

# Secondary Adrenal Insufficiency Presenting as Petrified Ear Accompanied by Patulous Eustachian Tube: A Novel Combination

Ji Hyun Bae<sup>1</sup> , Dongwon Yi<sup>1</sup> , Jung Hee Lee<sup>2</sup> , and Hyun Min Lee<sup>3</sup> 

<sup>1</sup>Division of Endocrinology and Metabolism, Department of Internal Medicine, Pusan National University Yangsan Hospital, Pusan National University School of Medicine, Yangsan; and  
<sup>2</sup>Departments of Pathology and <sup>3</sup>Otorhinolaryngology-Head and Neck Surgery, Pusan National University Yangsan Hospital, Pusan National University School of Medicine, Yangsan, Korea

## 개방성 이관을 동반한 골화 귀로 나타난 이차성 부신기능부전: 새로운 조합

배지현<sup>1</sup> · 이동원<sup>1</sup> · 이정희<sup>2</sup> · 이현민<sup>3</sup>

<sup>1</sup>부산대학교 의과대학 양산부산대학교병원 내과학교실, 내분비 분과,  
부산대학교 의과대학 양산부산대학교병원 <sup>2</sup>병리학교실, <sup>3</sup>이비인후과학교실

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**Address for correspondence**  
Hyun Min Lee, MD, PhD  
Department of Otorhinolaryngology-  
Head and Neck Surgery,  
Pusan National University  
School of Medicine, Pusan National  
University Yangsan Hospital,  
20 Geumo-ro, Mulgeum-eup,  
Yangsan 50612, Korea  
**Tel** +82-55-360-2132  
**Fax** +82-55-360-2162  
**E-mail** enthmllee@gmail.com

Petrified ear, characterized by auricular cartilage ossification, is a rare condition often associated with underlying endocrinopathies. We report a unique case of a 31-year-old male presenting with bilateral petrified ear accompanied by patulous Eustachian tube (PET), both manifestations of previously undiagnosed secondary adrenal insufficiency. The patient presented with progressive auricular rigidity, hearing impairment, and symptoms of PET including autophony. Endocrinological evaluation revealed central adrenal insufficiency, and the temporal bone CT demonstrated ossification of both auricular and Eustachian tube cartilage. Following the initiation of steroid replacement therapy, endocrinological parameters improved, and unexpectedly, the patient's hearing thresholds also showed improvement at one-year follow-up. This case highlights the importance of comprehensive endocrinological evaluation in patients with petrified ear and represents the first reported association between petrified ear and PET, suggesting the need for vigilant otologic assessment in such cases.

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## Introduction

Petrified ear, a rare condition characterized by auricular cartilage ossification, was first described by Bochdalek<sup>1)</sup> in 1866. Most cases are discovered incidentally and are either asymptomatic or present with mild symptoms such as otalgia or conductive hearing loss.<sup>2)</sup> Although uncommon, radio-

logic evaluation of 200 otologic patients demonstrated auricular calcification in approximately 20% of cases.<sup>3)</sup>

Recent comprehensive review literature categorizes the etiology of petrified ear into three main groups<sup>4)</sup>: local causes such as trauma<sup>5)</sup> and cold injury<sup>6)</sup>; endocrine disorders including adrenal insufficiency, hypopituitarism, and hypothyroidism<sup>2,7-11)</sup>; and idiopathic cases.<sup>12,13)</sup> Diagnosis typically involves clinical examination, radiography, temporal bone CT (TBCT), and endocrinological evaluation, with tissue biopsy performed in select cases. Currently, there is no method to reverse the ossi-

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fication, and management focuses on treating underlying conditions. Surgical intervention is considered only in cases with severe symptoms or significant quality of life impairment.<sup>4)</sup>

