A Rare Case of Spontaneous Pneumatocele due to Intra Sinus Bony Space Occupying Lesion

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A 21-year-old, male Hindu patient presented with headache and weakness in the distal part of the right upper limb for 10 days. The patient described that the nature of the headache was dull aching holo-cranial and intermittent, which was relieved by rest and medication. No specific aggravating factors or specific aggravating position of the head were found. The patient also described that weakness was gradually progressing proximally and was only in the right hand. Weakness began insidiously and started from the most distal fingertips of the right hand and then moved proximally upward towards the wrist. Later it became difficult for the patient to hold a pen and do the writing, and subsequently it progressed to difficulty in holding and lifting objects with the right hand. No such findings were seen in the left hand. There was no history of any sensory symptoms like tingling and numbness. The patient did not give any history of trauma in the recent past. No history of any fluid leak from the ear on either side or nose in either nostril. Also, there was no history of neck rigidity or fever. A patient gave a history of continent bowel and bladder function. He did not have an addiction. There were no significant histories of any major surgery, major illness or comorbidity. No significant drug or allergy history.
On Examination
The patient was examined in full light with proper consent. He was fully conscious, oriented to time, place and person, followed proper commands and had normal speech and comprehension. Memories of recent and past events were normal. Calculation and vision were found to be normal. Other cranial nerves were also found to be normal. On sensory examination, no focal neurological deficit was found at any dermatomal level. On the motor examination, there was a right-sided distal grip weakness in the upper limb having the power of 4/5 (according to MRC classification) in the fingers and wrist. Motor activities in rest of the body parts were normal. All superficial reflexes, as well as deep tendon reflexes, were found to be normal. The patient’s bowel and bladder activities were normal.

Imaging

The MRI is suggestive of an intense homogenous hypodense area both in T1W and T2W sequence in the left frontal region. This finding is suggestive of an air pocket inside the cranial cavity which is suggestive of left frontal pneumocele, which actually from the history given by the patient was suggestive of spontaneous onset. As the patient did not give any history of trauma or CSF leak, CT cisternography was suggested to find the cause of air in the cranial cavity. The findings of CT cisternography are as follows:

As shown above, CT cisternography did not show any obvious leak of CSF from any sinus or any discontinuity in bone i.e. fracture. But surprisingly it showed a bony lesion which was homogeneously enhancing, inside the left frontal air sinus which was solid and appeared to be a part of the inner table of the frontal air sinus.

Plan of Management
After evaluating all images and all routine blood investigations, it was thought to be due to this lesion which had produced a large pneumocele, leading to compression over the motor area and causing weakness of the opposite limb. So the patient was posted for exploration with left perional craniotomy and excision of intra sinus sol with exteriorization of left frontal sinus followed by repair of anterior cranial fossa with a pedicled pericranial graft which was augmented with fibrin glue.

Intraoperative Picture
Post-Operative Course
The postoperative course was uneventful and the patient was kept on routine antibiotics and anticonvulsants. Intraoperative subgaleal drain was put in to remove excessive collection which was removed on postoperative day 2 and post-operative NCCT brain with bony cuts at sinus level done. These cuts were suggestive as follows:

Post-operative Images

There was almost complete resolution of left frontal pneumatocele with complete removal of intra sinus bony SOL. The postoperative course of the patient was uneventful and the patient was discharged on postoperative day 5. Follow up was done for 3 months at regular intervals. There were no fresh complaints from the patient side and no complications were seen.

PATHOLOGICAL MANAGEMENT

On histopathological examination, the lesion turned out to be intra sinus osteoid osteoma that was arising from the posterior wall of the left frontal sinus.

Discussion
Generally, cerebral pneumatocele is due to a breach in either base of the skull or air sinus post any trauma and it usually presents as a CSF leak either from the ear (if the fracture is in the middle cranial fossa) or from the nasal cavity (if fracture is in anterior cranial fossa).

Various causes include:
1. Congenital: - Skull base defect, tegmen tympani defect
2. Traumatic: - The most common cause
3. Infectious: - Meningitis, chronic otitis media etc
4. Neoplastic: - Dermoid cyst rupture, tumour eroding skull base, skull base osteoma (very rare)
5. Iatrogenic: - Transcranial surgeries, endonasal surgeries, following lumbar puncture, ventriculostomy.

There are thought to be 2 mechanisms of development of pneumocephalus:
1. Ball valve theory of Dandy: - Unidirectional movement of air from the outside environment into the cranial cavity.
2. Inverted Soda bottle effect of Horowitz and Lunsford: - Excessive loss of CSF due to drainage in a physiological way during Valsalva or via lumber drain leads to low intracranial pressure and trapping of air in the vacuum created inside the cranium.

The presence of air is a source of infection and it can lead to meningitis.

Evaluation
There are various methods to evaluate which include the following things:-
- Skull X-ray: - Nowadays having a limited role as they miss a small quantity of air.
- Non-Contrast CT scan of Brain: - Usually the mainstay of diagnostic tool, which can identify even a small amount of air and also shows the site of fracture in case of trauma. It can also show the compressive effect of pneumatocele over brain parenchyma and is helpful to decide the further treatment plan.
- MRI Brain: - Not as useful as NCCT brain as air can be mistaken as flow voids or blood products. However, it can diagnose evidence of infection very well like sinusitis, meningitis etc.
- Cisternography: - particularly useful in presence of CSF rhinorrhoea to find the defect.

Management Options
Most of the time pneumatocele is managed conservatively.

Conservative Treatment Includes
A. Bed rest
B. Placing the patient in 30 degrees fowler position
C. Avoid Valsalva maneuver like nose blowing, coughing and sneezing.
D. Analgesics and Antipyretics
E. High flow Oxygen Therapy (5L per minute for five days at least) via a face mask or 100 % non-rebreathing mask.
F. Hyperbaric oxygen therapy.

**Indications for Surgical Intervention**
1. Symptomatic pneumatocele
2. Tension pneumatocele
3. Persistent traumatic pneumocephalus lasting for more than one week
4. Tension pneumoventricle.

**Surgical Options**
I. Needle tapping through a previous craniotomy burr hole.
II. Fresh burr hole and aspiration with insertion of the subdural drain.
III. Conventional craniotomy and repair of the dural defect.

**DIFFERENTIAL DIAGNOSIS**

Intracranial fat can sometimes be mistaken for air on a plain CT scan, and in MRI, pneumocephalus can be mistaken for blood products or flow voids.

**Complications**

Patients with pneumocephalus can develop the following complications:
1. Meningitis.
2. Seizures due to cortical irritation.
3. Brain abscess is more common in otogenic pneumocephalus.

4. Brain herniation syndromes may be due to raised intracranial pressure due to pneumocephalus.

Here, our patient presented with weakness of the right side distal upper limb which was due to the spontaneous onset of pneumatocele. This pneumatocele is thought to have occurred because of the presence of bony SOL in the frontal sinus. This caused a microscopic gap in the bony continuity. So it caused air entry during inspiration inside the cranial cavity and acted as a partial ball valve for air entry so complete removal from the cranial cavity was not possible which lead to pneumatocele formation.

**CONCLUSIONS**

From the above-mentioned case, it can be said that though pneumatocele is most commonly seen following trauma, it can also be a sign of such rare presentation of intra sinus bony SOL and this requires expertise for treatment of such condition.

**REFERENCES**

[1] https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4583710/