BANGLADESH JOURNAL OF NEUROSURGERY

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CONTENTS

| Editorial | 29 |
|---|----|
| Original Articles Primary bone tumour of the spine: Surgical treatment and Histopathological analysis Md. Shafiul Alam, Md. Shafiqul Kabir Khan, Monsur Ahmed, Muhammad Shamsul Arefin, Kazi Hafiz Uddin, Mohammad Hossain, Md. Rezaul Amin | 3 |
| Outcome of Conservative Management of Traumatic Acute Subdural Haematoma in Relation to Underlying Brain Injury Sanat Kumar saha, Mohammad Abu Sayed, Joynul Islam, Shafiul Alam, ATM Asadullah, Misbah Uddin Ahamed, Shamsul Islam Khan | 3 |
| Signs and Symptoms of Early VP Shunt Infection in a Tertiary Hospital Kaisar Haroon, Tania Taher, Mokhlasur Rahman, Kanak Kanti Barua | 4 |
| Assessment of Serum Homocysteine Level in Epileptic Patients on Carbamazepine Treatment Md. Abu Hanif, M A Hannan, M Iqbal Arslan, SK Mahbub Alam, Mohammad Hossain, Md. Aynul Hoque, Niloy Ranjan Roy | 44 |
| Case Reports Tubercular Spinal Epidural Abscess Involving the Lumbo-Sacral Region: A Case Report with Review of Literature Akhalaque Hossain Khan, Ranjit Kumar Chaurasiya, Narendra Shalike, Mohammed Aminul Hasnat, Bipin Chaurasia, Abhishek Chaturbedi, Asifur Rahman, Rezaul Amin | 48 |
| Giant Ganglioneuroma of Thoracic Spine: A Case Report Das S, Alam K, , Rashid MM, Islam MM, Mahbub H, Khan MSI | 5 |



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BANGLADESH JOURNAL OF NEUROSURGERY

Volume 5, Number 2, January 2016

OFFICIAL ORGAN OF BANGLADESH SOCIETY OF NEUROSURGEONS

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| |
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| or submitted for publication elsewhere. |
| We believe that this article may be of value to medical professionals engaged in Neurosurgery. |
| We therefore, hope that you would be kind enough to consider our manuscript for publication in your journal as original / Review article / Case Report. |
| |

Thanks and best regards

More than 17,000 people in the United States and 18,000 in India are diagnosed each year with a brain tumor. Unfortunately we do not have any accurate data for Bangladesh.

Some tumors are benign (noncancerous). Noncancerous tumors can usually be removed and are not likely to recur. Other tumors are malignant (cancerous). These tumors interfere with vital functions and are life threatening. Cancerous brain tumors usually grow rapidly, crowding and invading tissue.

Primary brain tumors (gliomas) start in the brain and affect the central nervous system (CNS). They can be noncancerous or cancerous. Secondary brain tumors, which are 10 times more common, are cancers that originated elsewhere in the body and have metastasized (spread) to the brain. Secondary tumors are about 3 times more common than primary tumors of the brain.

People with the following conditions or characteristics may be at risk for developing a brain tumor:

- · Radiation exposure
- · Exposure to pesticides, herbicides, fertilizer
- Certain occupations, such as lead, petroleum, plastic, rubber, and textile workers, as well as aircraft and vehicle operators
- · Exposure to electromagnetic fields
- · Certain viruses, especially Epstein-Barr virus
- · Certain genetic disorders
- People who have had transplants and individuals with AIDS
- A small percentage of brain tumors may be hereditary (approximately 5% of gliomas)
- People over age 45

Studies have not shown an association between brain cancer and cell phone use.

Now it will be the right time to ask all the neurosurgeons of Bangladesh to take an initiation in opening and maintaining a combined central brain tumor registry.

Original Articles

Primary Bone Tumour of the Spine: Surgical Treatment and Histopathological Analysis

Md. Shafiul Alam¹, Md. Shafiqul Kabir Khan², Monsur Ahmed³, Muhammad Shamsul Arefin⁴, Kazi Hafiz Uddin⁵, Mohammad Hossain⁶, Md. Rezaul Amin⁶

Abstract

Background: Primary bone tumour of the spine are rare but not uncommon. These include both benign and malignant lesions. Surgical resection and stabilization in some cases are the treatment of choice. Adjuvant radio and/or chemotherapy are needed in malignant cases.

Methods: We retrospectively reviewed all the primary bone tumour of the spine that were surgically treated in the department of Neurosurgery of Dhaka Medical College Hospital and National Institute of Neurosciences and Hospital Dhaka, Bangladesh between 2010 and 2015.

Results: Eighteen cases were included that presented at a median age of 36 years (range 14 to 49 years). Pain was the most common presenting symptom. Three patients had significant neurological deficits at time of presentation and in most cases there was an improvement after surgery. A variety of surgical strategies was employed with the use of adjuvant radio- or chemotherapy in six cases. Six benign and twelve malignant tumours were treated.

Conclusions: Primary bone tumours of the spine are often associated with a significant delay in diagnosis. Surgical strategy should be individualized for each case. Acceptable results can be achieved with early surgery and adequate chemo-radiotherapy.

Key words: primary bone tumour, spine, surgical treatment, histopathology.

Bang. J Neurosurgery 2016; 5(2): 31-36

Introduction:

Primary bone tumour of the spine is a heterogynous group of conditions that includes both benign and malignant tumours, with only 6% of these tumours considered malignant. The spine is a frequent site of metastasis, but primary bone tumours of the spine are rare and make up only 4.2% of spinal tumours. Eleven per cent (11%) of primary musculoskeletal tumours occur in the spine with certain tumours having an affinity for the spine. Although rare, these tumours offer a considerable diagnostic dilemma and therapeutic challenge. Timely diagnosis is crucial, as immediate attention could affect overall prognosis.³

The most common primary tumour of the spine is metastatic deposits. A number of both benign and

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malignant tumours may arise primarily from the spine. Among benign tomour osteoid osteoma, osteoblastoma, osteochondroma, giant cell tumour (GCT), aneurysmal bone cyst (ABC), eosinophilic granuloma (EG), haemangioma are common. Among malignant lesion malignant lymphoma, sarcoma, ewing sarcoma, osteosarcoma, chondrosarcoma, malignant fibrous histiocytoma (MFH), fibrosarcoma, chordoma, plasmacytoma/multiple myeloma and peripheral primitive neuroectodermal tumour (pPNET) are common.

Primary bone tumours are more common in the thoracic and lumbosacral regions than in the cervical spine.⁴ These tumours occur according to a typical anatomical distribution within the vertebra. In general, malignant tumours occur more frequently in the anterior elements and benign tumours in the posterior elements.^{3–5}

The aims of treatment of these axial neoplasms include complete resection if feasible, preservation of neurological function and stabilisation of the spine if needed.⁴ The aim of our study was to review the primary bone tumours of the spine treated at our unit regarding the presentation, the surgical strategy employed and the outcome.

Materials and Methods:

This is a retrospective study. All the primary bone tumours of the spine that received surgical treatment

at the department of Neurosurgery of Dhaka Medical College Hospital and National Institute of Neurosciences and Hospital Dhaka, Bangladesh from 2010 to 2015 were included in this study. We excluded all secondary deposition in the spine. We treated 18 cases during this period that were included according to these criteria. A case note and imaging review was done where patient demographics, details of presentation, clinical information and the management strategy were analyzed. All complications and mortalities were recorded.

Results:

Between 2010 and 2015, a total 18 patients were diagnosed as primary bone tumour of the spine. The median age at presentation was 36 years (range 14 to 49 years) with eleven(61.12%) male and seven(38.88%) female patients affected. The median follow-up was 18 months (range 3 to 60 months).

