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## Fibrous Dysplasia of the Temporal Bone Presenting with Chronic Suppurative Otitis Media

**Otorrhea** can result from middle ear discharge with tympanic membrane perforation, external ear canal pathology, skull injury and other serious diseases in trauma and immunocompromised patients.<sup>1</sup> Thorough investigation is warranted in cases without improvement after treatment. We present a case where fibrous dysplasia was the underlying cause and discuss its treatment.

### CASE REPORT

A 41-year-old farmer man consulted at our ENT Outpatient Clinic with a 1-year history of clear, non-foul-smelling discharge from his left ear, associated with intermittent pounding ear pain. A hard immobile nontender prominence was noted over the patient's left temporal area. (*Figure 1*) He was initially treated for chronic suppurative otitis media for 3 months. Severe ear pain was noted, 10/10 on pain scale, squeezing in character, radiating to the left temporal and parietal area associated with progressive hearing loss and ear fullness. Plain CT scans revealed a combined lytic and ground glass expansile lesion involving the left occipital, mastoid, and petrous temporal bone and portion of the parietal bone obliterating the external auditory canal and middle ear. (*Figure 2*)

On physical examination, a hard immobile nontender prominence was noted over the patient's left temporo-parietal area. No facial asymmetry or numbness was noted. On otoscopy, there was a skin-colored round smooth hard immobile nontender mass in the external auditory canal with non-foul smelling whitish to yellowish purulent discharge. A Weber tuning fork test lateralized to the left and a Rinne test revealed air conduction < bone conduction on the left, and air conduction > bone conduction on the right.

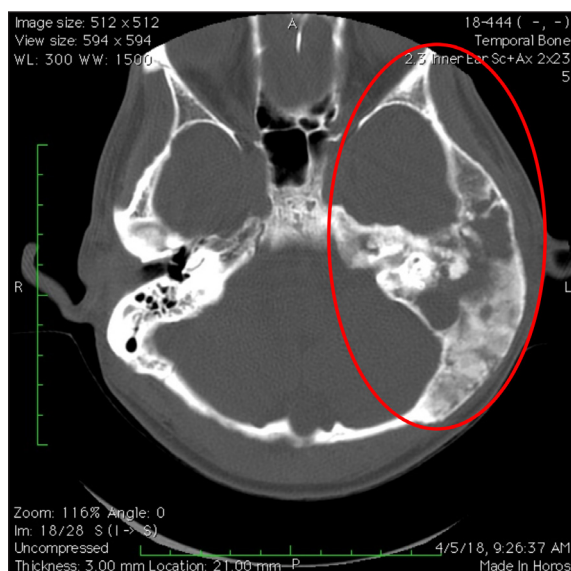
The patient was admitted with an impression of chronic suppurative otitis media, left; to consider fibrous dysplasia vs. tuberculosis. Aerobic culture of ear discharge revealed no growth. Canal down mastoidectomy was done. Intraoperative findings revealed granulomatous and cartilaginous lesions on the mastoid and temporal bone. (*Figure 3*) There were aural polyps and cholesteatoma with the ossicles apparently eroded by the lesion. (*Figure 4*) Temporal bone fluid was sent for GeneXpert MTB/RIF while the surgical specimen was sent for histopathology. GeneXpert MTB/RIF revealed negative results for tuberculosis. Final histopathology results showed fibrous dysplasia.

**Keywords:** *fibrous dysplasia; chronic suppurative otitis media; otorrhea*

The patient was discharged home with no complications. Our final diagnosis was chronic suppurative otitis media, left; secondary to fibrous dysplasia, left temporal, occipital and parietal bone. He followed up at the OPD with no complaints of ear pain, headache, no facial asymmetry, with minimal non foul smelling ear discharge, and prominence on the left was still observed on the left temporo-parietal area. (Figure 5)



**Figure 1.** Bony prominence over the left postauricular area. Note the protruding left ear.

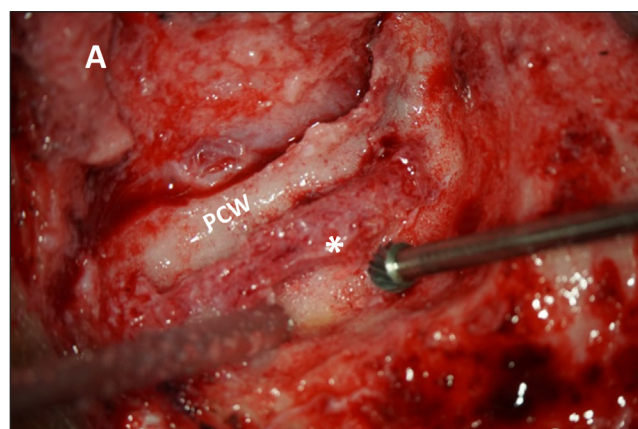


**Figure 2.** Axial CT Scan showing combined lytic and ground glass expansile lesion (encircled) involving the left occipital bone, mastoid and petrous temporal bone and portion of the parietal bone.

