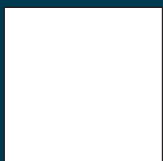




• **Editor & Publisher**

Dr. K. Madeswaran

Chairman - Consultant Neuro & Spine Surgeon



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CHAIRMAN'S COLUMN



New year wishes to all!

Hope the new year 2020 brings all the happiness and good health to everyone. A new year means new beginnings and fresh starts. We at Royallcare are constantly upgrading ourselves with new age cutting edge tools for better diagnostic services to the needy patients. Installation of advanced dual head SPECT scanner Discovery NM series is complete and open for commercial operations.

Consecration ceremony of newly constructed temple complex within the Royal Care Hospital premises is going to happen on 5th Feb'2020.

Hopefully we will be able to start the postgraduate DNB programs in medicine, surgery and radiology in this new year.

Health is of paramount importance, but many people do not make healthy food habits and exercising a priority until their health deteriorates. You must live in a balanced state so that you can be at your optimal level in all aspects of your life.

"The greatest gift you can give your family and the world is a healthy you"

Regards

Dr. K. Madeswaran

Founder Chairman

From The Editor's Pen..



"Watch your thoughts, they become words;
Watch your words, they become actions;
Watch your actions, they become habits;
Watch your habits, they become character;
Watch your character for it becomes your destiny"..

- Frank Outlaw

The royal care family walks into its fourth year of successful functioning and is constantly growing in stature in the hearts and minds of our clients across the land. Soon we hope to be able to expand to a magnitude where we reach out to the most needy public everywhere. The "Royal Fest 2019" was celebrated by all our staff and doctors with great jubilation and was an evening to remember.

We congratulate **Dr. Sandip Chandrasekar** for his international journal publication and also other consultants who presented in international & national conferences as invited faculty. Various camps and health awareness campaigns were conducted by our hospital at various locations.

The SPECT scan machine has been commissioned and shall start functioning from the end of January 2020 which is one of the steps to ensure that Royal care shall become an one stop destination hospital for all ailments soon. The state of the art PMR department with hydrotherapy and Hyper baric oxygen therapy is also being constructed here, which shall be one of the first facilities in this region.

In this issue, we have articles on OBG, a published journal and other interesting patient cases. Some unique cases are also showcased in this edition. We welcome the new consultants who have joined Royal care Hospital and wish them success in their endeavours..

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HYPOTHENAR HAMMER SYNDROME : NOT AN UNCOMMON CAUSE FOR DIGITAL ISCHEMIA.



Dr. C. Senthil Kumar
M.S, MRCS, MCH.,
Consultant Cosmetic Surgeon



Dr. B. Madan Mohan
MD, PDCC, FINR.,
Consultant Neuro Interventional
Radiologist

There are plethora of causes for an upper extremity digital ischemia such as Raynaud's disease, Raynaud's phenomenon associated with connective tissue disorders, vasculitis, arterial emboli from a cardiac source, thromboangiitis obliterans, atherosclerosis with secondary thrombosis and thoracic outlet syndrome. To this list there is also work-related and less common cause of digital ischemia, hypthenar hammer syndrome.

Typically, hypthenar hammer syndrome occurs in men with a mean age of 40 years involving the dominant hand and in occupational settings where the worker uses the hypthenar portion of the hand as a tool to hammer, push or squeeze hard objects. Nevertheless, Indian women with household work using the hand will also come in the list.

Although generally described as a rare condition, Hypthenar hammer syndrome may occur more frequently than acknowledged. Here we report such a case with a brief review.

Case study

A 51 years anxious female with a right handed dominance, doing routine household work, came to us with pain in the 3rd, 4th and 5th finger for 15 days. The pain was insidious and intermittent to begin with. Now it became more constant and severe. It was aggravated by working and reduced by dropping the hand down. She gave no history of trauma to the hand. Sensation over the hand was normal. She had no symptoms pointing towards cardiac issues.

Clinical Examination revealed discolouration of the ulnar three fingers, more so distally. However, the capillary fill was normal.

While doing the Allen test the pain in the fingers was aggravated.

X ray of the hand was within normal limits. Joints and movements were adequate. Sensory evaluation was fine.

Angiography

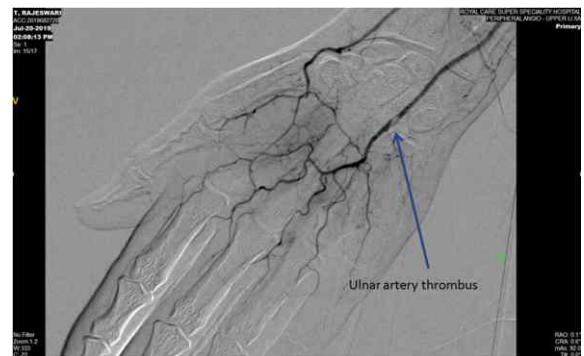


Fig : 1

Post thrombectomy



Fig : 2



Fig : 3



Fig : 4

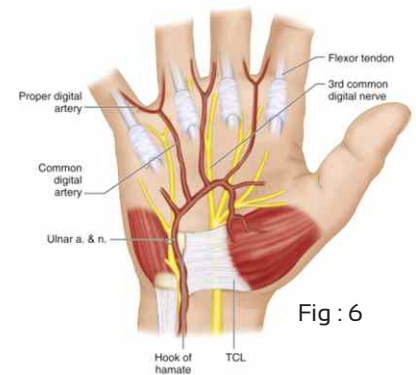


Fig : 6

Evaluation

We were working out in the line of Raynaud's or embolic disease. Hence the necessary investigations were undertaken. Xray revealed no bony lesions.