Table-IDistribution of patient by age.

| Age group | Frequency | Percentage |
|----------------|-----------|------------|
| 0 to 10 years | 0 | 0 |
| 11 to 20 years | 3 | 16.67 |
| 21 to 30 years | 7 | 38.89 |
| 31 to 40 years | 4 | 22.22 |
| 41 to 50 years | 4 | 22.22 |
| Total | 18 | 100 |

Table-IIDistribution of patient by sex.

| Sex | Frequency | Percentage | |
|--------|-----------|------------|--|
| Male | 11 | 61.12 | |
| Female | 7 | 38.88 | |
| Total | 18 | 100 | |

Table-IIIDistribution of patient by site of involvement.

| Site of involvement | Frequency | Percentage |
|---------------------|-----------|------------|
| Cervical | 1 | 5.55 |
| Dorsal | 7 | 38.89 |
| Lumber | 6 | 33.34 |
| Sacrum | 4 | 22.22 |
| Total | 18 | 100 |

The most utilized diagnostic tool was MRI and CT scan of the spine. It helped to diagnose the case as well as planning of surgery. X ray is no longer a standard diagnostic tool of spine tumour, it was used in all cases as c complimentary diagnostic tool. It also helped to find out the extra spinal involvement like rib cases in two cases. Pre-operative CT guided FNAC was done in seven cases. Post-operative histopathological study was done in all cases.

Primary bone tumour of the spine most commonly involved the dorsal (38.89%) and the Lumber (33.34%). It affect the sacral (22.22%) and the cervical spine (5.55%) less frequently. (Table-3)

Table-IVDistribution of patient by histopathological findings.

| Туре | Histopathology | Frequency | percentage |
|-----------|------------------------|-----------|------------|
| Benign | -Osteoid osteoma | 1 | 5.55 |
| | -Haemangioma | 1 | 5.55 |
| | -Giant cell tumour | 1 | 5.55 |
| | -Aneurysmal bone cyst | 2 | 11.11 |
| | -Esonophilic granuloma | 1 | 5.55 |
| Malignant | -Myxoid Chondrosarcoma | a 1 | 5.55 |
| | -ewing's sarcoma | 2 | 11.11 |
| | -Chondrosarcoma | 2 | 22.22 |
| | -Chordoma | 3 | 33.33 |
| | -Multiple myeloma | 2 | 22.22 |
| | -pNET | 2 | 22.22 |
| Total | | 18 | 100 |

Pain was the most common presenting symptom (15 from 18 cases) and four cases presented with pain alone. Pain was mostly axial pain in the affected region (11 of 18 cases), although three patients presented with chest pain and one with flank pain. Other presenting symptoms included neurological symptoms and odynophagia. One patient presented with a recurrence of a malignant aggressive tumour (Ewing sarcoma of sacral spine) following incomplete resection at another unit. Ten patients had no neurological deficit at time of presentation. Two patients presented with radicular symptoms and another three had significant neurological deficits at time of presentation. One case presented with significant neurological deficit with a diagnosis of benign aggressive lesions (Aneurysmal bone cyst). Two of these cases had significant improvement after surgery. We had no cases of neurological deterioration following surgery. We treated six benign and twelve malignant tumours. The histopatghological diagnoses are summarised in Table 4. In all cases the surgery was performed by the senior author. A

variety of surgical strategies was utilised. An intralesional resection or debulking of the tumour was performed in six cases and a marginal resection was performed in nine cases to correlate with diagnosis. In six patients a fusion was performed by transpedicular screws and rods. Adjuvant therapy was used where indicated. Two of the recurrent cases

were in malignant tumours and one in benign aggressive tumour. In the one recurrent malignant tumour a repeat attempted marginal resection was performed. The other two recurrent cases were not re-operated as resection was not possible with acceptable morbidity. We had three deaths in the series on further follow up.

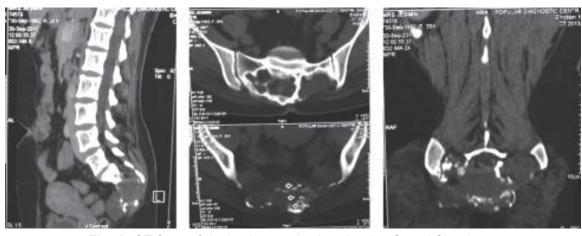


Fig.-1: CT Scan of the lumbosacral spine showing Sacral Chordoma.

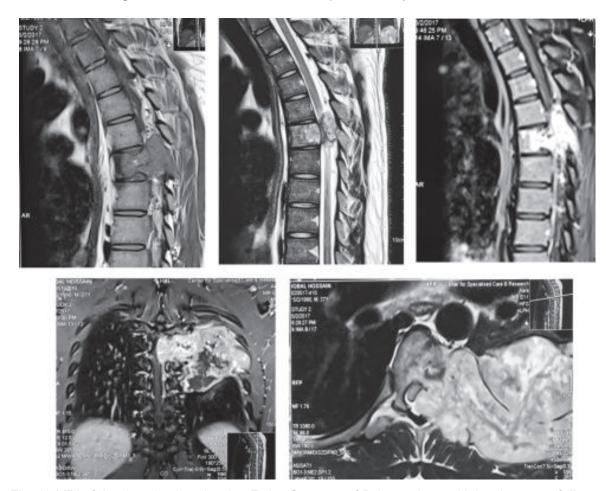


Fig.-2: MRI of the dorsal spine showing Ewing Sarcoma of D-9 vertebrae with involvement of ribs.

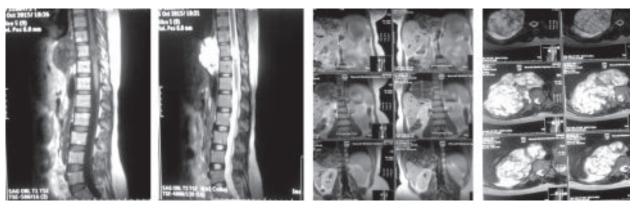


Fig.-3: MRI showing extensive Chondrosarcoma of the dorsal spine.

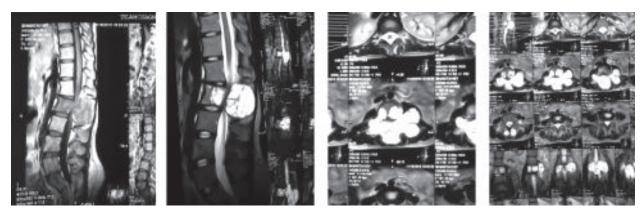


Fig.-4: MRI of the dorsolumber spine showing Aneurysmal Bone Cyst of the L-2 vertebrae.

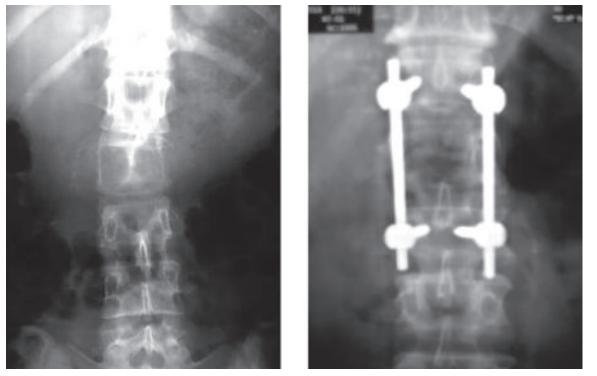


Fig.-5: Pre and post-Operative X-ray of L-2 Aneurysmal Bone Cyst.

Discussion:

The most common presenting complaint of patients with primary bone tumour of the spine is pain, with roughly 60% complaining of axial pain and 25% of radicular symptoms.3 The presence of a neurological deficit carries a worse prognosis. 11 Pain at rest or night pain is red flag symptoms that warrant further investigation. Spinal deformity is obvious when present, but occurs in less than 10% of patients. The most common cause of a painful scoliosis in adolescents is an osteoid osteoma.5 A delay in diagnosis is common and our finding is confirmed in other series. ⁴ This is due to the presenting symptoms often being vague, pain often being the only symptom, and the initial radiographs often only showing subtle or no abnormalities leading to the symptoms not being properly investigated. Most patients report a slow, gradual onset of pain in the involved area. In benign tumours of the spine an average of 26 weeks of symptoms before presentation are reported.4 This was confirmed in our series where the average delay in presentation was 7 months.

