ABSTRACT
Synovial Sarcoma is a soft tissue neoplasm having incidence 6%-10%. Malignant cells in synovial fluid aspiration is extremely rare. Only 5% cases have been reported to have joint cavity involvement. We report a case of synovial fluid malignant effusion of knee joint in a 35 year old male who presented with a left popliteal fossa swelling. Synovial fluid aspiration revealed tumor cell clusters with anaplastic morphology. Subsequent biopsy showed spindle cell tumor consistent with synovial sarcoma. This is a rare presentation of synovial sarcoma with the presence of malignant cells in synovial fluid.

Keywords: Synovial sarcoma, Synovial fluid, Spindle cell tumor.

INTRODUCTION
Synovial sarcoma is a soft tissue neoplasm with the incidence of 5-10%. It occurs commonly in adolescents and young adults. Although it is often found to be in close association with tendon sheaths, bursa, and joint capsule; it is unusual for it to involve the joint. It rarely involves synovial membrane. Less than 5% synovial sarcomas arise within joint space (1). Synovial sarcoma are often misdiagnosed as benign lesion on biopsy. Chow LT reported a case of epitheloid sarcoma of knee joint mimicking pigmented villonodular synovitis. Bergovec at al reported a case of lateral meniscus synovial sarcoma (2). Age, gender of patient, size, effusion and calcification are helpful in differentiating intraarticular synovial sarcomas from localized PVNS (3).

CASE REPORT
A 35 year old male presented with swelling at posterior aspect of left knee joint for 10 months & painful since 15 days. Clinical Examination showed a non tender 8 x 6 cm, firm to hard swelling, causing restriction of knee flexion. Routine hematological Investigations were within normal limit. X-ray showed Mass in popliteal fossa. Synovial fluid was aspirated and sent for cytology followed by biopsy from the tumor. Approximately 4 ml straw colored synovial fluid was received. Smears were made & stained with H & E. Microscopic examination was done. Cellular smears made from synovial fluid revealed clusters & singly scattered spindle to ovoid atypical cells admixed with lymphocytes. Cells had high nucleocytoplasmic ratio, irregular nuclear contour clumped chromatin & prominent nucleoli at places (Figure 1). Following which an impression of synovial fluid positive for malignant- spindle cell lesion was made and patient was advised biopsy for confirmation of diagnosis.

Fig 1:(H&E;10X)Photomicrograph from synovial fluid aspiration shows spindled to ovoid cells with high nucleocytoplasmic ratio irregular nuclear contour, clumped chromatin and prominent nucleoli at places.
spindle to ovoid cells arranged in interlacing bundles & fascicles. Tumor cells were moderately pleomorphic with hyperchromatic nuclei & finely clumped chromatin. Interspersed in these spindle cells were cleft like spaces (figure 2). Tumor cells did not stain for PAS stain. Immunohistochemistry revealed vimentin positivity in spindle cell elements and cytoplasmic expression of pancytokeratin in interspersed ovoid cells. A histopathological diagnosis of synovial sarcoma was made.

DISCUSSION

Synovial Sarcoma is a soft tissue neoplasm having incidence 5%-10%. Most cases present with a periarticular soft tissue mass, but now it is being observed that it can arise from any organ and anatomic site. Intraarticular synovial sarcoma is very rare. Aspiration biopsies of synovial sarcomas yield moderately and more frequently highly cellular smears. Each cell shows a solitary ovoid nucleus with finely granular, hyperchromatic chromatin with inconspicuous nucleoli. The neoplastic elements have mostly scant cytoplasm. A small number of neoplastic cells have more abundant cytoplasm which appears as bipolar tapered cytoplasmic tails. The shape of individual neoplastic nuclei vary from blunt and ovoid to elongated and spindled. In some cases the neoplastic elements exhibit a distinct epithelial differentiation as polygonal contours and solitary round hyperchromatic nuclei. The nuclei are central or eccentric and the cytoplasm is vacuolated. Immunocytochemically positive reactivity for epithelial markers (eg: the various cytokeratin) assists in the differential diagnosis with other spindle cell sarcomas. Histologically synovial sarcoma has two patterns monophasic & biphasic pattern. Monophasic pattern is characterized by spindle cell in bundles and fascicles with hemangiopericytoma like foci. Biphasic pattern is characterized by epithelial cell component disposed as glands and nests along with fibroblast like spindle cell. Poorly differentiated synovial sarcoma is difficult to identify due to considerable overlap with other variants. It is characterized by primitive round cell. Immunohistochemistry for EMA and CK is positive in both spindle and epithelial cell elements. It also shows positivity for keratins, desmoplakin, Leu-7, and S-100 protein.

Radiological findings are not pathognomonic but a calcified lesion near a joint is suggestive of diagnosis. MRI shows triple sign; bowl of grapes sign. On MRI, the lesion is hypointense on T1-weighted (T1W) and hyperintense on T2-weighted (T2W) images and demonstrates multilobulation and marked heterogeneity (creating the “triple sign”) with haemorrhage, fluid levels and septa (creating the “bowl of grapes” sign) (Figure 3) (2,11,12). Slow growth (average time to diagnosis, 2–4 years) and small size (< 5 cm at initial presentation) of the lesion may result in a mistaken initial diagnosis of a benign indolent process.

Aisner SC described two cases of synovial sarcoma diagnosed by aspiration cytology he showed the utility of aspiration cytology in diagnosing both unsuspected and recurrent synovial sarcoma (4). Kilpatrick SE studied 13 fine-needle aspiration specimens from 10 patients with histologically proven synovial sarcoma and concluded that synovial sarcoma could be diagnosed with FNAB but clinically and histological correlation is necessary to confirm the diagnosis especially in monophasic variants (5).