Non-invasive vascular examination, Doppler localised a circulatory compromise at a distal level. Detailed bilateral upper-extremity and hand angiography was done, as it is the "gold standard" in such situations. Angiographic findings revealed a subtle area of fusiform ectasia with intraluminal thrombus. (Fig 1 and 2) Changes were noted in the arterial segment related to the hook of the hamate. Other possible angiographic findings (not seen in this case) alternating areas of stenosis and ectasia (corkscrew) or Occlusion of the ulnar artery at the level of hamate.

Further, findings of a digital artery occlusion was seen in the 2nd (Ulnar aspect), 3rd and 4th digit in this patient, which is a critical finding to differentiate it from the Raynauds's disease. In Raynaud's all the 5 fingers are involved, but in hypothenar hammer syndrome thumb is usually spared.

Anatomy / Pathogenesis

The pathogenesis of the hypothenar hammer syndrome is related to the anatomy of the ulnar artery as it enters the palm. At Guyon's canal, the ulnar artery branches off to the deep palmar arch and exits the canal to form the superficial palmar arch. This superficial branch of the ulnar artery crosses the surface of the hypothenar muscles for about 2 cm before penetrating deeper. This arterial segment is very susceptible to injury. (Fig 6)

With palmar trauma, this vascular segment can be compressed against the adjacent bony hook of the hamate (also known as the hamulus).

With repetitive blunt trauma, the superficial palmar branch of the ulnar artery can develop intimal damage producing vasospasm and encouraging platelet aggregation and thrombus formation. If the damage spreads through the media into the arterial wall, aneurysm formation can occur as well. Microemboli can be generated that can occlude the digital arteries downstream, resulting in digital ischemia. Some studies finds that there are preexisting fibromuscular dysplasia of the palmar ulnar artery that Pre - exists in these patients.

Neurologic symptoms such as paresthasias and pain may also result from compression of the sensory branches of the ulnar nerve which run in close proximity to the ulnar artery.

Treatment

Optimal therapy for HHS has not been determined so far. Surgery is indicated for occluded palmar ulnar arteries in the presence of significant ischemic symptoms. In our case, we did excision of the diseased segment and vein grafted the (Fig 3 and 4) segment. In one way, this removes the occluded segment and in other, it prevents further micro embolic process.

Other published interventions are observation and risk-factor management, palmar ulnar artery ligation, cervical sympathectomy, and thrombolysis.



Fig : 7

REDO - AORTIC VALVE REPLACEMENT WITH ROOT ENLARGEMENT – A CASE REPORT



Dr. S. Krishna Kishor
MS, DNB(CTVS),
Consultant Cardiothoracic Surgeon

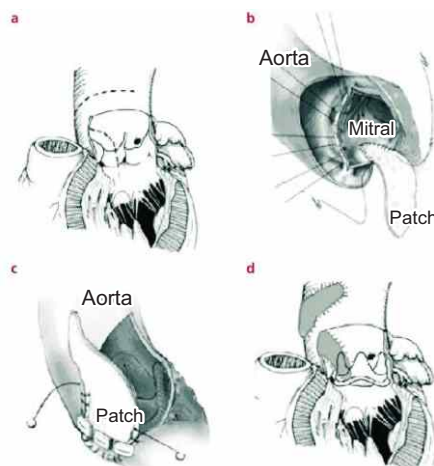
Introduction :

Bicuspid aortic valve is the commonest morphological anomaly seen in aortic valve disease with an incidence of 2%. There is a male predominance with 3% developing stenotic lesions, however women are not spared.

Case :

A young lady in the mid thirties presented with history of fever and shortness of breath since a fortnight. She had undergone Aortic valve replacement a few years ago with a mechanical prosthesis. At the outpatient clinic an echocardiogram was done which revealed a suspicious vegetation on the ventricular surface of the mechanical prosthesis. The peak and mean gradients across aortic valve were 90/40. A working diagnosis of prosthetic valve endocarditis was made.

Blood cultures were sterile for bacteria and fungi. She had received a 17 mm tilting disc prosthesis earlier, upon calculation for the current BSA of 2.04 she needed an aortic prosthesis of 21mm and above to prevent mismatch. Following counselling she was taken up for Redo aortic valve replacement with aortic root enlargement after a fortnight following medical optimization.



With initiation of femoral cardiopulmonary bypass, sternotomy was done using an oscillating saw and aorta was accessed after dissecting the adhesions. Using standard myocardial preservation techniques the heart was arrested and the existing prosthesis was explanted and sent for culture.

The annulus was sized to 17mm. In view of a narrow annulus Nick's root enlargement was planned. The right lateral end of aortotomy incision was extended into the non coronary sinus and over the annulus stopping short of the mitral valve. A diamond shaped prosthetic patch was used to augment the annulus, and a 21mm bioprosthesis was implanted.

Aorta was closed and she was weaned off cardiopulmonary bypass. She was extubated on day 1 in the postop ICU and was discharged on day 5. Post op echo revealed a gradient of 12 mm across the valve. On first follow-up she is in Class I.

NYHA Redo surgeries are technically challenging and carries a 5% risk of perioperative mortality. At Royal care redo cardiac surgeries are being performed on a regular basis with exemplary results.

"From every wound there is a scar.
And every scar tells a story.
A story that says, I survived."

- Craig Scott

LAPAROSCOPIC MANAGEMENT OF HUGE PRIMARY PARASITIC MYOMA



Dr. S. Kalyana Kumari

MD(OG), Dip.Gyn.Endoscopy(Ger), MBA,

Consultant Gynaecologist & Laparoscopic Surgeon

Introduction:

Parasitic leiomyomas (PL) are rare intra-abdominal tumours found in woman of reproductive age group, and they are of two types, primary and secondary.