A variety of classifications systems is used for primary bone tumours of the spine. The Enneking staging system¹² was initially described for primary bone tumours of the appendicular skeleton, but is applied to tumours of the spine as well. It prognosticates as well as guides surgical resection margins and has been validated in the literature. The Weinstein-Boriani-Biagini surgical staging system¹³ is an anatomical classification based on the Enneking system that describes the number of vertebra as well as the number of sectors within a vertebra involved. Recommendations are then made for the most appropriate approach to resection of the lesion. We used the Enneking staging system. According to this system we treated six benign and twelvw malignant tumours. Of the benign group five could be classified as active and seven as aggressive. Appropriate treatment may be observational (Enneking grade 0 latent lesions) or surgical (most other lesions), depending on the level of pain, instability, neurologic compromise and the natural history of the lesion.⁵ The aims of surgery are complete resection of the lesion where feasible with preservation of neurological function. We used a variety of surgical strategies customised to the patient and the tumour. In peripheral malignant sarcomas the most important factor affecting survival is complete resection of the tumour with a wide

margin. 14,15 There are however no reports prescribing surgical margins in spinal tumours. Traditional intralesional resection in a piecemeal fashion is thought to increase the likelihood of local recurrence. True wide resection would cause unacceptable morbidity as it would involve resecting the segment of spinal cord. Total en bloc spondylectomy in a single stage from an all posterior approach was described by Tomita. 14 Here wide margins are achieved except at the pedicles and occasionally the spinal canal. This approach decreases local recurrence rates. 16-18 In benign aggressive lesions surgical eradication also provides the best long-term cure.6,18 We had three cases of local recurrence in our series. Two were in malignant tumours where a marginal resection was performed. The other recurrent case was a thoracic giant cell tumour encapsulating the aorta and forcing the surgeon to perform an intralesional resection.

Recent advances in chemotherapy have led to improved survival in malignant primary bone tumours, including in the spine. ^{7,19} Adjuvant radiotherapy is indicated in some malignant and benign aggressive lesions. ^{6,9} The use of local radiotherapy does increase the risk of major local complications, including sepsis. ^{8,10} We used radio- and chemotherapy in a number of cases as guided by our local oncology unit. The main limitations of this study are the retrospective character and the relatively small numbers.

Conclusion:

Most primary bone tumour of the spine in our population occurred in adults and was malignant. Surgical treatment resulted in pain reduction and neurological improvement, allowing a rapid adjuvant therapy in good clinical condition. An experienced multidisciplinary team of tertiary care center should perform diagnosis and treatment of these extremely rare tumors. In order to optimize management and elaborate guidelines, we encourage national or international registries, so that correct epidemiology, better classification and understanding, improved diagnostics, superior treatment and an overall better outcome may be achieved. We conclude that primary bone tumours of the spine are often associated with a significant delay in diagnosis. Surgical strategy and adjuvant therapy should be individualized for each case. Acceptable results with minimal complications can be achieved with this approach.

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Outcome of Conservative Management of Traumatic Acute Subdural Haematoma in Relation to Underlying Brain Injury

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Abstract

Background: Acute subdural haematoma is haematoma within the dura & arachnoid mater presenting within 72 hours of injury. It is caused by high speed impact that accelerates the brain relative to the fixed dural structure tearing the bridging veins that traverse between the cortical surface & venous sinuses. Small Acute Subdural haematoma less than 5 mm thick on axial CT images, without sufficient mass effect to cause shifting of midline less than 5 mm can be managed conservatively.

Objective: The objective of this study is to asses the morbidity & mortality following conservative management of traumatic acute subdural haematoma.

Methodology: A total 40 patients of Traumatic acute subdural haematoma were selected from Neurosurgery Department, Dhaka Medical College Hospital, Dhaka for this study. All the cases were diagnosed with History, Clinical examination & radiologically by non-contrast CT scan of the Brain. The epidemiological data were recorded and categorized in groups based on some medical and physical features. Then all the categorized data were analyzed using Computer based software SPSS program.

Result: 40 patients were managed conservatively. Among them 30 were not associated with underlying brain injury and 10 were associated with underlying brain injury. In our series mortality of associated underlying brain injury group is 50% but mortality of without associated underlying brain injury is 6.6% and morbidity of associated underlying brain injury group was 40% but disability of without associated brain injury group was 13.33%,

Conclusion: Outcome of conservative management of Traumatic Acute Subdural Haematoma is directly related with underlying brain injury. That is, outcome is worse when there is associated underlying brain injury.

Key Words: Acute Subdural Haematoma, conservative treatment, Morbidity, Mortality.

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Introduction:

Head injury is the most common cause of death in the world. Acute subdural haematoma is much more common which approximately 30% of all severe head

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injuries¹. In our country there is no such epidemiological statistics but report of the Bangladesh Bureau of Statistics, 2001 identifies RTA with head injury as one of the major causes of death⁶. According to the records of two main neurosurgical centers of Bangladesh DMCH & BSMMU 12000-15000 head injury patients get admission every year & the number is increasing day by day. Acute Subdural Haematoma caused by high speed impact that accelerates the brain relative to the fixed dural structure tearing the bridging veins that traverse between the cortical surface & venous sinuses. It can occur due to brain laceration where it is associated with intracerebral contusion & haemorrhage. Acute Subdural Haematoma can result from injury to surface of brain with bleeding from cortical vessels into subdural space. In a general series mortality is around 60% in acute subdural haematoma but can be lowered by very rapid surgical intervention & aggressive medical management. Seelig et al. observed a marked decrease is mortality & morbidity rates in those patients with acute subdural haematoma undergoing craniotomy with evacuation

of haematoma within 4 hours of injury; 30% of the patient died & 65% had functional recovery². However when surgery was delayed for more than 4 hours post injury, the mortality rate increase to 85% & only 7% of the patients had functional recovery^{11,13}. Haselberger et al, documented that when the interval between the injury & operation exceeded 2 hours, the mortality rise from 47 to 80%3. According to Henry H. Schmidek, MD, FACS, 2000, acute subdural haematoma form against a broad spectrum of primary brain injury of diffuse axonal type⁴. According to Tom Scaletta et al January 2005, all patients of acute subdural haematoma with GCS score less than 12 should be managed in ICU (Intensive Care Unit) with endotracheal intubation⁵. But in our series it was not possible to manage all patients GCS less than 12 in ICU due to limited ICU facilities. We have managed in ICU, all patients GCS less than 9, with severe respiratory distress & falling blood O2 level with endotracheal intubations. Conservative management includes anticonvulsant, diuretics, maintain fluid & electrolyte balance, antibiotics and other supportive treatment were not taken in ICU in our present study^{7,8,9}. This study tried to find out a plan for management of these patients to reduce mortality & morbidity within an acceptable limit.

Methodology

This is a prospective study. A total 40 patients of Traumatic Acute Subdural Haematoma were selected randomly from Neurosurgery Department, Dhaka Medical College Hospital, Dhaka from July 2005 to June 2007. All the cases were diagnosed with History, Clinical examination & Radiologically by non contrast CT scan of the Brain. The epidemiological data were recorded. Functional recovery associated morbidity & mortality were assessed & recorded in every case as per Glasgow outcome scale (GOS). The collected data was edited, compiled & statistical analysis was done. The data were presented in tables & graphs. In our series 40 patients were managed conservatively. 35 of them was radiologically indicated for conservative management & rest 5 was managed conservatively either due to refusal of surgery or patient was not physically fit for surgical management. On the basis of type of conservatively management of patients some Criteria were followed such as in CT scan of Brain shows maximum thickness of acute subdural haematoma is less than 5 mm, little or no

midline shifting (Fig. 1), refusal of surgical management by the attendants, patient is not physically fit for surgery.