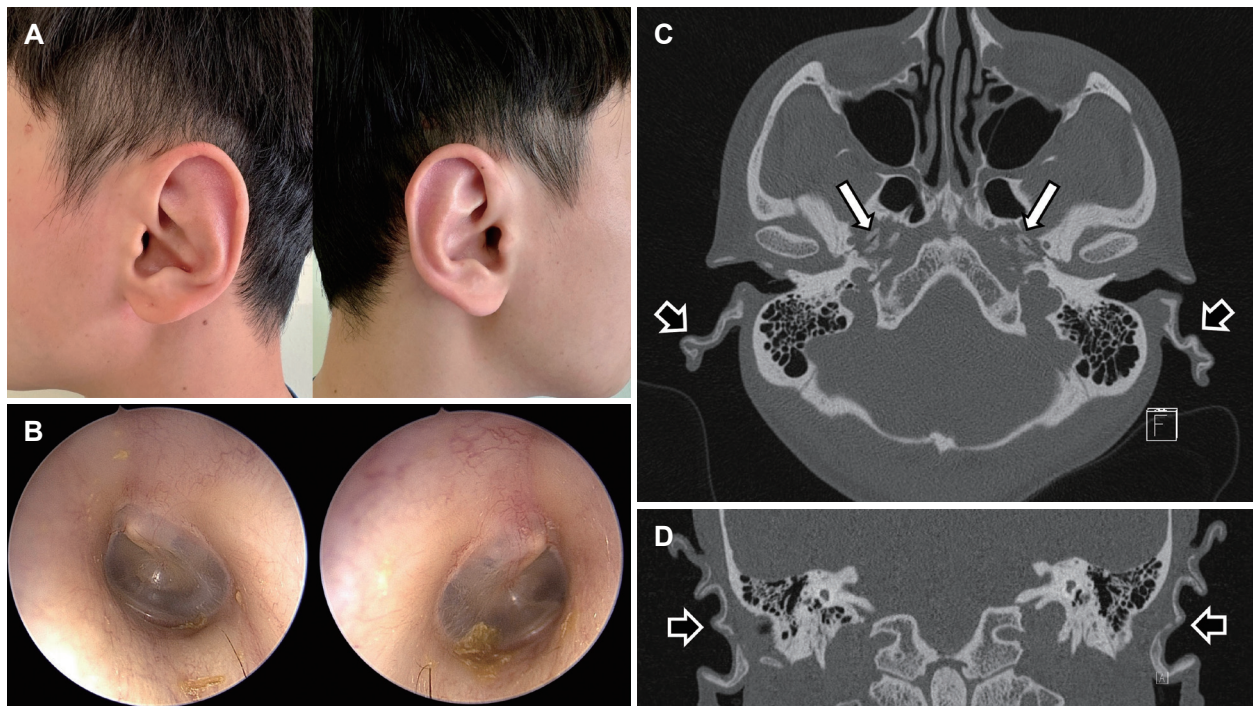
Patulous Eustachian tube (PET) is characterized by abnormal patency of the Eustachian tube, with patients typically presenting with autophony, aural fullness, and respiratory synchronous tinnitus. While PET can be associated with various conditions including weight loss, pregnancy, and neuromuscular disorders, the exact cause often remains unclear. The estimated prevalence ranges from 0.3% to 7.0%, with a female predominance. Management options range from conservative measures to surgical interventions, including tympanic membrane manipulation (patch apply), middle ear ventilation tube insertion, or transtympanic catheter insertion for refractory cases.<sup>14)</sup>

This report describes a case of progressive petrified ear where endocrinological evaluation revealed underlying adrenal insufficiency, accompanied by bilateral PET confirmed through physical examination and TBCT. This unique presentation emphasizes the importance of multidisciplinary evaluation, including endocrinological assessment and consideration of potential PET, in patients presenting with petrified ear.

## Case

A 31-year-old male presented with progressive bilateral auricular rigidity and hearing impairment that developed over four years. He also reported bilateral clicking sounds during swallowing and autophony. The patient had no history of auricular cold injury, trauma, otitis media, external otitis, or prolonged calcium supplementation. He had no chronic conditions such as hypertension, diabetes mellitus, or hepatitis. Five years prior, he was diagnosed with hypothyroidism due to fatigue and lethargy, for which he had discontinued thyroid hormone replacement therapy. Following military service in his early twenties, he had been socially isolated due to panic disorder and depression.

