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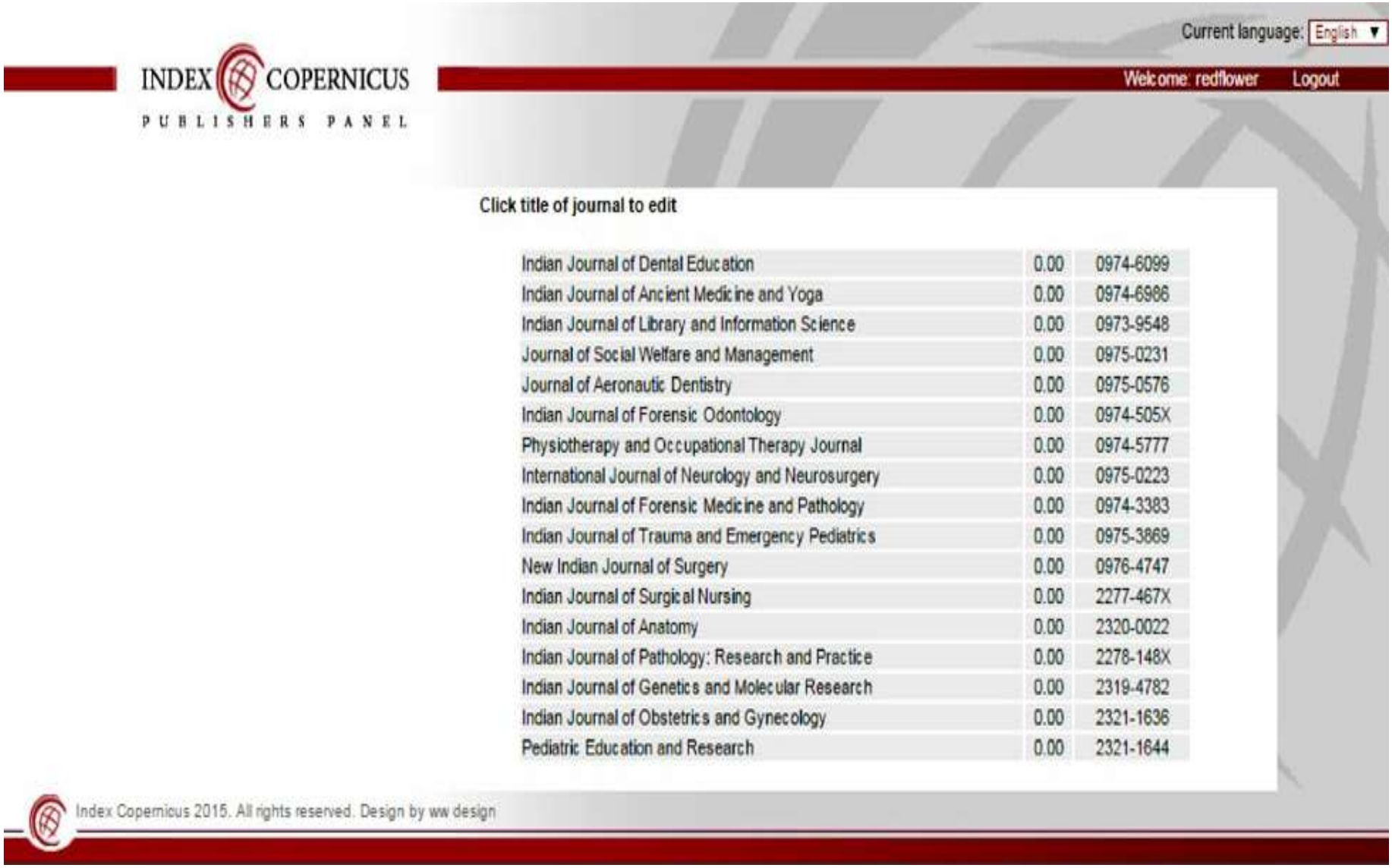

Review Article

Indian Journal of Trauma and Emergency Pediatrics
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Medical Education in India

Liza Bulsara*, Sunil Mhaske**

Abstract

The state of medical education in India presents a scenario marked by rhetoric and wishful thinking rather than concrete steps in right direction. The search for a need-based curriculum is not new. It has been felt for ages, but the curriculum has not really changed. It is an oft-repeated criticism that our medical colleges are producing graduates who are not well equipped to tackle the health care needs of the society. In India, a medical college is an educational institution that provides medical education. These institutions may vary from stand-alone colleges that train doctors to conglomerates that offer training related in all aspects of medical care. The Medical Council of India (MCI) is a statutory body with the responsibility of establishing and maintaining high standards of medical education and recognition of medical qualifications in India. It registers doctors to practice in India, in order to protect and promote the health and safety of the public by ensuring proper standards in the practice of medicine.

Keywords: Medical Education; Medical Council of India (MCI); Medical Training.

History of Medicine in India

Pre-Vedic period from 3000BC- Evidence of anatomic knowledge on cave paintings in Indus valley [1]. Vedic period (around 1500BC)- marked by writing of four Vedas, or sciences (describe the aspects of anatomy, medicinal herbs and plants). Post- Vedic period (800BC-1000AD)- golden age of Indian medicine (documented by writings of Charaka and Sushruta) [2] in form of Ayurveda and Siddha system. 13th century- Unani system of medicine was introduced by Muslim rulers. From 1810 Homeopathy gained foothold in India till the advent of British in 18th century. Western medicine-introduced by Portuguese in Goa (1840), started the Medicine and Pharmacy Licentiates (Goa Medical College). British established Madras Medical School in 1835. University affiliated medical education (1850s) - after opening of first 3 Indian universities in Chennai, Mumbai, and Kolkata.