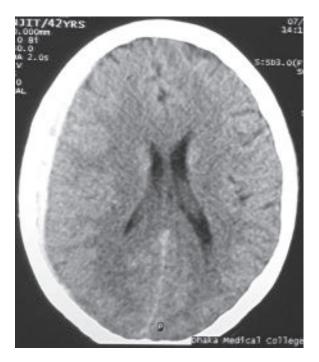


Fig.-1: CT Scan of the brain axial view showing acute subdural haematoma (Right) less than 5 mm without midline shifting.

Result:

In our study highest number of patient were in the most active period of life, 70% patients that is 28 patients out of 40 patients were found between 21 to 40 years of age. Below 20 years of age 5 cases (12.5%) were found & 7 cases (17.5%) were found above 40 years of age (Table I).

Table-IAge distribution in study group (n = 40)

| Age in year | No of cases | % |
|-------------|-------------|------|
| <20 | 5 | 12.5 |
| 21-30 | 13 | 32.5 |
| 31-40 | 15 | 37.5 |
| >40 | 7 | 17.5 |
| Total | 40 | 100 |

Out of 40 patients in our study 32 cases (80%) were male & only 20% (8 cases) were female. The number of occurrence of brain injury was approximately four times higher in male than in female (Table-2).

Table-IISex distribution in study group (n=40)

| Sex | No of cases | % |
|--------|-------------|-----|
| Male | 32 | 80 |
| Female | 8 | 20 |
| Total | 40 | 100 |

Highest numbers of head injury were found due to road traffic accident which was 50% (20 cases). Next to road traffic accident 2nd highest causes of head injury was fall from height which was 25% (10 cases) & 7 cases were due to assault (17.5%).

Table-III
Incidence of underlying brain injury according to mode of injury

| Mode of | ASDH | | ASDH | |
|------------------|-------------------------|-------|----------------------|----|
| injury | without Brain Injury | | with Brain Injury | |
| | No. | % | No. | % |
| RTA | 16 | 53.33 | 4 | 40 |
| Fall from height | 7 | 23.33 | 3 | 30 |
| Assault | 5 | 16.66 | 2 | 20 |
| Others | 2 | 6.66 | 1 | 10 |
| Total | 30 | | 10 | |

CT scan of Brain done in all 40 (100%) cases, as CT scan in the investigation of choice for its high

sensitivity & specificity. Plain X-ray skull (B/V) done in 32 cases (80%). Random Blood Sugar, S. Electrolytes & Blood gases analysis was done in some selective cases, according to need but not included in this study.

Out of 40, 10 patient of associated underlying brain injury, highest incidence of brain injury was underlying brain contusion which was 60% cases (6 patients). 3 patients had associated intracerebral clot which was about 30% & associated infarction was found in 1 patient (10%) (Table-4).

Table-IVType of underlying brain injury in study group (n = 10)

| Type of injury | No of patients | % |
|--------------------|----------------|-----|
| Brain contusion | 6 | 60 |
| Intracerebral clot | 3 | 30 |
| Infarction | 1 | 10 |
| Total | 10 | 100 |
| | | |

So, in conservatively managed group of patient, prognosis is directly related with underlying brain injury. That is, prognosis is worse when there is associated underlying brain injury.

Table-VFollow up of the conservatively managed patients as Glasgow Outcome Scale (n = 40)

| Total no of patients | | De | ath | Vege | tative | Sev | | Mode | | Go | |
|---------------------------|----|-----|-----|------|--------|-------|--------|-------|--------|-------|------|
| | | | | | | disal | oility | disal | oility | recov | very |
| | | No. | % | No | % | No | % | No | % | No | % |
| ASDH Without brain injury | 30 | 2 | 6.6 | 1 | 3.3 | 0 | 0 | 3 | 10 | 24 | 80 |
| ASDH With brain injury | 10 | 5 | 50 | 1 | 10 | 1 | 10 | 2 | 20 | 1 | 10 |
| Total | 40 | 7 | | 2 | | 1 | | 5 | | 25 | |

Table-VI

Outcome of conservative treatment (n = 40)

For analysis of data vegetative, severe disability & moderate disability is counted as single group as disability.

| ASDH | Death | Disability | Good recovery | Number |
|----------------------|-------|------------|---------------|--------|
| Without brain injury | 2 | 4 | 24 | 30 |
| With brain injury | 5 | 4 | 1 | 10 |
| Total | 7 | 8 | 25 | 40 |

Chi-Square Tests:

| | x ² | df | p value |
|--------------------|----------------|----|---------|
| Pearson Chi-square | 22.14 | 2 | p<.001 |

Discussion:

Acute subdural haematoma is a neurosurgical emergency worrying the surgeon with its malignant behavior. High mortality & morbidity of patients following acute subdural haematoma has led to search for better treatment modalities¹⁰. In advanced world despite of rapid transportation, emergency medical services, and improved radio imaging acute subdural haematoma continue to contribute significantly poor outcome is severely head injured patients.

Outcome of the conservative treatment was evaluated on the basis of Glasgow Outcome Scale (GOS)¹². According to Rengachary S.S 1994, the overall mortality rate of patients with a treated ASDH is roughly 50%, but in our series overall mortality is 17.5% (7 out of 40). In our series mortality of associated underlying brain injury group is 50% but mortality of without associated underlying brain injury is 6.6%. So mortality of associated underlying brain injury is much more higher. In our series overall disability was 20%. Disability of associated underlying brain injury group was 40% but disability of without associated brain injury group was 13.33%, So, disability of associated brain injury is also higher. On the other hand overall good recovery was 62.5%, in our series. Good recovery of without associated brain injury was 80% but with underlying brain injury 10%, so good recovery of associated underlying brain injury is worse than without underlying brain injury.

Morbidity & mortality following acute subdural haematoma is still much more higher. Mortality can be lowered by rapid surgical intervention & intensive medical management. According to Kotwica 1993, an acute subdural haematoma commonly is associated with extensive primary brain injury11. This diffuse parenchymal injury correlate strongly with the outcome of the patient. According to Henry H. Schmidek 2000, Rates of mortality & morbidity after an acute subdural haematoma are the highest of all traumatic mass lesions4. The poor outcome results largely from associated parenchymal injuries & subsequent intracranial hypertension. Approximately 50% of patients have associated lesions, but in our

series associated underlying brain injury was 25% (10, out of 40). So, according to different authors underlying brain injury is more common in acute subdural haematoma & morbidity, mortality is higher in underlying brain injury.

Conclusion:

Outcome of Traumatic acute subdural haematoma depends on underlying brain injury. Our study revealed that both mortality and morbidity are higher in traumatic acute sub dural haematoma with underlying brain injury than without underlying brain injury after conservative management.

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Signs and Symptoms of Early VP Shunt Infection in a Tertiary Hospital

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Abstract:

Background: VP shunt infection is a major concern for the treating surgeon. Shunt infection causes shunt malfunction and is source of much morbidity for the patient.

Introduction: Hydrocephalus was diagnosed in patients clinically by history, symptoms and signs. This was confirmed by CT and/or MRI scans and was treated with ventriculoperitoneal shunt system. In this study we have analyzed signs and symptoms of early shunt infection. That will help in early detection of shunt infection and faster management.

Methods: This is an observational study. The study was done from September 2008 up to January 2010 in BSMMU. Thirty patients were operated on. They were followed up to 15 days after surgery. They were evaluated with presence of non-remitting fever in the absence of other causes, indication of infection along the shunt tract or incision site and signs of meningeal irritation or peritonitis.

Result: Seven patients of the thirty patients treated with ventriculoperitoneal shunt surgery had VP shunt infection. That is 23.33% of patients. This is more in the young and elderly age group. It was also more at the lower socioeconomic age group. Most patients had fever, headache and neck rigidity.

Key Words: Hydrocephalus, ventriculomegaly, shunt infection, headache, papilledema, meningitis, ventriculitis, ventriculoperitoneal shunt.

