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Case Study

Vestibular Schwannoma in Pregnant Female: Anesthetic Management of 2 Case Studies

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Summary

The brain tumors are infrequent association with pregnancy, usually undiagnosed and worsen the outcome of mother as well as fetus. The tumor also takes aggressive course during pregnancy which may unmask a previously unknown diagnosis as the symptoms generally deteriorate particularly in this period. Here in our set up we reported two rare cases of vestibular schwannoma, case-1 was presented with history of severe headache, worsened nausea and vomiting, hearing loss with some neurological problems (facial weakness or gait imbalance) with 22 weeks pregnancy in outpatient department. Due to incomplete neural development and organogenesis she was advised to follow up in outpatient department and continue the treatment. At 32 weeks she developed premature rupture of membrane and gone into respiratory distress. She underwent emergency caesarean section under general anesthesia, had attack of seizures peri-operatively; non-contrast computerized tomography revealed gross obstructive hydrocephalus, managed by External Ventricular Drain (EVD) insertion followed by subtotal excision of tumor.

While case-2 came in early 3rd trimester (26 weeks by per abdominal examination and 22 weeks by USG), due to persistent symptoms of obstructive hydrocephalus with acute neurological deterioration craniotomy was performed uneventfully and tumor excised totally. After completion of full term, she delivered spontaneously a healthy male baby. There were no related maternal or fetal morbidity/ or mortality in both the cases.

Keywords

Anesthetic Management; Pregnant Female; Vestibular Schwannoma

Introduction

Pregnancy may aggravate the natural history of an intracranial tumor or increase the growth of a previously existing tumor, and unmask a previously unknown diagnosis [1]. During pregnancy the reported incidence of non-obstetric surgery is 0.75-2% [2] and intracranial neoplasm as 6.9 /100,000 in aged 24-35 years parturient [3]. In a population-based study; Haas et al., reported that the number of meningiomas, acoustic neuromas, and primary malignant intracranial neoplasms diagnosed during pregnancy was less than expected with the ratio of observed/expected tumors associated with pregnancy was found to be 0.38 [4]. Surgery for an intracranial tumor in parturient is even rarer.

Schwannomas (Neurilemmomas) are benign, encapsulated, slow-growing, and usually solitary tumors originating from Schwann cells of the peripheral nerve sheath with uncertain etiology [5]. Vestibular schwannomas are relatively common tumors that arise from vestibulocochlear nerve and represent ~80% of cerebellopontine angle masses. The clinical picture may include headache, worsened nausea and vomiting, tinnitus, hearing loss with some neurological problems (facial weakness or gait imbalance), in large tumors brain-stem and cerebellar compression with involvement of additional cranial nerves. Large vestibular schwannomas present a great challenge in peripartum management of both the mother and the foetus.

They impose a unique risk which needs attention and formulation of proper guidelines for the anaesthetic management. The aim of these rare case studies is to describe clinicopathologic and radiographic features of vestibular schwannoma involving the pregnancy stage and to review the literature, discuss this unusual clinical entity, and suggest evidence-based strategy for the perioperative anesthetic management of pregnant females with brain tumors.

Presentation and Management of Case 1

A 28-year-old primigravida, 24 weeks gestation came to neurosurgery Outpatient Department (OPD)-11, JNMCH, AMU with left ear hearing loss for the last 1 year and left facial weakness and gait imbalance for last 2 months. There is history of on and off nausea, vomiting and headache. She was neglecting these symptoms in early stage of pregnancy as she considers it as normal physiological changes of pregnancy. She got medical attention when her husband noticed abnormal facial pattern with recurrent left ear problems. General examination revealed moderate pallor, per abdominal examination showed a uterine size consistent with 22 weeks of gestation with normal fetal heart rate. Neurologically she was conscious and well oriented to time, place and person. She had bilateral papilledema, left facial paresis (House and Brackman grade [6] III), absent left corneal reflex and mild ataxia. Biochemical and hematological parameters were in normal range except hemoglobin-8.9 gm/dl.