- 1946- Bhore committee- Recommended major changes in medical education- 3 month's training in PSM to prepare "SOCIAL PHYSICIAN"
- 1975- Srivastava committee- Group on Medical Education and Support Manpower, Family and community oriented practitioner with social responsibility.
- Advocated and recommended reorientation of medical education according to national needs through a medical education commission on lines of UGC.

Medical Council of India

A governmental agency under the Ministry of Health and Family Welfare (1934). Indian Medical Council act 1956 is operational today. It was amended in 1956, 1964, 1993 & 2001.Statutory recommending body. It stipulates the rules for medical school curriculum, structure and content.

- 1977- Reorientation of Medical Education (ROME) scheme was launched to link community based facilities with medical colleges.
- 1983- National health policy provided directions

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to reconstruct the curriculum (train Undergraduate medical students as a Primary care physician).

- ❖ 1986- Bajaj committee report- emphasized on need of a Medical and Health Commission.
- ❖ In 1992- the National Institute of Health and Family Welfare carried out a study on the effectiveness of the training given in the graduate course on issues relating to Maternal and Child Health and Family Planning.
- ❖ In the same year MCI organized a National Workshop for debating a Need based Curriculum for UG Medical education
- ❖ 1997- GoI on recommendation of MCI promulgated the "Regulations on Graduate Medical Education" through a gazette notification.
- ❖ In 2011 the Board of Governors of MCI had announced a fresh set of curricular changes entitled 'Vision 2015' to re-look at the various aspects of medical education, training and practice for the country

Medical Colleges in India:

Total Number:

1947	1965	1980	1990	2006	2014
23	86	112	143	262	395

Private and Government Medical Colleges:

	1995	2006	2014
Private	47	131	214
Government	109	131	181

2014

Total MBBS seats- 49530

- Government Colleges- 181, Seats- 24610
- Private Colleges- 214 , Seats- 24920

MD/MS/Diploma

- Government Seats- 13913
- Private Seats- 8387

Reasons of Current Downhill

The medical education system in India may be on the verge of collapse.

1. An exploding number of medical colleges
2. A skewed distribution of these around the country
3. Devaluation of merit in admissions, particularly in private institutions; increasing capitation fees admission of suboptimal quality of students with poor motivation
4. An alarming shortage of teachers, with those who exist being untrained in modern teaching–learning technology.
5. Gross shortage of patients in many institutions.
6. A less than desirable evaluation system and poor internship supervision.
7. The curriculum is outdated, insensitive to modern concepts of the process of teaching–learning, rigid and discourages innovation.

Recommendations in Vision 2015

- Increasing seats of PG diplomas and degrees.
- Building competency based modules.
- Change of Nomenclature and introduction of M. Med Course - 2 year Master of Medicine (M. Med) program as the first level of specialists with focus on skill development and providing care to community.
- Career Pathways after M. Med.: Degree Course
  - i. A two year course on allied subjects
  - ii. A three year research path would lead to the MD/Ph.D degree on completion of requirements
- Fellowship programs
- Need based Assessment and Distribution of Diplomas and PG Courses.
- Training and Assessment- Extensive faculty development Training, Continuous formal structured assessment, a doctoral committee in every institute will constantly monitor the training of these students, Establishment of skill labs would be mandatory and Maintenance of log book.
- Rural Service criteria for entry in postgraduate courses.

Challenges in Medical Education

- Rapid and uneven growth of Medical school.
- Validity of student selection policies is questionable
- Curriculum goals are weakly focussed on health care needs with deficiency in internship year
- Lack of faculty development to meet the needs of

expanding number of medical schools.

Challenges to Dental Education in India

- The challenge of selecting students passionate enough to pursue dentistry.
- The challenge of bridging the theory - application gap:
- The challenge of having adequate number of competent faculty.
- The challenge of delivering oral care for the masses.
- The challenge of ensuring ethical dental practice in the country.
- The challenge of ensuring the quality of dental graduates practicing in the country.
- The challenge of suitable employment for graduate dental surgeon.

Reciprocal Selection of Students and Profession

- In India, medical students are selected on the basis of pre-medical tests (PMTs), held by each state for its residents. The tests, which take place once a year and are open to those who have graduated from high school, consist of multiple-choice questions (MCQs) covering physics, biology and chemistry.
- There is also a national examination which allows students from one state to apply for admission in another. About 85% of admissions are made on the basis of the tests administered by the states, while 15% are made on the basis of the 'all-India' entrance test.
- There are two issues involved here: selection of the profession by the students and selection of students by medical colleges.

Medical Training

- Teaching is based mainly on the traditional didactic pedagogic method and case discussions during clinical postings. The lack of coordinated teaching hampers learning to some extent.
- The inclusion of the relevant radiological and laboratory findings of the diseases under discussion could help to develop a systematic scheme by which the students could evaluate the results by themselves.
- Since students are exposed to these investigative modalities during their rotations through these specialties, they would be able to participate actively in the discussions making the learning experience more effective than one characterized by didactic lectures.
- Vertical integration is another method that is increasingly being introduced with classes on relevant pre- and Para- clinical topics during a clinical rotation being held independent of the clinical classes.
- Coordinated teaching can be introduced as early as the first year, with the same systems being covered under all three subjects during the same period—'horizontal integration'. Early community-based training for longitudinal clinical experiences.
- Community experiences contribute positively to the education, critical thinking and problem-solving skills of students. Value early clinical experiences, which help them acquire important clinical skills and knowledge.