Primary parasitic leiomyomas are usually pedunculated subserous or broad ligament myomas. They enlarge and get attached to adjacent visceral structures or omentum and derive their blood supply from these structures. Over the time these myomas may detach from their site of origin. According to the literature secondary parasitic myomas following laparoscopic power morcellation are common.

Due to their huge size and extensive vascularity, parasitic myomas are usually managed by open surgical approach.

Here we report a case of huge parasitic myoma managed by laparoscopic approach in a young infertile patient.

Case Report:

A 35 year old female on evaluation for infertility diagnosed to have huge pedunculated myoma. She presented with fullness and discomfort in the lower abdomen for 5 months period. There was no history of previous surgeries.

On per abdomen and per vaginal examination there was a huge mass occupying entire lower abdomen measuring about 28 weeks size. Uterus could not be made out separately from the mass lesion.

USG revealed large pedunculated subserous myoma 10.1x16.2x18.3cm arising from the fundus extending cranially upto supra umbilical region with few small intramural myoma. MRI was done to rule out sarcomatous change. She was posted for laparoscopic myomectomy after counselling.

As there was limited space available for port placement 3mm camera port is placed at palmer's point after creating pneumoperitoneum with veres needle. Under the guidance of thin 3mm lens other ports were placed safely. On entering, transverse colon along with mesocolon was found to be adherent to the anterior abdominal wall as the colonic loop was displaced upwards by the huge myoma.

On releasing the adhesions huge 25x20x15cm myoma with multiple dilated tortuous blood vessels on the surface found to be attached to anterior abdominal wall and omentum. Myoma was supplied by huge vessels from these structures. These vessels were coagulated meticulously and the myoma was detached from the adjacent vital structures.

The origin of the myoma was traced to the right cornual region where again multiple huge vessels supplying the myoma noted. Myoma was dissected from myoma bed in the cornual region with extreme care.

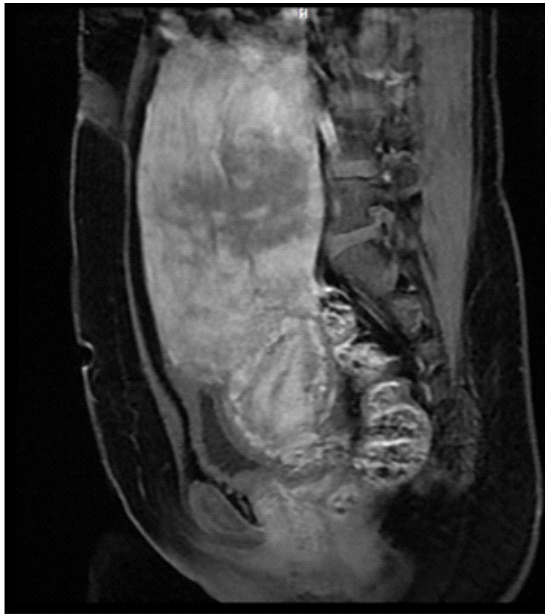
Specimen was retrieved by morcellation in endobag. Weight of the myoma was 2.2kg. Huge size along with extensive vascularity made each step starting from port placement to retrieval of specimen very challenging.

HPE report - Tumour composes of proliferated smooth muscle consistent with leiomyoma.

Discussion :

Parasitic leiomyoma is regarded as a subtype of subserosal myoma. The term parasitic myoma was coined by Kelly and Cullen in 1909.

When a subserous myoma outgrows its blood supply from uterus it acquires its blood supply from adjacent structures like bowel, omentum, broad ligament, peritoneum, common iliac artery



MRI showing 10*16*18CM sub serous myoma arising from fundus of the uterus

and inferior mesenteric artery. Though most of these myomas detach from the uterus over the course of time, some myomas may retain their attachment to their origin.

Most parasitic myomas are considered as a complication of morcellation of myomas during myomectomy.

Ours is a case of huge primary PL measuring about 20x25x15 attached to the omentum, sigmoid mesocolon and anterior abdominal wall with highly vascular dilated tortuous multiple feeding vessels. This myoma retained its attachment to the right cornual region of the uterus.

Only few cases of primary PL are reported. In review of literature by Clark NV et al six patients had undergone abdominal myomectomy without morcellation developed parasitic myomas. Peritoneal metaplasia is one of the theory which describes pathogenesis of myoma in unexpected locations.

There are other factors which influence the formation of PL which includes prolonged exposure to endogenous or exogenous gonadal steroids. There are case reports which show rapid growth of PL during pregnancy supporting the hormonal impact on parasitic leiomyomas.

Conditions that restrict the blood supply to the uterus such as GnRH agonist, radio frequency ablation, uterine artery embolisation may be associated with this process as pedunculated myoma seeks an alternate blood supply.

Lumsden et al and Vollenhoven et al state that association of polypoidal growth factors, such as platelet derived growth factor and transforming growth factor may stimulate growth of these type of myomas.

The sites of parasitic fibroids are para rectal fossa, eul-de sac, abdominal wall, appendix, paravesical space, intestinal serosa, rectus muscle and bowel mesentery. Usually PL present with pressure symptoms which include bladder and bowel symptoms rather than menstrual symptoms.

Aimal khan et al reports a case of parasitic leiomyoma of greater omentum presented as small bowel obstruction in journal of surgical case report. Though they are benign, PL may mimic malignant tumour and pose a diagnostic challenge. Radiology, particularly MRI is useful in differentiating from ovarian mass and broad ligament myomas.

