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Case Reports

Neurosurgical Management of Intractable Temporal Lobe Epilepsy by Amygdalohippocampectomy Plus Anterior Temporal Lobectomy: Report of Initial Three Pediatric Cases with Short Literature Review

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Abstract:
Temporal Lobe Epilepsy (TLE) is one of the most common surgical epilepsy that is usually resistant to antiepileptic drugs and surgery is the treatment of choice. This type of epilepsy may be due to Mesial Temporal Sclerosis (MTS), tumors [i.e. low grade glioma, Arterio-Venous Malformation (AVM) etc],trauma, infection (Tuberculosis),parasitic infestation (e.g. Schistosomiasis) etc. Here we report three cases of surgically treated TLE in pediatric age that was due to MTS and low grade ganglioglioma. In all three cases the only presenting symptom was complex partial seizure (Psychomotor epilepsy) for which all underwent scalp electro-encephalography (EEG) and Magnetic Resonance Imaging (MRI) of Brain. All three patients were managed by amygdalohippocampectomy plus standard anterior lobectomy. In post operative period the cases were seizure and disease free till last follow up. We did not face any nominal dysphasia, memory disturbances, hemi paresis or visual field defect.

Key Words: *Amygdalohippocampectomy, Intractable temporal lobe epilepsy, Neurosurgical management, Anterior temporal lobectomy.*

Introduction:

Epilepsy is a relatively common neurological disorder, even commoner in pediatric patients and that may occur from a number of etiologies or sometimes without any etiology. About 80% epilepsy patients can be effectively managed with antiepileptic drugs (AEDs) whereas 20% epilepsies are drug resistant and intractable.[10] In this group of patient some form of surgery is usually needed either to cure epilepsy or to improve living standard by better control of epilepsy with AEDs. The idea of surgical treatment for epilepsy is very old. However, widespread use and general acceptance of surgical treatment of epilepsy has only been achieved during the past three decades. Improvements in imaging technique resulted in an increased ability for preoperative identification of intracerebral, potentially epileptogenic lesions and epileptogenic zone or focus. High resolution magnetic resonance imaging plays a major role in structural imaging. EEG (Especially video EEG), MRI and other functional imaging usually help in identification of epileptogenic lesion, zone or foci. Today, surgery is more effective and conveys a better seizure control rate. It has become safer and less invasive, with lower morbidity and mortality rates.[21]

Here we report our initial experience of epilepsy surgery (amygdalohippocampectomy) in pediatric patients with divergent medial temporal lobe pathologies in Bangladesh.

Case presentation:

Case 1 (Figure1&2):

A 11 year old right handed high school girl presented with sudden development of uneasiness, perception of smell of kerosene, followed by loss of awareness about surroundings for a span of 1-2 minutes each time for last 20 months. Initially the episode of seizure was 3-4 attacks per week but later it increased up to 10-12 attacks per week. She had no significant past events related to present disease. There was no significant event in her birth process and family history is negative for such type of disease. She had no

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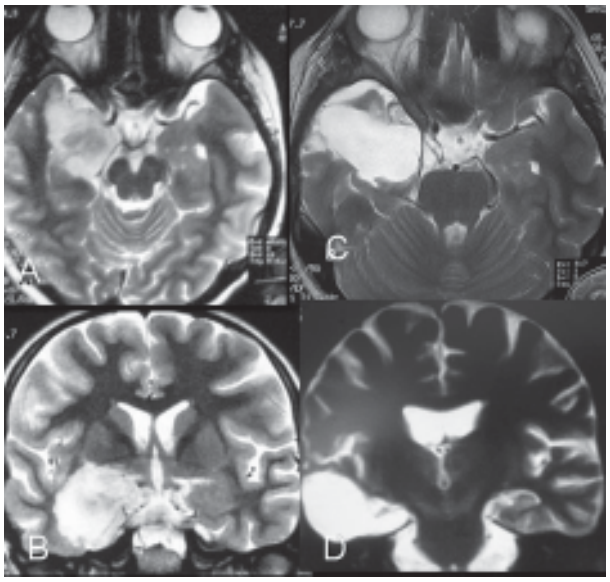

Fig.-1: *MRI of brain T2 weighted images. A-axial & B-coronal showing right amygdalohippocampal tumor. C&D-MRI T2 weighted axial and coronal images respectively after amygdalohippocampectomy and standard anterior temporal lobectomy.*