Evaluation

- The focus of evaluation should also shift to the practical aspects.
- Students need to be assessed continuously for an evaluation of their interest and involvement in healthcare, and provided feedback regarding the same.
- The frequency of tests also needs to be reduced to the minimum to reduce stress and facilitate training.
- It would also be useful to replace the traditional long-/short-case examinations with more valid and reliable instruments for the assessment of clinical skills, such as objectively structured clinical examination (OSCE).
- Subjective assessment of knowledge by the faculty has been shown to correlate only weakly with objective performance
- It would be useful to adopt a system similar to the United States Medical License Examination (USMLE), which assesses the student's ability to apply his knowledge in three parts:

Step 1 for pre- and Para-clinical fundamentals

Step 2 for clinical knowledge and skills

Step 3 for comprehensive evaluation of case management ability.

- Systematic and reliable programmes for evaluation are a must to prevent curricular changes from drifting away from the intended path.
- It is imperative to revise the evaluation methods

to permit other changes to be made without increasing the stress on students. Evidence-based medicine should be emphasized throughout.

- The training. The aim should be to stimulate the student to independently explore and assess various modalities of investigation or management in terms of their relative merits or demerits.
- It is mandatory for students to have publications to their credit before they can apply for the final degree.
- Students should be encouraged to attend workshops and seminars as these help them remain up-to-date with the current developments and expose them to potential areas of research.
- Students should be provided incentives, including financial ones, to undertake research.
- Programmes such as the 'Short-term studentship' of the Indian Council of Medical Research are a step in this direction.
- Programmes in which a student can choose a preceptor or a mentor under whose supervision he can do clinical rotations.
- By choosing a preceptor, a student is able to obtain better exposure to various specialties of interest, which can later help her/him make informed decisions regarding specialization

Faculty Development and Accreditation

- Any reform should begin with the systematic development of medical educators, The MCI has recommended the establishment of medical education units/departments in all medical colleges.
- These units can be utilized for the development of the faculty and for the provision of learning resource material to teachers.
- The development of faculty for medical education in India has been supported by the Foundation for Advancement of International Medical Education and Research (FAIMER), USA. This is a non-profit foundation designed to support programmes and research that improve medical education and healthcare worldwide.
- The term 'accreditation' can be defined as a process by which a designated authority reviews and evaluates an educational institution, using a set of clearly defined criteria and procedures.
- Despite these variations, India has a well-established policy on quality assurance of medical education.
- Mandatory accreditation is conducted by the MCI. As an outcome of the recommendations of the National Policy in Education, an autonomous body called the National Assessment and Accreditation Council (NAAC) was established in 1994.
- Accreditation of medical colleges by the MCI is compulsory, but the requested information emphasizes documentation of infrastructure and human resources rather than measures of the quality of medical education and outcome.

Recommendations

- Uniformity of basic level of competencies across the country and uniform standard for Indian Medical Graduates.
- Integrated curriculum
- More attention on development of various skills, viz., problem solving skills, psychomotor or performance skills, attitudinal and communicational skills
- Changing the schedule of PG Entrance.
- Introduction of evaluation at the end of internship
- Rationalize the examination system of medical students- assessment should be criterion-referenced and based on the core curriculum.
- Adoption of practice of Evidence Based Medicine.

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Review Article

Indian Journal of Trauma and Emergency Pediatrics
Volume 7 Number 3-4, July - December 2015

Modification in Indian Medical Education Plan: Need of the Decade

Bhawna Sharma*, Sudhir Dubey, Neha Gaur*****

Abstract

It is the obligation of Indian Government to serve the rural poor. Modification in the Indian Medical Education system is the need of the decade in order to fulfill this obligation and bridge the national urban-rural gap in health care. The recent introduction and subsequent withdrawl of rural posting after the basic medical education course (MBBS) in India introduced a new controversy to the medical education system. A 7-year integrated MBBS-MD course-MBBS (4 year) + MD (3 years) may bring much needed respite to the students aimimg for post-graduate medical education within India and a clinical posting of 3-months duration (depending on the intern's choice/availability) during internship may facilitate the interns/fresh medical graduates to get a better exposure to their chosen speciality, medical problems, management and technology. Introducing 3 or 6 month rural posting(s) during/at end of the integrated curriculum might be welcome as it would facilitate learning about community and social problems in India and to get good exposure of the countries' medical problems. Adequate provision of newer technologies and diagnostics in rural setups through NRHM and state/central/NGO fund will benefit the rural population as well as the medical recruits in continuing medical education. The community work in our country may be further addressed through increase in rural posts and better service regulations, which may be achieved by setting up of additional Rural Health Service (RHS) Cadre or the Rural Medical Service (RMS) Cadre with additional benefits for those opting for it. Provision of optional/voluntary rural posting clause in the existing services, with additional service benefits, may be another option to strengthen the rural healthcare sector.

Keywords: Medical Education; Rural Medicine.

The number of poor people in India, according to country's Eleventh National Development Plan, amounts to more than 300 million. Almost one-third of the country's population continues to live below the poverty line, and a large proportion of poor people live in rural areas. Hence, it has been the obligation of subsequent Indian Governments to serve the rural poor.

The recent introduction and subsequent withdrawl of rural posting after the basic medical education course (MBBS) in India introduced a new controversy to the medical education system of our country. The medical students and the new doctors are striving for higher education and excellence and trying to reach new heights of advanced patient care, while a rural/community medicine posting system is feared to create an educational time lapse (of the duration of rural posting) which might slow the progress of the individual and the medical education system, along with introducing a gender bias, as the women doctors may not be able to undertake higher specializations due to social factors such as marriage and family life.