We report this case as it was first suspected to be synovial sarcoma on synovial fluid cytology which showed spindle to ovoid malignant cells which was further confirmed as synovial sarcoma on biopsy from main tumor mass in popliteal fossa. Justin E et al reported two cases of primary intra articular synovial sarcoma of Knee (6). Ahana Gupta et al reported a case of monophasic synovial sarcoma in a 14 year old boy (7).

Synovial sarcoma metastasizes to lung and lymph nodes. Local recurrence is common and carries a poor prognosis. Detection of cytogenetic alteration t(X;18(p11.2; q11):SYT-SSX1 gene and t(X;18)(p11.21;q11):SYT-SSX2 fusion gene by PCR helps in further confirming the diagnosis (8,9).

CONCLUSION

Synovial sarcomas secondarily involve joint space, primary involvement of synovium by sarcoma is extremely rare. Mostly these cases are diagnosed by biopsy but in this case it was diagnosed first on synovial fluid cytology. Early diagnosis of primary
tumor & recurrence by synovial fluid cytology can lead to early detection of tumor which will lead to better prognosis.

REFERENCES


ABSTRACT

Spinal epidermoid are rare lesion of spine. We report a case of 12 year old female who presented with difficulty in walking and weakness in left lower limb. MRI spine showed epidermoid at level of L1-L2. Laminectomy and surgical excision of the cystic lesion was planned. Intra operatively a pearly white tumor adherent to cord was found and excised. Histopathological examination confirmed the diagnosis of epidermoid cyst. One year after surgery she again came with complaint of pain at the site of surgery following which MRI was done which showed recurrence of epidermoid, intra operatively it was found that it was a cystic cavity filled with hemolyzed blood mimicking as recurrent epidermoid radiologically.

Keywords: Spinal Epidermoid, MRI, Laminectomy.

INTRODUCTION

Spinal epidermoid are cystic lesions lined by squamous epithelium they account for less than 1% of spinal lesion. They were first described as tumeursperlées (pearly tumors) by Cruveilhier in 1835. Epidermoid can be congenital or acquired. They can be extradural, intradural or extramedullary, or intramedullary in the spine. Spinal epidermoids present with non-specific symptoms, MRI is helpful in arriving at diagnosis. Reoccurrence of epidermoid cyst are rare but are not uncommon after incomplete surgical excision of capsule.

CASE REPORT

We report a case of 12 year old female who presented to our hospital with history of trauma to back 3 years back and complaints of difficulty in walking and pain in back since 2 year along with weakness of left lower limb with foot drop since 1 year. Neurological examination revealed 4/5 power in left lower limb, power at ankle 2/5 with foot drop. Upper limb had normal power. Rest of the neurological examination was within normal limits. MRI spine was ordered which showed T1, T2 hyperintense lesion at level of L1& L2 - likely of epidermoid cyst was made. She underwent L1-L2 laminectomy along with surgical decompression of the sac. Intra operatively tumour was present just below the dura along with lipomatous tissue which was pearly white, non vascular and non suckable, capsulated adherent to cord and one of the nerve root. A small part which was adherent to nerve root was left behind. Histopathological examination was consistent with the diagnosis of epidermoid cyst and showed cyst wall lined by stratified squamous epithelium and abundant keratinous material (figure 1).

The patient was discharged after uneventful hospital stay. The patient came to neurosurgery OPD after 1 year with complaint of pain at the site of surgery. MRI was done which showed recurrence of epidermoid cyst. Intra operatively it was found that it was a cystic cavity filled with hemolyzed blood mimicking as recurrent epidermoid cyst.
year with the complaints of pain over the same site of surgery since 1 month. Again MRI was done with radiological appearance (figure:2)

Fig 2: MRI: T1,T2 hyperintense lesion at L1 & L2

Showing recurrence of epidermoid. Re exploration of the earlier postoperative site with excision of recurrent epidermoid was planned. Intra operatively it was found that cystic cavity filled with hemolyzed (motor oil like fluid) was present, it was conclude that intracystic blood was mimicking as recurrent epidermoid.

DISCUSSION

Congenital epidermoid occur as a result of inclusion of ectodermal tissue within the neural canal at the time of neural tube closure at fourth to fifth week of intrauterine life. Congenital epidermoids are usually associated with spinal dysraphisms such as syringomyelia, dermal sinus and spina bifida. Acquired epidermoid cysts are mostly iatrogenic occur as a result of implantation of ectodermal cells in spinal cord during surgery or spinal procedures like lumbar puncture or during trauma to spine. Very few cases of post traumatic epidermoid cyst have been reported in literature. Shengre reported a case of spinal epidermoid following burst fracture of lumbar vertebra.

The symptomatology in case of spinal epidermoid is non-specific and confusing. Patients generally present with non-specific features of numbness, weakness, spasticity, paraparesis of lower extremities and defecation disorders pose challenges in pre-operative diagnosis. Differential diagnosis in such cases include ependymomas, metastasis, astrocytomas and dermoid cysts. MRI is useful in differentiating epidermoid from ependymoma, astrocytoma and metastasis. A histopathological examination is necessary to confirm the diagnosis of dermoid cyst.

Intra operatively epidermoid can be ruled out by its gross appearance. Epidermoid cysts are grossly pearly white cysts filled with characteristics keratinous content. Total surgical excision is treatment of choice for epidermoid cyst but sometimes it is difficult to remove cyst in Toto due to adhesion of capsule to spinal cord and nerve roots. In such cases a sub total excision of cyst is done. Recurrence of spinal epidermoid is not uncommon after incomplete resection. Fleming reported a case in which he described recurrence of epidermoid cyst seven times at the same location. Cases with symptomatic recurrence should be retreated with surgery but surgery is often difficult due to formation of scar tissue. Delayed postoperative haemorrhage is seen as a complication of spinal epidermoid. Multiple relapse of epidermoid cyst have been satisfactorily treated in a case with radiotherapy. Recurrent cases can be managed by radiotherapy but is not used routinely in recurrent cyst cases, this modality of treatment is reserved for cases who refuse surgery or are inoperable due to medical reasons.