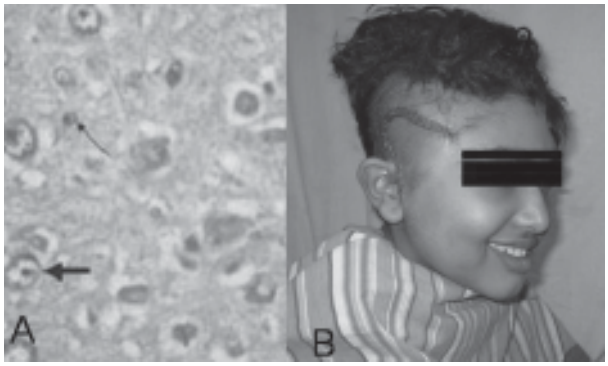

Fig.-2: *A-Histopathology (Microphotograph of ganglioglioma), B-Patient after operation showing operation site.*

complain suggestive of intracranial space occupying lesion or raised intracranial pressure. Physical and neurological examination including mental function revealed no abnormality. Initial 04 months she was on carbamazepine. Then she was on two Anti Epileptic Drug (AED) [Carbamazepine and sodium valproate] for last 16 months but seizure frequency was not reduced rather increased. Scalp EEG recording showed epileptic spikes originating from left temporal region. MRI of brain showed features of left amygdalo hippocampal mass lesion (1x1.5x1cm) suggestive of low grade glioma.

She underwent amygdalohippocampectomy with lesionectomy plus standard anterior temporal lobectomy. Histopathology revealed grade-1 ganglioglioma. Postoperatively she recovered uneventfully. She was put on carbamazipine 100mg tablet thrice daily that was tapered to 100mg tablet twice daily, 4 months after operation. She has been seizure free for last seven months. Post operative MRI of brain at the end of 4th months after operation showed no residual or recurrence tumor and there was no visual field defect or nominal aphasia.

Case 2 (Figure3):

A seven years old girl presented with sudden staring followed by unconsciousness and left sided tonic-clonic convulsion involving left upper and lower limbs for last 4 years. All these events persisted only 40-50sec.She had history of mild birth asphyxia. Her family history of such kind of illness is negative. Her parents visited several physicians both in the country and abroad. She took several AEDs in combination for several years without significant improvement in seizure control. There was 25-30 seizure attacks per day even with multiple AEDs (Carbamazepine + Phenobarbitone + Valproic acid). Scalp EEG revealed abnormal epileptic spikes originating from right centro-temporal region. MRI of brain showed features of right mesial temporal sclerosis.

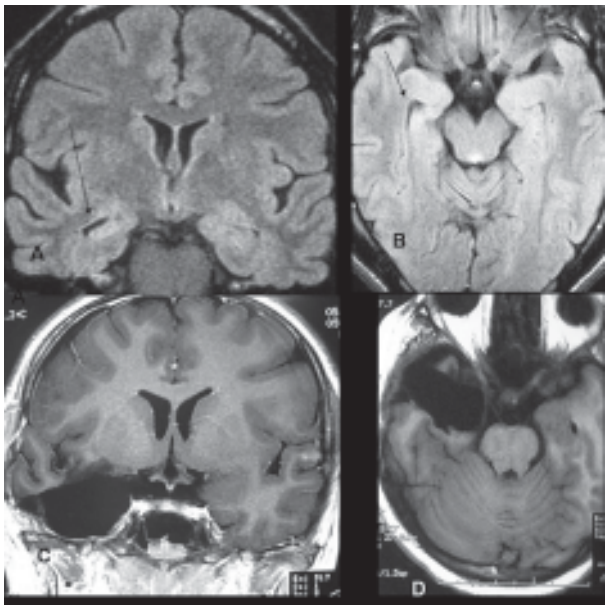

Fig.-3: *A-MRI of brain (FLAIR) coronal image and B-Axial mage showing right mesial temporal sclerosis (MTS) with compensatory dilatation of temporal horn in comparison to left temporal horn. C&D-MRI of brain T1 weighted images coronal and axial view respectively after amygdalohippocampectomy and standard anterior temporal lobectomy.*

She underwent right amygdalohippocampectomy plus standard anterior temporal lobectomy. Postoperatively she recovered uneventfully. She was given carbamazepine 100mg tablet thrice daily that was tapered 04months after operation. She is seizure free for last six month with carbamazepine tablet 100mg 12 hourly.