Laparoscopic myomectomy remains the gold standard treatment for women harbouring myomas in the reproductive age group. Myomectomy by laparoscopic approach needs surgical expertise. Very few cases of laparoscopic myomectomy for PL reported.

There are two different techniques regarding the surgical management. Some surgeons detach the myomas from the uterine attachment and proceed with rest of the surgery and others release the myoma from the adjacent structures from where it receives its blood supply and finally detach the myoma from the uterus.

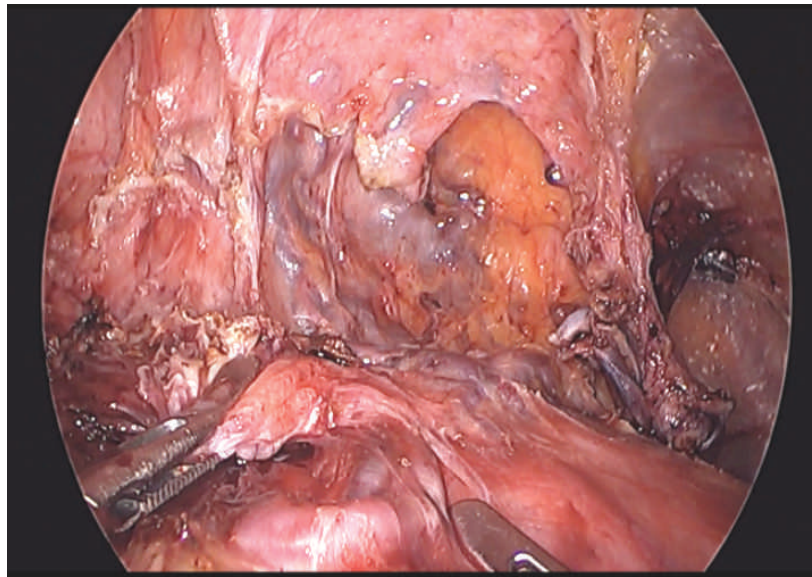
In our case because of huge size and broad base initially it was not possible to make out the site of uterine attachment. Only after detaching the myoma from the omentum, anterior abdominal wall and sigmoid, mesocolon the origin of myoma could be traced to the right cornual region with multiple feeding vessels.

The main challenges of removing parasitic myomas include limited working space due to

huge size, tackling the tortuous dilated vascular pedicles, dissection from adjacent vital structure like bowel and retrieval of the huge myoma. In addition to above mentioned risks our patient had a broad base of myoma attached to the right cornual region with extensive vascularity.

Though there are many limiting factors for laparoscopic myomectomy in patients with

huge parasitic myomas, surgical expertise, good anaesthetic back up, optimal theatre set up with good quality equipments and instruments can make this difficult surgery feasible without compromising patient safety. The advantage of laparoscopic approach is minimal postoperative adhesion which contributes to the favourable outcome in an infertile patient.



Extensive vascular supply from anterior wall of abdomen.

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Best Super Speciality
Hospital Award
by Radio City



Health Talk Programme
at BPCL
by Dr. P. Krishnananda



CME Programme at
P.Velur on 22.12.2019 by
Dr.Kirubanand Jaganathan,
Dr.Nachimuthu Kumar,
Dr.Dinesh Chidambaram



CME Programme
at Karur
by Dr. K. Madeshwaran



Breast Cancer
Awareness Camp
KPR College



74th IMA State Conference, Erode
on 14,15th December 2019



Organ Donation Awareness Campaign on 27.11.2019



Mock drill at
Coimbatore
Airport

Health Talk Programme at
Indoshell Mould Company Cbe
by Dr.Nachimuthukumar



Royal Fest- 3rd Year
Anniversary
Celebrations



SUCCESSFUL PREGNANCY OUTCOME WITH BI-CORPOREAL UTERUS, TRANSVERSE VAGINAL SEPTUM AND UNILATERAL RENAL AGENESIS



Dr. N. Premalatha

MBBS, DGO, MRCOG (UK), DIUI (France),
Consultant Obstetrician and Gynaecologist

Miss S a 25 year old healthy lady attended for a pre employment health check up. Routine bloods including renal function tests were normal. Ultrasound revealed agenesis of the right kidney with a uterine anomaly. Her periods were regular and normal and no dysmenorrhoea was reported. Subsequently a detailed 3D scan revealed a bi-corporal septate uterus with a transverse vaginal septum. She was also found to have multiple small fibroids. The lady was counselled thoroughly about the findings and the potential adverse fertility outcomes.

She subsequently attended the OPD after a year having got married 6 months ago with a positive pregnancy test. A confirmation scan at 6 weeks showed a single live intra uterine pregnancy in the left horn of the uterus. Her booking bloods including urea and creatinine were all normal. Subsequently she had another scan at 12 weeks. The Nuchal translucency was 1.36mm, nasal bone was seen, there was a single umbilical artery with no other anomaly in the foetus. There were 2 pedunculated fibroids, two from the posterior wall of the uterus and another fibroid in the anterior surface. Her PAPP-A (Pregnancy associated plasma protein) was borderline at 0.4mom. Having a low PAPP-A carries an increased risk for Pre eclampsia, intra uterine growth retardation, pre term delivery and increased incidence of aneuploidy in the fetus. However her first trimester combined screening results revealed that fetus

was at a low risk for Trisomy 21, Trisomy 18 and Trisomy 13. She was commenced on low dose Aspirin.

She had her routine pregnancy vaccinations that includes TT, DPT and influenza vaccine. She had regular calcium and iron supplementations. A glucose tolerance test done in the second trimester was entirely within normal limits. Her renal functions were checked every 6 weeks and they remained within normal limits.