Introduction of a clinical posting of 3-months duration (depending on the intern's choice/availability) during internship may facilitate the interns/fresh medical graduates to get a better exposure to their chosen speciality, medical problems, management and technology. Alternatively, introducing an 7-year integrated MBBS-MD course-MBBS (4 year) + MD (3 years) may bring much needed respite to the students aimimg for post-graduate medical education within India and 3 or 6 month rural posting(s) during/at end of such curriculum

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might be welcome as it would facilitate learning about community and social problems in India and to get first hand exposure of the countries' medical problems. The new system might give first hand insight to doctors and inspire them to choose a specialization according to their perspective after their rural stint and some young minds may opt according to community needs. This may be supplemented by 2-3 years super-speciality fellowships, as followed in USA. This may reduce the total duration to 9-10 years to prepare a super-specialist for the country, compared to the present 12 years for MBBS+MD+DM/Mch.

The Bhore committee (1946) had recommended the 3-month training in social and preventive medicine to prepare social physicians for India and the Mudaliar committee (1962) recommended strengthening of district hospitals with Specialist services and setting up of All India Health Service [1]. But despite subsequent efforts achievement of national health goals has been a far-fetched dream. The integrated course may benefit the country and the medical system by provision of much needed specialist services in rural health.

Practical learning about recent advances in medicine and new technologies, can continue by way of adequate provision of newer technologies and diagnostics in rural setups through NRHM and state/ central/NGO fund. This will benefit the rural population as well as the medical recruits.

The integrated system may have some additional advantages too. The financially disadvantaged students might benefit by the introduction of the integrated system as they can expect a better renumeration sooner (ie after 4 years of entering the course). The number and amount of scholarships may be increased in order to help the needy and disadvantaged students.

Goverments ought to view doctors as professional, practitioner, scholar and scientist and the medical professionals should also live up to such high standards in order to derive full benefits from the Government/ regulating bodies [3].

The trend towards early specialization and higher studies has indeed helped the country attain high standard of medical care and expertise. The high standards of medical care, specialization and education along with good community work attained by the Armed Forces in our country is a good example.

The community work in our country may be further addressed through increase in rural posts and better service regulations, which may be achieved by setting up of additional Rural Health Service (RHS) Cadre or the Rural Medical Service (RMS) Cadre with additional benefits for those opting for it. Introduction of Rural Medical Service (RMS) Cadre with special service benefits is expected to provide better stability of workforce in rural areas [4]. How the Indian Medical System (or the proposed Indian Medical Service) absorbs, retains, trains, encourages and motivates the new recruits to keep their morale high will determine the eventual success of rural health care. The rural service period or internship rendered by the individual may continue to be counted as an advantage towards further studies or training. This will help the government retain the young doctors in service in rural areas. Provision of optional/ voluntary rural posting clause in the existing services, with additional service benefits, may be another option to strengthen the rural healthcare sector. The provision of better infrastructure in rural areas will also help in retaining rural cadre manpower. The Integrated Action Plan (IAP) for selected Tribal and Backward Districts under the BRGF programs and other rural development programs cover many districts and require further expansion to cover the entire country.

The proposed system of extended 3-month clinical posting along with the integration of the MBBS-MD course and the efficient implementation of the National Rural Health Mission (NRHM) shall help to strengthen the community and social service sector in the country, along with providing an excellent career opportunity for young medical post-graduates.

Several countries such as Nigeria, South Africa too have a one –year community work requirement, 2 year rural service in Thailand and 3 year rural service in Myanmar. Bolivia too requires 3-month rural internship similar to India. Canadian colleges look for student's volunteer work along with a graduate course prior to admission in medical course. United States colleges look for activities such as leadership roles prior to medical college admission. Some offer the medical course with two-year internship after a 5-year basic medical course such as the Hong Kong University, South Africa and Tunisia. Norway and Sweden (with six-months of family medicine training) have 18-months internship for medical students. New Zealand places patient care and supervision responsibility on 'Trainee Interns'. Medical students in Italy and Romania write and discuss a medicine related 'thesis' over a period of one–year before their registration as medical practitioners. Interestingly, Brazil offers a shorter (4-year) medical course supplemented by 2-year internship [2]. Australia has a "rural medical course" to cater to the rural areas and the graduates are subsequently given rural

postings after its completion. They take up rural medicine by choice [6,7,8]. Such graduates have a lower attrition rate [9,10].

To allay the fears of young medical students and graduates and prevent attrition, these changes should be implemented as a priority. Addition and strengthening of Rural Service cadre with options for temporary postings will provide diversity to the Indian Medical Service. Amalgamating the changes in internship programme, rural postings and the setting up of Rural Medical Service (RMS)/Rural Health Service (RHS) Cadre may help achieve an amicable solution to the current situation.

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Penile Strangulation by a Metallic Encircling Body in a Child: An Emergency

Pranam G. M.*, Usha Pranam*, G. A. Manjunath**

Abstract

Penile strangulation is a rare pediatric emergency among children. Its usually anaccidental event than intentional. We report a case of 4yr old boy with accidental insertion of metallic encircling body onto the penis as an act of play. As per Bhat et al classification it was a grade 2 and was managed by string technique with no complications.

Keywords: Penile Strangulation; Metallic Encircling Body; Management; String Technique.