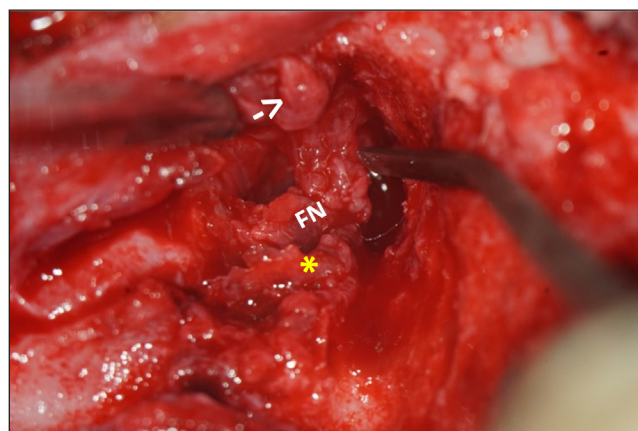
The patient was again seen at the OPD two weeks post operation where ear discharge was not observed. There were no complaints of ear fullness, tinnitus or dizziness.

## DISCUSSION

Otorrhea can result from different disease entities. A characteristic ear discharge is helpful in making a diagnosis. Clear discharge could signify cerebrospinal fluid leak, or water from recent swimming or bathing; bloody discharge could mean trauma to canal, barotrauma or skull fracture; mucoid discharge more from the middle ear due to absence of mucous glands around the ear canal; and purulent discharge could signify acute or chronic otitis media, auditory canal infection or perforated tympanic membrane.<sup>1,2,3</sup> Chronic otitis media is defined as an inflammation in the middle ear resulting in permanent or long-term changes of the tympanic membrane.<sup>4</sup> Changes of the TM include atelectasis, retraction, perforation, tympanosclerosis and



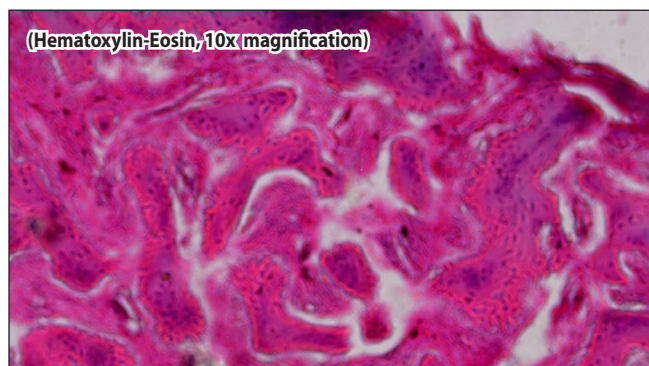
**Figure 3.** Intraoperative views showing (asterisk) granulomatous and cartilaginous lesions in mastoid. PCW, posterior canal wall; A. Anterior



**Figure 4.** Intraoperative view showing aural polyps (arrow) and cholesteatoma (asterisk). The facial nerve is labeled (FN). No ossicles were appreciated.



**Figure 5.** Two weeks post operative photo. Prominence on the left was still observed over the temporo-parietal area; note left auricle protruding laterally (dashed oval).



**Figure 6.** Histopathologic slide, H&E stain, low power view, 10X magnification. Note low to moderate cellular fibrous stroma surrounding irregular, curvilinear trabeculae of woven bone, exhibiting "Chinese alphabet" pattern.

cholesteatoma which could present as granulation tissue on otoscopy.<sup>4,5</sup> In this case, the patient initially presented with left purulent otorrhea for 1 year duration with a noted external auditory mass on otoscopy leading to the primary impression of chronic suppurative otitis media (CSOM).