Her anomaly scan at 21 weeks including a foetal echo was normal. The fibroids remained static in size during her subsequent scans. She had one positive urine culture at 25 weeks that was treated with appropriate antibiotics. A growth scan at 28 weeks revealed that the estimated fetal weight was on the 5th centile with normal amniotic fluid index and dopplers. A subsequent scan after 3 weeks revealed an Estimated Fetal Weight of 1.3 kg, that has fallen to the third centile. She was given 2 doses of 12mg Betamethasone 24 hours apart. She was counselled about the increased risk for preterm delivery.

Another scan at 33 weeks revealed that the Estimated Fetal Weight continued to remain at the 3rd centile at 1.6 kg with normal AFI and dopplers. The presentation remained persistently breech. 6 days later she presented with pre-term pre-labour rupture of membranes at 34 weeks. She was commenced on antibiotics and an emergency LSCS was done for footling breech. A footling

breach presentation with ruptured membrane poses an increased risk for umbilical cord prolapse. During Caesarean, the pregnancy was in the left horn of the uterus, the right horn was very small and non communicating, right adnexa was underdeveloped. An uneventful breech delivery was undertaken. There were 3 sub serous fibroids noted. Two measuring around 4x5 cm were seen in the posterior wall. Another fibroid was arising from the fundus. One of the posterior wall fibroids looked unhealthy with extensive degenerative changes. Therefore decision was made to remove it. An uneventful Caesarean myomectomy was undertaken. As the transverse vaginal septum was very vascular no intervention was done at that stage. An alive female baby weighing 1.7 kg was delivered in good condition. The baby had an uneventful early neonatal period and was discharged after 10 days.

Discussion- According to the new ESHRE/ESGE classification, this falls into Class U3c or bi-corporeal septate uterus characterized by the presence of an absorption defect in addition to the main fusion defect. In patients with bi-corporeal septate uterus (Class U3c) the width of the mid line fundal indentation exceeds by 150% the uterine wall thickness. With coexisting vaginal anomaly i.e, a transverse vaginal septum which falls into V3.

Uterine anomalies pose an increased risk for miscarriages, pre-term labour, malpresentation, Intra uterine growth restriction, placental problems and increased requirement for operative deliveries. Correction surgeries could be done in certain cases to improve the reproductive outcomes. Our lady here was already pregnant when she first visited the Obstetrician after her wedding. She was reviewed at 3 months postpartum. The septum has considerably shrunk in size. She was not keen on any intervention at that stage.



MULTIDISCIPLINARY WORKSHOP UNDERSTANDING AND SUPPORTING CHILDREN WITH ADHD



Dr. Mallikai Selvaraj
MBBS, DCH, PGD-DN.,

Consultant Developmental Paediatrician



Dr. Pavithra Mony, M.D. (PSYC),
Consultant Psychiatrist



Mr. D. Santhosh, M.SC, M.PHIL (CLIPSY), IMH.,
Clinical Psychologist

In anticipation of National ADHD awareness month, the Child Development Centre the Royal Care Super Speciality Hospital - City Unit conducted the ADHD awareness workshop on 5th October 2019. The workshop was open to the public and approximately 150 people which included teachers, social workers and parents with their children. Separate game activities was arranged for the children. During the workshop participants had the opportunity to question, share their opinions thoughts and suggestions with the speakers. It was a structured process of brainstorming and information exchange between the participants at the end of the session.

The keynote speaker was Dr. Mallikai Selvaraj, our Developmental paediatrician spoke on the topic "INSIDE THE ADHD MIND" as follows.

ADHD defines a type of behaviour that is an extreme end of the normal range of behaviour. Children with ADHD demonstrates problem levels

- Inattention (i.e difficulty concentrating)
- Hyperactivity (i.e disorganised excessive levels of activity)
- Impulsive behaviour

Some show all the symptoms, while others show only some. 4 -5 times more boys are affected than girls. When one gets older although the type of difficulties you experience make change.



The range of symptoms depends on which type of

ADHD the child has. Behaviour is usually first noticed at the age of 3 to 4 years. The behavioural problems must be present in more than one setting. For example, if symptoms are present at home there would be problems in school or in another social situations for diagnosis of ADHD. Parents/carers may notice that children is



constantly restless, irritable and emotionally immature. Aggressiveness and poor discipline are common. The child may show no sense of danger, for example, climbing fearlessly and ignoring repeated warnings. Sometimes children with ADHD are also clumsy. Starting school often highlights a problem or makes it worse. Learning is often impaired by the lack of attention and concentration. As a child grows older he/she may engage in antisocial behaviour and be unaware of the consequences, if not properly guided. ADHD is not related to intelligence - children with all levels of ability can be affected.

ADHD has prevalence of approximately 5-7

percent worldwide, i.e. 1-2 students in a class of 30. Boys are three times more likely to be diagnosed. Girls tend to show more symptoms of inattention and they appear as "day dreamers". When one gets older although the type of difficulties you experience make change.

The cause of ADHD is unknown and there are likely number of factors involved. There is a strong genetic element so many children with ADHD have another family member with the same problem. Children are more likely to have ADHD if they are born prematurely or if their mother smoked or misused alcohol during pregnancy. When these factors interact with non conducive environment symptoms worsen.

Neurotransmitters like dopamine and noradrenaline transfer messages from one cell to another. They act like postman to deliver the messages between the nerve cells. We need sufficient amount for effective transfer of messages. This helps to process our thoughts, plan and carry out the tasks. If there is faster recycling of neurotransmitters, the transfer of messages slow down leading to failure of executive function. Executive function for self regulating skills that we used to accomplish routine tasks like getting dressed to doing homework. The medical management block the recycling of chemical messengers and thereby result in effective message transfer.

