CASE REPORT/CAS CLINIQUE

CLINICO-RADIOLOGIC FEATURES AND SURGICAL TREATMENT OF BONE- INVASIVE DOMINANT HEMISPHERIC MENINGIOMAS - A REPORT OF 2 CASES IN YOUNG NIGERIANS

ASPECTS CLINICO-RADIOLOGIQUES ET TRAITEMENT CHIRURGICAL DES MÉNINGIOMES ENVAHISSANTS L'OS. A PROPOS DE 2 CAS OBSERVE CHEZ DES JEUNES NIGERIANS

ENOCH Uche¹ IKENNA Onwuekwe² NNEKA Iloanusi³ NKECHI Uche⁴ OLUCHI Ekenze²

- 1. Neurosurgery Unit, Department of Surgery, University of Nigeria Teaching Hospital, Ituku-Ozalla, Enugu, Nigeria
- 2. Neurology Unit, Department of Medicine, University of Nigeria Teaching Hospital, Ituku-Ozalla, Enugu, Nigeria
- 3. Department of Radiology, University of Nigeria Teaching Hospital, Ituku-Ozalla, Enugu, Nigeria
- 4. Department of Ophthalmology, University of Nigeria Teaching Hospital, Ituku-Ozalla, Enugu, Nigeria

E-Mail Contact - ENOCH Uche : kechyenny (at) yahoo (dot) com

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ABSTRACT

Background

Convexity meningiomas are the commonest type of intracranial meningiomas. There are no previous reports on the clinico-radiologic profile of patients with bone-invasive non-sphenoidal calvarial meningiomas from South East Nigeria.

Aim

This is the report of two young Nigerians with convexity meningiomas. One had been mismanaged as a case of epilepsy for 4 years. Both cases had successful neurosurgical intervention.

Methods

The clinical features of the patients are presented with relevant results of investigations including neuroimaging studies. Careful literature search was made using library and internet sources.

Results

The first case is a 20- year old woman with a 4 -year history of jacksonian seizures affecting the right limbs, chronic headache and progressive visual loss. She had been managed unsuccessfully with both orthodox and traditional herbal remedies prior to being referred to the University of Nigeria Teaching Hospital Enugu. The second case is a 26- year old man with one year history of chronic headache and jacksonian seizures of the right face, arm and leg. Sensorium was unimpaired in both cases and neurological deficits were present. Both patients had cranial computerised tomograms showing bone- invasive convexity meningiomas. Successful medical/ neurosurgical management was instituted in both instances.

Conclusion

The paucity of neurological/neurosurgical services in Nigeria makes for mismanagement of several disorders amongst the populace. Convexity meningiomas may be heralded by focal epilepsy. Neuroimaging helps to confirm suspected cases and appropriate medical/surgical intervention reduces the risk of mortality/ severe morbidity.

INTRODUCTION

Focal epilepsy especially with a jacksonian march of symptoms may underlie the existence of an intracranial tumour. Convexity meningiomas are the most common type of intracranial meningiomas and maybe bone-invasive. The first recorded surgery for removal of a brain tumour was for a convexity meningioma(14) There are infrequent reports on the clinico-radiologic profile of patients with bone-invasive non-sphenoidal calvarial meningiomas in the African literature and none identified from Enugu, South East Nigeria. These two cases of bone invasive meningiomas encountered at the University of Nigeria Teaching Hospital, Enugu, are quite illustrative and are thus presented.

CASE SUMMARY 1

O.E, a 20- year old right- handed female presented to the Neurology Clinic of the University of Nigeria Teaching Hospital (UNTH) Enugu in February 2010 with a 4-year history of right- sided Jacksonian seizures associated with hemi-body weakness. In the preceding 2 months she had developed progressive visual loss in both eyes.

Examination revealed a pale, young woman with no other signs of chronic systemic ailments. She was fully conscious and well oriented in time, place and person. She had impaired recent memory and a mild motor aphasia. There was anisocoria with a larger left pupil (5mm) which was also poorly reactive to light. Visual acuity was to hand movement on the right while the left eye had nil light perception. She had bilateral secondary optic atrophy. There was a right facioparesis upper motor neurone type and a right spastic hemiparesis with a right Babinski sign. She had a pulsating giant left hemicranial swelling measuring 13cm x15cm in its largest diameter associated with markedly distended scalp veins. Cranial computerised tomogram (CT) showed a giant left hemicranial extra-axial mixed density mass with significant contrast enhancement. There was associated hyperostotic and osteolytic skull changes with both extracranial and intracranial neovascularisation and severe mass effects. These findings are demonstrated in Figure 1 (A-C).