MRI revealed - A large axial well defined altered signal intensity solid cystic predominantly cystic lesion with fine septation and irregular eccentric solid component involving the Lt. Cerebellopontine Angle (CPA) which was hyperintense on T2-WI weighted image. There was mass effect in the form of compression and displacement of the 4th ventricle towards right side with mild dilatation of 3rd and B/L lateral ventricles suggestive obstructive hydrocephalus (Figures 1 and 2). The provisional diagnosis of cystic vestibular schwannoma was made.

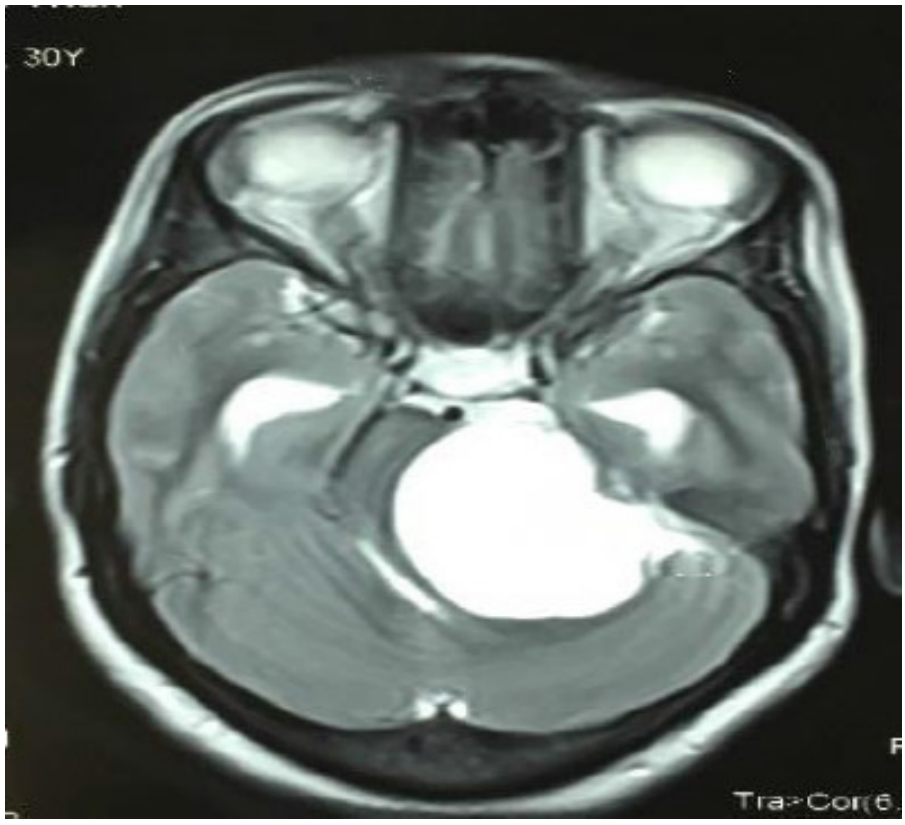

Figure 1: Axial T2 contrast MRI showing left cystic vestibular schwannoma measuring 4.6 X 4.2 × 4.8 cm (AP.TR, CC) in size with compression and displacement of 4th ventricle towards right side.

