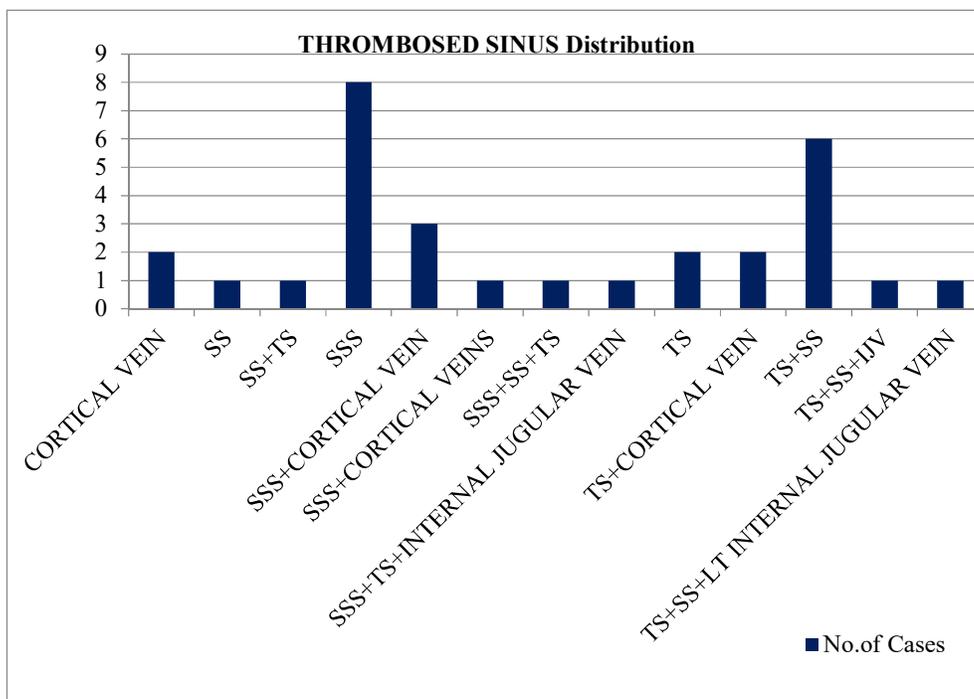


Figure 1. Graph showing distribution of thrombosed sinus

Surgical Management

All the 30 patients underwent decompressive craniectomy of adequate size, with duraplasty performed with a pericranial graft or Dura patch. Re-implantation of the bone flap was performed within 3 months. The

comparative radiological imaging pre and post decompression presented in Figure 2a & 2b. After Post operative day 2, patients were started on Low Molecular Weight Heparin followed by Oral Anti Coagulants.

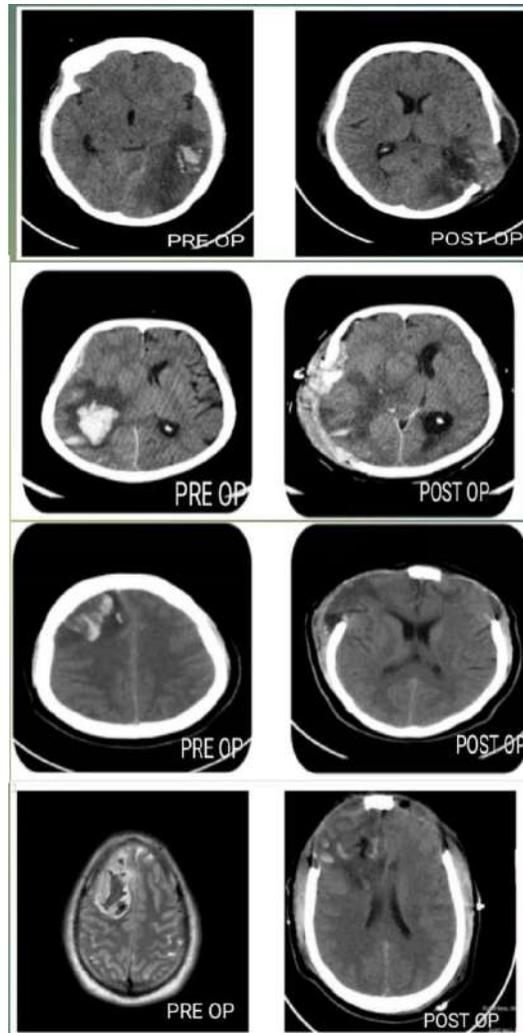


Figure 2a. Sshowing comparative images pre and post surgery.

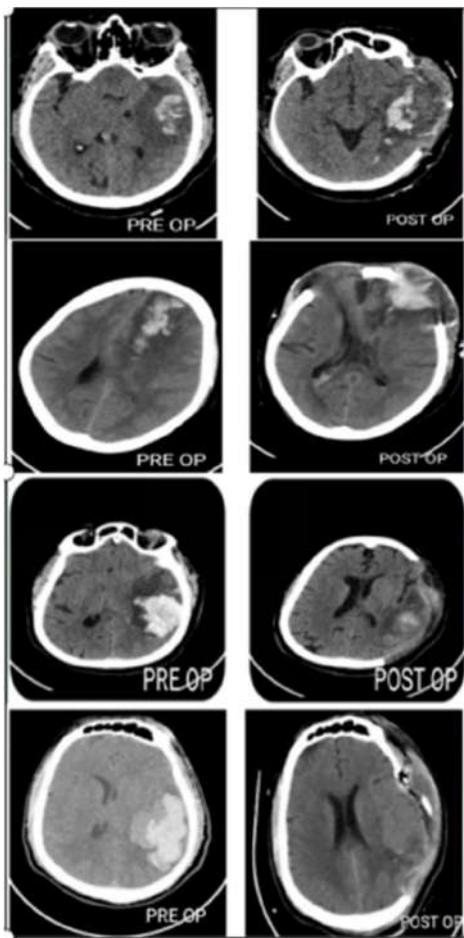


Figure 2b. Showing comparative images pre and post surgery.

Postoperative Course

The GCS score recorded 48 hours, 72 hours and 96 hours after surgery showed a mean of 10.13. Four patients showed no improvement in their GCS score, and in 1, there was further deterioration in the score. postoperative complications were seen in eight patients. Two patients had cerebrospinal fluid (CSF) leak, associated with wound dehiscence and flap necrosis. one patient developed urosepsis. Five patients died; among these, 4 died due to progression of the disease and one patient died due to chest infection.

Prognostic factors

Favourable outcome following surgery were seen in 25 patients (83.3%) and poor outcome (including death) in 5 patients (16.6%). Factors associated with mortality in our study were analysed. Midline line shift more than 10mm was shown to be significantly associated with mortality in Table 2. (p=0.006; considered extremely significant) Factor that did not predict outcome were preoperative GCS (p=0.56), Age > 40 (p=0.52), gender (p=0.65), multiple sinus involvement (p=0.75).

Table 2 shows the analysis of prognostic factors

Survival	Total	Alive (Count)	Death (Count)	MIDLINESHIFT <10	MIDLINESHIFT >10 nt)
	Count	25	5	22	3
	%	100.0%	100.0%	88.0%	12.0%
Chi-Square Tests					
Pearson Chi-Square	Value	10.770a			
	Df	1			
	Asymp. Sig. (2-sided)	.001			
	Exact Sig. (2-sided)	.006			
	Exact Sig. (1-sided)	.006			
Continuity Correctionb	Value	7.304			
	Df	1			
	Asymp. Sig. (2-sided)	.007			
Likelihood Ratio	Value	9.246			
	Df	1			
	Asymp. Sig. (2-sided)	.002			
	Exact Sig. (2-sided)	.006			
	Exact Sig. (1-sided)	.006			

Fisher's Exact Test					
	Exact Sig. (2-sided)		.006		
	Exact Sig. (1-sided)		.006		
N of Valid Cases					
	30				

Discussion

The present study included 30 patients with cerebral venous sinus thrombosis. The highest incidence of our cases was during the third and fourth decades of life, with a mean age of 45 years, which was coincident with the study by Mahale [18] et al. who had 36.7 years as a mean of age. Men represented about 63% and women represented about 37%. In our study, males formed the majority (63.3%) of the total 30 patients similar to that observed in Narayan et al. [30] who observed a male predominance (53.7%).

The presenting symptoms in upto 90% of patients with CVST complain of headache which is the most frequent symptom and often the initial one [31]. By ISCVT, headache was the only symptom in 9% of patients with CVST [13]. In our study headache was the most common symptom (50%) similar to the observations of most other studies [5,6,18,30].

Other common symptoms noted in our study were altered sensorium, focal neurological deficit and seizures similar to other studies [32,33]. Next to headache, most common symptom observed in our study was convulsion and focal deficit.

Seizures are more frequent in CVST than in other stroke types [31]. About 30 to 40% of patients present with seizures, either focal or generalized or with status epileptics. Seizures was observed in 33.33% of our patients however seizure incidence was slightly higher in other studies [18,26].

Focal neurological deficit, was observed in 33.33% of our patients however incidence was higher in 73% in Mahale et al. [18] and 52% in Aaron et al. [26].

Comparison of the common clinical presentations in our study with other studies is shown in the Table 3.

Table 3. Shows comparison of different studies

Symptoms	In present study	Mahale et al. [18]	Aaron s et al. [26]	Vivakaran et al. [5]
Headache	50%	86.70%	75%	82.40%
Seizures	33.33%	33.40%	68%	52.90%
Motor deficit	33.33%	73.40%	52%	88.20%
Altered sensorium	13.33%	63.40%	36%	97.10%

In the present study group, the most common sinus involvement was superior sagittal sinus in 26.7%. The study done by Mohindra et al. and Mahale et al. represented 30% and 43.4% respectively in their observations.

The information on decompressive surgery in CVST is limited to case reports and results from small series. A multinational registry of decompressive surgery in 38 patients with malignant CVST by Ferro et al., (2011) reported favourable outcome (mRS score of 0–2) in 21 patients (55.3%), unfavourable outcome (mRS score of 5–6) in 8 patients (21.1%) and mortality in 7 patients (18.4%) at the follow-up of 14.5 months.

They concluded that the decompressive surgery acts as a

lifesaving procedure in selected CVST patients [25]. A study by Aaron et al. [26] on 44 CVST patients who had decompressive surgery showed good outcome in 27 patients (61.4%) and mortality in 9 patients (20%). Another study by Vivakaran et al. [5] on 34 CVST patients who had decompressive surgery showed good outcome in 26 patients (76.4%) and mortality in 6 patients (17.6%).

In present study 30 patients with CVST studied over a period of 2 years who had decompressive surgery. Favourable outcome was seen in 25 out of 30 patients (83.33%) and were comparable to the other studies (Table 4). The mortality rate was 5 out of 30 patients (16.61%)

Table 4. Shows comparison of favorable outcome and mortality in various studies

Serial Number	Author	Favorable outcome (n/%)	Mortality (n/%)
1	Aaron et al. [26]	27/61.4	9/20.0
2	Vivakaran et al. [5]	26/76.4	6/17.6
3	Present study	25/83.33	5/16.61

This study shows that decompressive craniectomy in CVST is not only life-saving but that the survivors can have an excellent long-term outcome. Compared with arterial strokes where an mRs of 2 after 12 months was achieved only in 14%, we had 60% of survivors having an mRs of ≤ 2 by 6 months itself. It is interesting to note that the mRs improved maximally after the third month. Similar findings were observed by Bousser's [34].

In the present study, we found the GCS at presentation and GCS at 48 hours, 72 hours, 96 hours following decompressive craniectomy showed significant improvement ($p < 0.05$). The prognosis of patients in our case study with presenting GCS was compared with the final outcome at the end of 6 months. There was a statistically significant difference of the same comparison ($p < 0.05$).

The factors predicting poor outcome in our series were midline shift more than 10mm. Factors that did not predict outcome were presence of headache, seizures, preoperative GCS score, age, gender, low GCS at presentation and involvement of multiple sinuses. Lath et al. [6] reported GCS < 8 , deep venous system

involvement, malignancy among few factors affecting favourable outcome. Mahale et al. [18] found following factors predicting poor outcome were age more than 50 years, midline shift more than 10mm and total effacement of basal cistern.

The present study has limitations. The limitation of the study was its small sample size due to the rarity of malignant CVST. In conclusion, decompressive surgery should be considered in large malignant venous infarcts with midline shift. Survivors can expect excellent outcome.

Conclusion

CVST is not uncommon in males, contrary to the previous perception that it primarily affects females. The prompt diagnosis and early intervention were found to have a positive impact on the overall outcome.

Ethics declarations

Funding

This study did not receive any funding.

Conflict of interest

The authors declare that they have no competing interests.

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ORIGINAL ARTICLE

Evaluating Anemia Counseling: A Study among Pregnant Women and Adolescent Girls in Bhanvad Taluka, DevBhoomi Dwarka, Gujarat

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Abstract

Introduction: The iron requirement of the body increases during this period and resulting into anemia. National Family Health Survey-5 (2019-20), about 65% women (15-49 y) and about 62% of pregnant women were suffering from various degrees of anemia in Gujarat. Current study is the part of the innovation pilot project in which counselling was very important component. It motivated the beneficiaries to increase uptake of project services. Thus, the current paper is designed with the aim and objective to analyse the effect of counselling done for all the beneficiaries (Pregnant women and adolescent girls) in context for their improvement in anaemia status. **Materials and Methods:** The current study, conducted between June 2020 to March 2021 in Bhanvad Taluka of DevBhoomi Dwarka, Gujarat, is part of the innovation pilot Anaemia Project under ICDS. Utilizing a mixed-method approach, the study focused on 60 pregnant women and 304 adolescent girls, selected through simple random sampling. Verbal Informed Consent was obtained from each participant, and a semi-structured, pilot-tested questionnaire was administered in the local language by the evaluation team. The active involvement of counsellors, including FHW/ MS/ FHS/ ASHA, added a crucial dimension to the project, enhancing its effectiveness and outreach. **Results:** it was found that overall anemia burden among beneficiaries was reduced (9.34%) considering baseline and endline data taken during the study. This reduction was more seen among PWs (18.33%) as compared to AGs (7.56%). **Conclusion:** The results showed that nutrition education and counselling positively influenced the intake of dietary iron and vitamin C-rich foods.

Keywords: Counselling, Anaemia, IFA, Diet Diversification

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Graphical Abstract

Evaluating Anemia Counseling: A Study among Pregnant Women and Adolescent Girls in Bhanvad Taluka, DevBhoomi Dwarka, Gujarat

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Aims & Objectives

To analyse the effect of counselling done for all the beneficiaries (Pregnant women and adolescent girls) in context for their improvement in anaemia status in Bhanvad Taluka of Devbhoomi Dwarka, Gujarat.

Introduction

Health and Nutritional education and counselling have been defined as educational measures for inducing desirable behavioural changes for the ultimate improvement in the nutritional status of people. It considered as the cheap, highly efficacious health technology and services exist for many leading diseases in developing countries. Prevention and control of anemia is one of key strategies of the health, nutrition and population sector program for reducing maternal and child mortality and improving maternal, adolescent and childhood health status. With this in background, to combat anemia among adolescent girls and pregnant women, the study was conducted in Dev Bhoomi Dwarka District initiated innovation pilot project in Bhanvad Taluka.