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Introduction:

Hydrocephalus is defined as an abnormal enlargement of the ventricles due to an excessive accumulation of CSF from a disturbance of its flow, absorption or uncommonly, secretion¹. Ventriculoperitoneal shunt catheter placement is a relatively common neurosurgical procedure, performed for the treatment of hydrocephalus as well as for associated conditions in which the natural flow of cerebrospinal fluid is obstructed². According to McGirt et al. the signs of shunt infection are meningitis, ventriculitis, CSF leak from wound, skin infection—anywhere along the tract, peritonitis, seizure disorder³.

Trojanowski had commented that between 5 and 15% of the devices become infected, of which more than a half within the rest month after surgery⁴. Ahmed et al. have studied complications on fifty patients. Of these 6 (12%) patients had shunt infection⁵. Drake and lantosca reported that the incidence of shunt

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infection is approximately 5%- 10%⁶. Bokhary and Kamal incidence rate is nearly 10%⁷. Kinasha et al. in their study in Tanzania reported the shunt infection rate about 24.6%⁸. McGirt et al. had concluded from their observation that shunt infection occurred in 11% of cases (92 of 820)³. Leach and Kerr had reported that shunt infection should be within 1% and 7% of shunt insertions⁹.

Materials and Method:

This is an observational study. This was conducted from 01-07-2008 to 17-05-2010. All patients were admitted in the BSMMU Hospital for ventriculoperitoneal shunt surgery. Patients who met the inclusion criteria from all age groups were included in the study. Number of patients were thirty (*n*=30).

Shunt infection was defined if at least one of the following criteria was present:

- 1. Presence of non-remitting fever in the absence of other causes
- Indication of infection along the shunt tract or incision site
- 3. Signs of meningeal irritation or peritonitis.
- Shunt system used: In all patients we used Chhabra medium pressure ventriculoperitoneal shunt system (Surgiwear Inc.)

Inclusion criteria:

All patients with ventriculomegaly in imaging and signs of hydrocephalus undergoing ventriculoperitoneal shunt surgery were selected.

- Exclusion criteria:
 - o Patients with preoperative fever or any other focus of infection
 - Patients with other medical problems e.g., diabetes mellitus, valvular heart disease, renal impairment and ruptured meningocele.
 - Patients who had previous CSF diversion surgery (e.g. ETV, EVD)

Results:

After proper collection and processing of samples, the data is analyzed. The results and observations from the analysis were presented in tables and figures. Data were expressed in numbers or percentage as appropriate.

Table-IDistribution of the patients by age

| Age in years | Freq | Frequency | | | |
|--------------|-----------------|--------------------|----|--|--|
| | Shunt infection | No shunt infection | | | |
| 0-2 | 2 | 6 | 8 | | |
| 2-12 | 1 | 5 | 6 | | |
| 12-30 | 1 | 3 | 4 | | |
| 30-50 | 1 | 4 | 5 | | |
| >50 | 2 | 5 | 7 | | |
| Total | 7 | 23 | 30 | | |

From this table we can see that shut infection is more in the below 12 years and more than 50 years.

Table-IIDistribution of the patients by sex

| Variables | Male | Female | Total |
|--------------------|------|--------|-------|
| Shunt infection | 4 | 3 | 7 |
| No shunt infection | 16 | 7 | 23 |
| Total | 20 | 10 | 30 |

In this table we can see that 4 male patients and 3 female patients had shunt infection.

Table-IIIDistribution of the patients by post-operative morbidity

| Post operative complications | Total | Percentage |
|------------------------------|-------|------------|
| Persisting Fever | 07 | 23.33% |
| Surgical site infection | 05 | 16.66% |
| Shunt tract infection | 05 | 16.66% |
| Skin necrosis | 1 | 3.33% |
| Headache | 6 | 20% |
| Vomiting | 6 | 20% |
| Neck rigidity | 1 | 3.33% |
| Shunt block | 1 | 3.33% |
| Peritonitis | 2 | 6.66% |

From the above table it is seen that seven patients (23.33%) developed persisting fever. SSI occurred in 5 patients (16.66%). Four patients (13.33%) had shunt tract infection and only one patient had skin necrosis. Headache and vomiting was present in 6 patients (20%), neck rigidity was present in one patient and shunt block occurred in one patient. Two patients (6.66) developed signs of peritonitis.

Table-IVDistribution of the patients by post-operative vomiting, headacheand non-remitting fever

| Age in years | Vomiting | Headache | Non-remitting |
|--------------|----------|----------|---------------|
| | | | fever |
| 0-2 | 3 | 2 | 3 |
| 2-12 | 1 | 1 | 1 |
| 12-30 | 0 | 0 | 0 |
| 30-50 | 1 | 2 | 2 |
| >50 | 1 | 1 | 1 |
| Total | 6 | 6 | 7 |

From the above table it is seen that total 6 patients had vomiting in the post operative period. 3 patients were under the age of 2 years. In this table 6 patients also headache. Most patients were above 12 years of age (66.67%). It also shows that total 7 patients had non-remitting fever.

Table-VDistribution of the patients by surgical site infection (n=6)

| Age in years | SSI | Neck rigidity | Peritonitis |
|--------------|-----|---------------|-------------|
| 0-2 | 3 | 0 | 1 |
| 2-12 | 2 | 0 | 1 |
| 12-30 | 0 | 1 | 0 |
| 30-50 | 1 | 0 | 0 |
| >50 | 0 | 0 | 0 |
| Total | 6 | 1 | 2 |

From the above table we can see that six patients had surgical site infection, one patient had neck rigidity and two patients had signs of peritonitis.

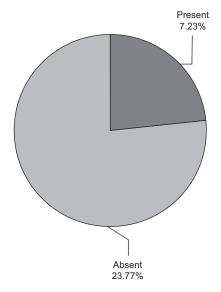


Fig.-1: Distribution of the patients by shunt infection in all patients (n=30).

From this above chart it is seen that shunt infection occurred in seven patients out of thirty patients. This was about 23.33%.

Discussion:

Choux et al. in 1992 had defined post-operative shunt infection as an infection confirmed within six months of operation and was diagnosed if there was inflammation along the length of shunt, wound discharge or wound dehiscence. Signs of meningitis, ventriculitis or non-focal systemic indication of infection were also investigated for shunt infection 10. In this study, 5 patients (16.66%) had these signs. Signs of meningitis was present in one patient. Five patients had headache and vomiting. Kontny et al. had mentioned that the main sign and symptoms were fever, shunt malfunction and meningeal irritation, and with VP-shunts only abdominal pain¹¹. Seven patients had fever in this study following shunt insertion. Pople in 2002 had suggested the following as most important clinical features of shunt infection: general malaise, pyrexia, headaches, vomiting, neck stiffness, abdominal tenderness or distension, recurrent lower end shunt obstruction, occasionally pain and erythema around the shunt¹². In our study seven patients (23.33%) had pyrexia, five patients (16.66%) had shunt malfunction and peritonitis in two patients

(6.66%). The distribution of the shunt infection is congruent with the patients of the other studies.

Conclusion:

Ventriculoperitoneal shunt surgery is a common procedure. Shunt infection can reduce the success of this surgery. If we can diagnose the shunt infection early, then we can reduce morbidity as well as mortality of these patients. Therefore, we have to diagnose shunt infection very quickly and accurately and take necessary measures for benefit of the patient.

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Assessment of Serum Homocysteine Level in Epileptic Patients on Carbamazepine Treatment

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Abstract:

Background: This comparative cross sectional study was carried out to evaluate the level of serum homocysteine with carbamazepine treatment in epileptic patients.

Materials and Methods: This study was conducted in the Department of Neurology, BSMMU, Dhaka, from March 2013 to December 2015 for the duration of two years and nine months period. Patients, who were diagnosed as epileptic and receiving Carbamazepine at least for six months and no other anti epileptic drugs (AEDs), were considered as case group and Patients other than epilepsy attending Neurology outpatient department (NOPD) were considered as control group.