CONCLUSION

Spinal epidermoid are rare, recurrence is rare but can occur before considering it as a case of reoccurrence. Complications associated postoperatively with epidermoid must be evaluated. As in our case it was delayed postoperative haemorrhage. MRI is helpful in arriving at the diagnosis. Surgical removal of cyst is treatment of choice.

REFERENCES


**ABSTRACT**

Methotrexate (MTX) is a folic acid antagonist with cytotoxic and immunosuppressant activity and with potent antirheumatic action. It is commonly a first choice Disease modifying antirheumatic drug (DMARD). There were some spurious reports of Adverse Drug Reaction (ADR) by this drug. Here we report a rare occurrence of Stevens-Johnson syndrome (SJS) / Toxic Epidermal Necrolysis (TEN) after the use of Methotrexate. Naranjo score for this adverse drug event was six, thereby making it a probable ADR. Symptomatic management of the patient was done and the offending drug was withdrawn. We are presenting this case to highlight the serious adverse reactions possible from a routinely prescribed drug.

**INTRODUCTION**

The World Health Organisation defines an adverse drug reaction (ADR) as “a response to a drug which is noxious and unintended, and which occurs at doses normally used in man for the prophylaxis, diagnosis, or therapy of disease, or for the modification of physiological function.” Methotrexate (MTX) is well-established as the “anchor drug” for patients with rheumatoid arthritis (RA), to be used early and aggressively, with higher long-term effectiveness, tolerability, and safety than any other disease-modifying antirheumatic drug (DMARD)(1). Methotrexate has greater affinity than folic acid for the dihydrofolate reductase enzyme. This mechanism of methotrexate reduces the tetrahydrofolate and attenuates the DNA synthesis in proliferating cells, making it an ideal disease modifying agent for rheumatoid arthritis. Methotrexate is metabolized into a series of polyglutamate derivatives that increase linearly with the concentration and duration of exposure. Therefore, higher doses of methotrexate administered for prolonged duration can result in greater toxicity(2). Other reported adverse effects are diarrhea, dizziness, stomach pain, fever and chills. Stevens-Johnson Syndrome (SJS) is a life-threatening, bullous cutaneous disease considered as immune-mediated reactions to drugs characterized by epidermal necrosis, extensive detachment of the epidermis, erosions of mucous membranes and severe constitutional symptoms(3). Here, we report a case of methotrexate induced Stevens Johnson syndrome, a clinical association that has been previously reported in very few cases in Indian population.

**CASE REPORT**

A 46-year-old female came to Dermatology outpatient department with chief complaints of fever, swelling of lips, oral ulcers and blisters all over the body. Fever was of high grade 39.4°C (103°F) and had a continuous pattern. Patient had noticed the rashes a few days after developing fever. She had been diagnosed with rheumatoid arthritis one month ago and had been taking oral methotrexate 7.5 mg per week for 2 weeks. No past history of allergic diathesis was reported. Gradually these rashes developed into bullae and blisters spreading all over the body from face to hands and legs, then to abdominal region and to the back. She also developed ulceration of buccal mucosa and desquamation of tongue.

She complained of pain in joints of both hands and legs. Physical examination revealed cutaneous involvement of trunk, face, lips, palms and soles. Patient had erythema and multiple vesicles in the oral mucosa and involving most of her face. Few disrupted vesicles with crusting were also present around the lips, nose and forehead. Conjunctival congestion was present. Bullous lesions with erosions and peeling were present bilaterally on both the legs and trunk. The vital signs recorded were as follows: temperature (axillary)-39.5°C; pulse rate- 92/min; respiration rate-16/min. Blood Pressure (BP) was 160/80 mmHg. On taking history, patient’s attendant told that she was...
taking Tab. Methotrexate 7.5 mg per week for 2 weeks as a treatment for Rheumatoid arthritis which was prescribed by a local practitioner. The patient was not suffering from any kind of infection or any other chronic disease. Patient was diagnosed with Drug Induced Stevens Johnson Syndrome. She was immediately admitted in emergency ward and given fluid resuscitation with normal saline, intravenous corticosteroids (Hydrocortisone Hemi succinate 100 mg I.V), antihistamines (Avil) and supportive medication. (Ranitidine 50 mg i.v, Benzocaine gel for oral ulcerations). All the previous medications taken by the patient were discontinued. Laboratory investigations revealed haemoglobin 10.04 g/dl, white blood cell count 5890 mm3, platelet count 180,000 mm3, serum creatinine 1.1 mg/dl, blood urea nitrogen 17 mg/dl, liver function test was normal. Pus culture was positive for gram positive cocci and coagulase negative Staphylococcus. Blood and urine cultures were sterile. Chest X-ray and ultrasound abdomen were normal. Gradually the condition of the patient improved and after 3 days, crusting of rashes, bullae and blisters started developing and itching, oedema and erythema subsided.

The Naranjo adverse drug reaction probability scale score of six indicated a 'Probable' relationship between Stevens Johnson Syndrome and Methotrexate therapy in this patient.(4) WHO Uppsala Monitoring Centre Causality Assessment Criteria (5) also indicated a 'Probable' association with Methotrexate.