Material and Methods:

The current study, conducted between June 2020 to March 2021, focused on Bhanvad Taluka of DevBhoomi Dwarka, Gujarat. Employing a mixed-method approach, the study targeted a sample of 60 pregnant women and 304 adolescent girls, selected through simple random sampling. Verbal informed consent was obtained from each participant to ensure ethical research practices. The evaluation team administered a semi-structured, pilot-tested questionnaire in the local language. Counsellors, including FHW/ MS/ FHS/ ASHA, actively participated in the project, contributing to the comprehensive nature of the study.

Distribution of counselling sessions in last month

Anemia	92.86%
IFA	92.86%
Diet Diversification	85.71%
other	35.71%

Results: The current study revealed a notable reduction in the overall burden of anemia among beneficiaries, amounting to 9.34%, when comparing baseline and endline data collected during the study. Notably, 22% of pregnant women (PWs) and 15% of adolescent girls (AGs) initiated the consumption of Iron and Folic Acid (IFA) with lemon. A significant rise in the daily intake of green leafy vegetables was observed among nearly 70% of beneficiaries, and 40% reported an increase in vitamin C intake.

Conclusion: The success of the intervention was attributed to effective communication between educators and participants. This communication emphasized an individualized approach, fostering two-way feedback and facilitating face-to-face counseling sessions. Such personalized communication strategies were instrumental in enhancing knowledge and promoting healthier behaviors among the targeted population.



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Introduction

Iron deficiency anaemia is the most common form of malnutrition among pregnant women and adolescents. The iron requirement of the body increases during this period and resulting into anemia [1,2]. Anemia is associated with poor cognitive and motor development outcomes in children, can cause fatigue and low productivity, and, when it occurs in pregnancy, is associated with poor birth outcomes (including low birth weight and prematurity) as well as maternal and perinatal mortality [3]. Anemia is a health problem that caused most disability in India over a decade (2005-2016) according to the Global Burden of Disease study. India has the maximum number of anemic women and children in the world. As per the recent National Family Health Survey-5 (2019-20), about 65% women (15-49 y) and about 62% of pregnant women were suffering from various degrees of anemia in Gujarat [4]. Therefore, the importance

anemia as a public health problem has been increasingly recognized by health authorities and policy makers.

Health and Nutritional education and counselling have been defined as educational measures for inducing desirable behavioural changes for the ultimate improvement in the nutritional status of people. It considered as the cheap, highly efficacious health technology and services exist for many leading diseases in developing countries. Prevention and control of anemia is one of key strategies of the health, nutrition and population sector program for reducing maternal and child mortality and improving maternal, adolescent and childhood health status. With this in background, to combat anemia among adolescent girls and pregnant women, the study was conducted in Dev Bhoomi Dwarka District initiated innovation pilot project in Bhanvad Taluka. Various strategies like screening of beneficiaries,

distribution of Iron utensils, counselling, development of kitchen garden, drug distribution etc were implemented under this innovation pilot project. Prevalence of anemia in pregnant woman and adolescent girls can be decreased by some actions, start from promotive, preventive, and curative actions. The preventive and curative action cannot run smoothly without the promotive action. Therefore, counselling was chosen as the main action in promoting healthy life which could directly increase participant's awareness regarding diet for anaemia, so that the each participant become more concern about their health and disease status.

Current study is the part of the innovation pilot project in which counselling was very important component. It motivated the beneficiaries to increase uptake of project services. Thus, the current paper is designed with the aim and objective to analyse the effect of counselling done for all the beneficiaries (Pregnant women and adolescent girls) in context for their improvement in anaemia status.

Methodology

The research was conducted in Bhanvad Taluka of DevBhoomi Dwarka, Gujarat, spanning from June 2020 to March 2021, and forms a segment of the innovative Anaemia Project under ICDS. Utilizing a mixed-method approach, the study focused on 60 pregnant women and 304 adolescent girls, selected through simple random sampling. This initiative aimed to comprehensively address anemia and contribute valuable insights to inform future interventions and public health strategies. The inclusion criteria for the study encompassed individuals identified

as anemic during baseline screening. Additionally, participants who demonstrated a willingness to provide consent were considered eligible for inclusion. This dual criterion aimed to ensure that individuals with a specific health condition, anemia, were included in the study, and that their active consent was obtained, emphasizing both health status and voluntary participation. The exclusion criteria for the study involved individuals who were not identified as anemic during baseline screening and those who did not express a willingness to provide consent. This twofold criterion was established to exclude participants who did not meet the specific health condition of anemia and those who chose not to participate voluntarily. By delineating these criteria, the study aimed to ensure a focused and willing participant pool, emphasizing the relevance of anemia status and active consent in the research. Each participant provided Verbal Informed Consent, and a Semi-Structured, Pilot-Tested questionnaire was administered in the local language by the evaluation team. The project involved the participation of counsellors such as FHW/ MS/ FHS/ ASHA, who underwent training before being instructed to provide counseling to beneficiaries at their respective premises. The evaluation team actively engaged with counsellors during field visits, assessing the quality of counseling services. Additionally, the team conducted inquiries with beneficiaries to gather feedback on project activities. Counsellors implemented Nutrition Counselling as an intervention for the study participants, emphasizing nutrition education. The focus of education included information on anemia, identification of sources of iron-

rich foods, guidance on iron-enhancing and iron-inhibiting foods, and proper hygiene practices. Participants, organized in groups of 6–8, received education through interactive discussions facilitated

by tools such as pamphlets, flip books, play cards, etc. Each counselling session lasted for 30 minutes per participant, carefully designed to ensure a conducive and informative environment (Figure 1).

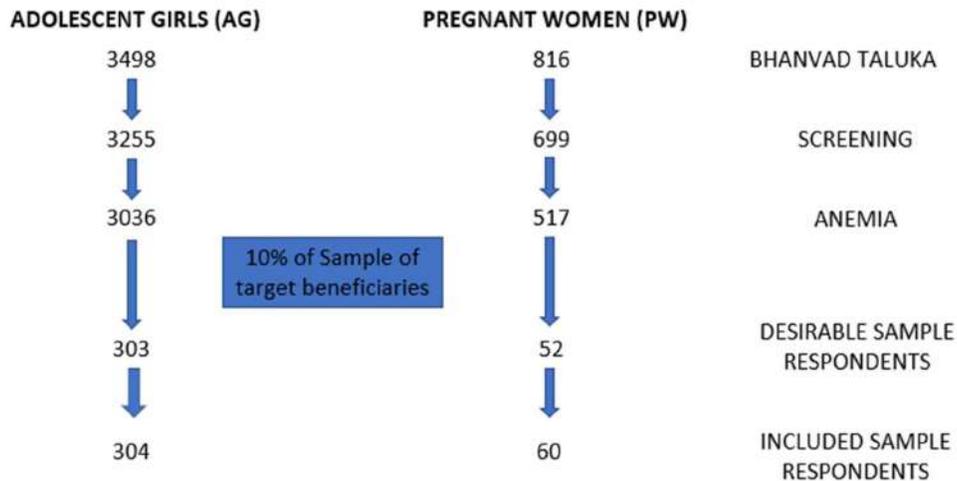


Figure 1. Sampling frame

During the counselling sessions, participants were directed to recognize locally available iron-rich and iron-enhancing foods, along with guidance on the optimal ways to combine them for enhanced iron absorption. Emphasis was placed on promoting proper dietary habits and discouraging poor dietary practices. End-line data collection took place immediately after the intervention to assess the impact of the guidance provided. The data entry process utilized MS Excel, and subsequent analysis was carried out using the Statistical Package for Social Science (IBM SPSS Statistics version 26) and MS Excel. The presentation of data included statistics such as frequency, percentages, mean, standard deviation, and mean difference. Statistical significance was determined by a p-value of <0.05 in all analyses.

Results

The current study revealed a notable reduction in the overall burden of anemia among beneficiaries, amounting to 9.34%, when comparing baseline and endline data collected during the study. This reduction was more pronounced among Pregnant Women (PWs) at 18.33% compared to Adolescent Girls (AGs) at 7.56%. Furthermore, a comparison with baseline screening findings indicated a substantial decrease in anemia prevalence by 22.91%, 7.64%, and 9.74% among PWs, AGs, and both groups collectively, respectively, by the end of the project. The consistent provision of regular counselling by counselors to all beneficiaries played a crucial role in achieving these positive outcomes. Table 1 reflects comprehensive counselling efforts reaching almost all beneficiaries, spearheaded by AWW and supported by FHW (Figure 2). The sessions covered crucial subjects including

Iron and Folic Acid (IFA) supplementation, diet diversification, and the intricate connection between diet and anemia (Figure 4). The emphasis on these topics during counselling aimed to empower beneficiaries with knowledge and practices that contribute to mitigating anemia prevalence. Table 2 illustrates the utilization of pamphlets, flip books, and badges as Information, Education, and Communication (IEC) materials during counseling sessions. The stock of Iron and Folic Acid (IFA) tablets, along with albendazole tablets, consistently ranged between 90% and 95%, ensuring regular availability. Counselors exhibited diverse experience levels, with 29% having less than 5 years, 36% possessing 5 to 10 years, and approximately 7% boasting more than 25 years of counseling experience. This varied experience contributed to the overall effectiveness of

the counseling intervention. (Table 3) (Figure 1). Table 4 illustrates the impact of counseling on dietary behavior within the study population. Notably, 22% of pregnant women (PWs) and 15% of adolescent girls (AGs) initiated the consumption of Iron and Folic Acid (IFA) with lemon. A significant rise in the daily intake of green leafy vegetables was observed among nearly 70% of beneficiaries, and 40% reported an increase in vitamin C intake. Moreover, 33% of PWs and 55% of AGs experienced a heightened daily protein intake. Figure 3 delves into the training status of counselors, revealing that approximately 86% received proper project training, and about 57% reviewed the relevant documents. This data underscores the positive influence of counseling interventions on dietary habits among the target population.

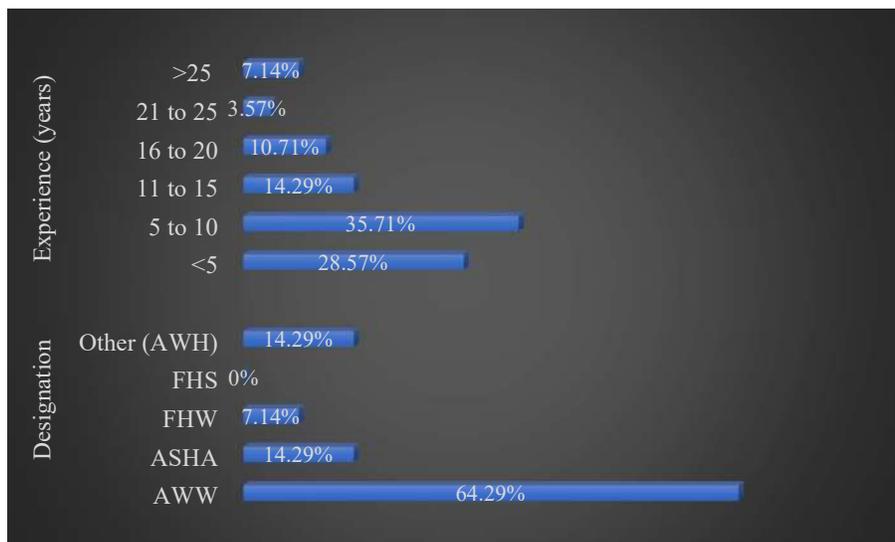


Figure 2. Designation and experience in jobs of counselors

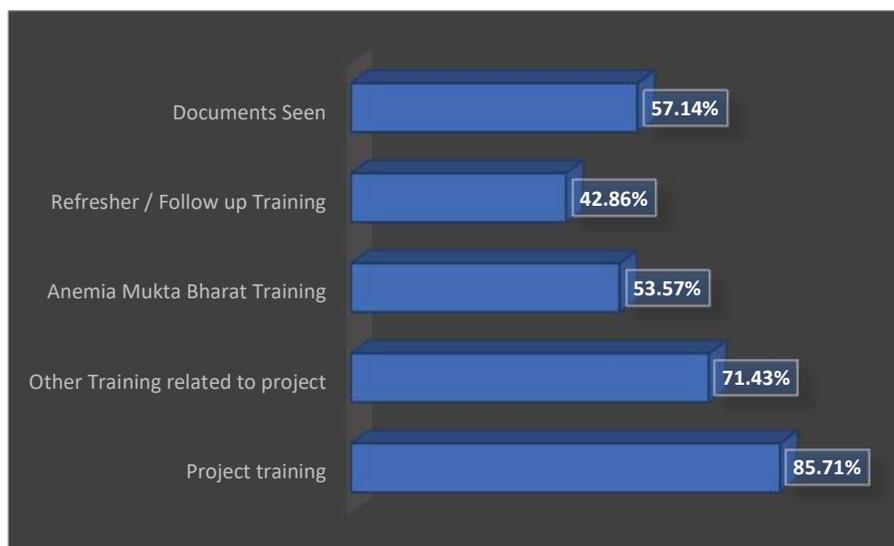


Figure 3. Training status of counselors

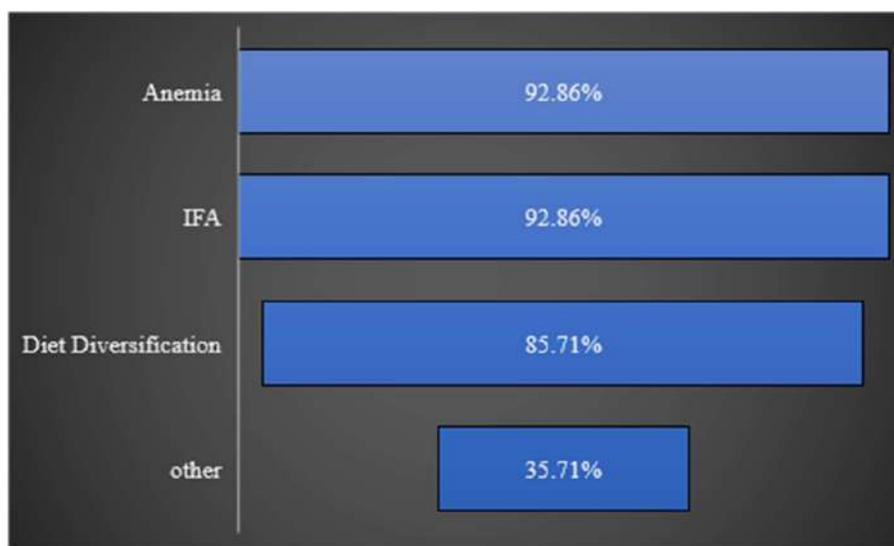


Figure 4. Distribution of counselling sessions in last month

Table 1: Details of Counselling done among Study Sample

		PW		AG	
		N=60	%	N=304	%
Counselling	Counselling done	60	100.0	294	96.71
	Beneficiaries benefitted	58	96.67	274	90.13
	Any IEC Material given	40	66.67	198	65.13
Counselling done by	AWW	51	85.00	270	88.82
	ASHA	18	30.00	70	23.03
	FHW	25	41.67	67	22.04
	Other	1	1.67	-	-
Counselling session in last month	0	9	15.00	21	6.91
	1	36	60.00	154	50.66
	2	14	23.33	83	27.30
	3	1	1.67	22	7.24
	4	-	-	14	4.61
Topic covered in counselling session	Anemia	40	66.67	243	79.93
	IFA	53	88.33	244	80.26
	Diet Diversification	30	50.00	98	32.24
	Diet	58	96.67	181	59.54
	other	5	8.33	9	2.96
Mean Counselling session in last month		1.12		1.5	
SD		0.67		0.91	