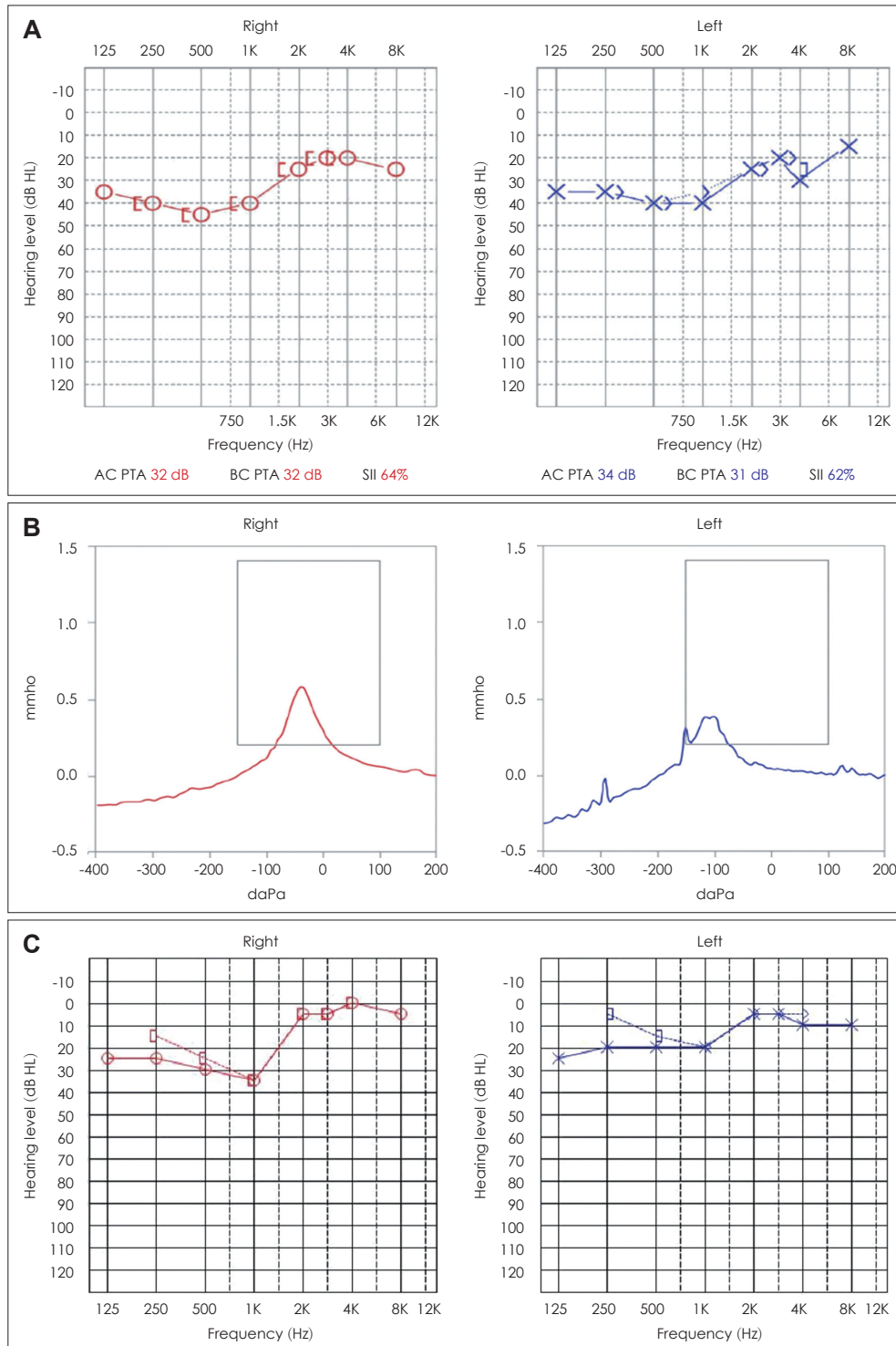
Physical examination revealed symmetrical external ears without morphological abnormalities. Palpation demonstrated complete rigidity of the auricular cartilage without elasticity (Fig. 1A), while the ear lobules maintained normal flexibility. The nasal and costal cartilages exhibited normal elasticity. Otoscopic examination showed normal tympanic membranes (Fig. 1B); however, the left tympanic membrane demonstrated respiratory movement, and both membranes showed significant inflation during the Valsalva maneuver. Pure tone au-



**Fig. 1.** Clinical and temporal bone CT findings of the petrified auricle. A: External appearance of the patient's ear demonstrates normal morphology without visible deformity, although palpation revealed complete rigidity with absence of elasticity. B: Bilateral tympanic membranes exhibit normal appearance on otoscopic examination. C: Transverse view demonstrates bilateral ossification of the auricular cartilage (black arrows) and partially ossified Eustachian tube cartilage (white arrows). D: Coronal view also confirms bilateral ossification of the auricular cartilage (black arrows).

diometry revealed mild low-frequency hearing loss bilaterally, with four-frequency (0.5-1-2-4 kHz) averages of 32 dB and 33 dB in the right and left ears, respectively (Fig. 2A). Tym-

panometry showed type A patterns bilaterally, with a characteristic saw-tooth pattern of PET in the left ear (Fig. 2B). TBCT demonstrated bilateral auricular ossification and par-