**DISCUSSION**

Stevens-Johnson syndrome (SJS) is a rare but severe cutaneous adverse reactions (SCARs) which cause significant morbidity and mortality (6). The aetiology of SJS and TEN is not clear and could be due to drug induced immunological mechanism. CD8 T-cells as well as the cytolistic molecules Fas ligand (FasL) and granulysin are key substances in the pathogenesis of SJS/TEN, but the manner in which a culprit drug regulates the function of these key substances in a patient who develops SJS/TEN is the subject of ongoing research (7). There are four causative categories which includes (a) infectious (b) drug-induced (c) malignancy-related (d) idiopathic (8).

SJS is classified as (7) –
- Stevens-Johnson syndrome: A minor form of toxic epidermal necrolysis (TEN), with less than 10% body surface area (BSA) detachment
- Overlapping Stevens-Johnson syndrome/toxic epidermal necrolysis: Detachment of 10-30% of the BSA.
- Toxic epidermal necrolysis: Detachment of more than 30% of the BSA.

The most frequently involved groups of therapeutic agents cited in the literature that induce SJS include sulphonamides, anticonvulsants, non-steroidal anti-inflammatory drugs beta lactam antibiotics, carbamazepine, valproic acid, lamotrigine, barbiturates etc (8). It has been recognized that drug-induced SJS is a severe hypersensitivity reaction which involves major histocompatibility class -I (MHC) restricted drug presentation and cytotoxic T lymphocytes (CTLs) expansion, which further leads to extensive keratinocyte death in skin lesions. (9)

Methotrexate has greater affinity than folic acid for the dihydrofolate reductase enzyme. This mechanism of methotrexate reduces the tetrahydrofolate and attenuates the DNA synthesis in proliferating cells, making it an ideal disease modifying agent for rheumatoid arthritis. Methotrexate is metabolized into a series of polyglutamate derivatives that increase linearly with the concentration and duration of exposure (2). Therefore, higher doses of methotrexate administered for prolonged duration can result in greater toxicity. Methotrexate induced SJS is a rare adverse drug reaction which we have highlighted in this case report. Cases of SJS/TEN are primarily induced by medications.

ALDEN (Algorithm for Drug causality in Epidermal Necrolysis) has been used to provide structured assistance for the assessment of culprit drugs in SJS/TEN patients (10).

A disease severity scoring system called SCORTEN (SCORE of Toxic Epidermal Necrolysis) built on seven independent variables (11) -
- Age more than 40 years
- Malignancy
- Heart rate >120/minute
- Initial epidermal detachment >10% of BSA
- Serum urea level >28 mg/dl (40 mg/dl in Indian settings)
- Serum glucose levels >250 mg/dl
- Serum bicarbonate levels <20 mEq/dl.

The probability of death predicted by this score is as follows: 0-1 points - 0.03; 2 points - 0.12; 3 points - 0.35; 4 points - 0.58; 5 to 7 points - 0.90. A probability of 0.90 means approximate 90 of 100 patients with TEN are expected to die.

SJS and TEN may be a dose dependent adverse drug reaction in susceptible individuals. In spite of MTX having a narrow therapeutic index, serum plasma concentration of MTX is not regularly monitored by the treating physicians. Folic acid should be supplemented along with methotrexate therapy is recommended by American College of Rheumatology to prevent the complications of methotrexate therapy (12).

CONCLUSION

The main intention of this case report of Methotrexate induced SJS is to make the clinicians as well as patients aware of this ADR occurrence. It is advocated that clinicians take a proper history before prescribing Methotrexate as a DMARD and early diagnosis and treatment might improve the outcome and decrease mortality in many patients of SJS. Patient education regarding the possibility of adverse drug reaction is essential to minimize the use of the drug.

REFERENCES


ABSTRACT
Renal cell carcinomas (RCC) are the most common solid lesions of kidney with commonest subtype being clear cell type. Very few studies have reported synchronous presentation of three different morphological variants of RCC. We present a case of renal cell carcinoma in a 50 year old female presenting with renal mass. Microscopic examination showed presence of papillary, clear cell and collecting duct types of morphologies, which is a rare finding. Hence thorough sectioning and microscopic examination should be done to rule out possibility of simultaneous presence of different morphological varieties of RCC.

KEYWORDS: Renal cell carcinoma, Papillary type, Clear cell type, Collecting duct type.

INTRODUCTION
Renal cell carcinoma (RCC) is the most common solid lesion of the kidney and accounts for approximately 2-3% of all malignancies in adults. (1)

The commonest subtypes of renal cell carcinoma are clear cell, papillary, and chromophobe type RCC and account for approximately 80%, 10%, and 5%, respectively. Among the benign neoplasms, renal angiomyolipoma (AML), and renal adenomas, oncocytoma are common. There are a few of studies that define bilateral synchronous malignant renal tumors (2–4) or coexisting benign and malignant tumors arising within the same kidney (5). Collecting duct carcinoma (CDC), also known as Bellini duct carcinoma is thought to arise from the collecting ducts of renal medulla, however it is a rare tumour and comprises less than 1% of renal epithelial neoplasms. (6,7)

To the best of our knowledge, Collecting duct RCC, clear cell RCC and papillary RCC arising within the same kidney are very rare in the literature.

Herein, we describe a case of a 50-year-old Female who had 3 different subtypes of renal cell carcinoma in the same kidney who underwent radical nephrectomy (RN).

CASE REPORT
A 50-year-old Female was admitted to another medical center with right flank pain lasting for more than one month. An ultrasound scan revealed a mass in the right kidney.

Thorough history taking and examination was performed along with relevant investigations, and surgical resection was planned.

The tumor was clinically diagnosed as a right renal tumor and classified as cT1bN0M0, according to tumor- node-metastasis system. Patient underwent right RN and adrenalectomy. The specimen was sent for histopathological examination.