Table 2. Logistics and drugs availability with counselors

		Counselor	
		(n=28)	%
IEC	Pamphlets	26	92.86
	Badges	12	42.86
	Flip Books	25	89.29
	Play cards	18	64.29
	Other counselling materials (Specify)	19	67.86
IFA	IFA tablets	25	89.29
	IFA Syrup	22	78.57
	Stock adequate	25	89.29
	Supply regular	27	96.43
	Knows correct Dose for treatment	21	75.00
	Knows correct Dose for prophylaxis	23	82.14
	Knows Iron absorbers	25	89.29
	Knows Iron Inhibitors	23	82.14
Albandazole Tab	Albandazole tablets	27	96.43
	Stock adequate	27	96.43
	Supply regular	28	100.00
	Knows correct Dose for prophylaxis	25	89.29
other supplements	Available	24	85.71
	Stock adequate	24	85.71
	Supply regular	24	85.71

Table 3. Counselling details in last month

		Counselor	
		(n=28)	%
Experience (years)	<5	23	82.14
	5 to 10	1	3.57
	11 to 15	1	3.57
	16 to 20	0	0.00
	21 to 25	1	3.57
	25 to 30	2	7.14
Topics Covered in Last Month	Anemia	27	96.43
	Diet Diversification	28	100.00
	Diet in Anemia	27	96.43
	Use of Iron utensil	28	100.00
	Role of Kitchen Garden	26	92.86
	IFA and Albendazole	27	96.43
	Compliance of IFA	27	96.43

Table 4. Effect of counselling on Dietary behavior among study population

		PW		AG	
		N=60	%	N=304	%
After diet Counselling	IFA taken with Lemon Juice	13	21.67	45	14.80
Daily intake of GLV like Palak, amarnath, methi, Bhaji etc.	increase	42	70.00	209	68.75
	decrease	1	1.67	0	0.00
	no change	17	28.33	95	31.25
Daily intake of Vit C rich foods Like Lemon, amala, orange etc.	increase	26	43.33	124	40.79
	decrease	1	1.67	0	0.00
	no change	33	55.00	180	59.21
Daily intake of Proteinrich food like pulses, dals, ground nut etc.	increase	33	55.00	166	54.61
	decrease	1	1.67	0	0.00
	no change	26	43.33	138	45.39

Discussion

As per the current study, one to one interview was performed with semi structured assessment checklist on total 304 adolescent girls (AGs) and 60 pregnant women (PWs). Most of PWs (81%) and AGs (88.16%) were vegetarian while were having mixed diet. Regular counselling was done to all beneficiaries under the project. Frequent training sessions are arranged for the counsellors (FHS, MS, AWWs, ASHAs and FHWs etc) for sensitization of project activities including health education and counselling of beneficiaries. All are ever counseled regarding project activities during any time of project period and majority stated that counselling is benefited them. Almost two third (2/3) of them stated that IEC materials are given to them. Most of beneficiaries stated that their counselling is mainly done by AWW and at least one time in the last month. IFA, Anemia related information, Diet and Diet diversification were included in the counselling. According to Ogunbile et al. [5] and Wang [6] implementation of nutrition education and counselling by counselors might have led to behavior change and increase in the intake of iron-rich foods which leading to improved healthy eating. According to the study done in China [6-9] nutrition behavior in adolescents was significantly improved after 6-month nutrition education program done in remote rural areas of China.

As per the current study counselors used pamphlets and flip books maximum as IEC material for the counseling. Intensive Counseling was used as an intervention to change the dietary behavior among study population. The outcome of the study showed the improvement in daily

intake of GLV, Vitamin C and Protein intake. Similar results were also consistent with Wiafe et al. [10], Sunuwar et al. [11] and Otoo et al. [12]. Those showed improvement in dietary iron intake within the study groups. Improvement in dietary iron intakes has been reported after the implementation of nutrition education which resulted into drop in the prevalence of anemia. Perhaps the implementation of nutrition education and counselling by counsellors might have led to behavior change as a result improving in iron-rich foods intake. Moreover, the wide variety of iron-rich foods may have increased compliance. Hence, the increased intake of this micronutrient rich foods. According to Kamalraja et al. [13] and Aazam doust Mohammad et al. [14] the nutrient like fat, energy, iron intake was significantly higher in experimental group when compared with the control group. The post intervention nutrient intake except for the intake of iron and folic acid continued to be below the RDA in both the groups. The results indicated that the lack of attitude, knowledge and practice on dietary habits and health has definitely had a negative effect on their nutrient intake.

As per Egryani et al. [15] intervention through one-to-one counseling using pamphlet as one of many methods used to promote healthy lifestyle especially prevention of anemia in pregnant women was successfully increased participant's knowledge about anemia and there was a significant difference between pregnant women's pre and post knowledge about iron tablet ($p = 0,000$) while as per Ghimire et al. [16] pregnant women's knowledge were correlated with high level education ($p = 0.002$) and ANC visits more than four

times ($p = 0.007$). Similarly, another study in Nepal [17] showed that pregnant women who were not consuming regular iron supplements were 18.38 times more likely to develop anemia than who had consumed regularly (AOR=18.380, with 95% CI =3.687- 91.624, $p=0.005$).

Conclusion

The study aimed to evaluate, how nutrition education and counseling impacted the nutritional status, specifically targeting anemia, among pregnant women (PWs) and adolescent girls (AGs). The results demonstrated a significant positive effect of the intervention on the consumption of iron and vitamin C-rich foods, leading to notable improvements in nutrient intake and a reduction in anemia prevalence among both PWs and AGs. Additionally, the incorporation of weekly iron-folic acid supplementation, along with semi-annual deworming, emerged as a feasible and cost-effective strategy for preventing anemia in AGs across various settings, be it institutional or community-based.

The success of the intervention was attributed to effective communication between educators and participants. This communication emphasized an individualized approach, fostering two-way feedback and facilitating face-to-face counseling sessions. Such personalized communication strategies were instrumental in enhancing knowledge and promoting healthier behaviors among the targeted population.

These findings underscore the importance of tailored interventions and personalized communication strategies within nutrition education and counseling programs. By addressing the specific needs

and circumstances of PWs and AGs, these programs can effectively contribute to positive health outcomes, particularly in terms of improving nutritional status and combating anemia. This highlights the significance of investing in targeted interventions and communication approaches to address nutritional challenges and promote overall well-being among vulnerable populations.

Limitations and Recommendations

The evaluation of the innovation pilot project utilized an exploratory study design to conduct a quick assessment. The study acknowledged the limitation of a relatively small sample size involving 14 AWCs, highlighting that a larger sample size would enhance the external validity of the findings. Recommendations were made for multiple training sessions for both counselors and beneficiaries, recognizing that behavioral change and lifestyle modification require time and motivation. The possibility of recall bias regarding daily diet frequency, type, and the intake of IFA or albendazole was also acknowledged. The importance of extending nutrition education and counseling to the male partners of pregnant women was emphasized, focusing on the complications of the disease and the utility of supplementary diets during pregnancy. Additionally, the need for counseling on good hygiene practices to reduce infestations and infections among beneficiaries was highlighted. These suggestions aimed to enhance the effectiveness and comprehensive impact of the intervention.

Authors Contribution

The manuscript has been read and approved by us and we believe that the manuscript represents honest work.

Conflicts of interest

The authors declares that they do not have conflict of interest.

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ORIGINAL ARTICLE

Endoscopic Pilonidal Sinus Treatment {Epsit} vs Limberg Flap for Pilonidal Sinus: A Single Center Experience

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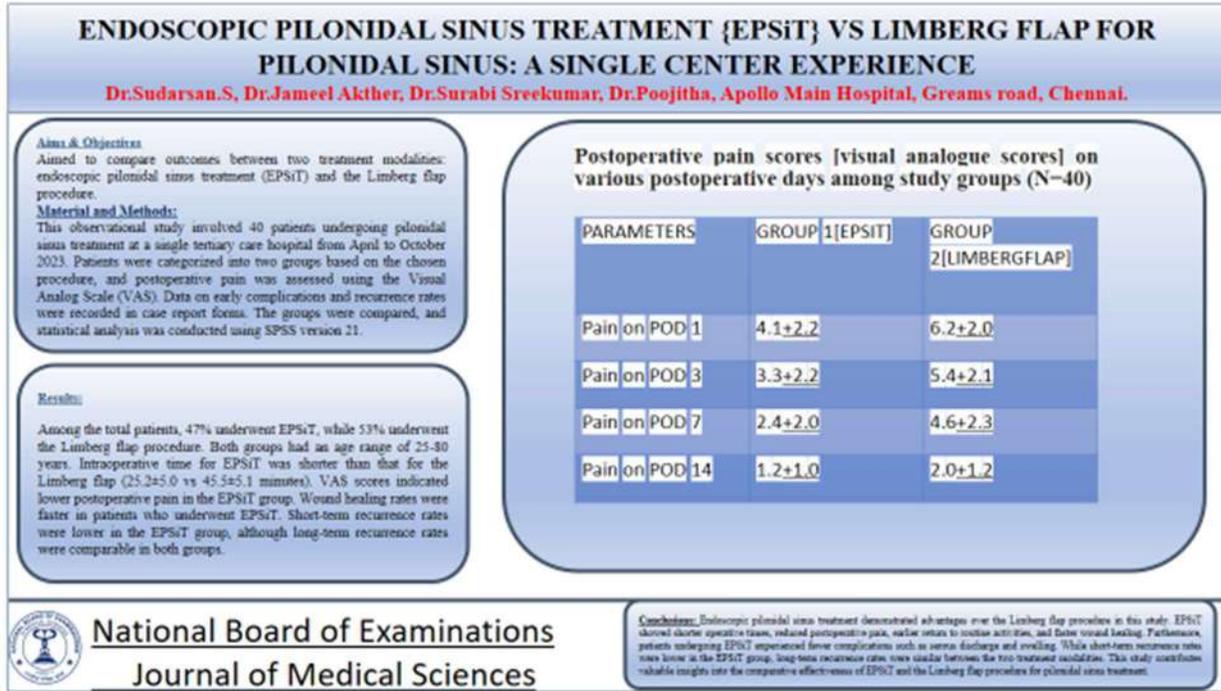
Abstract

Background: Pilonidal sinus, a condition characterized by chronic irritation in the gluteal cleft, predominantly affects males and is often associated with a higher body mass index and a sedentary lifestyle. Despite its prevalence, a universally accepted gold standard for treatment is yet to be established. This study aimed to compare outcomes between two treatment modalities: endoscopic pilonidal sinus treatment (EPSiT) and the Limberg flap procedure. **Materials and Methods:** This observational study involved 40 patients undergoing pilonidal sinus treatment at a single tertiary care hospital from April to October 2023. Patients were categorized into two groups based on the chosen procedure, and postoperative pain was assessed using the Visual Analog Scale (VAS). Data on early complications and recurrence rates were recorded in case report forms. The groups were compared, and statistical analysis was conducted using SPSS version 21. **Results:** Among the total patients, 47% underwent EPSiT, while 53% underwent the Limberg flap procedure. Both groups had an age range of 25-80 years. Intraoperative time for EPSiT was shorter than that for the Limberg flap (25.2±5.0 vs 45.5±5.1 minutes). VAS scores indicated lower postoperative pain in the EPSiT group. Wound healing rates were faster in patients who underwent EPSiT. Short-term recurrence rates were lower in the EPSiT group, although long-term recurrence rates were comparable in both groups. **Conclusion:** Endoscopic pilonidal sinus treatment demonstrated advantages over the Limberg flap procedure in this study. EPSiT showed shorter operative times, reduced postoperative pain, earlier return to routine activities, and faster wound healing. Furthermore, patients undergoing EPSiT experienced fewer complications such as serous discharge and swelling. While short-term recurrence rates were lower in the EPSiT group, long-term recurrence rates were similar between the two treatment modalities. This study contributes valuable insights into the comparative effectiveness of EPSiT and the Limberg flap procedure for pilonidal sinus treatment.

Keywords: Endoscopic Pilonidal Sinus Treatment (EPSiT), Limberg Flap Procedure, treatment Outcomes, pilonidal sinus, surgical modalities

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Graphical Abstract



Introduction

Pilonidal sinus is characterized as an inflammatory condition affecting the skin and subcutaneous tissue in the region of the gluteal fold [1]. The disease is estimated to have an incidence of 26 per 100,000 individuals, with a male-to-female ratio of 2.2:1 [2]. Various theories exist regarding the etiology of pilonidal sinus, including the foreign body response [3], Bascom's 'midline pits' hypothesis [4,5], and Stelzner's theory of retention dermatopathy [6].

Typically, surgical intervention for pilonidal sinus involves the radical excision of the sinus with primary wound closure or secondary healing through granulation, leading to extended wound healing times [7,8]. Off midline closure, such as with a Karydakias flap, is a preferred surgical

technique that appears to reduce recurrence rates in uncomplicated cases [9]. In instances of larger and more complex sinuses, a rhomboid Limberg flap may be necessary due to the extensive wound [10].

A minimally invasive alternative, endoscopic pilonidal sinus treatment (EPSiT), has been developed by Meinero et al. [11]. This technique enables the direct vision debridement of sinus tracts using a specially designed fistuloscope. While studies, including a recent systematic review, have reported positive outcomes for EPSiT, most of these cases involved simple, uncomplicated sinuses [12]. Complex cases with branched, purulent, or recurrent sinuses often necessitate major surgery involving flap rotation.

The focus of this observational study was to compare the treatment outcomes of pilonidal disease using either the endoscopic device (EPSiT) or a Limberg flap, exploring the feasibility of the endoscopic approach in addressing challenging cases.

Materials and Methods

Between April 2023 and October 2023, individuals diagnosed with pilonidal sinus were offered a choice between two treatment options: the minimally invasive EPSiT and the traditional Limberg flap. The surgeon thoroughly discussed the pros and cons of each method with every patient, and the decision on the treatment approach was based on the individual patient's preference. Ethical approval for the study protocol was obtained from the ethics committees, and informed consent from each patient was secured before the operation. All patients were admitted to the hospital one day prior to the surgery, receiving a single dose of antibiotics (2 g intravenous cephazolin, administered 30 minutes before surgery) and antithrombotic prophylaxis (following the Caprini Score). The procedures were conducted under epidural analgesia.

For the endoscopic procedures, the technique described by Meinero et al. was followed. A fistuloscope (STORZ GmbH; KARL STORZ SE & Co. KG, Tuttlingen, Germany) was inserted either through the primary opening (enlarged with forceps if necessary) or one of the side openings, as appropriate. Saline was used to flush the tracts, and under direct visual guidance, hair and necrotic debris were removed using forceps. Subsequently, electrocautery probe was employed for thorough ablation of all

sinus tracts, and a dedicated brush passed through the fistuloscope cleaned the sinus of necrotic tissues. The sinus orifice was left open for drainage, and patients were discharged the day after the procedure with a prescription for paracetamol (1 g orally four times a day). For patients opting for the flap procedure, the sinus was completely excised, including all side branches, and the wound was closed using a rhomboidal cutaneo-subutaneo-fascial flap rotated from the left or right buttock, depending on the configuration of the sinus and its branches. The flap and the secondary wound were sutured in three layers (fascia, subcutaneous tissue, skin) using absorbable polyglactin 2-0 sutures. No drains were left in the wound, and patients were discharged two days after surgery with the same paracetamol dosage prescription.