Case 3 (Figure4):

A 12 years old boy presented with sudden transient global headache, nausea followed by unconsciousness for 1-2 minute, after that he regains consciousness, but he remains unaware of place and person for another 3-5 minutes. He was suffering from this event repeatedly for the previous five years. Initially there was 12-15 attacks per year that increase to 3-5 attacks per day even with adequate AEDs treatment (Carbamazepine + Sodium Valproate + Phenytoin). His family history is negative for such kind

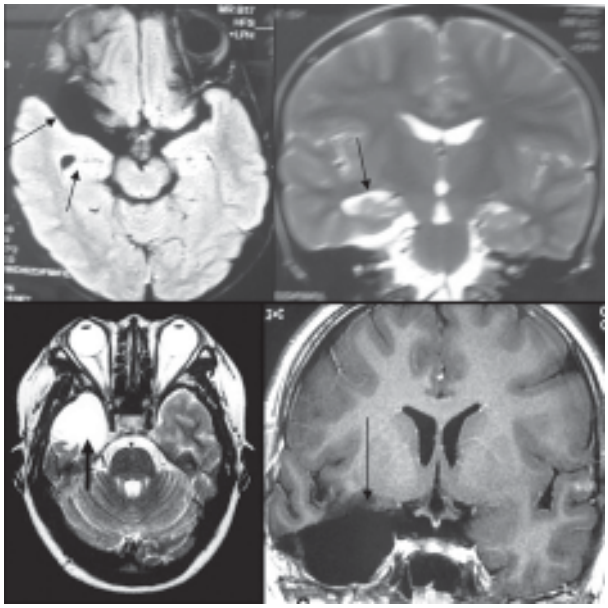

Fig.-4: *MRI of brain A-Axial mage (FLAIR) & B-Coronal image(T2W) showing right mesial temporal sclerosis (MTS) with compensatory dilatation of temporal horn in comparison to left temporal horn with anterior temporal arachnoid cyst and right temporal hypoplasia. C&D-MRI of brain T2 weighted axial image and T1W coronal image respectively after amygdalohippocampectomy with excision of arachnoid cyst and standard anterior temporal lobectomy*

of illness and his birth history was normal vaginal delivery with no significant event. Scalp EEG revealed abnormal epileptic spikes originating from right posterior centro-temporal region along with right anterior temporal slowing. MRI of brain showed features of right mesial temporal sclerosis (Right temporal horn apparent dilatation, decreased hippocampal volume and punctuate hyperintensity in hippocampus) and right temporal lobe hypoplasia along with large anterior temporal arachnoid cyst (3x2x3cm) without mass effect.

He underwent right amygdalohippocampectomy plus standard anterior temporal lobectomy with excision of arachnoid cyst. Postoperatively she recovered uneventfully. He was put on carbamazepine 100mg tablet thrice daily after operation. He is seizure free for last three month with carbamazepine. He has no clinical or perimetric visual field defects.

Discussion:

Epilepsy surgery is a interesting and fascinating subject to a neurosurgeon. William MacEwen (1848–1924) and Victor Horsley (1857–1916) in London were the first to localize and remove epileptogenic lesions, as identified by their symptomatogenic zone, according to the pioneering work of John Hughlings Jackson (1835–1911).[2]After a long dormant stage(due to invention of wide range of AEDs) epilepsy surgery developed tremendously in last three decades.[2] Among the drug resistant intractable epilepsy, temporal lobe epilepsy is the commonest one (about 80%) in both pediatric and adult patient and it responds beautifully to surgery.[1,2] Like obstructive jaundice (which is called surgical jaundice) temporal lobe epilepsy is sometimes called the surgical epilepsy. Common causes of intractable surgical epilepsy in childhood is cortical dysplasia, MTS, temporal lobe and other lobe tumors(DNET, ganglioglioma and others),cortical atrophy, stroke, trauma, vascular lesion(AVM, cavernoma)etc.[1,2,3]

In case-1 the patient was suffering from intractable TLE. Imaging confirmed a epileptogenic lesion in the amygdala and hippocampal area which was a low grade ganglioglioma. Low grade ganglioglioma is one of the common benign lesion in this area causing TLE specialy in children for which surgery is needed.[3] Appropiate surgical intervention is usually curative.

In case-2 intractable TLE was due to MTS. This girl had history of birth asphyxia, so this MTS can be considered due to birth asphyxia which is a common cause of MTS. In case-3 intractable epilepsy was due to MTS with temporal lobe hypoplasia rather than arachnoid cyst. Epilepsy associated with MTS is

usually AEDs resistant and needs surgery. Result of surgery in these cases is excellent. In these cases combination of AEDs with adequate dose failed to control disabling epilepsy, but surgical intervention brought a significant symptomatic improvement. Literature suggests that in most of the cases surgery is curative if there is no second focus or at least good control of epilepsy can be achieved with AED.[2,3]