On macroscopic examination the kidney was distorted with attached ureter altogether measuring 14.10.8 cm. Cut surface showed a mass at one pole measuring 6.5X7.5cm. which was well circumscribed with grey white to grey brown areas of necrosis. It appeared to push the other half of the kidney and pelvis which were compressed. Outer fatty tissue was grossly free from tumour invasion. Multiple sections were taken.

Sections from the tumor showed atypical cuboidal cells arranged in the form of papillae. These atypical cells had increased nucleocytoplasmic ratio, nuclear pleomorphism, clumped chromatin and scant cytoplasm, nuclear morphology was consistent with Fuhrman grade 2. The intervening stroma showed fibrocollagenous core which was infiltrated by lymphocytes and macrophages.

Focal area showed large clear cells which had shortly outlined boundaries interspersed by prominent network of delicate blood vessels and had Fuhrman grade 2 morphology with finely granular chromatin but small nucleoli that were not discernible at 10x magnification. Also seen were atypical cells in the form of branching tubules. Large areas of necrosis were also seen. Section from Perinephric fat showed infiltration by similar...
atypical cells. The tumor invaded the renal capsule and extended into the perirenal fat.

Sections from pelvis, renal artery and vein were free from tumor.

**DISCUSSION**

RCC comprises 2-3% of all cancers (1). The incidence of RCC has also risen over the past several decades due to incidental detection [8]. The best known etiological factors for all types of RCC are smoking, obesity, and hypertension (9). Among the documented etiologic causes that were described above, no known etiologic factor was present in our case.

RCC comprises several subtypes with specific histopathology and genetic characteristics, the most commonly diagnosed including clear cell, papillary, and chromophobe, collecting duct. Clear cell RCC has clear cytoplasm with solid, tubular, or cystic growth pattern. Two different papillary tumor subtypes have been defined. Both clear cell and papillary types of RCC originate from proximal tubules. Collecting duct RCC shows tubulopapillary architecture, atypical hyperplastic changes, clear cytoplasm, evident stromal reaction with fiber hyperplasia and detached single cells with a hobnail surface (10).

CDC is a rare pathologic type of RCC, with a tendency towards early dissemination and high mortality rates (11,12) The majority of CDC tumors have been found to demonstrate focal cortical extension, while perirenal invasion was also common in large tumors [13]. This was a case of papillary and collecting duct and clear cell RCC in the same kidney and in a single tumor mass which also involved the perinephric fat.

AML and RCC have been defined many times in the literature in tuberous sclerosis (TSC) and non-TSC patients.

Billings et al. defined an 86-year-old woman without TSC with a coexisting 7 cm clear cell RCC and 9.5 cm AML in the same kidney that were treated with right RN successfully. (5)

Khallouk et al. defined a case report of a 35-year-old male with TSC and bilateral massive AML. They performed right radical nephrectomy successfully and pathology revealed AML and clear cell RCC in the same kidney. (14)

In addition to coexisting benign and malignant tumors in the same kidney, there have been described some reports that define the coexisting 2 different types of RCC.

Simhan et al. reported the data of 97 patients who had multifocal renal tumors. They reported 8 patients who had mixed (papillary and clear cell) RCC, all of which were treated by partial nephrectomy (15).

In the case reported by Kawano et al. the most predominant histological component was the chromophobe renal cell carcinoma(CRCC). The chromophobe cells also showed dedifferentiation. Besides this component, the CDC component was also noted, and the CRCC and CDC elements showed obvious transition to each other (16).

Roehrl et al. reported another case of RCC that exhibited the features of both chromophobe and papillary carcinoma within the same tumor (17).

Capaccio et al. found 7 patients who had unilateral synchronous tumors with different subtypes. One of them had oncocytoma and one had clear cell RCC with synchronous AML.

Remaining 5 patients had different histological subtypes of RCC, 3 of which had synchronous papillary and clear cell type RCC. The other 2 patients had chromophobe subtype RCC with unilateral synchronous papillary type in 1 patient and clear cell type RCC in 1 patient (18).

In fact, there is not sufficient data to compare the different types of RCC in the same kidney.

Patel et al (19) found that malignant concordance was 89% among the patients who had bilateral synchronous renal tumors. On the other hand, there is no such data for unilateral synchronous different type of RCC.

**CONCLUSION**

Since three different subtypes of RCC have seldom been reported, our case presents a rare finding. So proper and thorough grossing should be done in cases of suspected RCC. However large number of studies are needed to make a comparison and comment on course of disease because this is a case without any controls or comparisons, so clinical implication is limited. However, synchronous 3 different types of RCC in same kidney should not change the management approach.

![Fig 1: Gross appearance of the renal mass showing complete loss of normal renal contour.](image)
Fig 2: Gross appearance of cut surface of the renal mass showing loss of cortico-medullary differentiation and a yellowish mass occupying almost whole of kidney.

Fig 3: Photomicrograph showing collecting duct type of morphology (H&EX100)

Figure 4: Photomicrograph showing collecting duct type of morphology (H&EX400)

Fig 5: Photomicrograph showing presence of papillae lined by 2 cell thick layer and a core of macrophages (H&EX100)

Fig 6: Photomicrograph showing papillae in high power view with core of macrophages (H&EX400)

Fig 7: Photomicrograph showing nests of clear cells with clear cytoplasm and central nuclei (H&EX100)
REFERENCES


ABSTRACT

Two staged bilateral total hip replacement (THR) is commonly performed for bilateral hip end stage arthritis and is preferred as THR is a complex planned surgery and performing both sides simultaneously may be fraught with risks and complications. However, many studies now indicate that in carefully selected patients, single stage or simultaneous bilateral THR can be performed with successful and cost effective results. We report a case of one-stage bilateral THR performed in a 22 year old with bilateral severe arthritis due to ankylosing spondylitis. Patient was severely disabled due to pain and was only ambulating on wheelchair. After a successful single stage bilateral THR, patient recovered fully and after 6 weeks was walking independently without any pain, with full function of both hips and performing his occupation normally. The surgical costs to the patient and hospital were both economical. We conclude that single stage bilateral THR is a better surgical option for young and fit patients with bilateral hip arthritis.