Case Report

We report a case of penile strangulation in a 4yr old boy. The child was brought to the private OPD at 10pm in the night by the parents with complains of inserting a metallic screw on to the penis as an act of play (act of curiosity). On examination child had Injury to skin with Distal penile edema with decreased penile sensation, but no evidence of urethral injury (grade 2 of penile injury as per Bhat et al classification [3]). Foley's catheter was inserted and the screw was removed by the string technique with no significant damage to the penis.

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Discussion

Penile strangulation also called penile incarceration is very rare emergency in pediatrics. Penile strangulation injuries range from simple penile engorgement to ulceration, necrosis, urinary fistula, or even gangrene [1] . Strangulation injuries of the penis can occur by self-infliction or accidentally. Self-inflicted strangulation is usually the result of self-mutilation or sexual stimulation more seen in adults and adolescent age group; however accidental strangulation may also occur in children [2]. The most common cause of penile strangulation among children are hair tie next is rubber bands. These injuries are graded by Bhat et al as follows [3]:

- Grade I. Edema of distal penis. No evidence of skin ulceration or urethral injury.
- Grade II. Injury to skin and constriction of corpus spongiosum, but no evidence of urethral injury. Distal penile edema with decreased penile sensation.

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- Grade III. Injury to skin and urethra but no urethral fistula. Loss of distal penile sensations.
- Grade IV. Complete division of corpus spongiosum leading to urethral fistula and constriction of corpora cavernosa with loss of distal penile sensations.
- Grade V. Gangrene, necrosis, or complete amputation of distal penis.

Silberstein et al. [4], simplified grading system proposed by Bhat et al., by dividing it into two broad categories. Low grade injuries corresponds to Grade 1-3 injuries and most of the time requires no further intervention after removal of encircled object, while High grade injuries corresponds to Grade 4,5 injuries and require surgical intervention.This grading helps in management. The management mainly includes urinary retention treatment, removal of the cause. Foley's catheter is recommended for grade I and II while suprapubic aspiration in higher grades. Removal techniques are of four types string techniques with or withour aspiration of blood from glans, cutting techniques, aspiration techniques, and surgical techniques [5,3,6].

String Technique

Flatt [7] devised string technique for removal of rings from traumatized fingers while Bucy used it to first in 1968 to remove a metal ball bearing from an incarcerated penis [8]. Turgor needs to be relieved first following to it a string needs to be passed proximally below the ring. The string can be a thread, Intravenous drip [9], suture [10], etc. The proximal end of the suture is lifted so that the encircling object is gently gently over the edematous penis [11,12]. Some authors have chosen to aspirate blood from glans before starting or during string procedure. String technique is effective in grade 1-3 injuries [5].

Cutting Devices

Cutting methods are often the first method for dividing an encircling device that cannot be removed with sequential compression. Its mainly a surgical procedure. Grade 1-3 can be treated by this technique provided appropriate cutting tools are available [5].

Aspiration Techniques

Aspiration of congested blood from penis by using 18-gauge needle insertion into the corpora or glans. Multiple punctures of distal penis also aids in relieving the edema and thus aids easy removal. Its usually combined with other technique. This technique is useful in grade 2 and 3 injuries [5].

Surgery

Surgical technique by dorsal slit, removal of edematous prepuce skin or degloving with circumcoronal incision, retrieval of ring and subsequent approximation can be used in grade 2-3 injuries [1], concurrent or delayed skin grafting can be done if defect is large due to skin excision. Advanced grade injuries can be treated with wide tissue debridement of devitalized tissue and partial thickness cutaneous graft. Penile amputation with re-implantation using microsurgical technique for grade IV and V has been suggested [13]. In case of gangrene of penis partial or total amputation of penis can be done [1]. After care: closely examine the penis for vascular or tissue injury, which can be confirmed by colour Doppler if necessary. Heparinization if any injury is noted.

Complications are directly related to duration and grade of incarceration include [11,3,1]: urinary retention, urethral stricture, urethral fistula, skin ulcerations; decreased or loss of penile sensation, priapism, gangrene of penile skin and subcutaneous tissue, gangrene of penis. Complications noted in two patients of our series were skin necrosis and urethral fistula. Long term follow-up with micturating cysto-urethrogram and uroflowmetry if necessary.

Conclusion

Prompt recognition, urgent decompression of involved tissue is the key of penile strangulation thus preventing from morbidity.

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Case Report

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Acute Presentation of Hepatoblastoma in a 9 Years Old Girl

Balaji M. D.*, Shivaprakash N. C., Akkammal Sathyabama K.*****

Abstract

Hepatoblastoma is the most common primary malignancy of the liver encountered in children. Primary tumors of the liver account for approximately 1% of malignancies in children. 50-60% of hepatic tumors in children are malignant, with >65% of these malignancies being hepatoblastomas. Hepatoblastoma occurs predominantly in children <3 yr of age. We present a 9 years old girl, who presented to our emergency ward with Jaundice and abdominal distension since 3 months. Examination showed pallor, icterus, hepatomegaly and ascites. Laboratory investigations revealed hyperbilirubinemia(TB:16.6, DB: 9) with thrombocytopenia (80000 cells/cumm). USG abdomen showed a mass lesion in right lobe of liver with irregular and nodular. CECT abdomen revealed a mass lesion of size 6.8x 5.5 x 5.9 cm in segment of right lobe with cirrhosis and metastatic nodules in the liver with moderate ascites and right pleural effusion. Spleen was moderately enlarged measuring 14 cm with infarcts. USG guided liver biopsy was done to confirm the diagnosis which showed histology pattern suggestive of Small Cell Undifferentiated Variant of Hepatoblastoma. Even a short period of history of jaundice and abdominal mass should rise the suspicion of hepatoblastoma, which help in early diagnosis and treatment.