Results: A total of 104 patients were recruited as study population. Of them 52 patients grouped into case and the rest 52 patients in control. The mean (SD) value of serum homocysteine in epileptic patients using carbamazepine as anti-epileptic drugs found 16.33 (5.34) ½mol/L which was highly statistically significant difference than control group measuring 8.77 (3.75) ¼mol/L. Considering the normal level 5-15 ¼mol/L, in epileptic patients with carbamazepine therapy, increased level was found in 53.8% patients. On the other hand, in control group, 13.5% had increased value of serum homocysteine.

Conclusion: Carbamazepine therapy which is usually given to epileptic patients may interfere with metabolic pathways of homocyateine (Hcy) as well as may lead to an alteration of its serum levels.

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Introduction:

Many developing countries such as Bangladesh, India are are densely populated. Like tuberculosis, malaria epilepsy is also a burden for our country. Anti- epileptic drugs are commonly used to treat epilepsy.

Commonly used first-generation antiepileptic drugscarbamazepine, sodium valproate, phenytoin and phenobarbitone. Second-generation antiepileptic drugs are- lamotrigine, gabapentin, topiramate etc. Oxcarbazepine and levetiracetam- available but are highly expensive

However consumption of carbamazepine causes significant elevation of serum homocysteine level¹.

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Elevated serum homocysteine (Hcy) is responsible for ischaemic heart disease, deep vein thrombosis, pulmonary embolism and stroke². Near about 10-40% of epileptic patients develop hyperhomocysteinaemia³. High plasma homocysteine is an independent risk factor for cardiovascular disease and thrombosis⁴. Anti-epileptic drugs raise serum homocysteine (Hcy) by reducing blood folate levels⁵.

Atherosclerotic vascular diseases are associated with well-known risk factors such as systemic arterial hypertension, diabetes mellitus, smoking and obesity, but in the last decade other emerging risk markers have been identified, one of them is being Hcy. Atherosclerosis is a chronic inflammatory disease of the blood vessel walls, in which deposits of lipids, cholesterol, calcium and other substances build up in the endothelial layer of the arteries. Many studies have identified moderate elevation of serum Hcy as an independent risk factor for atherosclerotic vascular disease⁶.

Lowering of serum folate and elevation of Hcy concentrations in blood are associated with poor cognition in general population. High levels of Hcy result in neurotoxic and vasotoxic effects in dementia and Alzheimer's disease, suggesting that Hcy is a direct marker of early cognitive deficit⁷.

The pathogenic role of high serum Hcy is in vascular dementia is yet no clear. The levels of serum Hcy, vitamin B12 and plasma folate were studied in Alzheimer's disease and vascular dementia, and it was found that Hcy was increased, while vitamin B12 and folic acid were decreased in both of these conditions. This suggests that supplementation of vitamin B12 and folate may be beneficial in vascular dementia and Alzheimer's disease ⁸.

Materials and Methods:

This study was conducted in the Department of Neurology, BSMMU, Dhaka, from March 2013 to December 2015 for the duration of two years and nine months period. Cases were selected by simple random sampling. Age ranges of the patients were 14 to 60 years. Because baseline serum Hcy level have similarity in these age group. Moreover we have collected cases from epilepsy clinic of adult neurology. Epileptic patients with other systemic diseases such as: Chronic kidney disease, Hypothyroidism, chronic liver disease, Leukemia, Inflammatory bowel disease, psoriasis and homocysteinuria were excluded from this study9. Patient with Diabètes Mellitus, Smoking, Pregnancy, age less than 14 years and greater than 60 years, taking Methotrexate, Isoniazide or other AEDs, folic acid supplement, receiving vit-B6, Vit- B12 or on folate antagonists were also excluded from this study

Data were collected by a pre-designed proforma. Detailed history and clinical examination were carried out and structured questionnaire were used to collect the necessary information. Similar numbers of controls were also selected. Relevant base line investigations were done which included Complete blood count with peripheral blood film where applicable, Urine R/M/E, Serum creatinine, ALT, Thyroid function tests in suspected hypothyroidism.

Serum Hcy assay was done from venous blood of both cases and control in the Department of Biochemistry, BSMMU on the day of sample collection. The ARCHITECT Homocysteine assay, a chemiluminescent micro particle immunoassay (CMIA) technology was used to measure homocysteine value. Statistical analysis of data was carried out with appropriate techniques and systems.

Results:

A total of 104 patients were recruited as study population. Of them 52 patients grouped into case and the rest 52 patients in control.

Table-IDistribution of patients (Case group) according to epilepsy type (n= 52)

| Type of Epilepsy | Frequency (n) | Percentage |
|-----------------------|---------------|------------|
| Simple partial | 17 | 32.7 |
| Complex partial | 5 | 9.6 |
| Primary generalized | 26 | 50.0 |
| Secondary generalized | d 4 | 7.7 |

This table I shows distribution of Case group patients according to epilepsy type. Here, half of the patients, 50% had primary generalized epilepsy which was followed by simple partial type 32.7%. Complex partial was accounted 9.6% whereas secondary generalized 7.7%.

Table-IIDistribution of case group patients according to duration of diagnosis and treatment (n=52).

| Duration (Years) | Mean | SD |
|-----------------------|------|------|
| Duration of diagnosis | 4.29 | 4.51 |
| Duration of treatment | 4.05 | 3.59 |

This table II shows distribution of case group patients according to duration of diagnosis and treatment. The mean (SD) value of duration of diagnosis was 4.29 (4.51) years while duration of treatment was 4.05 (3.59) years.

Table-IIIDistribution of case group patients according to Carbamazepine dose level (n=52)

| Carbamazepine | Frequency (n) | Percentage |
|----------------|---------------|------------|
| dose (per day) | | |
| 200 mg | 2 | 3.8 |
| 400 mg | 18 | 34.6 |
| 800 mg | 30 | 57.7 |
| 1000 mg | 2 | 3.8 |

This table III shows distribution of case group patients according to Carbamazepine dose level. Out of all patients, 57.7% patients were taken 800 mg carbamazepine daily. The second highest portion of patients 34.6% was taken 400 mg of carbamazepine daily.

Table-IVDistribution of patients according to serum level of homocysteine (N=104)

| Type of | Serum Hon | p-value* | | |
|----------|-----------|----------|----------|--|
| Patients | (micro | mol/L) | | |
| | Mean | SD | | |
| Case | 16.33 | 5.34 | <0.0001s | |
| Control | 8.77 | 3.75 | | |

S=significant

Table IV: shows distribution of patients according to serum homocysteine level. The mean (SD) value of serum homocysteine in epileptic patients using carbamazepine as anti-epileptic drugs found highly increased than control group [16.33 (5.34) vs. 8.77 (3.75)]

Table-VDistribution of patients according to range of serum Homocysteine level (N=104).

| Type of Patients | Serum Homocysteine (1/4mol/L) | |
|------------------|-------------------------------|---------|
| | Minimum | Maximum |
| Case | 8.45 | 30.28 |
| Control | 5.30 | 19.00 |

The table II shows distribution of patients by range of serum homocysteine level. Here, in case group, the range was 8.45 to 30.28 ¼mol/L and that of 5.30 to 19.00 ¼mol/L in control group

Discussion:

Carbamazepine (CBZ) is a potent antiepileptic drug. It is commonly used in partial onset and secondary generalized seizures. It has also good efficacy in the treatment of primary generalized tonic-clonic seizures. Some studies showed in epileptic patients receiving CBZ, there is significant elevation of serum homocysteine levels¹⁰. No previous study has done in Bangladeshi subjects. It is one of the foremost studies of epilepsy patients taking carbamazepine therapy to consider their serum homocysteine level in Bangladesh context. We compared our study findings with result of some other published articles elsewhere in the world.

According to gender distribution, no statistically significant difference was observed between two groups. Out of all patients, 67.3% were male and 32.7%

female in carbamazepine treated patients. In control group patients, 63.5% was male and 36.5% female.