KEYWORDS: Ankylosing spondylitis, bilateral hip end stage arthritis, Bilateral total hip replacement, Single stage bilateral total hip replacement

INTRODUCTION

Total Hip Replacement (THR) has become one of the most satisfying surgeries for patients with end stage arthritis of hip joint to enable them to lead a painless, independent and fully functional life. The increasing incidence of bilateral hip involvement especially in younger population due to diseases like avascular necrosis of femoral head (AVN) and inflammatory arthritis like ankylosing spondylitis (AS) and rheumatoid arthritis have led to a significant painful disability in this age group (1). The incidence of bilateral involvement of hip joint ranges from about 40-70% in AVN (2) and about 80-90% in AS patients (3). These diseases have led to advent of the idea to perform single stage or simultaneous un cemented bilateral THR surgery to give a one stop solution to these severely disabled young patients. There is still no clear clinical consensus in the literature on which is better, between single stage bilateral versus staged bilateral THA procedures. Some studies have reported higher incidence of complications after single stage bilateral THR (4). Increased blood loss, heterotopic ossification, higher prevalence of deep vein thrombosis and greater risk of pulmonary complications are amongst the main reported complications after single stage bilateral THR (5-7). These results have become much better with improved anaesthetic and surgical techniques and postoperative care. The major benefits of bilateral single stage uncemented THR include single anesthesia, shorter surgical time, more efficient use of hospital resources, reduced hospitalization, cost effectiveness and shorter rehabilitation (6). Also, the improvements in various elements of walking are reportedly higher in patients with bilateral THR than in those with unilateral staged replacement with resultant better hip function (7).

We hereby present a case of single stage bilateral cementless THR performed in a young patient suffering from ankylosing spondylitis. This bilateral THR in a single anesthesia was performed first time in our region. We hereby report the surgical details and the functional results after 2 years of follow up.

CASE REPORT

A 22 year old patient, school clerk by profession, belonging to Hardoi district came to Era's Lucknow medical college orthopedics outdoor service on a wheelchair with severe bilateral hip end stage arthritis due to ankylosing spondylitis. He had early involvement of cervical spine and severe back pain too. He was not able to ambulate independently since about 6 months and was in disabling pain. He was diagnosed with ankylosing spondylitis in 2014 and due to improper treatment and disease severity his hips were involved early and were totally arthritic. In September 2016, he was admitted to the indoor service for detailed examination and proper management planning.
On examination, he had severe bilateral hip arthritis with beginning of fusion in right hip joint. Both hips were painfully stiff with left hip having some limited range of motion and very poor Harris hip scores of 17 in right hip and 19 in left hip. (Table 1).

The antero-posterior radiograph of both pelvis with both hips and thighs in neutral rotation showed end stage arthritis of both hips (Figure 1).

As the patient was young, healthy and with normal hematological investigations after a detailed examination by anesthetist, he was graded as ASA (American Society of Anaesthesiologists) I fitness. A joint decision of surgical team, the anesthetist team, the operation theater staff, implant team and patient with his family members to perform a single stage bilateral uncemented total hip replacements.

It was deemed that the surgery would be single stage, single anesthesia, cost effective and with a single post op physical therapy plan offsetting the need of second surgery after 3 months and leading to a complete functional recovery.

**Operation**

Under Spinal Epidural anesthesia, first the left hip was operated with a standard posterolateral approach in lateral position and uncemented total hip replacement was done with total surgical time of 65 minutes. Then the patient was made supine again with careful monitoring of the patient by the anesthesia team and after a break of 15 minutes, the right hip was prepared and scrubbed and uncemented total hip replacement surgery was performed on the right hip with total surgical time of about 63 minutes. The total blood loss in the whole procedure was about 500 ml and there was a single unit transfusion given to promote early recovery. The patient tolerated the whole procedure very well and was shifted to the post op recovery room in a stable condition.

**Post op care and follow up**

He was given post op antibiotics for a period of 5 days and under adequate pain control he was able to perform bilateral quadriceps strengthening exercises from Day 1 of surgery and by day 4, he was ambulating pain free with walker, he was discharged on Day 5 and had complete functional recovery with joining of his clerical duties after 6 weeks of surgery.

On his 6 weeks post op evaluation, he had full functional recovery with the Harris hip score of 100 (Table 2) and at the last 2 year follow up, he was doing extremely well with full function and is very happy with his recovery. His latest radiograph showed well aligned and well fixed acetabular and femoral prosthesis in situ on both sides (Figure 2).