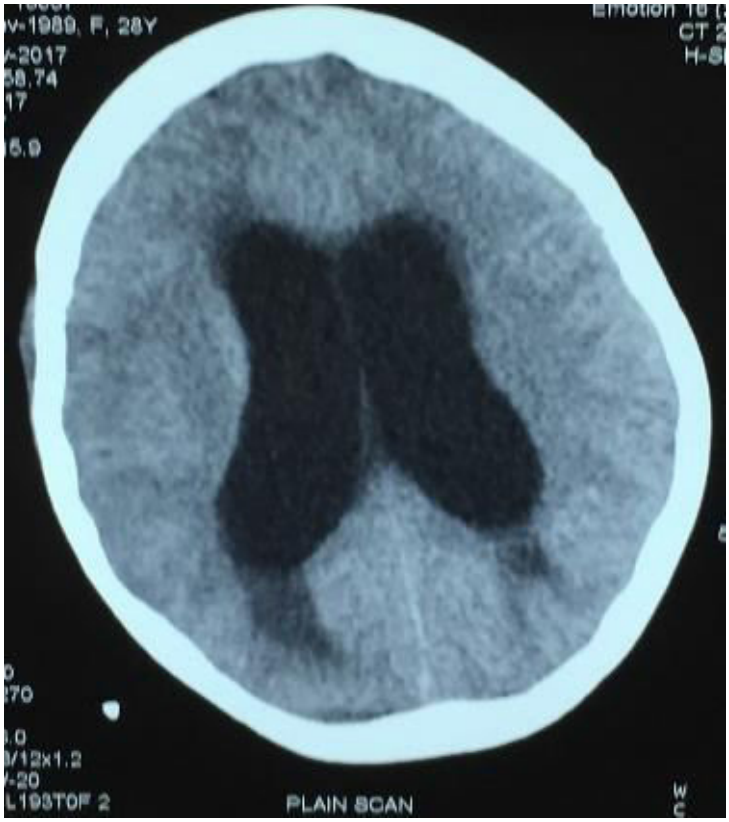

Figure-2: Axial noncontrast CT scan showing dilatation of b/l lateral ventricles s/o obstructive hydrocephalus.

USG-abdomen showed a normal viable fetus corresponding to the duration of gestation. As the patient was received at 22 weeks, the period of neural cerebrum development and incomplete organogenesis was neurologically stable was advised to follow regularly in neuro and Obstetric OPD till 32 weeks with a plan for early delivery after 32 weeks followed by tumor resection. However, at 31 weeks of gestation, headache and neurologic symptoms worsened and she was admitted in neurosurgery ward, discussed with Obstetric and Gynae (OBG) unit for Cesarean Section (C/S) at 32 weeks and tab. dexamethasone 4 mg qid was started for fetal lung maturity.

In neuro ward she had Premature Rupture of Membrane (PROM) with respiratory distress, for this an emergent Cesarean Section (C/S) was performed under GA. However, during C/S she had an episode of seizure, for this an emergency Non-Contrast Computerized Tomography (NCCT)-Head was done; revealed gross obstructive hydrocephalus. For which External Ventricular Drain (EVD) was inserted and therapeutic drainage of CSF was done. Patient was shifted to ICU in intubated state. In ICU bedside fundoscopy found grade III papilledema on Frisen scale. Case discussed with anesthesiologist for excision of tumor tomorrow.

On next day she was taken under General Anaesthesia (GA), preop bilateral Optic Nerve Sheath Diameter (ONSD) measurement by USG, mean was found to be 5.6 mm. Following scalp block for perioperative pain management, left retro-auricular sub-occipital craniotomy with subtotal excision of tumor was done successfully. In per-op findings the tumor was densely adhered to lower cranial nerves with the tonsils herniated below foramen magnum. Tumor mass sent for histopathological evaluation at pathology department. On the same day after 4 hours of surgery she was successfully extubated, and fundus examination as well as ONSD measurement by USG done post-op, the mean ONSD was found to be 4.9 mm and 5.2 mm, at 6 and 12 hours. Histopathology confirmed the diagnosis of schwannoma with mixed Antoni A and B component. Patient had an uneventful post-op course with minimal facial paresis (House-Brackmann score was grade II). Post-op NCCT head showed a complete excision of tumor (Figure 3). On follow up 1 month later, mother and child were doing well.