In analysis of epilepsy type, half of the patients, 50% had primary generalized epilepsy which was followed by simple partial type 32.7%. Complex partial was accounted 9.6% whereas secondary generalized 7.7%.

The mean (SD) value of duration of diagnosis was 4.29 (4.51) years while duration of treatment was 4.05 (3.59) years. Out of all patients, 57.7% patients were taken 800 mg carbamazepine daily. The second highest portions of patients 34.6% were taken 400 mg of carbamazepine daily.

The present study showed that epileptic patients receiving carbamazepine as AEDs have increased serum levels of homocysteine. The mean value of serum homocysteine in epileptic patients taking carbamazepine as anti-epileptic drugs found 16.33 (5.34) 1/4 mol/L which was highly statistically significant difference than control group measuring 8.77 (3.75) ½mol/L. However, in case group the highest number of 13 patients had their value in 15 – 20 1/4 mol/L and highest value 30 1/4 mol/L had only in 1 patient. At the same time, in control group highest number of 21 patients had their value in 5 – 10 1/4 mol/L and highest value near about 20 1/4 mol/L had in 2 patients. In case group, the range of serum homocysteine was found 8.45 to 30.28 1/4 mol/L and that of 5.30 to 19.00 1/4mol/L in control group.

Our results are in consistent with the previous reports on homocysteine concentrations in patients with epilepsy taking AED specially CBZ. Paknahad et al, also recorded mean of serum Hcy concentration in epileptic patients receiving CBZ was significantly higher compared to that in the controls [13.66 (0.95) vs. 12.97 (0.46) $\frac{1}{4}$ mol/L, p = 0.04]⁴. The similar result was obtained if patients with only CBZ monotherapy were included, Minitzer et al, found 11.1 (4.2) $\frac{1}{4}$ mol/L¹¹. Epilepsy patients switching from the enzyme inducing AEDs, phenytoin or CBZ to the non inducing AEDs drugs, LTG or levetiracetam results in significant declines in serum homocysteine level.

Conclusion:

In conclusion, our data demonstrate that serum homocysteine level significantly increased in carbamazepine treated epileptic patients. Therefore,

^{*} P-value derived from independent sample t test

it should be monitored the level of homocysteine in epileptic patients and managed accordingly.

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Case Reports

Tubercular Spinal Epidural Abscess Involving the Lumbo-Sacral Region: A Case Report with Review of Literature

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Abstract:

Tubercular spinal epidural abscess (SEA) without bony involvement is a rare condition but may be devastating and fatal.

This is a report of a rare case of a tubercular spinal epidural abscess in a 60 years old man who presented with paraparesis and Cauda-Equina Syndrome with Fournier's gangrene, but without vertebral bony involvement. MRI of the lumbosacral spine revealed huge collection of thick pus extradurally and posteriorly extending from L2 vertebra to sacral region. The abscess was treated surgically by posterior surgical decompression that involved laminectomy of L2 to L5 vertebra and pus was completely drained. Histopathology demonstrated Mycobacterium Tuberculosis bacteria in the drained pus. Anti- tubercular drugs were started postoperatively. The patient showed progressive improvement in his neurological deficits and started walking without support at 6 months of follow-up after hospital discharge.

Keywords: Epidural; abscess; paralysis; lumbar-sacral; laminectomy; ATT; Ambulation

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Introduction:

Spinal epidural abscess represents a severe pyogenic infection of the epidural space which may compress neural elements and require urgent surgical intervention to avoid permanent neurological deficits.[1] It is a rare variety reported with an incidence of 0.2 -2 cases per 10000 admissions at

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Address of Correspondence: Dr. Akhlaque Hossain Khan, Associate Professor, Department of Neurosurgery, Bangabandhu Sheikh Mujib Medical University, Dhaka, Bangladesh. hospital.[2] The incidence of spinal epidural abscess is increasing in developing country which usually occurs secondary to tubercular spondylitis but may rarely develop by haematogenous spread by any foci. Tubercular spondylitis and its resulting complications are common in developing nations. [3] Early diagnosis with advanced imaging techniques, proper surgical intervention and effective coverage of broad spectrum antibiotics have led to gradual decrease in mortality rates but still data show high mortality rates of 5-33% in some series .[4] The purpose of this study was to analyse the clinical manifestations, prognostic indicators, source of infection and final outcomes in patients with SEA.[4]We present a rare case of spinal epidural abscess of tubercular origin involving the lumbar and sacral spine without osseous involvement.

Case Report:

A 60 years old man, farmer by occupation, developed pain, tingling and numbness in his both lower limbs for 2 months which was insidious in onset. After one and half months later, he developed dribbling of urine and slowly over one week, he developed incontinence of urine. During the course of his illness, he had history of intermittent type of fever and weight loss, for which he visited a general practitioner. He was

diagnosed as a case of UTI and was advised some drugs. But the patient condition got worsened over time and got admitted at nearby hospital. A selfretaining urinary catheter with some medicines were advised and discharged after two weeks. After 2 weeks, patient developed Fournier's gangrene with bed sore in left sacral region. Surgical wound debridement and S.P.C was done under spinal block in September 2014 and was put on anti-tubercular regimen but patient later developed drug induced hepatitis. The drug was stopped and referred to higher centre and the patient was admitted in our hospital. MRI of the lumbosacral spine with whole spine screening film was done which showed there was significant compression over thecal sac extending from L2 to sacral region by huge collection of pus extradurally and posteriorly (Figure 1). Chest x-ray showed some patchy infiltrates around the mediastinal-hilar region. Neutrophil was 88%, ESR

was 85 in 1st hour and was diagnosed as a case of epidural abscess from L2 to sacral region with lower limbs paraparesis and incontinence of urine.

Posterior surgical decompression was done by laminectomy of L2 to L5 vertebra. A thick collection of pus was found extraduraly and posteriorly which was sticky, yellowish in colour and was evacuated and thorough irrigation done with normal saline mixed with antibiotic Gentamycin. Dural sac was made free and pus was sent for Gram stain, culture and sensitivity, and Acid Fast Bacillus (AFB). Wound was closed over a suction drain.

Pus for AFB was found to be positive. Again, antitubercular therapy (ATT) was started following the report and was discharged under supervision. After one week, patient developed ulceration in the genitoperineal region which was treated conservatively. Postoperatively no significant improvement was noted and passive physiotherapy was advised. Gradually,

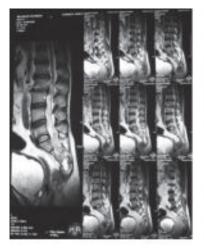






Fig.-1. Preoperative MRI of Lumbosacral spine with different views showing significant compression over thecal sac extending from L2 to sacral region by huge collection of pus extradurally and posteriorly.

patient regained improvement in power of his lower limbs and at 6 months of follow up, patient was found to have muscle power of 4 out of 5 in both his lower limbs and also control over his bladder function.

Discussion:

Most spinal infections in developed countries are due to pyogenic infections where as non pyogenic organisms are responsible for most spinal infections in developing countries and also in the immunocompromised patients in the developed nations.^{1,3} Mycobacterium Tuberculosis is the most important organisms in developing nations. 1 One of the common cause of death in the world is tuberculosis (TB) which is assumed that two billion people are infected with TB, who can develop TB disease in their course of illness. TB mostly affects children and young adults. The risk factors regarding the development of the disease depends on the patient compliance and the geographical variations because of the different incidence of TB in each country. [5,9] Infection of the spine can pose a wide range of problems for both patient and the surgeon. Tuberculosis demonstrates a varieties of clinical and radiological findings and has a known propensity for dissemination from its primary site; therefore it can mimic a number of disorders. The microbiological diagnosis of spinal infection is very important because despite the surgical treatment, the medical management with ATT drugs is essential postoperatively for resolution of infection. An estimated 20% - 40% involved spinal tuberculosis patients have another focus of infection and due to lack of proper antimicrobial treatment, poor compliance of the patients and proper duration lead to flare of the infection. Despite proper management, the resistance to spinal tuberculosis is increasing.[6] Innumerable patients and lack of risk factors in most of the patient results in diagnosis delay during the process of the disease¹¹.