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**Table 1. Functional Evaluation Of The Patient, 22 Years Male (preoperatively)**

<table>
<thead>
<tr>
<th>Particulars</th>
<th>Right Hip</th>
<th>Left Hip</th>
</tr>
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<tbody>
<tr>
<td>Flexion</td>
<td>30°</td>
<td>50°</td>
</tr>
<tr>
<td>Extension</td>
<td>0°</td>
<td>0°</td>
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<tr>
<td>Abduction</td>
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</tr>
<tr>
<td>Adduction</td>
<td>10°</td>
<td>10°</td>
</tr>
<tr>
<td>Internal Rotation</td>
<td>0°</td>
<td>0°</td>
</tr>
<tr>
<td>External Rotation</td>
<td>10°</td>
<td>10°</td>
</tr>
<tr>
<td>Flexion Deformity</td>
<td>20°</td>
<td>20°</td>
</tr>
<tr>
<td>Harris Hip Score</td>
<td>17</td>
<td>19</td>
</tr>
</tbody>
</table>

**Table 1. Functional Evaluation Of The Patient, 22 Years Male (postoperatively: 6 Weeks Follow Up)**

<table>
<thead>
<tr>
<th>Particulars</th>
<th>Right Hip</th>
<th>Left Hip</th>
</tr>
</thead>
<tbody>
<tr>
<td>Flexion</td>
<td>120°</td>
<td>120°</td>
</tr>
<tr>
<td>Extension</td>
<td>30°</td>
<td>30°</td>
</tr>
<tr>
<td>Abduction</td>
<td>40°</td>
<td>40°</td>
</tr>
<tr>
<td>Adduction</td>
<td>40°</td>
<td>40°</td>
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<tr>
<td>Internal Rotation</td>
<td>30°</td>
<td>30°</td>
</tr>
<tr>
<td>External Rotation</td>
<td>30°</td>
<td>30°</td>
</tr>
<tr>
<td>Flexion Deformity</td>
<td>0°</td>
<td>0°</td>
</tr>
<tr>
<td>Harris Hip Score</td>
<td>100</td>
<td>100</td>
</tr>
</tbody>
</table>
DISCUSSION

THR is one of the most versatile orthopedic procedures with gratifying results for both the surgeons and the patients. With serious joint diseases like AVN and ankylosing spondylitis affecting bilateral hips in young patients, the need for bilateral THR is increasing exponentially for these patients to lead a pain-free and independent life.

Recent literature evidence suggests that simultaneous bilateral total hip arthroplasty is a valuable therapeutic option in appropriately selected patients with bilateral hip arthritis. We believe that in properly selected patients with severe bilateral arthritis and after close consultation with our anesthetic and specialized joint replacement operative team, doing one-staged THRs were better, since both hips were equally arthritic, and replacing one hip would not have achieved the results that we otherwise did, since the gaits would have still been compromised after unilateral surgery. Secondary benefits reported from studies included less use of resources, quicker recovery, less hospital stay, and hence more economical. The cost data showed that in a theoretical model, comparing one- and two-staged procedures showed a 24% reduction in hospital and sick-leave costs in favour of the one-stage bilateral THRs in healthy patients. Also improved balance and walking ability is noted as early as 4 weeks after bilateral THR.

Our patient returned to work within 4 weeks with independent ambulation and had a complete recovery at the follow-up period of 6 weeks with Harris hip scores improving from below 20s to a normal 100. There were no complications observed till the latest follow-up of 2 years.

CONCLUSION

We recommend single stage bilateral uncemented THRs in cases of severe functional disability due to bilateral end stage arthritis of both hips in young patients (< 45 years) who are medically fit, well informed, motivated and well versed with the
postoperative rehabilitation plan. The added benefit of single anesthesia, cost savings for both the hospital and patient and rapid functional recovery is very important in our socio-economic conditions.

Conflict of Interest
The authors declare that they have no conflicts of interest in relation to this article.

REFERENCES
SPHENOCHOANAL POLYP RIGHT WITH LEFT SEPTAL SPUR: RARE ENTITY

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ABSTRACT
Choanal polyps can be classified as antrochoanal, sphenochoanal or ethmochoanal polyps depending upon the site of origin, in comparison to common nasal polyps and antrochoanal polyps, reports of sphenochoanal polyps are relatively rare. Authors present a case of a 18 year old boy presenting to the clinic with unilateral nasal obstruction and nasal tone in voice, which was a diagnostic dilemma for the radiology department as it was misdiagnosed unable to be differentiated by CT scan. MRI scan of the patient confirmed the diagnosis of a sphenochoanal polyp. We aspire to widen the horizon of the knowledge by discussing the case and enabling timely diagnosis and treatment.

KEYWORDS: Sphenochoanal polyp, Sphenoid sinus, Endoscopic sinus surgery.

INTRODUCTION
Choanal polyps are benign soft tissue masses arising from isolated paranasal sinuses, emerging through the ostia and extending to the posterior choanae. They are classified as antrochoanal, sphenochoanal or ethmochoanal polyp depending on the site of origin(1,2). Choanal polyps represents 3-6% of nasal polyps(3). These polyps, which occur in the setting of an otherwise normal sphenoid sinus and are more aptly, termed sphen-o-ostio-choanal which are extremely rare and are a curious entity. Clinically, the glistening and pale masses are similar to a typical nasal polyp and nasal endoscopy may show a site of origin. Here, we report a new case of a right sphenochoanal polyp that was surgically managed by a microdebrider assisted functional endoscopic sinus surgery.

CASE REPORT
An 18 year old boy presented to the ENT outpatient department with right sided nasal obstruction and change in voice. The patient was apparently asymptomatic 2years back when he developed a gradually worsening right sided nasal obstruction. There was also complained of nasal twang in voice for the past 1 month. There was no history of epistaxis, hyposmia or anosmia, diminished vision, facial pain. On anterior rhinoscopy there was a single glistening polypoidal painless mass present in the right nasal cavity which did not bleed on touch along with left side deviated nasal septum. Endoscopy showed a glistening polypoidal mass seen occupying the sphenoeethmoidal recess in the 2nd PASS area. 1st and 3rd PASS areas were found to be normal. Contrast enhanced computer tomography of the para nasal sinuses showed a homogenous space occupying lesion arising from the sphenoid sinus, extending through the sphenoid ostium, across the sphenoid recess, and into the choana without any bony erosion. Magnetic resonance imaging showed an area of altered signal intensity arising from sphenoid with extension into the oropharynx, nasopharynx, and nasal cavity, suggestive of sphenoidal polyp.