Presentation and Management of Case 2

26 yr/F, in her third trimester came to JNMCH, AMU

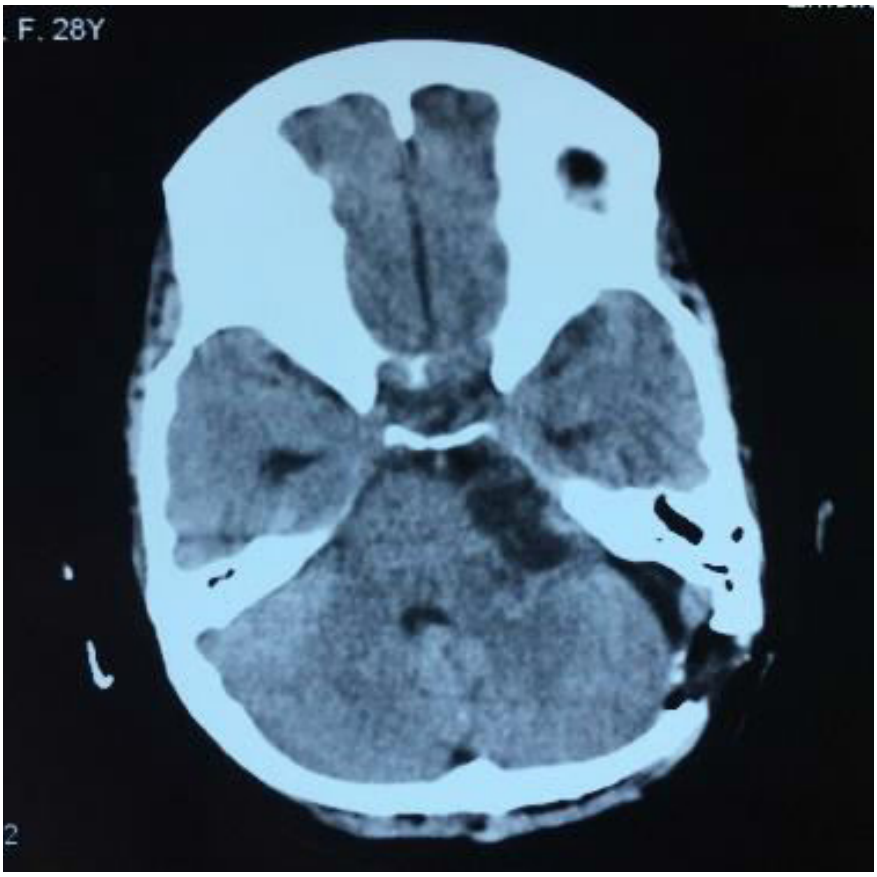

Figure 3: Non-contrast CT brain imaging after resection of vestibular schwannoma with no evidence of residual tumor.

presented with complains of headache, intermittent vomiting and progressive hearing loss on the right side for 6 months. For the past 3 months, she also had intermittent diplopia, gait ataxia with right sided swaying and facial paresis and numbness on the right half of face. Her general physical examination was unremarkable. Per abdominal examination revealed uterine size 26 weeks of gestation with normal fetal heart sounds. Neurological examination revealed bilateral severe papilledema and gaze evoked nystagmus, right lateral rectus paresis, about 50% sensory loss in trigeminal nerve distribution, absent corneal reflex, lower motor neuron facial paresis (House-Brackmann grade III), sensory neural deafness and gait ataxia. Hematological and biochemical parameters were within normal limits.

USG abdomen showed a single live fetus of 22 weeks gestation. MRI scan showed a mixed intensity mass in the right CP angle, measuring 4.54 cm, centered on the internal auditory meatus, causing severe brain stem compression and mass effect on the fourth ventricle, producing obstructive hydrocephalus. Pre-op ONSD measurement by USG found to be 5.8 mm and fundus examination showed Frisen scale of papilledema grade III. Audiometry confirmed the sensory neuronal deafness with poor speech reception and discrimination. In view of the persistent symptoms of obstructive hydrocephalus with neurological deterioration she was operated under GA via a right sub occipital craniotomy in left lateral position. Total excision of tumor was done. It was soft, friable and extremely vascular with hemorrhages inside. Facial nerve was very thin but could be preserved anatomically. Fetal heart was monitored during perioperative period by obstetric resident. Uterine relaxants were given to prevent spontaneous abortion. Patient had an uneventful postoperative recovery, was kept in ICU for further monitoring and maintenance, mean ONSD significantly reduced (4.9 mm) in post-op, 6 hour, and 12-hour intervals and fundus examination showed resolving papilledema. Histopathological examination confirmed the diagnosis of neurinoma/schwannoma as in histological sections the spindle cells with thin wavy nuclei arranged as typical Antoni A (with Verocay bodies) and Antoni B areas, nuclear palisading distribution (typical of a schwannoma). Postoperative CT scan showed complete excision of tumor. Pregnancy continued at term finally delivered a male baby spontaneously through vaginal route. At follow up after 5 months, mother and baby were doing well, but residual facial palsy still persisted in the mother.