Cauda equina syndrome is uncommon; accounting for an incidence of 1–5% of spinal pathology in the literature. Acute trauma is a rare cause of this syndrome. The most common presenting features are bladder symptoms, saddle anaesthesia and loss of motor function. Early surgical decompression has been suggested to obtain a satisfactory recovery^{7, 8, 10}. Patient with spinal epidural abscess (SEA) may have unknown focus of infection with pain and tenderness of the spine clue to proper investigation lead to early diagnosis of the disease¹².

Conclusion:

Spinal epidural abscess without bony and pulmonary involvement is uncommon but potentially devastating condition that challenge many clinicians regarding

its proper diagnosis in timely fashion for appropriate management. MRI of the spine is a noninvasive and sophisticated tool for the early diagnosis of SEA due to spinal TB. Early diagnosis and adequate surgical decompression of the SEA compressing the thecal sac and nerve roots along with proper postoperative anti tubercular therapy leads to cure of the patient having good chances of recovery of neurological functions.

Conflict of interest

Authors declare no conflict of interest.

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Giant Ganglioneuroma of Thoracic Spine: A Case Report

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Abstract:

Ganglioneuromas most commonly arise from sympathetic ganglia. These neoplasms may be located wherever ganglion cells are normally found from the skull base to the pelvis, including the adrenal medulla. We describe a 15-year-old girl with giant Ganglioneuromas in the thoracic spine, who underwent successful resection (T4-5 level) of the tumor. Histopathological examination confirmed the diagnosis. Ganglioneuromas should be considered in the differential diagnosis of any paraspinal mass. A high index of suspicion and correlation of clinico-radiological findings is necessary in differentiating a large benign tumor from a malignant growth. Complete surgical excision is the treatment of choice; however tumor size and location need to be considered for the surgical approach (one-step or multiple surgeries). Close follow-up after surgery is mandatory.

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Introduction:

Ganglioneuromas are rare benign tumors that originate from a neural crest or sympathetic ganglion¹. They most commonly appear in the posterior mediastinum and abdomen². The patients exhibit no obvious symptoms upon nervous system examination. The ganglioneuromas are often found in females, while the male/female ratio is approximately 2/3³. The incidence of ganglioneuroma is not well documented, but it is estimated to characterize 0.1 to 0.5 % of total central nervous system (CNS) tumors⁴. Paravertebral ganglioneuroma and scoliosis is rarer and has only been sporadically reported. We report a giant paraspinal Ganglioneuroma extending into extradural space and thoracic cavity.

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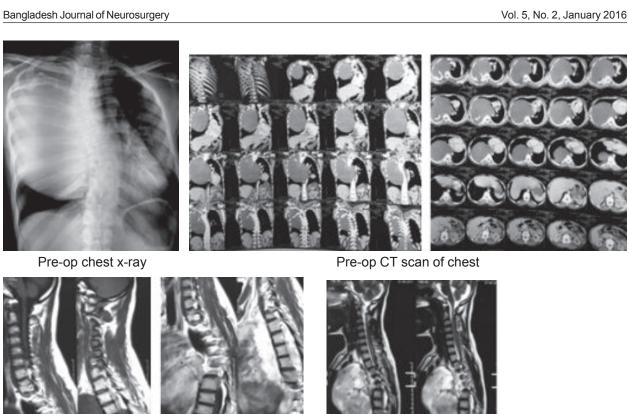
Case Report:

A 15-year-old girl presented with a one month history of progressive onset lower extremity weakness. The patient had no other significant complaints. On examination, she was unable to stand or walk without support.

A neurological examination revealed 3/5 strength in all muscle groups in the patient's lower extremities. Sensory was markedly decreased below the nipple line. Deep tendon reflexes were hyperreflexic throughout the lower extremities along with markedly increased tone, right greater than left. Sustained clonus was noted at both ankles.

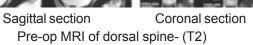
Magnetic resonance imaging showed a huge well defined mixed signal intensity oval lesion in posterior mediastinum predominantly on the right side. The lesion predominantly hypointense in T1WI and predominantly hyperintense in T2WI. Foci of signal void areas within the lesion represents calcification T1WI and T2WI increased signal intensity areas represents fatty component. The lesion causes collapse of right lung sparing basal segments of right lower lobe. Mediastinal structure are pushed of left. Epidural extension of the lesion is noted into the spinal canal causing widening of the right neural foramen resulting compression over the spinal cord and corresponding nerve root compression at D4-D5 level. Spinal cord pushed to left.

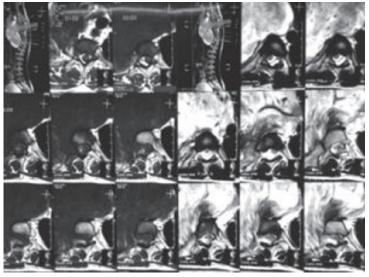
Right Posterolateral thoracotomy was done. A huge tumor occupying right paravertebral space pushing the right lung medially and inferiorly. It has a broad base, Posterior end of right 4th rib thin out and became triangular shaped. Tumor invaded in between the space.



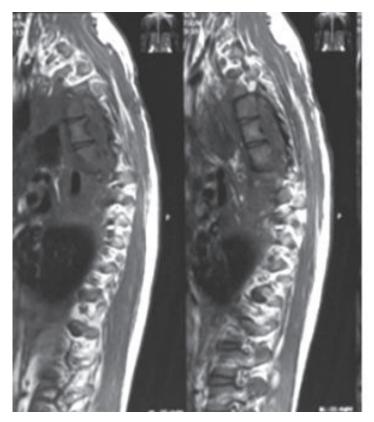
B. Contrast A. Non-contrast Pre-op MRI of dorsal spine- sagittal (T1)



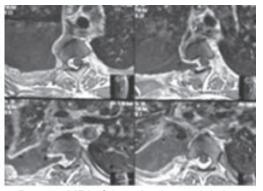




Pre-op MRI of dorsal spine Axial section







Post-op MRI of dorsal spine- axial view

Post-op MRI of dorsal spine- coronal view

Discussion:

About 10% of Ganglioneuromas may involve the spinal canal⁵. Paraspinal Ganglioneuromas can extend into the spinal canal, forming dumb bell shaped tumor. However, in rare cases intradural extension has been reported. Most Ganglioneuromas are incidentally detected, and the symptoms, if any, are usually due to the mass effect. Rarely, the tumor may secrete vasoactive intestinal polypetide. resulting in diarrhea. As this slow growing tumor extends through the neural foramen into the spinal cord, some patients may present with neurological deficits or scoliosis⁶. Thoracic intradural extramedullary Ganglioneuromas are very rare. In our patient, the characteristic feature was the remarkably large tumor size infiltrating into the thoracic cavity along with an intradural component. Microscopically, these tumors contain large ganglion cells and show areas with smaller lymphocyte-like cells within a matrix of fibrous stroma and schwann cells. The distinction from malignant tumor is based on the absence of necrosis or presence of any immature ganglion cells⁷.

It is usually safe and feasible to perform complete excision of Ganglioneuromas. However, in case of multiple and/or large-sized tumors, multi-stage dissection should be considered. In the present case, there were dense adhesions of the tumor with the nerve roots at the foraminal portions, which were left undisturbed during dissection. Ganglioneuromas generally has a favorable prognosis given its low metastatic potential⁸.

Conclusion:

This report describes a rare case of multiple Ganglioneuromas of the thoracic spine with intradural and thoracic extension. Ganglioneuromas should be considered in the differential diagnosis of a paraspinal mass. Although complete surgical resection is the best treatment option, stage-wise surgical resection should be considered in large-sized and/or multiple tumors, with close follow up.

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