Comorbidities of ADHD was explained in detail by DR Pavithra Moni, our psychiatrist. The common comorbid conditions are oppositional defiant disorder, specific learning difficulties, autistic spectrum disorder, anxiety, tourette syndrome, OCD, developmental coordination disorder,

sensory integration disorder and depression.

The cognitive behavioral therapy for ADHD management was explained by our clinical psychologist Mr. Santosh. He gave tips for the parents and the school teachers how to help the child by providing a structured environment with clear rules of acceptable behaviour. Good behaviour should be reinforced with rewards and bad behaviour should be ignored. When punishment is necessary that should be time limited, for instance set periods of time alone in a separate room "time out". Parents should make sure that their children's teacher is well informed about the condition. Structured and orderly class room without too many distractions together with the understanding teacher are helpful. Often small group of individual teaching helps. Special help should focus on particular problem areas such as difficulties with reading and spelling.

Finally Dr. Mallikai Selvaraj ended up by saying, ADHD brains have lot to offer the world. They can be generous, funny and creative. 300% more likely to start of their own business. They not only think outside the box but often not even aware that there is a box. ADHD brains struggle with the brains that not engage but they are great at tackling tasks that are urgent, working with new ideas, wrestling with problems that are challenging and dedicate themselves to the project that are of personal interest. Hence it is important to understand the ADHD brain and to find the strengths they have and learn the strategies for challenges. ADHD is not a knowledge deficit. It's a performance deficit.



MUCINOUS CYSTIC NEOPLASM OF THE LIVER WITH BILIARY COMMUNICATION: AN EXCEPTION TO THE CURRENT CLASSIFICATION



Dr. A. Sandip Chandrasekar

MS, M.Ch (SCE), DNB (SCE), FALS (Bariatric),

Consultant Surgical Gastro, Advanced Laparoscopic, HPB, GI
Oncology & Bariatric Surgeon

SUMMARY

Cystic neoplasms of the liver are rare tumours. According to the recent WHO classification, they are classified into mucinous cystic neoplasm and intraductal papillary neoplasm based on the presence of ovarian-like stroma and biliary communication. We report two rare cases of mucinous cystadenoma of the liver with biliary communication and discuss the shortcomings of current classification.

BACKGROUND

Cystic neoplasms of the liver are rare tumours and constitute less than 5% of cystic lesions of the liver.¹ Initially, they were termed as biliary cystadenoma and cystadenocarcinoma.² Recently, WHO had classified the cystic neoplasm of the liver into mucinous cystic neoplasm (MCN) and intraductal papillary neoplasm of the bile duct (IPN-B) similar to the classification used in pancreas.³ MCN is characterised by the presence of ovarian stroma and absence of bile duct communication. Whereas,

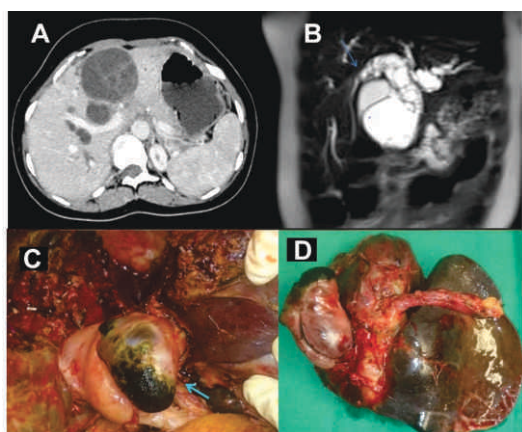


Figure 1 (A) CECT abdomen showing hypodense lesion in segment IV of the liver with few enhancing internal septations. (B) MRCP showing bilobar IHBRD with a heterogeneous T2 hyperintense lesion, extending into the proximal CBD causing abrupt narrowing at the mid-CBD level (arrow). (C) Intraoperative photograph showing portion of the tumour within the CBD milked out (arrow). (D) Resected specimen showing both hepatic and biliary components. CECT, contrast-enhanced CT; IHBRD, intrahepatic biliary radical dilatation; MRCP, MR cholangiopancreatography.

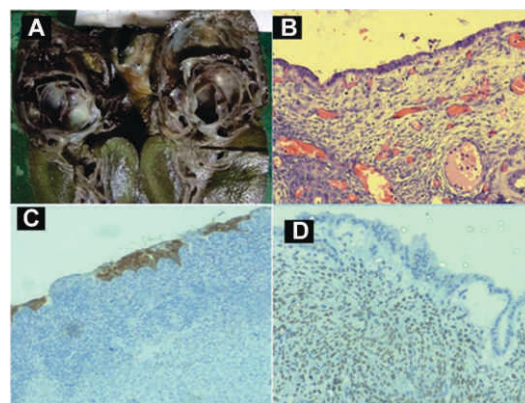


Figure 2 (A) Cut section of specimen showing solid cystic lesion. (B) Photomicrograph showing low columnar epithelial lining cells with underlying ovarian-like stroma (H&E, magnification x400). (C) Immunohistochemistry shows columnar lining epithelium with cytoplasmic staining of cytokeratin and (D) underlying ovarian stroma with nucleus staining positive for oestrogen receptor.

IPN-B lesions lack ovarian-like stroma and have predominant intrabiliary component. We report two rare cases of MCN of the liver with biliary communication and discuss the limitations of current WHO classification of cystic neoplasm of the liver.

CASE PRESENTATION

Case 1

A 20-year-old woman presented with intermittent upper abdominal pain, jaundice, fever and pruritus for 1 month. She had a firm tender mass arising from the left lobe of the liver extending 6 cm below the xiphoid in midline.