She was commenced on anticonvulsants (tabs. carbamazepine) and steroids and referred to the Neurosurgery Unit where she had a left fronto-temporo-parietal craniotomy and Simpson grade 1 microsurgical excision of the tumour in 2 stages with duraplasty using fascia lata graft. Operative findings included a massively neovascularised left hemicranial scalp with enlarged left superficial temporal and left occipital arteries; a highly vascularised yellowish- gray extra-axial left hemispheric convexity tumour with multiple foci of irregular lytic and hyperostotic calvarial bone changes. The gross findings are as shown in Figure 2 (A -C). Histology of tumour specimen showed fibroblastic meningioma (WHO Grade 1). Post-operatively, the patient's visual acuity improved bilaterally to counting fingers. She however had residual dysphasia and right hemiparesis.

CASE SUMMARY 2

D.E, a 25- year old right handed male undergraduate student, presented to the Neurosurgery Clinic of UNTH Enugu in September 2010, with 1-year history of headache with right hemi-body weakness and a 6- month history of right simple motor seizures with a Jacksonian march from the face and sequentially involving the upper and lower extremities.

Examination showed a young adult male in no obvious respiratory distress and with normal vital signs. He was fully conscious but had expressive dysphasia. Both pupils were 3 mm in size and had equal and normal direct and consensual reaction to light. There was a right facioparesis, upper motor neurone type. He also had a right spastic hemiparesis worse distally. Power in the finger flexors was grade 0 on the right but normal on the left. He had a right extensor plantar response. There was a 10cm x 8cm frontal scalp/calvarial mass with enlarged left superficial temporal vessels and dilated scalp veins.

Cranial CT with contrast showed a left fronto-parietal convexity extra-axial brilliantly contrast enhancing mass with overlying calvarial hyperostosis, peri-lesional oedema and mass effects. The patient after medical stabilization had microsurgical excision of the tumour with attached dura/ overlying bone including 1 cm of surrounding normal dura and bone. Duraplasty was performed with galeopericranial graft. Figure 3 (A-F) shows some of the pre- and post-operative findings for case 2. The patient had wound sepsis post-operatively which was managed with antibiotics and daily wound dressings. At discharge there was remarkable improvement in the hemiparesis with power in the finger flexors improving to grade 4+ on the

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right. There was however residual dysphasia. Patient is being followed up at the Surgical Outpatient Clinic, awaiting acrylate cranioplasty.

DISCUSSION

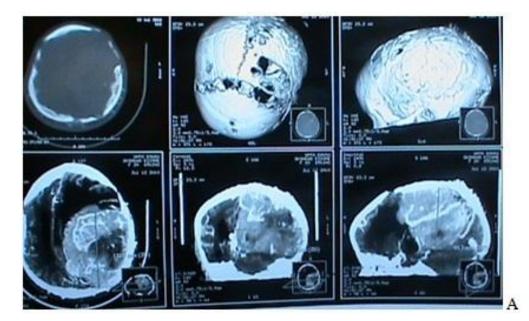
The first recorded cranial surgery performed for the treatment of brain tumour was for the excision of a left frontal convexity meningioma in a 14- year old female(14). However, the first published series on meningiomas was the seminal work by Harvey Cushing who also coined the term 'meningioma' to designate a group of previously disparate tumors arising from the same mesothelial mother cells-'the meningocyte'(5). Meningiomas based on location have been classified into convexity, midline, skull base and other. Although skull base meningiomas were more common in Cushing's series(6), convexity meningiomas were seen more commonly in Black's more recent series of 807 meningioma patients(2,11). However reports on the clinicoradiologic profile of patients with bone-invasive non-sphenoidal convexity meningiomas are infrequent in reported literature especially from our sub-region. This is without prejudice to the fact that convexity meningiomas are among the most common intracranial meningiomas(2,4,11). Although invasive behaviour is associated with WHO grade 2 and 3 meningiomas, histo-morphologically benign meningiomas (WHO grade 1) have occasionally been associated with invasion of the overlying skull and sometimes also of the underlying brain. These sub-sets of tumours are thought to express in addition to other humoral markers associated with benign meningiomas(3), the marker 'SPARC', which is currently thought to be a potential marker for the heightened invasive phenotype in meningiomas irrespective of the histological grade(15). The bone invasive phenotypes of meningioma encountered our case series invites the speculation of a possible role of the marker 'SPARC' especially as these two cases were WHO grade 1 tumors occurring in young adults.