Data from all patients operated on using both methods during the specified period were prospectively collected in an anonymized electronic database. The primary outcome was healing, categorized into three groups for patients with a follow-up of 12 months or more: healed (with no recurrence), recurrent (requiring another pilonidal sinus operation after initial healing), or persistent (not healed within 6 months). Secondary outcomes included surgery-related complications (wound infection, wound dehiscence, bleeding). At the time of database closure, complete follow-up data of 12 months or more were available for 40 patients: 21 treated with the Limberg flap technique and 19 with the EPSiT method. The data were analyzed using SPSS version 21.

EPSiT Technique

We employed a fistuloscope, a monopolar electrode, and an endoscopic grasping forceps in the procedure. The fistuloscope features an 8-degree angled eyepiece, an optical channel, and a working and irrigation channel. With a diameter of 3.2x4.8 mm and an operative length of 18 cm,

it is equipped with a removable handle for enhanced maneuverability and improved ergonomics for the surgeon. Pre-operative antibiotic prophylaxis was given, and patients were positioned in a prone posture with separated buttocks. The EPSiT procedure was conducted under general anesthesia (Figures 1 and 2).

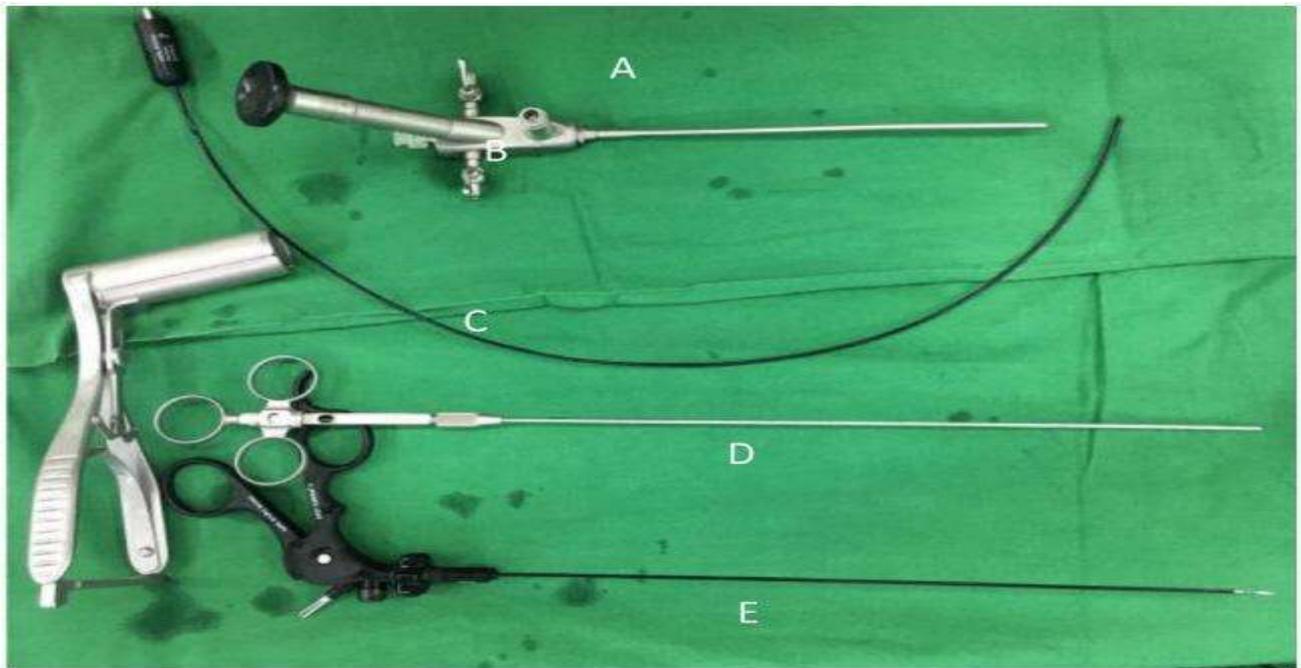


Figure 1. Equipment used for EPSiT. **A** Angled fistuloscope -30 degree scope, **B** Speculum, **C**, monopolar electrocautery wire, **D**, aspiration syringe, **E**, atraumatic non toothed forceps



Figure 2. Interior of pilonidal sinus when viewing with fistuloscope - cauterization of tract

Limberg Flap

The limberg flap was marked on the skin for a rhomboid-shaped excision, and dissection was carried out until reaching the presacral fascia. The procedure involves incorporating the gluteal fascia, forming an inferior border, and delivering it medially to

cover the rhomboid defect. A negative suction drain is inserted, and the wound was closed using nylon sutures. The drain was removed based on decreasing output, and the sutures are taken out on the 16th day after the operation (Figures 3 and 4).



Figure 3. Rhomboid shaped excision area marked and area for skin flap



Figure 4. Final outcome of limberg flap

Results

Patients who underwent endoscopic pilonidal sinus were grouped as group 1 and

patients who underwent limberg flap were grouped as grouped 2.

Table 1. Baseline characteristics of study population (N=40)

PARAMETERS	GROUP 1[EPSiT]	GROUP 2[LIMBERGFLAP]
Total number of patients	19	21
Age range (in years)	25-75	25-80
Mean age ± SD (in years)	50 ± 2.5	53 ± 3.2
Male /female	14/5	15/6
Mean Intraoperative time[mins]	25.2±5.0	45.5±5.1

The study involved a total of 40 patients, with 19 individuals undergoing Endoscopic Pilonidal Sinus Treatment (EPSiT) in Group 1 and 21 patients opting for the Limberg flap procedure in Group 2. In terms of age distribution, patients in the EPSiT group ranged from 25 to 75 years, with a mean age of 50 years and a standard deviation of ±2.5 years. The Limberg Flap group had patients aged between 25 and 80 years, with a mean age of 53 years and a

standard deviation of ±3.2 years. Gender distribution revealed that in the EPSiT group, 14 patients were male and 5 were female, while in the Limberg Flap group, 15 were male and 6 were female (Table 1).

One notable finding is the difference in mean intraoperative time between the two treatment methods. The EPSiT group demonstrated a substantially shorter average intraoperative time of 25.2 minutes, with a standard deviation of ±5.0 minutes, in

contrast to the Limberg Flap group, where the mean intraoperative time was 45.5 minutes, with a standard deviation of ± 5.1 minutes. These results suggest that the EPSiT

approach may offer a more time-efficient alternative for pilonidal sinus treatment compared to the traditional Limberg flap procedure. (Table 1)

Table 2: Postoperative pain scores [visual analogue scores] on various postoperative days among study groups (N=40)

PARAMETERS	GROUP 1[EPSiT]	GROUP 2[LIMBERGFLAP]
Pain on POD 1	4.1 \pm 2.2	6.2 \pm 2.0
Pain on POD 3	3.3 \pm 2.2	5.4 \pm 2.1
Pain on POD 7	2.4 \pm 2.0	4.6 \pm 2.3
Pain on POD 14	1.2 \pm 1.0	2.0 \pm 1.2

Table 2 displays the postoperative pain scores, measured on a visual analogue scale, for both study groups (N=40). In Group 1, which underwent Endoscopic Pilonidal Sinus Treatment (EPSiT), the pain scores were 4.1 \pm 2.2 on postoperative day (POD) 1, 3.3 \pm 2.2 on POD 3, 2.4 \pm 2.0 on POD 7, and 1.2 \pm 1.0 on POD 14. Comparatively, Group 2, which underwent the Limberg flap procedure, exhibited higher pain scores with 6.2 \pm 2.0 on POD 1, 5.4 \pm 2.1 on POD 3, 4.6 \pm 2.3 on POD 7, and 2.0 \pm 1.2 on POD 14.

These results suggest that patients in the EPSiT group generally experienced lower postoperative pain levels across various postoperative days compared to those in the Limberg Flap group (Table 2).

Resuming routine activities among groups

Patients in group 1 who underwent EPSiT returned to routine activities within 3.4 \pm 0.5 days and patients in group 2 who underwent limberg flap returned to routine activities within 5.6 \pm 2.2 days.

Table 3: Early complications among study groups (N=40)

PARAMETERS	GROUP 1 [EPSiT]	GROUP 2: [LIMBERG FLAP]
Swelling	2	4
Serous discharge	2	5
Purulent discharge	2	2

Table 3 outlines the early complications observed among the study groups (N=40) following different treatment modalities for pilonidal sinus. In Group 1, which underwent Endoscopic Pilonidal Sinus

Treatment (EPSiT), two cases reported swelling, two cases exhibited serous discharge, and two cases showed purulent discharge. In contrast, Group 2, which underwent the Limberg flap procedure,

experienced a slightly higher incidence of early complications, with four cases of swelling, five cases of serous discharge, and two cases of purulent discharge. These findings indicate that the EPSiT group had a relatively lower occurrence of early complications, specifically in terms of swelling and serous discharge, when

compared to the Limberg flap group. (Table 3)

Complete wound healing

Patients who underwent EPSiT had earlier wound healing rates 20.1 ± 2.3 days. The patients who underwent limberg flap had longer wound healing time 25.8 ± 2.9 days.

Table 4: Recurrence rates among study groups (N=40)

PARAMETERS	GROUP 1 [EPSiT]	GROUP2 [LIMBERG FLAP]
Recurrence <45 days	0	0
Recurrence in 3 months	1	2
Recurrence in 6 months	2	2

Table 4 outlines the recurrence rates observed among the study groups (N=40) following different treatment modalities for pilonidal sinus. In Group 1, which underwent Endoscopic Pilonidal Sinus Treatment (EPSiT), there were no reported cases of recurrence within the first 45 days postoperatively. However, one case of recurrence was observed within 3 months, and two cases were noted within 6 months. In Group 2, which underwent the Limberg flap procedure, similarly, there were no instances of recurrence within the initial 45 days. However, two cases of recurrence were reported within both 3 and 6 months postoperatively. These results suggest that both treatment groups had no recurrences within the first 45 days, but there were some occurrences within the subsequent 3- to 6-month period (Table 4).

Discussion

This study aimed to compare the outcomes of Endoscopic Pilonidal Sinus Treatment (EPSiT) and the Limberg flap

procedure in the management of pilonidal sinus. The baseline characteristics of the study population revealed a comparable distribution between the two groups, with Group 1 (EPSiT) consisting of 19 patients and Group 2 (Limberg Flap) comprising 21 patients. The age range and mean age were slightly lower in Group 1, indicating a relatively younger cohort. Additionally, the male predominance was observed in both groups, reflecting the typical demographic profile of pilonidal sinus patients. Nasr et al. [3] noted that the disease was common among both males and females with equal distribution, however in our study we noted that there was more persistence of disease among the males. But the study conducted by Nasr et al. was done in a pediatric group of patients. The same male to female equal distribution was noted in a study conducted by Sequeira et al. [13].

A valuable finding in this study was the substantial difference in mean intraoperative time between the two treatment methods. EPSiT demonstrated a

notably shorter average intraoperative time of 25.2 minutes compared to the Limberg flap procedure, which had a mean intraoperative time of 45.5 minutes. This was in accordance with study by Meinero et al [11]. This suggests that EPSiT may offer a more time-efficient alternative for pilonidal sinus treatment, potentially contributing to reduced surgical stress and operative costs.

Postoperative pain scores, measured on various days, revealed a consistent trend favoring the EPSiT group. Patients undergoing EPSiT experienced lower pain levels across different postoperative days compared to those undergoing the Limberg flap procedure. This indicates that the endoscopic approach may lead to a more comfortable postoperative recovery for patients. This was along the lines of study by Esposito et al. [14]

Resumption of routine activities further supported the advantages of EPSiT, with patients in Group 1 returning to routine activities within 3.4 ± 0.5 days, while those in Group 2 took longer, requiring 5.6 ± 2.2 days. This suggests that EPSiT may facilitate a quicker recovery and earlier return to daily life. The return to work was shorter in our study in patients who underwent limberg flap when compared with study by Kuvvetli et al. [15].

Early complications, including swelling and serous discharge, were less frequent in the EPSiT group compared to the Limberg flap group. This highlights the potential benefits of the endoscopic approach in minimizing early postoperative issues. The complications among patients underwent limberg flap was lesser when compared to study by Ozcan R et al. [16].

Wound healing rates also favored EPSiT, with patients in this group experiencing earlier complete wound healing at 20.1 ± 2.3 days, in contrast to the Limberg flap group, which had a longer healing time of 25.8 ± 2.9 days. This indicates that EPSiT may contribute to a more expedited and efficient wound healing process. The wound healing in patients who underwent limberg flap was in accordance with study done McCallum et al. [17].

The recurrence rates within the first 45 days were zero for both groups, indicating an immediate success in preventing early recurrences. However, within the subsequent 3- to 6-month period, there were some occurrences in both groups, suggesting that ongoing surveillance and follow-up are essential in assessing the long-term efficacy of these treatments.

Minimally invasive treatment offers numerous advantages, with several studies demonstrating a shorter post-treatment recovery period after Endoscopic Pilonidal Sinus Treatment (EPSiT) compared to more extensive surgical procedures [13–18]. Patients undergoing EPSiT often report experiencing less pain and enjoy a better quality of life when compared to those undergoing traditional flap surgery for pilonidal sinus [19]. Despite these benefits, one drawback of EPSiT is the requirement for specific equipment, including a fistuloscope and necessary instruments, incurring higher procedural costs, including the initial equipment purchase, and expenses associated with disposable elements and equipment sterilization. In contrast, the flap procedure can be performed using a standard set of

surgical instruments and several packages of surgical sutures.

The literature consistently reports favorable outcomes for the EPSiT procedure. A significant multicenter study, led by the inventor of EPSiT, involving 250 patients, predominantly with uncomplicated pilonidal sinus, demonstrated a notable 94.8% success rate in terms of healing [11]. Additionally, a study by the same authors focused on evaluating EPSiT in patients with recurrent pilonidal sinuses, revealing a similarly high effectiveness rate of 95% [20].

Acknowledging the limitations of our study, the absence of randomization stands out as a notable drawback. The choice of the surgical procedure was based on patients' decisions, potentially influenced by information provided by the surgeon, introducing a potential source of bias. Furthermore, the relatively small size of the patient groups is another limitation that should be considered when interpreting the results.

Conclusion

In conclusion, the findings from this study suggest that EPSiT may offer several advantages over the traditional Limberg flap procedure, including shorter intraoperative time, reduced postoperative pain, quicker resumption of routine activities, fewer early complications, and faster wound healing. However, long-term recurrence rates warrant continued monitoring. Overall, these results contribute valuable insights into the comparative effectiveness of these treatment modalities for pilonidal sinus, providing a foundation for informed clinical decision-making.

Conflicts of Interests

The authors declares that they do not have conflict of interest.