Nevertheless, both the patients had noticeable significant improvement in hearing, facial weakness, vertigo and abnormal gait. The seizure activity was controlled by levetiracetam. They were doing well with their babies without any new neurologic deficit.

Anesthetic Management

We managed both our patient under GA, one delivered by caesarean section and other by spontaneous vaginal delivery after completion of full term. Patient-1 had probable diagnosis of vestibular schwannoma, at 31 weeks. gestation d/t features of severe obstructive hydrocephalus, was admitted in neurosurgery ward for close observation and fetal monitoring, tab. dexamethasone was started for fetal lung maturity. She had Premature Rupture of Membrane (PROM) gone into respiratory distress in the neurosurgery ward. Following emergency caesarean section, a healthy male child was delivered with Apgar score of 10/10, managed in ICU by EVD insertion for acutely raised ICP, followed by planned craniotomy on the 2nd postpartum day.

2nd Patient came in early 3rd trimester (26th week by per abdominal examination and 22 weeks by USG), urgent right sub occipital craniotomy was performed in left lateral position due to acute neurological deterioration.

A multidisciplinary approach was planned, apart from anesthesiologist and neurosurgeons, the obstetrician was called for foetal wellbeing monitoring. Patient positioned by putting the wedge under the right buttock to produce pelvic tilt of 15 degrees to the left (effectively displace the gravid uterus to the left) minimize aortocaval compression and supine hypotension syndrome. Noninvasive monitors were attached to record the baseline BP (to keep systolic >100 mmHg in perioperative period and for prophylactic/ early use of vasopressor drugs), HR, ECG, SpO_2 and temperature. Antibiotic prophylaxis was administered IV one hour prior to surgery. She was premedicated IV levetiracetam 500 mg slowly, inj. ranitidine 50 mg IV, inj. dexamethasone 8 mg IV, inj. metoclopramide 10 mg IV and inj. midazolam 0.04 mg/kg IV slowly were given.

Patient was preoxygenated with 100% oxygen for 5 min followed by induction with propofol (2 mg/kg). Under smooth rapid sequence induction and cricoid pressure, crash endotracheal intubation by using armoured tube was performed following relaxation with suxamethonium 75 mg IV. Pharmacological ablation of pressor response to laryngoscopy was done by IV lignocaine 1.5 mg/kg and fentanyl 1.5 mcg/kg IV. Tracheal tube taped on the contra lateral side to operation. She was put on controlled ventilation, O_2 and N_2O in 50:50 ratio, ventilatory setting were made to keep carbon dioxide tension ($Etco_2$) at steady-state (30-32 mm Hg) and sevoflurane (0.5-1%) concentration to provide balanced anaesthesia and vecuronium bromide infusion after bolus dose of 0.08 mg/kg for muscle relaxation. She was catheterized to monitor the urine output hourly. Quickly observed and treat the hemodynamic changes to preserve both cerebral and uteroplacental perfusion, maintaining hemodynamic stability is important, the Intra-Arterial BP (IABP), CVP line was put to achieve large bore IV access (administration of appropriate fluid, concentrated vasoactive drugs, central venous pressure monitoring, and aspiration of air emboli). Intraoperative hypotension was managed by inj. 6 mg of mephentermine IV stat. Scalp block was given to prevent scalp pin placement response, other haemodynamic changes and for postoperative analgesia. She was positioned supine with the head turned to contralateral side, shoulder roll was put, chin was assessed, and it was 2 finger breadths away from the sternum (flexion should not be severe enough to compromise venous return from the head). The head was placed in pins in the Mayfield skull clamp and the navigational frame is anchored to this and approached through right sub occipital craniotomy and total excision of the tumor was done. Inj. mannitol 0.5 mg/kg IV started slowly at the time of drilling with burr before the opening the dura. Arterial sample send to measure the PaO_2 and $PaCO_2$. IV fluid therapy consisted normal saline and lactated ringer to reduce the risk of cerebral edema and hyperglycemia, colloid solution (gelofusine or voluvin) used when blood loss was over 1 liter and blood loss replaced timely and accordingly. The propofol infusion was used to control the intraoperative hypertensive events (compensatory hyperactivity of the sympathetic nervous system in response to elevated ICP and compression of the brain stem (Cushing's reflex) [7]. Inj. acetaminophen 1 gm IV infusion was given for perioperative analgesia. Patient was extubated smoothly and monitored closely in high dependency unit.