Our patient also presented with progressive conductive hearing loss, and bony prominence on the temporal area and ear pain radiating to the left temporal, parietal and occipital area. Bony prominence and a CT scan result of ground glass appearance led us to a diagnosis of fibrous dysplasia. Fibrous dysplasia is a genetically based developmental anomaly of the bone-forming mesenchyme

with a defect in osteoblastic differentiation and maturation that leads to replacement of normal bony tissue by fibrous tissue of variable cellularity and immature woven bone.<sup>6</sup> It represents about 2 - 3% of benign bone lesion and usually occurs throughout the skeleton. The craniomaxillofacial bones commonly involved were the ethmoids (71%), the sphenoid (43%), frontal (33%), maxilla (29%), temporal (24%), parietal (14%) and occipital bone (5%).<sup>7</sup> It has a male predilection with a male: female ratio of 2:1.<sup>7</sup> Common signs and symptoms of fibrous dysplasia of the temporal bone include stenosis of external auditory canal, progressive conductive hearing loss, bulging of temporal area, unilateral otorrhea, and otalgia<sup>8</sup> which were manifested by the patient. Fibrous dysplasia of the temporal bone can also lead to complications in the middle and inner ear resulting in chronic otitis media, facial nerve palsy or sensorineural hearing loss.<sup>9,10</sup> Cholesteatoma can be a complication which has been observed in 40% of patients<sup>11</sup> and was also present in our case intraoperatively. CT scans would show bony expansion with either heterogeneous "ground-glass" appearance which was consistent with our case.<sup>12</sup>

Due to chronic ear discharge not responding to antibiotics, tuberculous otitis media (TOM) was considered. There is a high prevalence of tuberculosis in our country, 500/100, 000.<sup>13</sup> TOM should be considered in patients with known or suspected active pulmonary tuberculosis and a chronic ear infection; however, lack of pulmonary tuberculosis does not exclude the possibility of TOM.<sup>14</sup> Many of these cases would remain asymptomatic and would require further evaluation and diagnostics hence our request of AFB and GeneXpert which revealed negative results. TOM was also considered due to ossicular destruction, opacification of the middle ear and mastoid.<sup>15</sup>

Fibrous dysplasia usually requires histopathologic confirmation for diagnosis. Histopathology reveals low to moderate cellular fibrous stroma surrounding irregular, curvilinear trabeculae of woven bone, with a "Chinese alphabet" pattern as shown in the patient's histopathologic report. (Figure 6) The stroma may be collagenized, and the ratio of fibrous tissue to bone can appear as being totally fibrous to being densely packed with dysplastic trabeculae.<sup>16</sup>

The management of fibrous dysplasia of the temporal bone is directed towards symptomatic relief of bone pain, removal of ear canal obstruction, correction of deformities, and treatment of infection.<sup>9,10</sup> In the case presented, these were addressed through modified mastoidectomy. We opted for a more conservative approach to preserve structures such as the facial nerve, semicircular canal and the cochlea. The patient did not complain of ear pain, there was no obstruction of the ear canal noted, and no ear discharge observed after the second week of follow up. However, we should be ready for recurrence of symptoms with disease progression.





Bone pain can be managed with bisphosphonate (intravenous pamidronate), with remarkable results in improvement of bone pain and healing of bone lesions demonstrated by Plotkin *et al.*<sup>17</sup> Bisphosphonate inhibits osteoclastic activity and has been used with success in patients with extensive lesions associated with significant disfigurement and pain.<sup>18</sup> However, bisphosphonate was not readily available in our area.

We should monitor our patient closely for recurrence which was observed by Linder *et al.* in 35% of patients with a primary lesion who underwent surgery.<sup>19</sup> Although recurrence was observed more in patients under the age of 20,<sup>19</sup> (our patient was 41 years old), our conservative surgery may predispose more to recurrence. Valentini *et al.* reported that there was no recurrence in patients with fibrous

dysplasia of the mandible where radical dissection was done, but was observed in patients who received conservative treatment.<sup>20</sup> In our case, our patient was advised frequent follow ups because conservative surgery was performed, but he was not able to comply.

Our experience in this case taught us that further investigation of chronic ear discharge is very essential. Ancillary tests like CT scans were helpful in making our diagnosis and planning treatment for the case. Mastoidectomy helped in abrupt relief of the symptoms presented by the patient. It is also important to emphasize the need for follow up due to possible recurrence of the disease, especially in cases where conservative treatment was done. The patient's life-situation and ability to consult and avail of further treatment must also be considered.

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