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ORIGINAL ARTICLE

Oral health situation analysis and integration of Massive Open Online Course Modules on Oral Health Promotion in WHO SEAR Countries: A Comprehensive Report

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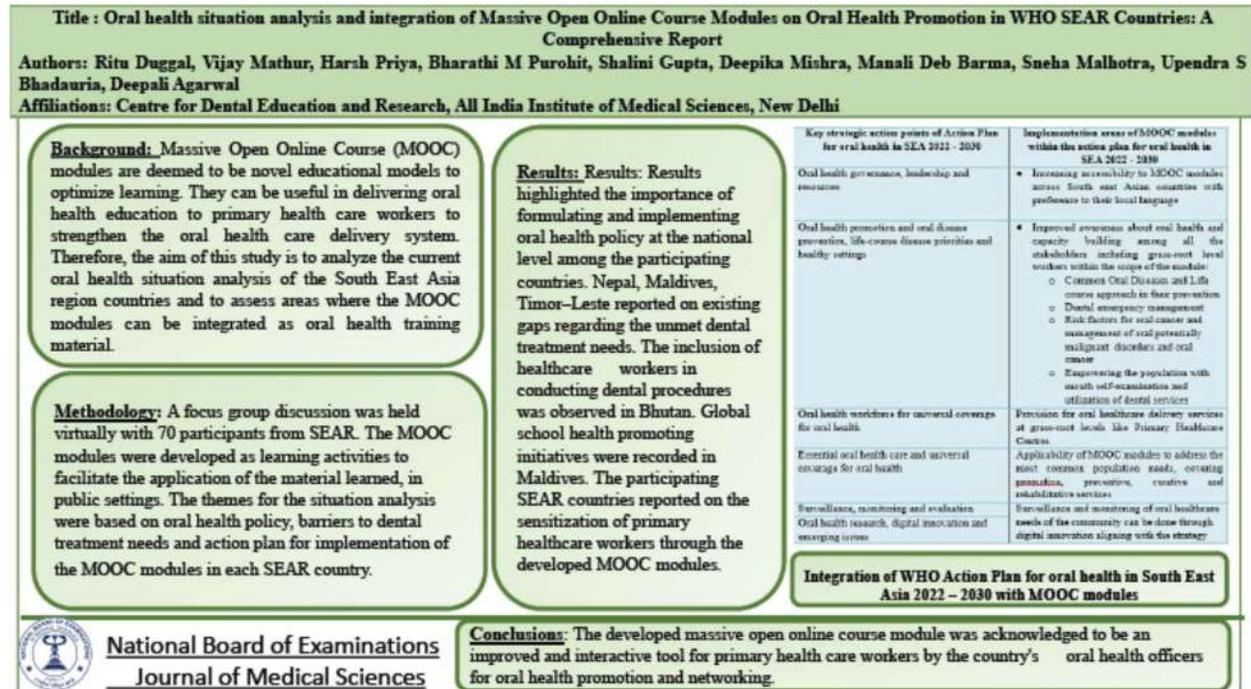
Abstract

Background: Massive Open Online Course (MOOC) modules are deemed to be novel educational models to optimize learning. They can be useful in delivering oral health education to primary health care workers to strengthen the oral health care delivery system. Therefore, the aim of this study is to analyze the current oral health situation analysis of the South East Asia region countries and to assess areas where the MOOC modules can be integrated as oral health training material. **Methodology:** A focus group discussion was held virtually with 70 participants from SEAR. The MOOC modules were developed as learning activities to facilitate the application of the material learned, in public settings. The themes for the situation analysis were based on oral health policy, barriers to dental treatment needs and action plan for implementation of the MOOC modules in each SEAR country. **Results:** Results highlighted the importance of formulating and implementing oral health policy at the national level among the participating countries. Nepal, Maldives, Timor–Leste reported on existing gaps regarding the unmet dental treatment needs. The inclusion of health care workers in conducting dental procedures was observed in Bhutan. Global school health promoting initiatives were recorded in Maldives. The participating SEAR countries reported on the sensitization of primary healthcare workers through the developed MOOC modules. **Conclusion:** The developed massive open online course module was acknowledged to be an improved and interactive tool for primary health care workers by the country's oral health officers for oral health promotion and networking.

Keywords: oral health, WHO SEARO, barriers to oral health, health education, primary healthcare workers

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Graphical Abstract



Introduction

Massive Open Online Course (MOOC) was first developed in the early 2008, as an educational tool with the intention of making education more effective among students [1]. Unlike other traditional courses, the MOOCs are designed to accommodate huge numbers of students, frequently in thousands with technology assisting the educators in facilitating large scale learning [2]. Most of the evidence available to date does not specifically address MOOCs even though they offer an opportunity to deliver content online and to contribute to a blended learning design that could produce an equal or better learner outcome. MOOCs are typically lecture based offerings, using short video presentations and independent formative assessments, to allow large numbers of learners across the globe to obtain high-quality education without direct instructor feedback [3]. The courses may not

be the primary mode of teaching for traditional medical courses which requires face to face practical and skill training with patients but act as an educational supplement through information propagation to many participants at the same time [4]. One of the examples in India is the Study Webs of Active Learning for Young Aspiring Minds (SWAYAM) portal [5] which is one of the government's efforts to establish a large number of online courses, practically all of which are free. Implementation of the courses is not a problem because the government has extensive infrastructure and resources. As online education continues to grow, it is important that it finds its use in various sectors especially health to facilitate learning. The health sector of India consists of a mix of public and private healthcare systems and is fundamentally divided into: primary, secondary and tertiary [6]. The healthcare system is designed such that at grassroot

level, primary health care workers provide preventive and curative services and become the first point of contact with the population [7,8]. Studies have been conducted to assess the oral health knowledge among primary healthcare workers, some of which have shown a good knowledge regarding oral health [9,10]. Online programs have demonstrated positive results for online learning in the areas of knowledge acquisition and retention, concluding that online education is at least equal to or superior to traditional educational methods for acquisition of knowledge and improving skill performance in nursing [11]. Various oral health training programs have also proved to be effective for primary healthcare workers [12-14], however this study was first of its kind to be conceptualized where an online educational resource has been used as a training material. The objective of this study is to analyze the current oral health situation analysis of the South East Asia region countries and to assess areas where the MOOC modules can be integrated as oral health training material.

Methodology

A Massive Open Online Course platform consisting of modules on basic oral health promotion targeted for the primary health care workers was developed by a team of faculties from interdisciplinary departments in Centre for Dental Education and Research, AIIMS New Delhi in 2019. A detailed content outline was drawn covering key topics in oral health promotion, preventive care and relevant interventions. A team of website developers were also approached to structure the module and

incorporate multimedia elements for engagement. A proper script and storyboard was developed for video lectures so as to ensure a cohesive and well-paced presentation of content. In order to promote this tool and enhance its usage, this study was conceptualised.

The participants were included in the study through nominations which were provided by WHO technical staff. Nominations were received from all SEAR countries except DRP Korea and Indonesia. The invitations were sent through mail and repeated reminders were also sent to the participants. The interviews were conducted through an online platform. A series of meetings were conducted throughout September, 2021. The online meetings were scheduled every Wednesday in September and were timed for approximately 2 hours. The study was conducted as an in-depth interview among 70 representatives from participating countries. Since the series of meetings were as a part of the Massive Open Online Course modules, the meetings started with basic knowledge on each module, followed by which the participants were divided into breakout rooms in the online platform for the focus group discussions. In total, four focus groups were created where an equal number of participants were enrolled. Representatives from each participating country were allocated to every group so as to maintain homogeneity. The discussions in the focus groups were regulated by a moderator and a reporter. The broad topics which were discussed are the following:

A) Oral health policy at the national level

- B) Barriers to dental treatment needs
- C) Action plan for MOOC modules

The topics were further probed through guiding questions by the moderator. Each breakout room session continued for approximately 30 minutes. All the sessions were video recorded with consent of the participants. The recordings were transcribed into verbatim format. Based on their responses from the participants, we created a SWOT (Strengths, Weaknesses, Opportunities, Threats) analysis of the current situation regarding the oral healthcare system of the SEAR countries. SWOT analysis was adopted as it is effectively used as a strategic analytical method, for systematically analyzing organizational environments.

Results

The SWOT analysis has been depicted in Table 1. Among all the SEAR countries Nepal and Thailand had an existing national oral health policy. However Nepal reported the data incorporated was not evidence based and the formed policy objectives were not practical as no clear definitive directives were present. An organized system consisting of strategic plans for oral cancer, oral health care for elderly are present along with Universal Health Coverage in Thailand. Whereas, Bangladesh, India and Sri Lanka are in the process of drafting a national oral health policy. The MOOC modules could be used as a training component and integrated into the national oral health policy, as suggested by the country representatives. The auxiliary dental workforce was reported to be present

in all SEAR countries. It was reported that Bhutan has introduced a Comprehensive School Health Program in all the districts whereby a group of health providers (general physician, dental surgeon, ENT technician and a lab technician) screen the school students and near-by communities. As per the Ministry of Health, the group is supposed to visit twice a year but due to budget constraints the visit is limited to just once in a year. Most of the dental workforce is composed of dental auxiliaries like dental hygienists who carry out oral prophylaxis, ART restorations, and dental extractions at remote places. Similarly, in India to bridge the gaps between urban and rural health disparities, there is inclusion of primary health care workers (ASHA, Anganwadi, ANM workers) but training them for oral healthcare services needs to be conducted through training programs, workshops. Bangladesh also reported presence of auxiliary healthcare workers but number of dental auxiliary workforce needs to be strengthened. Under this context, the modules were developed and intended to be used by them. Since the global school promoting initiatives are strong in Maldives, the school nurses also conduct oral health screening for the children and deemed to be an integral part of the healthcare workforce. Basic Package of Oral Care was implemented through auxiliary dental manpower in Nepal; however that led to malpractice in many areas. The weakness observed in Timor Leste was the low number of dental professionals therefore; there is an urgent need to strengthen the oral healthcare system through training of the medical workers for dental services as well.

Initiatives like National Oral Health Programs are required to provide infrastructure support and dental manpower in order to enrich the oral health services in a country. Out of all SEAR countries, only India, Sri Lanka and Thailand reported having specific programs for oral health services. This was an important area which needs focus for the rest of the South East Asian countries.

There are no National Oral Cancer screening programs in Bangladesh but screening of oral cancer is carried out by separate dental functional bodies, NGOs. Integration of oral cancer screening with NCDs has been proposed to the government. Similarly, no national oral cancer screening program is present in Nepal. However, integration with other cancer programs is done. Also, oral cancer screening is part of the PEN (Package of Essential non-communicable disease) package in Nepal. Since the PHC workers in any of the South East Asia Region countries are not trained in early detection for oral cancer, the modules can be used for the same since a major part of the MOOC modules focus on oral cancer detection. As Thailand reported, the National Cancer Control program is non-existing currently; however a pilot model has been initiated. Thailand feels an urgent need for early detection of oral cancer, since prevalence is on the higher side. It was also noted that among all SEAR countries, Thailand was the only country to have population based cancer registry. No countrywide oral cancer screening facilities are present in spite of the high tobacco usage in Bangladesh, Maldives, and Myanmar. In India and Bhutan, the National Cancer

Control Program exists where oral cancer screening programs are also integrated; therefore, nationwide coverage for oral cancer screening is present. However, India reports a need for opportunistic screening for oral cancer. The future National Oral Health Policy in SEAR countries reportedly has provisions to focus on early detection of oral cancer along with the need to strengthen the existing National Cancer Registry. The possible areas of integration have been tabulated in Table 2. Table 3 indicates the integration of MOOC modules in the action plan for oral health in South East Asia 2022 – 2030.

Table 1. Strength, Weakness, Opportunities, and Threats of present oral health situation analysis of South East Asia Region countries*

Country	Strengths	Weakness	Opportunities	Threats
Bangladesh	Oral health is included in certain NCD programs	Unaffordability of dental services	National Oral Health Policy in drafting stage	Symptom oriented view of utilizing dental services
Bhutan	Inclusion of dental hygienist to conduct OH procedures	No existing oral health policy	National Oral Health Survey is being conducted to get evidence based data	Betel nut chewing habit is culturally embedded leading to more cases of OPMDs and OCs
India	Various national oral health programs (NOHP) and integrations are existing	Uneven distribution of dental services	National Oral Health Policy is in the drafting stage	Perceived need related to oral health priority is lacking
Maldives	Global school health promoting initiatives are strong	Geographical barrier due to topography	The recently created Maldives Dental Council can initiate oral health promotional activities	Tobacco habits start at as early as 10 years of age
Myanmar	National Health Plan	Health Financing	Auxiliary dental workforce	Poor execution of anti tobacco law
Nepal	Existing National Oral health policy	Lack of evidence based data	Proportional dentist and auxiliary workforce	Poor health education information communication campaigns
Sri Lanka	Established referral and back referral system	Limited fund allocation towards oral health	Proposals for establishments of dental clinics at medical centres	Inadequacy in advocacy of oral health at all levels
Thailand	National Oral Health survey conducted every 5 years	Lack of integration of medical and dental services	Enhancing the public private partnerships for oral health programs	Unequal distribution of dental services
Timor Leste	Poor oral health awareness	Only 8 dentists for the entire population	Effort to train nurses, medical auxiliary workforce	Advocacy for poor health is poor

*Source: Information provided by the national oral health focal points attending the discussion

Table 2. Action plan for integration of the Massive Open Online Course modules

Country	Action plan for integrating MOOC modules
Bangladesh	<ul style="list-style-type: none"> ● Sub district dental surgeons, medical consultants, school teachers at district level should be targeted with these modules followed by monitoring and surveillance
Bhutan	<ul style="list-style-type: none"> ● Can be integrated into National Cancer Program ● It can also be used as a training Module for all health professionals.
India	<ul style="list-style-type: none"> ● Integration can be done with NCD programs, ongoing oral health programs. ● ASHA workers, Anganwadi workers would be the primary choice for implementing the modules ● Strengthening of school oral health programs through training of teachers through these modules
Maldives	<ul style="list-style-type: none"> ● Since, the school health programs are running successfully, implementation of the MOOC modules will work the best in that specific area. ● Oral health is a component in the school curriculum as part of their health system too, therefore these modules can be used to strengthen the system ● Even dental professionals can use these modules for oral health promotion
Myanmar	<ul style="list-style-type: none"> ● The modules can be integrated in tobacco control programs ● Translation into Burmese language required ● Modules will be more helpful in physical format rather than online link based
Nepal	<ul style="list-style-type: none"> ● The MOOC modules can be used in NCD programs, PHC workers. ● Sensitization through training, hands on workshop followed by implementation ● Training can be completed in 2-3 days ● Basic survey to see number of cases being reported pre and post the training of HCWs using the modules
Sri Lanka	<ul style="list-style-type: none"> ● Training of public health inspectors can be done using the modules. ● In school health programs ● Dental therapist posted in school health clinics ● Practical demonstration will be best
Thailand	<ul style="list-style-type: none"> ● As part of training programs for health professionals ● Certification programs, workshops, hands on training will be more beneficial in disseminating these modules.
Timor Leste	<ul style="list-style-type: none"> ● Health care workers constitute more in TL, therefore the modules can be used to sensitize them. ● Questionnaires along with practical demonstrations would be appropriate

Table 3. Integration of WHO Action Plan for oral health in South East Asia 2022 – 2030 with MOOC modules