We went for amygdalohippocampectomy with plus standard anterior temporal lobectomy in two cases and amygdalohippocampectomy with plus standard anterior temporal lobectomy with excision of arachnoid cyst in one case but selective amygdalohippo-campectomy (with lesionectomy where applicable) could be another surgical option to these lesions [2]. Size of the lesion, possibility of different pathology, narrow space through trans-sylvian route and possibility of manipulation and damage of critical vessels and neural structures and most importantly less familiarity of the approach along with less success rate of amygdalohippocampectomy in controlling epilepsy helped us to take decision in favour of trans middle temporal gyrus approach amygdalohippocampectomy with lesionectomy plus standard anterior temporal lobectomy instead of selective amygdalohippocampectomy in the hope to cure epilepsy as well as the lesion. It has been suggested that the amount of tissue resected in mesio-temporal operations is crucial for surgical success in mesial TLE [4,5,6,7]. In a randomized, prospective study comparing the trans-sylvian with the transcortical approach, seizure outcome was similar[8,9]. Middle temporal gyrus approach does not usually damage the optic radiation (Meyer loop) unless excessive backward and upward retraction or misdirection of transcortical incision[10]. In all of three cases we found no visual field defect both clinically and in perimetry.

The rate of minor complications was 3.6%, and the rate of major complications was 1.26% in the Zürich series of 478 amygdalohippocampectomies. Persisting hemiparesis occurred in 0.84% as a result of choroidal infarcts of the internal capsule[11]. Generally, complication rates of epilepsy surgery are relatively low and thought to be acceptable, with approximately 1 to 2% permanent morbidity[12]. Typical neurological complications after surgery for TLE include temporary dysphasia or hemiparesis as caused by manipulation-induced brain swelling or brain contusion, small vessel infarction, and hemorrhage. There are the classic surgical problems such as infection, thrombosis etc., in the range of 2 to 4%, which rarely cause permanent damage[12]. The mortality rate is clearly below 1% in most series.

In certain developmental tumors, e.g., ganglioglioma and dysembryoplastic neuroepithelial tumor that usually occurs in pediatric patient can be treated with excellent seizure-control and disease control rates in most patients [6] . Seizure control rate in MTS after surgery is excellent [2,3]. Certain types of low-grade gliomas (e.g., isomorphic subtype of low-grade astrocytoma, pilocytic astrocytoma) causing intractable epilepsy can be operated on with excellent results[13,14,15,16]. In a series limited to preoperatively tailored resections for lesional (nonsclerotic) mesial TLE, satisfactory seizure control was obtained in 86% of patients[17]. Outcome with lesionectomy and corticectomy was excellent, especially when a tumor was present (95% satisfactory seizure control) [18]. Although it depends more on patient selection than surgery, it should be noted that operating on children and adolescents with epilepsy is extraordinarily promising with respect to seizure control and neuropsychological and psychosocial outcomes [19,20,21,22,23]. Outcome of epilepsy surgery is assessed by 'Engel grading' system. In point of epilepsy control in our cases final comment can not be made as follow up period is still small for Engel grading but in this small follow up period control of seizure is excellent.

For successful initiation, development, implementation and up-gradation of 'epilepsy surgery program' by a team formed of epileptologist, neuropathologist, neurophysiologist, neuroradiologist and neurosurgeon, the facilities of video EEG, MRI and other imagings are very essential. Finally it should be realized that an 'expert' epilepsy neurosurgeon is helpless without an epileptologist who will at first get hold of the patients and play a key role in the effective medical and surgical management of epilepsy. Epilepsy surgery today is more effective with better seizure control rates in indicated cases; it is safer and less invasive with lower morbidity and mortality rates.

Conclusion:

Though follow up period is not adequate and cases of number is only three, even then our initial experience with epilepsy surgery in pediatric patients suggests that surgical result is very good in intractable TLE with concordant lesion.

References:

1. Greenberg MS, Seizures. In: Greenberg MS, (eds). Hand book of Neurosurgery. 5th edition, Newyork, Thieme, 2001. P. 254-284.
2. Schramm J, Clusmann H. Surgery of epilepsy. Neurosurgery 2008, 62[SHC Suppl 2]: S463–481.
3. Harvey S., Cross JH, Shinnar S., Matheren BW., The ILAE Pediatric Epilepsy Surgery Survey Taskforce. Defining the spectrum of international practice in pediatric epilepsy surgery patients.Epilepsia 2008; 49(1): 146-155.
4. Awad IA, Katz A, Hahn JF, Kong AK, Ahl J, Lüders H. Extent of resection in temporal lobectomy for epilepsy. I. Interobserver analysis and correlation with seizure outcome. Epilepsia 1989; 30: 756–762.
5. Bonilha L, Kobayashi E, Mattos JP, Honorato DC, Li LM, Cendes F. Value of extent of hippocampal resection in the surgical treatment of temporal lobe epilepsy. Arq Neuropsiquiatr 2004; 62:15–20.
6. Nayel MH, Awad IA, Luders H. Extent of mesiobasal resection determines outcome after temporal lobectomy for intractable complex partial seizures. Neurosurgery 1991; 29:55–61.
7. Renowden SA, Matkovic Z, Adams CB, Carpenter K, Oxbury S, Molyneux AJ, et al. Selective amygdalohippocampectomy for hippocampal sclerosis: Postoperative MR appearance. AJNR Am J Neuroradiol 1995; 16: 1855–1861.
8. Lutz MT, Clusmann H, Elger CE, Schramm J, Helmstaedter C. Neuropsychological outcome after selective amygdalohippocampectomy withtranssylvian versus transcortical approach: A randomized prospective clinical trial of surgery for temporal lobe epilepsy. Epilepsia 2004; 45:; 809–816.
9. Schaller C, Jung A, Clusmann H, Schramm J, Meyer B. Rate of vasospasm following the transsylvian versus transcortical approach for selective amygdalohippocampectomy. Neurol Res 2004; 26: 666–670.
10. Chowdhury FH, Khan AH. Anterior and lateral extension of optic radiation and safety of amygdalohippocampectomy through middle temporal gyrus: A cadaveric study of 11 cerebral hemispheres. Asian Journal of Neurosurgery 2010; 4: 78-82.
11. Wieser HG. Selective amygdalohippo-campectomy has major advantages. In: Miller JW, Silbergeld DL (eds): Epilepsy Surgery: Principles and Controversies. New York, Taylor & Francis, 2006, p. 465-478.
12. Behrens E, Schramm J, Zentner J, König R: Surgical and neurological complications in a series of 708 epilepsy surgery procedures. Neurosurgery 1997; 41:1–10.
13. Blümcke I, Luyken C, Urbach H, Schramm J, Wiestler OD. An isomorphic subtype of long-term epilepsy-associated astrocytomas associated with benign prognosis. Acta Neuropathol (Berl) 2004; 107:381–388.
14. Daumas-Duport C, Scheithauer BW, Chodkiewicz JP, Laws ER, Vedrenne C. Dysembryoplastic neuroepithelial tumor: A surgically curable tumor of young patients with intractable partial seizures. Report of thirty-nine cases. Neurosurgery 1988; 23: 545–556.
15. Luyken C, Blümcke I, Fimmers R, Urbach H, Elger CE, Wiestler OD, et al. The spectrum of long-term epilepsy-associated tumors: Longterm seizure and tumor outcome and neurosurgical aspects. Epilepsia 2003; 44: 822–830.
16. Morris HH, Matkovic Z, Estes ML, Prayson RA, Comair YG, Turnbull J, et al. Ganglioglioma and intractable epilepsy: Clinical and neuro-physiologic features and predictors of outcome after surgery. Epilepsia 1998; 39: 307–313.
17. Clusmann H, Kral T, Fackeldey E, Blümcke I, Helmstaedter C, von Oertzen J, et al. Lesional mesial temporal lobe epilepsy and limited resections: Prognostic factors and outcome. J Neurol Neurosurg Psychiatry 2004; 75: 1589–1596.
18. Schramm J, Kral T, Grunwald T, Blümcke I: Surgical treatment for neocortical temporal lobe epilepsy: Clinical and surgical aspects and seizure outcome. J Neurosurg 2001; 94: 33–42.
19. Blume WT: Temporal lobe epilepsy surgery in childhood: Rationale for greater use. Can J Neurol Sci 1997; 24: 95–98.

20. Clusmann H, Kral T, Gleissner U, Sassen R, Urbach H, Blümcke I, et al. Analysis of different types of resection for pediatric patients with temporal lobe epilepsy. Neurosurgery 2004; 54:847–859.

21. Gleissner U, Sassen R, Lendt M, Clusmann H, Elger CE, Helmstaedter C: Pre- and postoperative verbal memory in pediatric patients with temporal lobe epilepsy. Epilepsy Res 2002; 51: 287–296.

22. Kral T, Kuczaty S, Blümcke I, Urbach H, Clusmann H, Wiestler OD, et al. Postsurgical outcome of children and adolescents with medically refractory frontal lobe epilepsies. Childs Nerv Syst, 2001;17:595–601.

23. Mathern GW, Giza CC, Yudovin S, Vinters HV, Peacock WJ, Shewmon DA, et al. Postoperative seizure control and antiepileptic drug use in pediatric epilepsy surgery patients: The UCLA experience, 1986–1997. Epilepsia 1999; 40: 1740–1749.