Case 2

A 28-year-old woman presented with jaundice, pruritus and anorexia for 2 months. She was initially evaluated elsewhere, where investigations suggested a simple hepatic cyst with compression of the bile duct. The patient underwent laparotomy and subtotal excision of the cyst elsewhere which on histopathological examination (HPE) was reported as simple hepatic cyst. Postoperatively, the patient had persistent jaundice.

INVESTIGATIONS

Case 1

Her liver function test was suggestive of obstructive jaundice. Ultrasound abdomen revealed a 6×5 cm multiloculated cystic lesion in segment IV of liver with bilateral intrahepatic biliary radical dilatation (IHBRD). Common Bile Duct (CBD) was normal, and gall bladder was collapsed. On contrast-enhanced CT (CECT) abdomen, there were hypodense lesions predominantly in segment IV with internal septations, however there were no solid components or calcifications (figure 1A). MR cholangiopancreatography (MRCP) revealed a multiloculated 7.0×6.2×5.3 cm heterogeneous predominantly T2 hyperintense lesion in segment IV of the liver. Bilobar IHBRD with probable cystobiliary communication was present (figure 1B).

Case 2

She was evaluated with MRI abdomen and MRCP which revealed cystic lesion in segment IV with obstruction at the level of the proximal CBD and bilateral IHBRD. Slide review of previous operative specimen at our institute was reported as mucinous cystadenoma of the liver. On CECT abdomen, there was a hypodense lesion in the segment IV with communication with the left biliary system. Endoscopic ultrasound revealed a 2.4×1.9 cm cystic lesion with soft tissue thickening compressing the common hepatic duct and another 2.8×1.4 cm multiseptated cystic lesion seen along the left ductal system.

TREATMENT

Case 1

The patient underwent open left hepatectomy. On laparotomy, there was a 6×6 cm solid cystic lesion in segment IVb of the liver with a mobile mass within proximal CBD. Choledochotomy was performed at the level of the dilated portion of CBD, where communication of the cyst with bile duct through left hepatic duct was seen (figure 1C,D). HPE of the resected specimen confirmed the diagnosis of MCN (figure 2).

Case 2

On laparotomy, there was a 5×4 cm cystic lesion in segment IVb of the liver, adherent to the left hepatic duct. Intraoperative cholangiography and choledochoscopy confirmed communication of the cyst with bile duct. Complete excision of the hepatic portion and the biliary component along with the sleeve of the left hepatic duct was performed. Choledochotomy was closed primarily over T tube. HPE of the resected specimen confirmed MCN. The T tube was removed 4 weeks later, after T tube cholangiogram did not show any evidence of leak or obstruction.

OUTCOME AND FOLLOW-UP

Case 1

At the 2-month follow-up, the patient is asymptomatic with a normal liver function test.

Case 2

At the 12-month follow-up, the patient is asymptomatic with a normal liver function test.

DISCUSSION

Cystic neoplasms of the liver are rare tumours and constitute less than 5% of symptomatic cystic lesions of the liver.¹ In the WHO classification of liver tumours proposed in 2000, they were termed as biliary cystadenoma (benign) and

cystadenocarcinoma (malignant).² Biliary cystadenoma were further classified into mucinous (common type) and serous types (rare). The term biliary papillomatosis was used for tumours with predominant intrabiliary growth. Recently in 2010, WHO proposed a new classification system for cystic neoplasms of the liver similar to the terminology used for pancreatic cystic tumours and classified them into MCN and IPN-B.³ The diagnostic criteria for MCN were presence of ovarian-like stroma and absence of bile duct communication in addition to the presence of typical mucin-secreting biliary type cuboidal or columnar epithelium. IPN-B lesions were characterised by absence of ovarian-like stroma and predominant intraductal growth pattern. Similarity in clinical pathological features of MCN of the liver, pancreas and ovary suggests an association.⁴ During embryogenesis, right gonad lies dorsolateral to the liver, and left gonad lies dorsal to the pancreas and spleen until 8 weeks of gestation. Close relation of these structures to gonad could explain the similarity in microscopic features of these tumours.⁴

MCN commonly affects females in their reproductive period. Small lesions are usually asymptomatic while large cysts present with abdominal pain or mass. Jaundice is rare, and it occurs when there is biliary communication.⁵ Preoperative diagnosis is difficult, and they are easily mistaken for simple hepatic cysts or hydatid cysts. Hydatid serology can help in differentiating from hydatid cyst. Elevated levels of CA 19-9 (serum and cyst fluid) have been suggested by some authors to help in the diagnosis of MCN.⁶ On ultrasound, MCNs appear as anechoic lesions with internal septations.⁷ Intracystic haemorrhage or papillary projections appear as focal hyperechoic areas. Features of MCN on CECT abdomen are hypodense lesions with internal septations which enhance with contrast.⁸ Presence of irregular wall thickening, mural solid nodules, thick calcification and papillary projections are suggestive of a cystadenocarcinoma. On MRI abdomen, these lesions appear hyperintense in T2-weighted images and hypointense on T1-weighted images. Use of the biliary-specific contrast agent gadobenate dimeglumine can help in preoperative diagnosis of biliary communication.⁸ On HPE, MCN are lined by biliary type mucus secreting cuboidal or columnar epithelium. Characteristic feature of MCN is presence of dense subepithelial ovarian-like stroma with spindle cells expressing female sex hormone receptors.^{1,9} Historically, MCN were treated by marsupialisation, internal Roux-en-Y drainage, aspiration, sclerosis or partial resection. Considering their malignant potential and risk of recurrence complete excision is the treatment of choice.¹⁰