Key strategic action points of Action Plan for oral health in SEA 2022 - 2030	Implementation areas of MOOC modules within the action plan for oral health in SEA 2022 - 2030
Oral health governance, leadership and resources	<ul style="list-style-type: none"> ● Increasing accessibility to MOOC modules across South east Asian countries with preference to their local language
Oral health promotion and oral disease prevention, life-course disease priorities and healthy settings	<ul style="list-style-type: none"> ● Improved awareness about oral health and capacity building among all the stakeholders including grass-root level workers within the scope of the module: <ul style="list-style-type: none"> ○ Common Oral Diseases and Life course approach in their prevention ○ Dental emergency management ○ Risk factors for oral cancer and management of oral potentially malignant disorders and oral cancer ○ Empowering the population with mouth self-examination and utilization of dental services
Oral health workforce for universal coverage for oral health	Provision for oral healthcare delivery services at grass-root levels like Primary Healthcare Centres
Essential oral health care and universal coverage for oral health	Applicability of MOOC modules to address the most common population needs, covering promotive, preventive, curative and rehabilitative services
Surveillance, monitoring and evaluation	Surveillance and monitoring of oral healthcare
Oral health research, digital innovation and emerging issues	needs of the community can be done through digital innovation aligning with the strategy

Discussion

The current global health crisis as a result of the COVID-19 pandemic has expressed the need, now more than ever, for interprofessional collaboration among health professionals and health workers [15]. Alternative modes of oral health education and training delivery, such as online and remote learning, are thus becoming more common, and programmes must plan ahead to ensure high-quality healthcare delivery. With the same view, the MOOC modules were conceptualized to empower the healthcare workers especially at the primary healthcare level to understand the basics of oral healthcare (anatomy of oral cavity, common oral diseases, prevention and management of oral cancer). The current study conducted an oral health situation analysis among South East Asian Region countries and from our findings it may be interpreted that oral health is still an under-explored issue in these developing countries. The lack of an oral health policy was observed in all countries except Nepal and Thailand, leading to low oral health literacy [16] and an unorganized oral healthcare system. In 2007, the WHO World Health Assembly passed Resolution (WHA60.17) on oral health, urging countries to implement public health initiatives for disease prevention and health promotion [17]. This study identifies that low-income nations reported fewer preventive activities than middle-income countries, and especially when compared to high-income countries, indicating a significant difference in preventive care delivery. Under the same context, the MOOC modules were made to train the primary healthcare workers in South

East Asia Region countries. A few alterations to the module like translation into regional language were suggested by the oral health focal points participating in the study. The oral health situation analysis identified various gaps in the oral health system of the SEAR countries mainly at the policy level and effective integration of oral health into primary health care. Through brainstorming it was suggested that guidance for the formulation of national oral health policy should be given by the nations with an existing national oral health policy (Nepal, Thailand) to the nations where the policy is at the drafting and conceptualizing stage.

The lack of sufficient dental manpower was also observed among Myanmar, Timor Leste, Maldives. Similar results have been reported in previous studies [18,19], therefore there is an urgency to initiate dental auxiliary training programs and eventually to make oral healthcare accessible at grass-root level. The use of the MOOC modules as training material was envisioned to act as a leverage point and promote public private partnerships which can dramatically bridge this barrier.

The oral health infrastructure was also reported to be lacking, and working conditions are inadequate in almost all SEAR member countries. This could be a significant impediment to providing proper oral health care to those who require it most. Harmonizing the oral health-care delivery system is another priority that came up during the discussion with the study participants.

In spite of existing anti-tobacco laws, poor vigilance and monitoring was reported by all member country participants. The mortality to incidence ratio of oral cancer is

reported to be highest in SouthEast Asia, globally [20]. As the practice of consuming tobacco was observed in all SEAR countries, it must be ensured that anti-tobacco laws be refined and regulated strictly. Taxation on both forms of tobacco, fine based laws were some of the suggested resolutions that were inferred.

The WHO-HQ brief on the implementation of global targets to monitor progress toward 2023 and frequent reporting systems, similar to those for NCDs, will help countries build and maintain momentum in the oral healthcare delivery system. The distance between the current system and the ideal system can be bridged through sufficient public demand, political will and leadership [21]. Based on the key objectives of the Action Plan for oral health in South East Asia 2022 – 2030 [22], the MOOC modules can be an excellent tool of implementation to achieve advocacy, community involvement, and oral health promotion at community level with focus on the under privileged level.

Conclusion

The comprehensive report on the current oral health situation among the SEA countries shows that there is a need to strengthen the oral healthcare delivery system in majority of the countries. The formulation and implementation of an efficient National Oral Health Policy involving all the stakeholders of the community is best suited for an equitable, affordable oral health system. The incorporation of the Massive Open Course modules for oral health promotion will aid the oral healthcare services at community

level and enrich the oral health related quality of life among the underserved population as well.

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Statements and Declarations

Conflicts of interest

The authors declares that they do not have conflict of interest.

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REVIEW ARTICLE

A Comprehensive Review of Polatuzumab vedotin: Mechanisms, Clinical Applications, and Future Prospects

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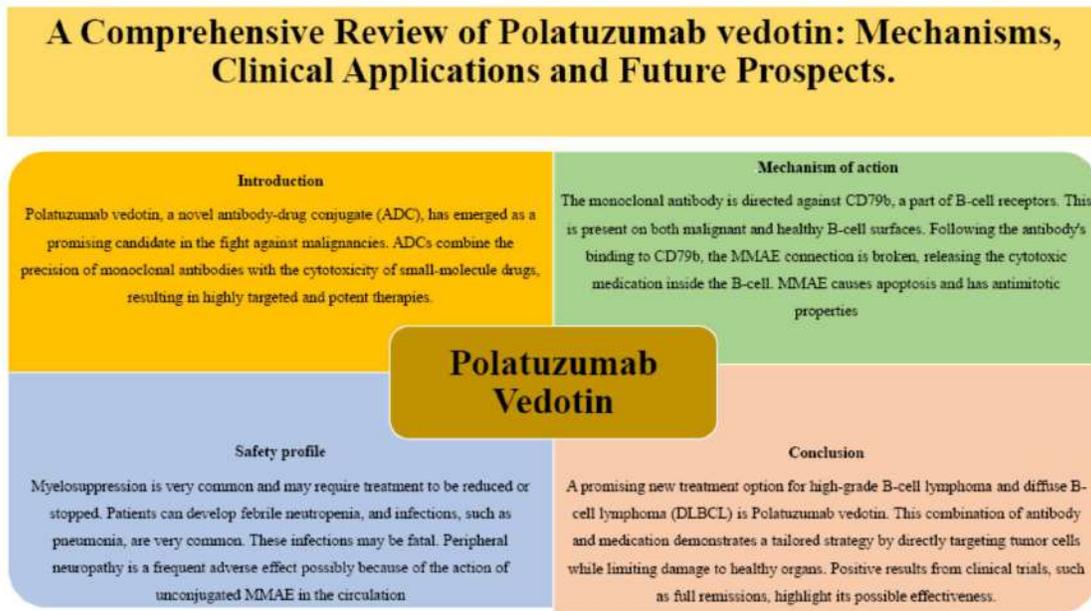
Abstract

A novel antibody-drug conjugate (ADC), Polatuzumab vedotin has emerged as a promising player in the oncology field. This comprehensive review aims to elucidate the intricate details of Polatuzumab vedotin, encompassing its molecular structure, mechanisms of action, clinical applications, therapeutic efficacy, and ongoing research endeavors. With a focus on precision and targeted therapy, Polatuzumab vedotin stands as a beacon of hope in the battle against various malignancies. It has been regarded as a significant advancement in the field of targeted treatment of diffuse large B-cell lymphoma. As research continues to expand, our understanding of Polatuzumab vedotin, it is evident that its potential to improve patient outcomes and reduce the burden of cancer will remain a topic of great interest for the medical community.

Keywords: Antibody-drug conjugate (ADC), Polatuzumab vedotin, Diffuse large B-cell lymphoma (DLBCL), Relapsed/refractory, CHOP

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Graphical Abstract



Introduction

Cancer remains a global health challenge, demanding innovative treatment strategies. Diffuse large B-cell lymphoma (DLBCL) is the most prevalent form of Non-Hodgkins lymphoma. Until 2000, the main chemotherapeutic agents used in the treatment of DLBCL and other aggressive NHL was cyclophosphamide, doxorubicin, vincristine, and prednisone every 21 days (CHOP-21); however, the treatment's cure rates was only 35%. Then, they could be given transplant consideration for stem cells [1].

Lymphoid malignancies treatment was certainly changed with the advent the first anti-CD20 monoclonal antibody, Rituximab. In 1997, rituximab was licensed for the treatment of follicular B-NHL; however, its use with CHOP-21 (Cyclophosphamide, Doxorubicin, Vincristine, and Prednisone) extended to include newly diagnosed DLBCL. More than twenty years later, rituximab (R) is still the gold standard for treating DLBCL; A significant number of patients

experience initial refractory illness or recurrence following R-CHOP treatment, even though there was initial high cure rates [2,3]. Role of prognostic biomarkers in the identification of high-risk cases has been highlighted in many studies. Polatuzumab vedotin, a novel antibody-drug conjugate (ADC), has emerged as a promising candidate in the fight against malignancies. ADCs combine the precision of monoclonal antibodies with the cytotoxicity of small-molecule drugs, resulting in highly targeted and potent therapies. This review examines Polatuzumab vedotin's pharmacological and therapeutic attributes, encompassing its mechanism of action, clinical applications, safety profile, and ongoing research efforts [4].

Mechanism of Action

Immunoconjugates also called as antibody-drug conjugates (ADCs) are made up of a monoclonal antibody and a cytotoxic drug, which is referred to as the payload (toxic at their therapeutic dose

levels in untargeted form are highly active molecules) connected by a chemical linker. The purpose of the ADC is to target cancer cells directly with an extremely toxic payload that is delivered selectively. Cytotoxicity and cell death occurs when the ADC/antigen combination binds to the appropriate antigen of tumor cell surface, after which internalization occurs and its payloads are released. At the moment, loncastuximab tesirine, brentuximab vedotin, polatuzumab vedotin, and inotuzumab ozogamicin are the four ADCs that are licensed for the treatment of lymphoid malignancies; numerous more compounds are being studied in clinical studies [5,6].

Polatuzumab vedotin (PolaV) is an ADC targeting, a component of the B-cell receptor, CD79b invariably expressed in B-NHL; A monoclonal antibody and monomethyl auristatin E (MMAE) which is cytotoxic combined to form Polatuzumab vedotin. Following the antibody's binding to CD79b, the MMAE connection is broken, releasing the cytotoxic medication inside the B-cell. MMAE causes apoptosis and has antimetabolic properties [7,8].

In the treatment of adult patients with refractory DLBCL who are not eligible for hematopoietic stem cell transplant (as a second line treatment) European Medicines Agency (EMA) approved PolaV in combination with Bendamustine and Rituximab (BR). FDA granted PolaV accelerated approval in June 2019 when used in conjunction with BR, for the treatment of refractory DLBCL after at least two previous therapies [9,10].

Polatuzumab vedotin's influence on the tumor immunological microenvironment is quite significant apart from its direct cytotoxic effects. Immune

microenvironment, a complex network of immune cells, cytokines, and signalling chemicals inhibit or stimulate the tumor growth. New research highlights the complex interplay between immunotherapeutic drugs' ability to modify this immune milieu and how well they work, such as Polatuzumab vedotin.

Studies have shown that the targeted administration of monomethyl auristatin E by Polatuzumab vedotin to B-cells expressing CD79b not only induces the demise of cancer cells but also may affect immunological responses. Polatuzumab vedotin may lessen the immune suppression brought on by tumors by eradicating malignant B-cells, and enhance the tumor antigens presentation to immune cells. This might then set off a series of immunological activation events, such as improved T-cell recruitment and recognition, which would ultimately strengthen and prolong the antitumor immune response [11].

Furthermore, there is strong evidence to support the conduct of clinical trials combining immune-stimulating antibodies or checkpoint inhibitors with Polatuzumab vedotin. These combos take use of the medication's ability to alter the immunological milieu, which may lead to increased response rates and extended illness control. Examining how immune cells and Polatuzumab vedotin interact in the context of tumors may provide important new insights into therapy approaches that use the immune system to improve patient outcomes [12].

Pharmacokinetics

The administration of Polatuzumab vedotin must be intravenous. The medication is diluted and infused after it has been reconstituted. Over ninety

minutes pass during the initial infusion. If this is well tolerated more infusions may be administered over a 30-minute period. Dose of Polatuzumab vedotin is 1.8mg/kg IV. The majority of MMAE in the bloodstream is coupled to an antibody. The half-life of this conjugated form is roughly 12 days.

Antibody degradation is anticipated to occur akin to that of other proteins, with the majority of the dosage likely to be excreted in the faeces. MMAE concentrations can be altered by cytochrome P450 3A4 inducers and inhibitors, because unconjugated MMAE is a substrate for cytochrome P450 3A4. Pharmacokinetics of Polatuzumab vedotin is not well-documented in liver and kidney disease patients [13].

Clinical Trials & Studies

Patients with Relapsed/Refractory (R/R) B-cell non-Hodgkin's lymphoma received intravenous Polatuzumab vedotin in cycles of 21 days as part of a Phase I open-label research (NCT01290549). 45 patients with evaluable NHL received 2.4 mg/kg Polatuzumab vedotin during the exacerbation or expansion phase. Among them, objective response (OR) occurred in 23, comprising 7 complete responses (CR) and 16 partial responses (RP).

Patients receiving 2.4 mg/kg of Polatuzumab vedotin experienced 6.2 months as median duration of response (DoR) and 5.7 months as median progression-free survival (PFS). Comparable results is also produced in a Japanese experiment (JO29138) [14].

The main study done in patients not eligible for transplants was an open-label, phase II trial involving 80 participants. The participants were divided into two groups (40 in each group).one

group received treatment with bendamustine, rituximab & Polatuzumab vedotin. Another group received bendamustine and rituximab. Significant improvements were seen in objective response rate (ORR-45% vs. 18%), complete response rate (CRR- 40% vs. 17.5%), overall survival (OS- median 12.4 vs. 4.7 months), and PFS (median 9.5 vs. 3.7 months) with the addition of PolaV. The Polatuzumab vedotin regimen was a 6 cycles infusion for every twenty one days. Using positron emission tomography, 16 patients showed improvement with Polatuzumab vedotin plus bendamustine and rituximab, while only 7 individuals showed improvement with bendamustine and rituximab.

The three-drug regimen's progression-free survival after a median follow-up of 22.3 months was 9.5 months, while bendamustine and rituximab's was 3.7 months. The chance of dying was decreased by adding Polatuzumab vedotin; the median overall survival was 12.4 months as opposed to 4.7 months when bendamustine and rituximab were used.

The greatest overall response (BOR) was 56.6% in the extension of this research, which involved 106 patients who took PolaV-BR; the ORR was 41.5% with 38.7% CRs. The median response time was 9.5 months, with corresponding PFS and OS times of 6.6 and 12.5 months. Lower PFS and median duration of response were found in a subgroup analysis of patients with primary refractory disease, those who were refractory to the last treatment, and those who had received more than one prior therapy [15].

In fact, PolaV-containing regimens were employed as a bridging therapy to allo-HSCT or CAR T-cell therapy in a recent trial by Liebers et al. The latter was

carried out effectively in 28 out of 41 patients, with an OS of 57.5% at 12 months and 77.9% at 6 months, respectively. By comparison, only 22% of patients who did not advance to CAR T had a 6-month OS. There is relatively limited data about the use of PolaV following CAR T-cell treatment. A recent retrospective analysis with 44 patients in relapse following CAR T showed a median PFS of 9 weeks and an ORR of 45% with only 14% CRs.