Keywords: Hepatoblastoma; Jaundice; Thrombocytopenia.

Introduction

Hepatoblastoma is the most common primary malignancy of the liver encountered in children [1]. Primary tumors of the liver account for approximately 1% of malignancies in children. 50-60% of hepatic tumors in children are malignant, with >65% of these malignancies being hepatoblastomas. Rare hepatic malignancies include embryonal sarcoma, angiosarcoma, malignant germ cell tumor, rhabdomyosarcoma of the liver, and undifferentiated sarcoma [2].

Hepatoblastoma occurs predominantly in children <3 yr of age. The etiology is unknown. Hepatoblastomas are associated with familial adenomatous polyposis. Etiology is thought to be alterations in the antigen-presenting cell (APC)/â-catenin pathway[2,3,4]. Low birthweight is associated with increased incidence of hepatoblastoma, with the risk increasing as birthweight decreases. Because of its rarity and inherent malignant nature, early diagnosis and treatment is difficult.

Case Report

9 years old girl, born to a 3rd degree consanguineous married couple, presented to our emergency ward with history of yellowish discoloration of eyes since 3 months, passing high colored urine since 3 months and progressive distension of abdomen since past 3 months. Child also presented with history of swelling of legs since 1 week with painful skin lesions, associated with itching. There was history of passing high colored urine. There was no history of passing clay colored stools, bleeding manifestations or convulsions. She had insignificant family history. For the above complaints child was treated by a quack with oral medications for 2 months.

On examination, vitals were stable and blood pressure was more than 95th centile. Anthropometry

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measurements revealed weight less than 10th centile and height between 25th to 50th centile. Pallor and icterus was present(Fig 1). Itchy, scaly skin lesions were seen in lower limbs up to knee (Fig 2). Pitting Pedal edema was present up to knee. In per abdomen examination, upper abdomen was distended with umbilicus pushed downwards and dilated veins were seen over abdomen(Fig 3). Shifting dullness was positive. Liver was palpable 3 cm below right costal margin and span was 12cm. liver was firm to hard in consistency There were no other signs of liver cell failure. Other systems were within normal limits.

Laboratory investigations are as follows: Hb: 10.6, TC: 9600, DC: N80 L19 E1, ESR: 30, platelets: 80000, total bilirubin: 16.6mg/dl, direct bilirubin : 9 mg/dl, SGOT- 330, SGPT: 171, ALP- 171, albumin- 2.4,RFT & Electrolytes: within normal limits, urine analysis: bile salt and bile pigments-present, HIV and Hepatitis B and C were negative. PT, APTT and INR were within normal limits. USG abdomen showed a mass lesion in right lobe of liver with irregular and nodular surface suggesting multiple heterogeneous nodules diffusely involving the liver. Further investigation with CECT abdomen revealed a mass lesion of size 6.8x 5.5 x 5.9 cm in segment of right lobe with cirrhosis and metastatic nodules in the liver with moderate ascites and right pleural effusion (Fig 4, 5). Spleen was moderately enlarged measuring 14 cm with infarcts (Fig 6).

USG guided liver biopsy was done to confirm the diagnosis. The microscopic examination of the liver biopsy showed undifferentiated cells with minimal cytoplasm and round chromatic nuclei with inconspicuous nucleoli. Histology also revealed discrete small nests associated with minimal embryonal elements suggestive of Small Cell Undifferentiated Variant of Hepatoblastoma (Fig 7).

Fig. 1: Eyes showing deep icterus

Fig. 2: Itchy scaly skin lesions with pedal edema

Fig. 3: Distended abdomen with umbilicus pushed down

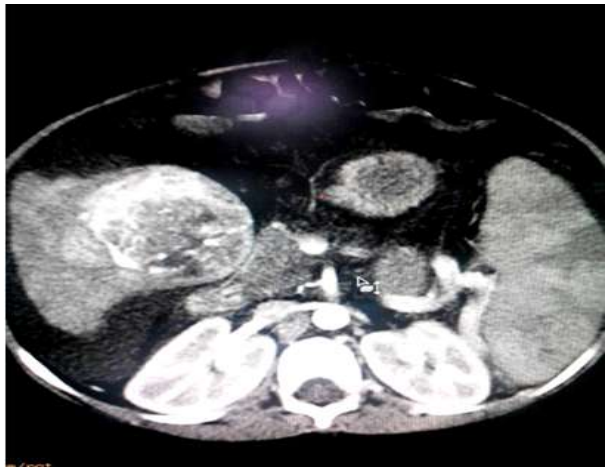

Fig. 4: CT scan showing mass lesion of size 6.8x 5.5 x 5.9 cm in segment of right lobe

Fig. 5: CT scan showing metastatic nodules in liver

Fig. 6: CT scan showing splenomegaly with infarcts

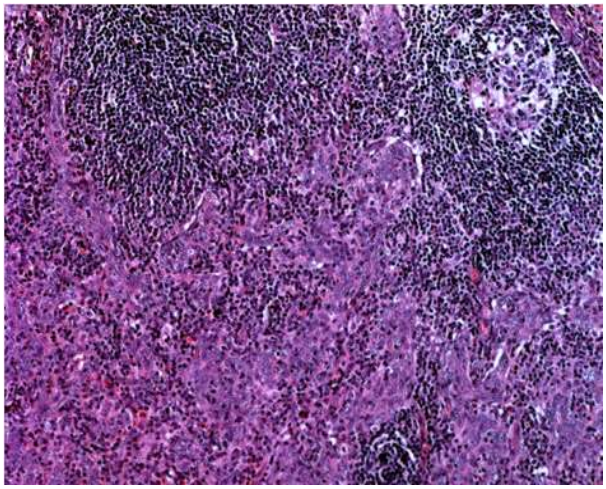

Fig. 7: Histology picture showing undifferentiated cells with minimal cytoplasm and round chromatic nuclei