The patient was planned for a functional endoscopic sinus surgery under general anaesthesia. A microdebrider assisted Functional endoscopic sinus surgery was done which included Anterior ethmoidectomy with Posterior ethmoidectomy and Sphenoid sinusotomy with removal of sphenochoanal polyp. The complete polyp was delivered via the oral route, followed by inspection of sphenoid sinus which was found to be clear of any polypoidal mass. Specimen was sent for histopathological examination, which showed an inflammatory polyp without any signs of malignancy. Intranasal packing was done, and was removed 48 hours after the surgery following which the patient was discharged from the hospital.
Nasal endoscopy was done as part of follow up at 1 week, 2weeks and 4weeks post surgery. The patient did not have any nasal complaints and nasal cavity showed healthy tissue with no signs of disease recurrence even after 1 month of follow up.
DISCUSSION

The first description of sphenchoanal polyp was attributed to ZuckerkandL in 1892 (2,4). Sphenchoanal polyps originate from the sphenoid sinus and present with nasal obstruction. They are rare as compared with antrochoanal polyps and are mostly seen in adolescents and young adults without gender predominance (5). Unilateral nasal obstruction is the most common presenting symptom, other presenting symptoms include purulent nasal discharge, epistaxis, retro orbital pain, epistaxis. Eustachian tube dysfunction, snoring and obstructive sleep apnoea have been reported (6-9).

A mass arising from sphenoid sinus must raise suspicion of neoplasm. A review of series of isolated sphenchoanal masses found that pathologically, 3 cases were of inflammatory polyps and the other 3 of neoplasms 2 cases of inverted papilloma and 1 of pituitary adenoma (10). This high percentage of neoplasia illustrates the importance of obtaining a histological diagnosis in cases of isolated nasal cavity masses that involve sphenoid sinus. Other diagnosis to consider in the older patients include squamous cell carcinoma, adenocarcinoma, lymphoma or metastatic disease (3).

Although the exact pathogenesis of sphenchoanal polyps is unknown they seem to have same pathogenesis of antrochoanal polyps (4). Inflammation caused by the sinus infection is supposed to be the main triggering causative factor. Since maxillary sinusitis is more common than sphenoid sinusitis sphenchoanal polyps are seen less commonly seen than antrochoanal polyps (11). Allergic rhinitis is not a predisposing factor for choanal polyps. Scarcity of eosinophil infiltration, higher number of other inflammatory cells, normal appearance of both basement membrane and surface epithelium with transmission electron microscopic evaluation may indicate that the pathogenesis of choanal polyps is chronic inflammation rather than allergy (12). Occasionally, these choanal polyps may undergo angiomatous degeneration as a result of vascular compromise due to passing through a tight ostia. Compression of a feeding vessel induces stasis of blood flow and subsequent oedema and dilatation of the polyp. Hypervascular state of some of these choanal polyps needs differential diagnosis of an angiofibroma (13).

Choanal polyps have been usually observed in three forms: antrochoanal polyp, sphenchoanal polyp and ethmoido-choanal polyp, the latter being very rare. However, choanal polyps have not been reported to originate from frontal sinus. It is difficult to identify the sinus of origin of choanal polyp with anterior rhinoscopy or plain radiography. Site of origin of choanal polyps are mostly evaluated by nasal...
endoscopic examination and CT findings. Nasal endoscopic examination demonstrates a solitary solid polypoid lesion in the posterior part of the nasal passage but with only nasal endoscopy it is hard to differentiate a sphenochoanal polyp from other polyps(4). CT is helpful for differentiating a sphenochoanal polyp from and antrochoanal polyp. In antrochoanal polyp, maxillary sinus is full filled since the polyp extends to nasopharynx lateral to the middle concha, the area between the middle concha and nasal septum is spared. For sphenochoanal polyp cases the polyp occupies sphenethmoidal recess and area between the nasal septum and middle concha is occupied (14,15). CT and MRI are important in the evaluation of other sphenoid sinus pathologies that mimic sphenochoanal polyps. The differential diagnosis should include mucocele, inverted papilloma, Juvenile Nasal Angiofibroma and meningoencephalocele (6,7,9). Misdiagnosis may result in inadequate treatment and surgical complications.

Though there have been reports of regression of choanal polyps using medical treatment(16,17), it is generally agreed that surgical intervention is the current standard management(1,7,17,18). Most authors recommend total removal under endoscopic guidance, as simple polypectomy alone carries a higher risk of recurrence(1,4,6,7,18). Preceding correction for deviated nasal septum and resection of anteroinferior part of superior turbinate affords sufficient exposure of the sphenoid sinus. After widening the sphenoid sinus ostium, any cystic component of the polyp attached to the sinus wall must be totally removed to prevent recurrence. A microdebrider can achieve this objective and preserve adjacent normal mucosa. As for the location of the polyp origin in the sinus most cases have been found to originate from the floor(19) or the latero-inferior aspect(20), with only one case originating from the roof(9). In our case the origin of pedicle was found to be rom the lateral wall. Great attention should be exercised to avoid violating the vital structures such as the optic nerve, internal carotid artery and pituitary gland.

CONCLUSION

Patients complaining of progressive unilateral nasal obstruction should be evaluated on the basis of choanal polyps. Antrochoanal polyps being the commonest entity. Sphenocoanal polyp is a rare occurrence and should be distinguished from other commoner disorders leading to proper management of patient and preventing unnecessary exploration of the sinuses.

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