The first phase III experiment to use PolaV in the treatment of newly diagnosed DLBCL patients is called POLARIX (NCT03274492). In this trial, 440 patients were randomized to receive PolaV-R-CHP whereas 439 received standard R-CHOP. Every patient had six cycles of the prescribed regimen, which were then followed by two more cycles of rituximab monotherapy. Patients treated with PolaV-R-CHP showed a substantially longer PFS (76.7 vs. 70.2% at 2 years, $p: 0.02$) after a median follow-up of 28.2 months, but no change in OS was seen. Interestingly, despite the fact that the two groups' CR rates were similar, the statistically significant variations in Disease free survival (DFS) imply that PolaV-R-CHP produced more persistent responses. Patients over 60 years old, those with International Prognostic Index (IPI) scores ≥ 3 , and those with the activated B-cell-like subtype are those that may benefit more with PolaV-R-CHP [16].

Safety Profile

In terms of tolerability, PolaV-BR was linked to an increased incidence of grade 3–4 cytopenias, but not an increased risk of transfusion requirements or infections. Interestingly, low-grade,

reversible peripheral neuropathy was observed in the PolaV arm.

Bendamustine and rituximab become more hazardous when Polatuzumab vedotin is added because CD79b is not only exclusive to cancer cells. Because myelosuppression is so prevalent, medication may need to be lowered or discontinued. Pneumonia and other infections are prevalent, and patients may experience febrile neutropenia. These illnesses could be lethal.

One common side effect may be peripheral neuropathy, which could be brought on by unconjugated MMAE acting in the bloodstream. This may also be a reason to cut back on or discontinue treatment. When bendamustine and rituximab are combined with Polatuzumab vedotin, additional side effects that occur more frequently include fever, diarrhea, decreased appetite, hypokalaemia, hypoalbuminaemia, and hypocalcaemia. Hepatotoxicity is a possibility, so liver function should be examined in addition to the patient's blood count. An infusion-related response will occur in about one-third of individuals. Prior to each infusion, each patient should get an antihistamine and an antipyretic [17].

Challenges and Opportunities

Overcoming challenges such as resistance and optimizing dosing regimens are key to maximizing its therapeutic potential. Antibody-based treatments like ADC are thought to be intriguing and promising for enhancing cancer therapy. As others have said, the pharmaceutical industry's interest in funding research and development in the field is reflected in the growth in the number of registered ADCs in clinical trials.

Although an ADC's design may not appear extremely complicated, there are a number of factors that need to be taken into account in order to fully realize an ADC's potential as a cancer therapeutic agent. This could be the primary cause of the situation where just a small number of ADCs have made it to market. The main problems with ADC development appear to have their roots in the things that impede their effectiveness and cause off-target cytotoxicity.

It is not necessary for ADC tumor markers to be connected to tumor growth. As a result, ADC has a wide spectrum of therapeutic applications in malignancies. An ADC tumor marker must, however, satisfy three requirements: it must have a significant expression level in tumor cells relative to normal cells, it must have a cell surface immunogen, and it must be able to internalize ADCs [18].

The main considerations when selecting an antibody are receptor-mediated internalization, sufficient affinity, and high specificity. It would be a fantastic idea to optimize the antibody component to produce better ADCs. Indeed, antibody improvement can solve some of the main drawbacks of ADCs, such as limited internalization, low efficiency, and off-target effect (heterogeneous expression of the target in tumors, and target expression in normal tissues. Numerous preclinical models have demonstrated the efficacy of antibody engineering technology in producing alternative antibodies for the purpose of designing more efficient ADCs.

This technology is justified by the fact that the aforementioned ADC's shortcomings can be addressed by designing better ADCs that function as better antibodies in terms of internalization activity, affinity, and specificity, either by

increasing the therapeutic activity or reducing the adverse effects of the ADC [19].

The search for cytotoxic payloads with restricted drug-to-antibody ratio (DAR) 195 that are strong enough to perform therapeutic activity is a major concern in the development of ADCs. Cytotoxic payloads are often selected based on their stable storage and circulation properties, non-immunogenic nature, and acceptable aqueous solubility.

On the other hand, the second fascinating topic is the development of novel techniques to alter the cytotoxic payloads of ADCs with adaptable functional groups (such as amine or thiol groups), which facilitate conjugation. The inability of linking and conjugation chemistry to uniformly bind an optimal number of payloads to the antibody in a predetermined place presents another difficulty for ADCs.

The most promising methods for reaching the objective of site-specific conjugation and homogenous ADCs involve interdisciplinary and multidisciplinary works, as well as associated studies like recombinant DNA technology, bioconjugation, and chemistry. According to promising data from research to synthesize homogenous ADCs, it is possible that the first ADC products created via site-specific conjugation will be made for cancer therapy. This may hold the promise for the future employment of ADCs.

All things considered, despite difficulties with ADC design, the future of ADCs appears to be very bright since additional clinical trials and fundamental research on ADCs now in use will open the door to addressing problems with

tumor marker, antibody, cytotoxic payload, and linkage method [20].

Conclusion

A promising new treatment option for high-grade B-cell lymphoma and diffuse B-cell lymphoma (DLBCL) is Polatuzumab vedotin. This combination of antibody and medication demonstrates a tailored strategy by directly targeting tumor cells while limiting damage to healthy organs. Positive results from clinical trials, such as full remissions, highlight its possible effectiveness. However, the rise of side effects such as peripheral sensory neuropathy and neutropenia highlights the need for careful monitoring and all-encompassing management approaches. Polatuzumab vedotin presents a potential breakthrough by bringing hope back to those who have had setbacks with previous treatments. Nonetheless, thorough research is necessary to fully understand its therapeutic environment. With future research, this innovative treatment strategy might reveal further aspects of its efficacy, which would further refine its function in transforming the paradigms around the treatment of high-grade B-cell lymphomas.

Conflicts of interest

The authors declare that they do not have conflict of interest.

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CASE REPORT

Isolated Polycystic Disease of the Pancreas

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Abstract

Isolated polycystic disease of the pancreas is an extremely uncommon condition, with very few examples documented in the literature. Pancreatic cystic lesions are usually associated with other genetic disorders that involve other organs as well. However, isolated pancreatic polycystic disease is rare. We reported a symptomatic case of polycystic disease of the pancreas without evidence of cystic lesions in other organs.

Keywords: Pancreatic cysts, Pancreatic insufficiency, polycystic disease, polycystic pancreas

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Introduction

Polycystic disease of the pancreas is also known as dysontogenic cysts of the pancreas [2,3] It usually occurs with cysts in other organs such as the kidneys, liver, spleen, and central nervous system. 10 percent of cases of Polycystic disease of the kidney show cysts in the pancreas [1,3]. This suggests the embryologic connection with different cystic subtypes. Pancreatic cysts can be broadly classified as congenital cysts, developmental cysts, retention cysts duplication cysts, pseudocysts, neoplastic cysts, and parasitic (hydatid) cysts. True cysts of the pancreas occur as a developmental anomaly due to the sequestration of primitive pancreatic ducts [1].

Case Report

A 26-year-old female presented with a complaint of recurrent upper abdominal pain for five months. The pain was vague

and located in the epigastric region mainly. Her physical and systemic examination was unremarkable. The bowel sounds were normal. There was tenderness in the epigastric region with no guarding or rigidity. The patient's vitals were normal. Laboratory tests showed a white blood cell count of $10,102 / \text{mm}^3$, a hemoglobin level of 12.9g/dl , and a platelet count of $255,600 / \text{mm}^3$.

An ultrasound of the abdomen showed multiple variable-sized cystic lesions involving the head, body, and tail of the pancreas. There was no evidence of cystic lesions in the kidneys and liver as shown in the images. The possibility of Pancreatitis was ruled out as patients had no history of pancreatitis or blunt abdominal trauma. Serum Amylase and Serum Lipase were done and found within normal limits (Serum Amylase – 73 U/L and Serum Lipase – 88 U/L .) (Figures 1 to 3).



Figure 1. Ultrasound of upper abdomen showing multiple well-defined variable sized anechoic lesions involving almost all of the pancreatic head body and tail. Doppler study reveals absent color flow within the anechoic lesions suggestive of Multiple cystic lesions likely.



Figure 2. Ultrasound of spleen, right and left kidneys respectively showing normal parenchyma and no evidence of anechoic cystic lesions on either side.



Figure 3. Ultrasound of liver showing normal liver parenchyma with no evidence of focal or diffuse lesion seen.

Contrast enhanced computed tomography of abdomen was performed for further investigation and differential diagnosis. Plain images demonstrated variable sized well well-defined rounded fluid density (10-15 HU) lesions seen in the

head, body, and tail region of the pancreas. On arterial and venous phases, the lesions show mild peripheral wall enhancement as shown. The rest of the abdominal study was found unremarkable (Figure 4).

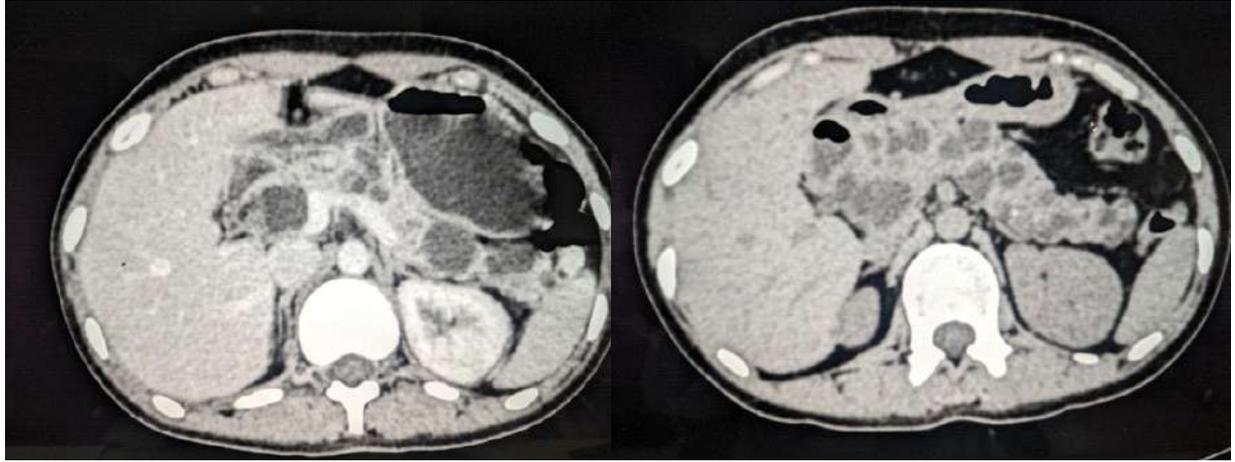


Figure 4. Contrast enhanced CT scan shows mild peripherally enhancing variable sized cystic lesions.

On Magnetic resonance imaging of the abdomen, the lesions appeared well-defined variable sized T2 hyper-intense lesions in the head body, and tail region of the pancreas. The lesion appeared hyper-

intense on T2 fat sat images. There was no dilatation of the pancreatic duct and no visible communication of the pancreatic duct with the cysts (Figure 5).

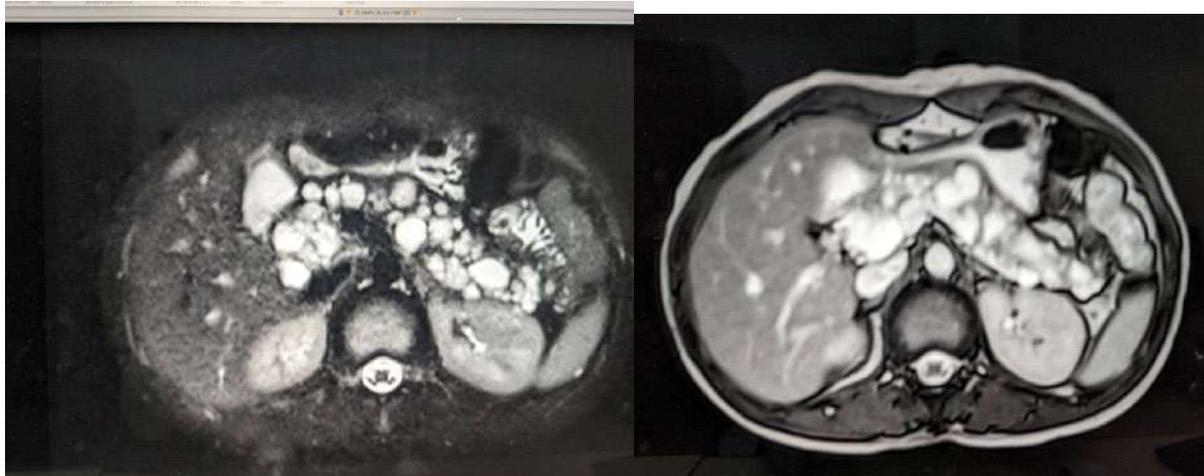


Figure 5. Magnetic resonance imaging of abdomen T2 weighted image and T2 fat sat images showing multiple well defined rounded variable sized thin-walled hyper intense lesions are seen in the head, body, and tail of the pancreas.

MRI brain was normal. Tumor markers (CA – 19-9 and CEA) were normal. A mutation of the VHL tumor suppressor gene on the short arm of chromosome 3 was

not found. So, Von Hippel – Lindau disease was ruled out.

Discussion

A vast and intricate network of ducts in the liver and pancreas carries bile and pancreatic secretions to the gut lumen. The epithelial cells that make up the ductal trees originate from the endoderm germ layer lining the primitive gut. Polycystic disease in the liver and pancreas can result from abnormal growth of these duct networks, such as ducts with expanded lumens and multiple cysts. Primitive duct sequestration brought on by developmental abnormalities results in the formation of true cysts [1].

True or congenital pancreatic cysts are distinguished from pseudo cysts by the presence of a true cuboidal or columnar epithelial lining on their inner surface. Thus, histological analysis is the basis for differentiating between actual cysts and other cystic lesions. Pancreatic enzymes are abundant in the cyst fluid, and pseudocysts frequently experience episodes of acute pancreatitis. Single or multiple congenital pancreatic cysts can occur. They could be associated with other systemic diseases or isolated. Multiple cysts are often associated with cystic disease of other organs. Autosomal dominant polycystic kidney disease, Von Hippel-Landau disease, cystic fibrosis, and Beckwith-Wiedemann syndrome are syndromes linked to numerous pancreatic cysts [1].

Although they seldom cause symptoms, congenital cysts can cause vomiting, distension in the abdomen, and vague abdominal pain. Pancreatitis or hepatitis. Particularly useful for differentiating between solid and cystic conditions is ultrasound. Color Doppler imaging can establish if the lesions are

vascular because cystic lesions do not exhibit color flow. At some point, characterization and localization of the cysts depend on cross-sectional imaging. The cystic nature of the lesions, as well as the interaction between the cyst and surrounding tissues and the expansions of the cysts, can be established by CT and MRI scans. But MRI does a better job of showing these things than a CT scan does. Simple cysts on MRI appear hyper-intense on T2 weighted images and hypo intense on T1 weighted images. Features of computed tomography include round or oval, homogenous, hypodense appearance, no discernible wall, internal septa, calcification, hemorrhage, or mural nodule, and an attenuation coefficient of 0–20 HU. Patients who have no symptoms can still be treated with follow-up and no interventions. However, a segmental pancreatectomy is required to remove the dominant cyst in symptomatic patients [4].