Discussion

The most common primary malignant liver neoplasm in children is hepatoblastoma. Its incidence is approximately 1 to 2 in 1 million births. 2/3^{rds} of cases occur before 2 years of age and 90% of the cases are found below 5 years [5]. Males are affected two times more compared to females. Hepatoblastoma in adolescent and adults are diagnosed late when compared to children and heralds poor prognosis. Our patient was also diagnosed as hepatoblastoma at 9 years of age which was over a short period and associated with metastasis.

There are several environmental risk factors associated with hepatoblasoma. Premature and very low birth weight have been found to be associated with the later appearance of hepatoblastoma [2]. But our case did not have either of these risk factors.

A large number of congenital syndromes have been described in patients with hepatoblastoma, but only Edward's syndrome, familial adenomatous polyposis and Beckwith Wiede mann syndrome have been clearly shown to increase the risk of hepatoblastoma.

Most patients present with an enlarging abdominal mass. Our case presented with jaundice and distension of abdomen. The right lobe is involved three times more commonly than involvement seen in 20%-30%, and multicentric involvement in 15%. In our case also, lesion was seen in right lobe of the liver. Abdominal ultrasound is the first technique of choice as the initial diagnostic procedure for abdominal mass.

Magnetic resonance imaging (MRI) is the best modality for revealing morphologic details and to differentiate liver tumors. Computed tomography (CT) is better for detecting metastasis [6]. In our patient CECT was done which showed metastatic nodules in liver and infarcts in spleen.

Although thrombocytosis is commonly reported , low platelet counts can also occur in hepatoblastoma [6,7,8]. Our patient had thrombocytopenia with platelet count 80,000/cmm. Anemia is also described in previous studies [9].

Hepatoblastoma Mainly has 2 Variants

- The epithelial type, composed of small polygonal fetal cells or smaller embryonal cells forming acini, tubules, or papillary structures.
- The mixed epithelial and mesenchymal type, which contains foci of mesenchymal differentiation that may consist of primitive

mesenchyme, osteoid, cartilage, or striated muscle. The most common mesenchymal elements are osteoid and cartilage [5,10].

Epithelial hepatoblastoma is further broken down to pure fetal (31%), embryonal (19%), macrotrabecular (3%) and small-cell undifferentiated (3%). Our patient had small-cell undifferentiated type of hepatoblastoma which usually has bad prognosis. Our patient died within a week after the diagnosis was made. Death was due to hepatic encephalopathy. Our case is reported because of short duration history with acute presentation.

In general, the cure of malignant hepatic tumors in children depends on complete resection of the primary tumor. Even if 85% of the liver is resected hepatic regeneration is noted within 3-4 mo after surgery. Cisplatin in combination with vincristine and 5-fluorouracil or doxorubicin is effective treatment for hepatoblastoma and increases the chances of cure after complete surgical resection [2].

In low-stage tumors, survival rates >90% can be achieved with multimodal treatment, including surgery and adjuvant chemotherapy. With tumors unresectable at diagnosis, survival rates of approximately 60% can be obtained. Metastatic disease further reduces survival, but complete regression of disease often can be obtained with chemotherapy and surgical resection of the primary tumor and isolated pulmonary metastatic disease, resulting in survival rates of about 25%.

Liver transplant is a viable option for unresectable primary hepatic malignancies and results in good long-term survival [2,5].

Conclusion

Hepatoblastoma is the most common primary malignant tumour of liver seen in children and the main stay of treatment is complete surgical removal of the tumour mass which has a better prognosis when diagnosed early. Hence, even a short period of history of jaundice and abdominal mass should rise the suspicion of hepatoblastoma, which help in early diagnosis and treatment.

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Case Report

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A Rare Case of Meckel Gruber Syndrome

Liza Bulsara*, Sunil Mhaske, R. B. Kothari*****

Abstract

Meckel-Gruber syndrome, also known as 'Dysencephalia splanchnocystica', is a rare lethal autosomal recessive disorder consisting of central nervous system malformation- mainly posterior encephalocele (80%), multicystic kidneys (95%) and polydactyly (75%). Besides the classic triad of neural tube defects, polydactyly and cystic dysplasia of the kidneys, other abnormalities can occur in association with the syndrome, which may be detected sonographically include micrognathia, cardiac abnormalities, syndactyly, clinodactyly and clubbed foot. The worldwide incidence varies from 1 in 13,250 to 1 in 140,000 live births. Highest incidence was reported in Gujarati Indians.We report a case of a 26-year-old woman with previous LSCS referred from a private practitioner with abnormal ultrasonographic findings. She was diagnosed to have Meckel-Gruber syndrome. Woman and her husband were counseled regarding this lethal condition incompatible with life and after proper consent and information, pregnancy was terminated.

Keywords: Meckel-Gruber Syndrome; Encephalocele; Polydactyly; Cystic Kidneys.