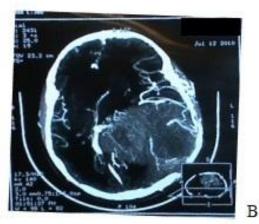
The association between Jacksonian seizures and convexity meningiomas occurring in proximity to the motor strip is well reported in literature(17). In one series, 40.7% of patients with convexity meningiomas had seizures(9). Both patients in our report had Jacksonian seizures and hemiparesis as main presenting features. Whereas seizures were adequately controlled with anticonvulsants, motor weakness improved in the post-operative period.

Although the median age of occurrence of meningiomas in many clinical series is \geq 40 years(13), the occurrence in our patients both < 30 years of age, invites the speculation of a possible correlation between young age and invasiveness in convexity meningiomas. However any such association will require a larger sample size for statistical validation.

Computed tomography (CT) is very useful in the diagnosis of meningiomas. It can demonstrate bony alterations (hyperostosis, bone invasion), significant contrast enhancement due to hyper-vascularity and in 25% of instances calcification of the tumour(13). Our patients were evaluated with computed tomography. In case 1 with CT evidence of focal hyperostosis, there was also significant areas of bone invasion, osteolysis and hyper-vascularity of the overlying scalp (Figure 1 A-C) depicting that the tumour derived supplementary blood supply from the scalp. Magnetic resonance imaging (MRI scan) is very useful in further evaluation of patients with intracranial meningiomas and in the planning of operative approach and corridors to tumour excision. However our patients had serious financial constraints and could not afford MRI scan.

Although most meningiomas are benign tumours, they have significant rates of recurrence, 5 - 15% (1,7,10,16). The most important determinant factor in the recurrence of meningiomas is the extent of tumour excision(8,16). Simpson grade 1 tumour excision, which was performed in case 1 of this study and entails total excision of tumour along with any bony, dural and sinus attachment, is associated with the lowest rate of recurrence(8). To further reduce the rate of recurrence of meningiomas, some authors advocate, where possible, excision of the tumour along with a margin of normal surrounding bone and dura (grade 0 tumour excision). This grade of tumour excision has been associated with zero recurrence rate(8), and was performed in case 2 of this report. Although complete resection is usually the goal in the surgical treatment of convexity meningiomas, the ultimate decision, however is best determined by assessing the patients surgical risks. Ojemann and Black have highlighted some of the management decision challenges in the management of these tumors(12).





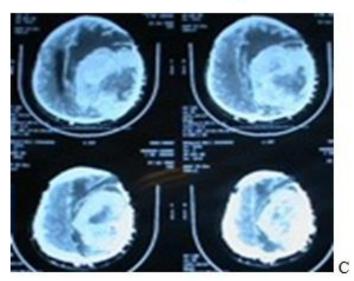
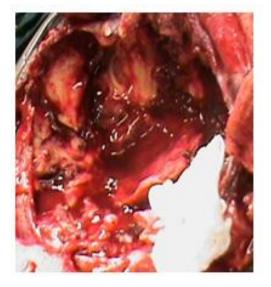
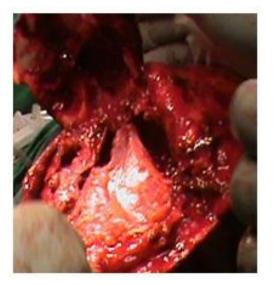


Figure 1(A-C)
 showing neuroimaging findings on cranial CT for case 1.
 A- contrast enhancement and mass effect of a huge left temporo-parietal convexity meningioma
 B- significant mass effect with mid-line shift.

• C- significant contrast enhancement of the tumour.



A

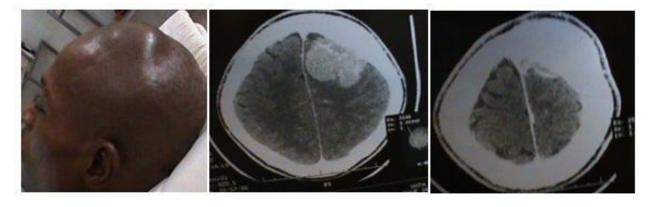


В



C

Figure 2 (A- C) Ahowing gross findings at neurosurgery for case 1.



A

В

С



D

E

F

Figure 3 (A -F)

Illustrating pre-operative and operative features in case 2.

- ▶ A- shows the pre-operative photograph of a 26- year old male with left fronto-parietal tumour.
- B and C- show contrast cranial CT studies revealing an extra-axial dural based tumour with calvarial hyperostosis.
 - D- shows the patient 2 weeks post- operatively.
 - E- demonstrates the intra-operative craniotomy flap showing bone invasion.
 - F- shows intra-operative tumour bed and adjoining normal brain at the end of tumour excision.

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