In conclusion, we reported a rare case of isolated polycystic pancreatic disease on magnetic resonance imaging, contrast-enhanced tomography, and ultrasonography. It is essential to exclude related syndromes before making a final diagnosis [4,5]. When diagnosing isolated polycystic pancreatic disease, MRI is significant.

Conflicts of interest

The authors declares that they do not have conflict of interest.

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CASE REPORT

Cryptorchidism with Duplicate Vas Deferens and Epididymis: A Rare Case with Literary Review

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Abstract

Background: Incidence and recognition of the congenital anomalies involving vas (or) ductus deferens, also called as sperm duct, are low, particularly when duplication of the sperm duct and epididymis occurs together. These anomalies, although of rare incidence, should be considered while performing surgeries related to the spermatic cord, to prevent inadvertent damage.

Case presentation: We, here, present a case of a seventeen year old boy with undescended testis on left side, where laparoscopic evaluation revealed polyorchidism. The accessory testis was removed, and histo-pathological examination confirmed duplication of the sperm duct and the epididymis.

Conclusion: This case scenario highlights the importance of recognizing and managing rare urogenital anomalies during procedures involving handling of spermatic cord structures.

Keywords: Cryptorchidism, polyorchidism, duplicate vas deferens, duplicate epididymis, vanishing testis

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Background

Cryptorchidism is not an uncommon presentation. But rare is polyorchidism with an estimate of less than 200 cases reported in the medical literature. Prevalence of sperm duct anomalies is quite less and estimated at less than 0.05 % of the general population. Varieties of anatomical variations were recognized in the sperm duct and epididymis but are quite rare [1]. Duplication is such a rarer entity. It is defined as the identification of a second sperm duct in the spermatic cord contents, and it should not be mixed-up with double vas deferens, an entity which describes ipsilateral agenesis of the kidney with a blind ureter that ends in ejaculatory system. It has been encountered in various surgical procedures involving spermatic cord, which include inguinal hernia management, orchidopexy, varicocelectomy, vasectomy and radical prostatectomy. We operated a case of undescended testis found to be polyorchidism following laparoscopic examination and inguinal exploration. Histo-pathological examination revealed the presence of duplication of the sperm duct and the epididymis with possible vanishing testis. Here, we present our case scenario with reference to the pre-existing literature.

Case Presentation

History and clinical findings: A seventeen year old male with otherwise no known comorbidities came to our out-patient clinic with the complaint of absent left testis. He had no genito-urinary or abdominal complaints. Surgical, medical, and family histories were non-contributory. He was a term baby delivered through normal vaginal delivery with uneventful perinatal period. On examination his right testis was present in

normal position. Left hemi-scrotum was empty, but well developed with good rugosities. Left testis could be identified as a bulge in the left inguinal region more towards deep ring with preserved testicular sensation. On duplex ultrasound examination, the position of left testis was confirmed and the size and vascularity were comparable with the right testis. Basic preoperative workup, which included baseline investigations, cardiac and anaesthesia consultation were done and found to be normal.

Therefore, we proceeded with laparoscopy followed by inguinal exploration and orchidopexy.

Operative findings: Laparoscopic examination was normal and no abdominal cryptorchidism or complete urogenital non-union was noted and fibrous adhesions were released around the cord and vessels till they entered deep ring to provide adequate length for scrotal fixation of testis. A solitary spermatic cord was noted entering the left deep inguinal ring. Then inguinal incision was given and layers dissected. Spermatic cord was identified; internal spermatic fascia was laid open. We could see two testes in the spermatic cord (Figure 1). Proximal testis, which was normal in size for the age, was connected to spermatic cord which contained testicular vessels whereas distal small testis had connection with a second spermatic cord which had only a sperm duct (Figures 2 and 3). We removed the distal testis along with its vas deferens because of its smaller size as compared to the proximal testis and absence of its to the blood vessels. The proximal testis was mobilised into scrotum and fixed in the sub-dartos pouch. Wounds were closed with absorbable sutures, procedure was uneventful and scars healed well (Figure 4).



Figure 1. Intra op –Duplication of left testis with normal looking proximal testis and abnormal distal accessory testis

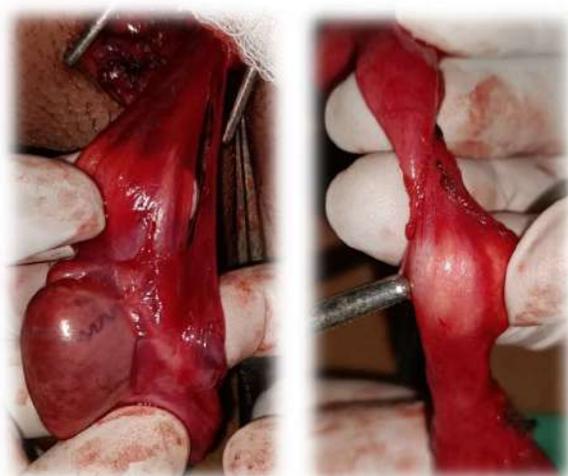


Figure 2. Intra op – normal proximal testis and its vas



Figure 3. Intra op – accessory distal testis and its vas



Figure 4. Post op day zero & after 6 weeks

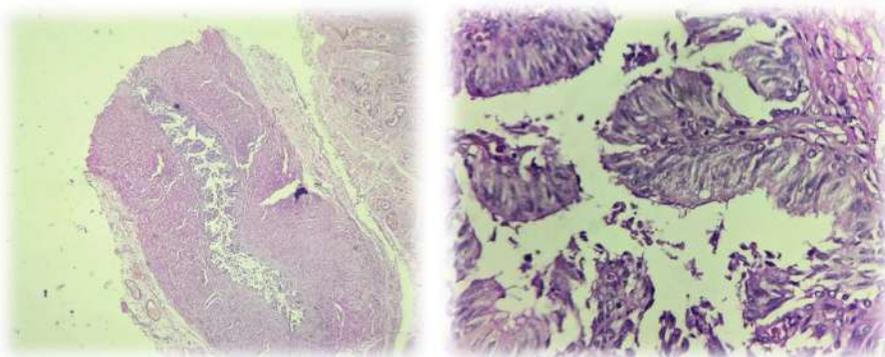


Figure 5: A - Vas deferens 40 x, B - Vas deferens 400 x

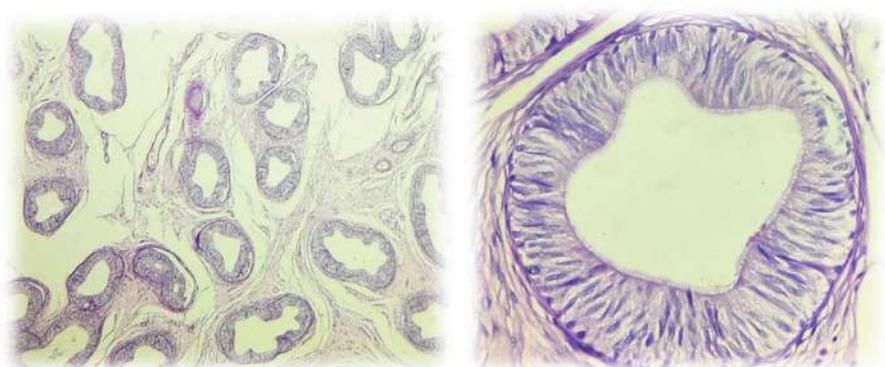


Figure 6: A - Epididymis 40 x, B - Epididymis 400 x

Histo-Pathological examination

findings: The resected distal testis showed only fibrous tissue was found and it had no seminiferous tubules, sertoli cells or sperm cells. But normal epididymal and vas deferens micro-anatomy was identified (Figures 5 and 6).

Discussion

The case presented here involved the presence of a duplicate sperm duct and epididymis presenting as polyorchid testis in a cryptorchid patient. The primary sperm duct in communication with the proximal testis was normal. This duct was seen arising directly from the epididymis, ran through the inguinal canal straight into the left deep inguinal ring. Distal supposedly “duplicate testis” had a sperm duct which was connected to the primary sperm duct at the deep inguinal ring proximally. This accessory testis was found to have only epididymis along the tunica vaginalis.

There is no exact explanation for the formation of polyorchidism, but various theories were proposed in the literature.

Leung, in 1988, described the variations in the anatomy (Table 1 and Figure 7) by a possible embryological origin. According to him, the type II variation is most common and types II and III clubbed together account for almost more than 90 % of cases of polyorchidism [2].

Singer et al., in 1992, suggested a different classification of polyorchidism which was based on both anatomy and function. [3].

- **I:** The supernumerary testes are attached to the epididymis and vas deferens with proper drainage and have reproductive potential (Similar to Leung Types II, III and IV).
- **II:** Testes with lack of such contiguity or attachment, hence they have no reproductive potential (Similar to Leung Type I).

Bergholz et al. presented an anatomical classification system of polyorchidism according to testicular reproductive function (Table 2 and Figure 8). Here anomalies of the sperm duct and the epididymis were grouped as anomalies in number, like absence or duplication, variation in location, like ectopia, abnormality in continuity, like segmental hypoplasia, and aberrations in integrity, like a diverticulum [1].

Liang et al. developed a classification system exclusively for poly-vasa deferentia [4].

- **I** - Duplicate vas deferens in the spermatic cord, but no polyorchidism.
- **II** - Multiple vas with presence of polyorchidism.
- **III** - False poly-vasa deferentia. Here an ectopic ureter is noted which drains into ejaculatory system.

Based on the above classification by Liang and his team, our patient belongs to either Type I poly-vasa deferentia or Type II poly-vasa deferentia with vanishing testis.

Table 1: The Leung classification of polyorchidism [2]

- **Type-I:** supernumerary testis lacks an epididymis or vas and has got no attachment to the usual testis. (Division of genital ridge only).
- **Type-II:** the supernumerary testis drains into epididymis of usual testis and they share a common vas. (Division of genital ridge occurs in the region where the primordial gonads are attached to the metanephric ducts, although the mesonephros and metanephric ducts are not divided, i.e.-incomplete division).
- **Type-III:** the supernumerary testis has its own epididymis and both epididymis of the ipsilateral testes draining into one vas. (Complete transverse division of mesonephros as well as genital ridge).
- **Type-IV:** complete duplication of testes, epididymis and vas. (Vertical division of genital ridge and mesonephros).

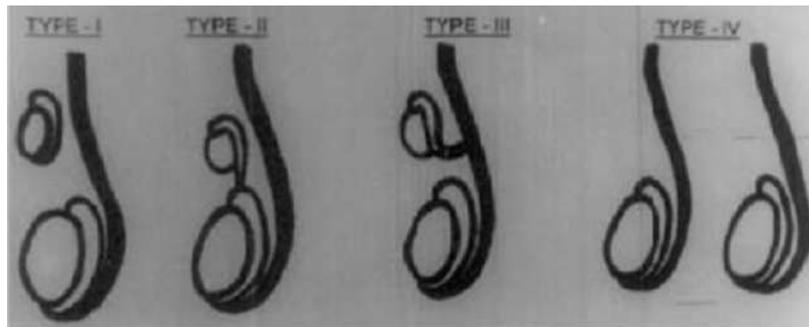


Figure 7. The Leung classification of polyorchidism [2]

Table 2: The Bergholz classification of polyorchidism [1]

Type A	Type B
Testis drained by outflow path (vas deferens)	Testis not drained by outflow path
A1 - Testis with its own epididymis and vas deferens	B1 - Undrained testis with its own epididymis
A2 - Testis with its own epididymis but shares a vas deferens with neighboring testis	B2 - Undrained testis with no epididymis
A3 - Testis shares a common epididymis and vas deferens with neighboring testes	
A4 - Testis has its own vas deferens but shares a common epididymis with neighboring testis	

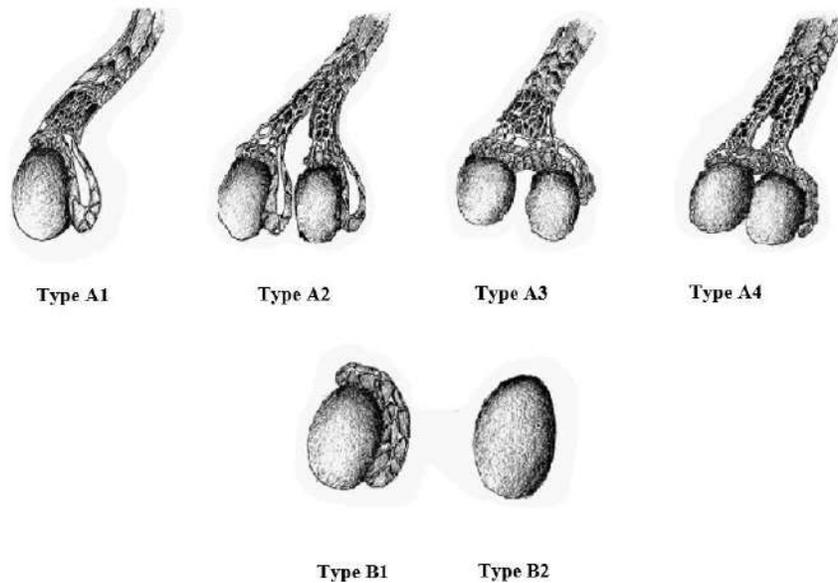


Figure 8. The Bergholz classification of polyorchidism [1]

Patients with cryptorchidism may have frequent association with anomalies of the vas deferens and epididymis. To our knowledge, there are very few reported cases of duplication of sperm duct and epididymis. One such case scenario is of an infant with left cryptorchidism who underwent left orchidopexy at an age of twelve months and intraoperatively they found two testes within the spermatic cord. In this child, the distal testis was connected to the testicular vessels along with a sperm duct whereas the proximal testis was communicating with only a sperm duct. The distal testis was preserved and orchidopexy was done. The other (proximal) testis was removed and sent for histo-pathology, which revealed only grouped tubes covered with a simple columnar epithelium. This was surrounded by spindle cells with no identifiable testicular tissue [5]. The other documented case is that of a four year old boy whose left testis was impalpable. He underwent laparoscopy which revealed, not only the sperm duct entering the internal inguinal

ring, but also a small intra-abdominal testis which was supplied by the testicular artery and vein. Inguinal canal, on exploration, revealed that the vas terminated in a nubbin of tissue. Histology showed epididymal structures in the areas both adjacent to the testis as well as in the terminal nubbin of the sperm duct. This case scenario is an example of the urogenital non-union. A blind-ending sperm duct found intra-operatively on exploration of inguinal canal might be considered as an evidence of vanishing testis syndrome [6].

We assume that the aetiology of our case is polyorchidism with left duplex testis accompanied by vanishing distal accessory testes. This is supported by the presence of well-developed rugosities of left hemi-scrotum which may be because of the distal testis which later atrophied. Urogenital non-union has an embryological explanation and it is frequently associated with a cryptorchid testis. Hence, we suggest that patients with complete urogenital non-union should be

evaluated further by diagnostic laparoscopy, to either identify the presence of an intra-abdominal testis or confirm total absence of the testis. Our patient did not have a complete urogenital non-union.

Conclusion

Here, we report one of the rarest cases – A patient with undescended testis found to be poly orchid during surgery but confirmed as duplication of vas and epididymis on histo-pathological examination. Surgeons must be aware of this abnormality while doing surgeries involving inguinal exploration or handling of spermatic cord in order to avoid any iatrogenic injury to cord contents.

Conflicts of interest

The authors declares that they do not have conflict of interest.

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