Introduction

MKS was first described by Johann Friedrich Meckel in 1822 in two siblings who died of identical malformations of occipital encephalocele, polycystic kidneys, and polydactyly. George B Gruber, in 1934, reported many familial cases with similar features and coined the term "dysencephalia splanchnocystica [1, 2].

It is a lethal autosomal recessive disease with gene locus mapped to chromosome 17q21-q24 [2] . The MGS is characterized by triad of features having occipital encephalocele, cystic kidneys and polydactyl [3-8]. The incidence of this rare syndrome is 1 per 1 300 live births in Gujarati Indian families, 1 per 3 000 in Belgium, and 1 in 9 000 in Finland. The disease affects all races with males and females being equally affected. Once diagnosed, the chances of MKS in subsequent pregnancy are 1 in 4 (25%). The diagnostic criteria for MKS is presence of at least two of the three classic features like cystic renal dysplasia, occipital encephalocele, and polydactyly, which are observed in 100%, 90%, and 83.3%, respectively [1–3]. As observed in our cases, our case number 1 showed minute cyst in the kidney on microscopic examination, indicating the need of neonatal autopsy in syndrome diagnosiIt has wide phenotypic variations showing abnormalities of lip, palate, eye, ductal plates of the liver, cardiovascular and genital systems [3,5,7,8]. Other variants described include MKS2 in the Middle East and Northern African families, and recently MKS3. A high incidence of MKS has been reported in Gujarati Indians [3].

We report on a MGS diagnosed in the 2nd trimester antenatal ultra sonogram examination and subsequently confirmed in the abortus for its rarity

Case Report

A 26-year-old primigravida presented with 19 weeks of gestation for routine antenatal examination. There was history of second-degree consanguineous marriage. Past and family histories were noncontributory. She was not on any teratogenic drugs. Routine antenatal scan done showed features of anencephaly.

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Fig. 1,2,3,4: Anetenatal scan

Fig. 5: Ventral surface of fetus showing encephalocele

Fig. 6: Gross features of Polycystic kidney

Ultrasound scan, done at 15 weeks gestation revealed encephalocele. Detailed ultrasound scan was repeated at 24 weeks gestation which confirmed posterior encephalocele, bilateral cystic kidneys, talipes, and polydactyly. The possibility of MGS was raised, the natural course and the prognosis was explained to the parents who opted to terminate the pregnancy. Following further extensive genetic counseling and approval of the hospital ethics committee, termination of pregnancy was carried out at 25 weeks gestation. The baby was born alive but gasping and died in 15 minutes, no resuscitation was

offered.he birth weight was 1 kg, length was 32cm, and head circumference was 20cm all <3rd centile. Clinical examination revealed occipital encephalocele, microcephaly, cleft palate, post axial polydactyly, bilateral talipes, short limbs, and grossly enlarged abdomen with bilaterally palpable kidneys (Figures 1 and 2). The genitalia were undetermined with 1 cm phallus with a single opening, labioscrotal folds and no palpable inguinal mass. Chromosomal analysis of the baby revealed 46 XY karyotype. Further detailed DNA study was not available.

Discussion

Meckel-gruber syndrome (MGS) is otherwise called as Dysencephalia splanchnocystica [9]. It was first described byJohann Friedrick Meckel and it is a rare autosomal recessive lethal disorder.

The incidence worldwide has been reported as 1:13,250 to 1:140,000[10]. Finnish and Gujarati Indians show an increased incidence of this condition [11].

Meckel-gruber syndrome is characterized by occipital myeloencephalocele, bilateral renal cystic dysplasia, hepatic ductal proliferation, fibrosis and cysts, and polydactyly[11]. But the characteristic clinical triad consists of occipital encephalocele, polycystic kidneys and postaxial polydactyly. At least two of these features are essential for the diagnosis. Microcephaly, cleft palate and ambiguous genitalia may also be present in Meckel-gruber syndrome [13].

Meckel-gruber syndrome can be diagnosed prenatally by ultrasound findings at 11 to 14 weeks of gestational age. Alfa fetoprotein can also be measured in the maternal serum but it is not elevated when the encephalocele contain a closed sac [14].

It can occur following artificial insemination like in vitro fertilization. Celentano et al reported a case of MGS diagnosed at 17 weeks in a pregnancy obtained with intracytoplasmic sperm injection (ICSI) [15]. Three genes (MKS1, MKS2 and MKS3) have been identified. MKS1 located on chromosome 17q, MKS2 is on chromosome 11q and MKS3 is on chromosome 8q or 13 q. Locus heterogeneity is a feature of Meckel-gruber syndrome since the presence of phenotype variability [16].

This case report highlights the importance of antenatal scan in diagnosis of inherited diseases such as MGS. This helps physicians and parents to plan further management of pregnancy. Following extensive genetic counseling the parents opted to terminate this pregnancy. Termination of pregnancy raises complex ethical, legal, social and religious issues. As MGS is not compatible with life, the opinion of parents was adhered to. In cases of unborn babies affected by conditions leading to developmental delay and other morbidities which are not lethal, termination of pregnancy is generally not indicated.

Recently, primary prevention of genetic diseases became a reality by introduction of preimplantation genetic diagnosis (PGD) [15- 16]. In order to help families to make use of PGD, physicians need to confirm the diagnosis of the genetic disease at the DNA level in the index case. We were unable to perform mutation analysis for our patient because of lack of facilities. It was a missed opportunity to prevent possible recurrence of MGD for the third time in this unfortunate family.

Conclusion

In Meckel Gruber syndrome most infants are still born or die within few hours or days after birth. Review of literature reveals only ten neonates surviving beyond birth.

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