Discussion

Wang and Paech [8] reported the "literature regarding neuroanesthesia for the pregnant woman is generally unhelpful with respect to evidence-based neuroanesthetic management for pregnant patient," and so the planning and decision-making must be based largely on general principles of neurosurgical and obstetric anaesthesia. They further advocated the case reports and small studies or case series thus form an important source of knowledge and experience [8].

In clinical scenario like headache, fullness in ear or vomiting or seizure (characteristics of CP Angle tumor) in early or late pregnancy can easily misdiagnosed as a case of hyperemesis gravidarum or eclampsia. During pregnancy symptoms may be exacerbated because of increased tumor growth or edema,

increased vascularity or pregnancy-related immunotolerance [9]. It has been reported that acute neurological deterioration of suprasellar and cerebellopontine angle tumors mandating resection during pregnancy [10,11]. Similarly, our both the patient presented in critical condition and tumor excision was done on emergency basis. Cerebellopontine Angle (CPA) is an area of neurological tissue. Tumor in this area might present with varied signs and symptoms. Vestibular schwannoma seldom presents during pregnancy, however the symptoms of the same can be exaggerated or worsened during the pregnancy [12] due to exaggerated growth and edema of the surrounding tissue lead to compression of the cerebellum and brain stem. MRI imaging is the gold standard for vestibular schwannomas [13]. The recent treatment options include microsurgical excision, stereotactic radiosurgery or fractionated radiotherapy [14]. However, large tumors with compression effect on these vital areas are difficult to manage, needs urgent intervention and complete excision.

In 1917, accelerated growth of vestibular schwannomas during pregnancy was first reported by Harvey Cushing [15]. The correlation of increase in size of the tumor with the pregnancy led to research of presence of Estrogen Receptor (ER) on the tumor. It is found that expression of ER-α is increased in cases of sporadic vestibular schwannomas [14]. The reported preferred operative time of acoustic schwannomas in pregnancy is the second trimester [16]. Our patient operated on emergency basis in 3rd trimester (late reporting) subsequent to worsening of symptoms; on severely increase ICP was confirmed by USG guided ONSD measurement [17]. ONSD showed good correlation with directly measured ICP [18]. However, mean ONSD was 5.6 mm 1st case and 5.8 mm in 2nd case, in isolated eye, on compression side it was found > 6.2 mm on both the cases, indicating severely increased ICP.

Therefore, emergency craniotomy for acutely symptomatic Cerebello-Pontine (CP) angle tumor (mean optic nerve sheath diameter >5.5 mm) proved successful with uneventful recovery of the mother and safe delivery of the baby in third trimester of pregnancy. Excision of CP angle tumor during late pregnancy demands timely collaborative management inclusive of neurosurgeons, obstetricians and anaesthesiologist in order to ensure the safe delivery of baby and uneventful recovery of mother.

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