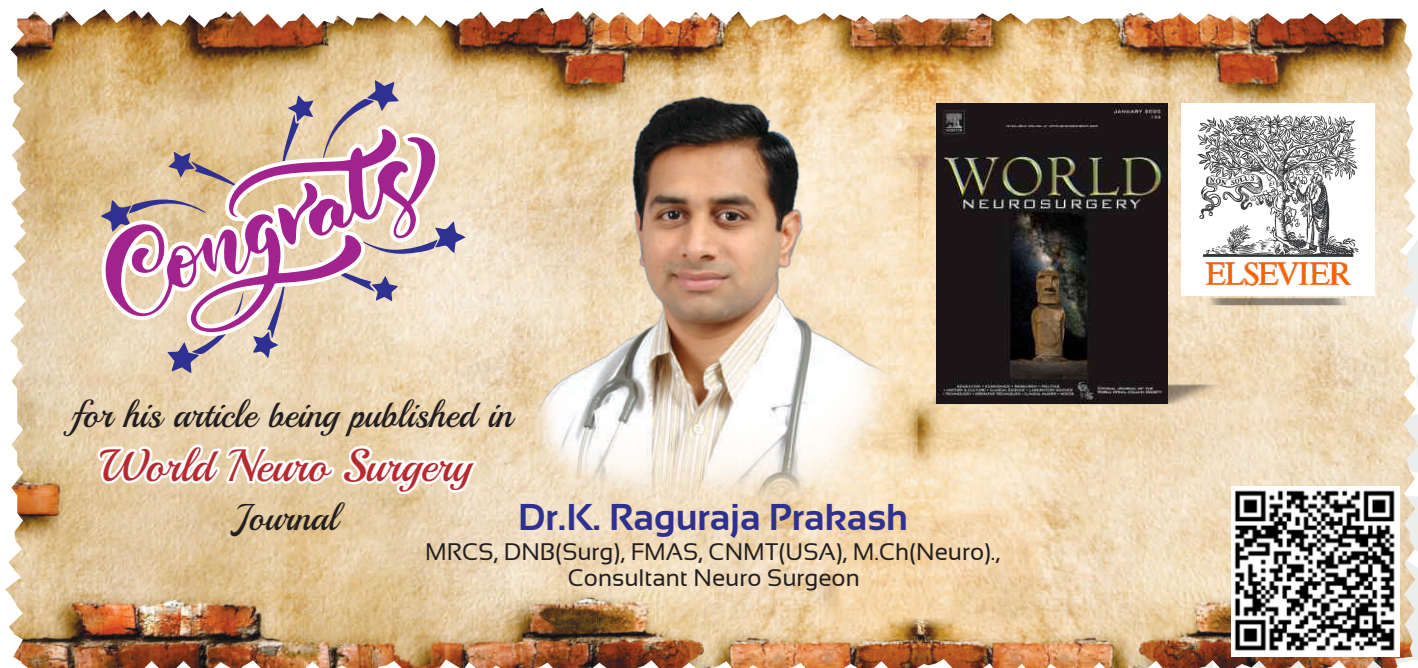
Peculiar features of the present cases are presentation with jaundice which is a rare clinical manifestation of MCN. Possible mechanisms for jaundice are intraductal extension, intracystic haemorrhage or mucin secretion. Very few case reports have described cystadenoma prolapsing into bile duct.¹¹ In these two cases, it is due to intraductal extension of tumour which is a rare feature of MCN. We also highlight the problem with current classification. Since ovarian-like stroma was present, a diagnosis of MCN was made. However, bile duct communication was also present which is a feature against MCN according to current diagnostic criteria. Hence, we suggest that MCN should be further subclassified into MCN with or without biliary communication.

Learning points

- Mucinous cystic neoplasm of the liver is a rare tumour and should be considered in the differential diagnosis of patients with cystic neoplasm and jaundice.
- Current classification needs to be modified to include these rare cases of mucinous cystic neoplasm with biliary communication.

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Dr. Neha Chauhan

MBBS, MS, FVRS.,
Consultant Ophthalmologist, Retina & Phaco Surgeon

Completed MBBS from Lady Hardinge Medical College, New Delhi in 2004. She has obtained M.S.(Ophthalmology) G.R. Medical College, Gwalior in 2008. She worked in Vasan eye care hospital erode, before joining our Royal Care Super Speciality Hospital.



Dr. P. Kiruthika

MBBS MD(Radio diagnosis)
Consultant Radiologist

Completed MBBS from Stanley Medical College in 2012 and MD (radiodiagnosis) from PGIMER Chandigarh in 2015. Worked as senior resident in PGIMER till 2017. Worked in SSRD Centre in Chandigarh (2017-2018). and house of Diagnostics In Delhi (2018-2019). Now she has joined our Royal Care Super Speciality Hospital as Consultant Radiologist.



Dr. Pavithra Mony

MBBS, MD(Psych).,
Consultant Psychiatrist

Completed MBBS from Saveetha Medical college in 2016. and she has achieved MD(Psychiatry) from Pondicherry Institute of Medical sciences in 2019. Now she Joined as Consultant Psychiatrist at our Royal Care Super Speciality Hospital - city unit.



Dr. A.S. Rahul

MBBS, DNB, (Nuclear Medicine).,
Consultant in Nuclear Medicine

Completed MBBS from Azeezia Institute of Medical Science and Research, Meyyanoor, Kollam, Kerala in 2008-2014. and he achieved Primary DNB trainee in Nuclear Medicine from Seth G.S Medical College and K.E.M Hospital, Parel, Mumbai in 2016-2019. Now he joined as a Consultant in Nuclear Medicine department at our Royal Care Hospital.





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