**Fig. 2.** Pre- and post-treatment audiologic evaluation results. A: Initial pure tone audiometry (PTA) demonstrates mild low-frequency hearing loss bilaterally with four-tone averages of 32 dB and 33 dB in the right and left ears, respectively. B: Tympanometry reveals type A patterns bilaterally with a characteristic saw-tooth pattern of patulous Eustachian tube in the left ear. C: Follow-up audiometry at one year post-treatment shows improved hearing thresholds with four-tone averages of 17 dB and 13 dB in the right and left ears, respectively.

tially ossified Eustachian tube cartilage, with normal external auditory canals, middle ear cavities, and mastoid air cells (Fig. 1C and D).

Given the suspected endocrine disorder, endocrinological evaluation was performed. Laboratory findings revealed markedly decreased serum cortisol (0.2 µg/dL; normal range: 4.46–22.7), with non-elevated adrenocorticotrophic hormone (ACTH) (5.1 pg/mL; normal range: 0–46), normal triiodothyronine (1.03 ng/mL; normal range: 0.64–1.52), decreased free thyroxine (0.57 ng/dL; normal range: 0.70–1.48), elevated thyroid-stimulating hormone (36.01 µIU/mL; normal range: 0.35–4.94), and elevated prolactin (144.7 ng/mL; normal range: 3.7–17.9). Brain MRI revealed slight sellar widening with flattened pituitary gland. The patient was diagnosed with idiopathic central (secondary) adrenal insufficiency and primary hypothyroidism. He started on hydrocortisone replacement (10 mg morning, 5 mg evening) and subsequent thyroid hormone replacement therapy.

We performed a biopsy to determine whether the ossification lesion was due to frostbite, chronic traumatic change, or chondritis. Additionally, histological examination was necessary to differentiate between true ossification and calcification processes in the cartilaginous tissue. Histopathological examination of the left conchal cartilage biopsy revealed extensive ossification with residual cartilage tissue only at the peripheral margin (Fig. 3). The patient was diagnosed with bilateral petrified ear accompanied by PET, which developed from ossification of the Eustachian tube cartilage, all manifestations of underlying idiopathic central adrenal insufficiency.

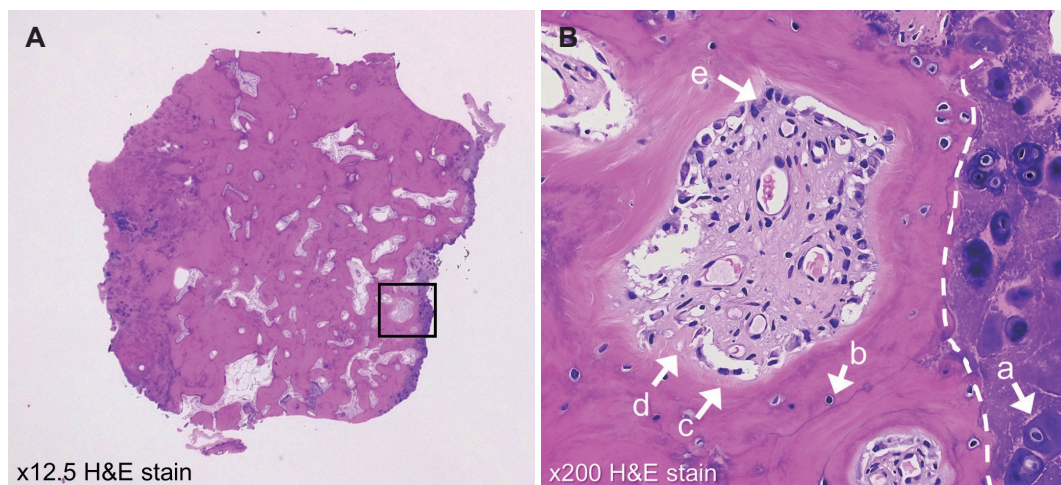
At one-year follow-up, endocrinological parameters showed improvement. While auricular rigidity persisted, audiometric thresholds improved to 17 dB and 13 dB in the right and left ears, respectively (Fig. 2C). Given the mild nature of his PET symptoms, conservative management was chosen. The patient continues endocrinological follow-up every four months with ongoing steroid and thyroid hormone replacement therapy, and annual otolaryngological monitoring of external ear, middle ear, and hearing status.

## Discussion

This report describes a young male patient initially presenting with bilateral petrified ear, which led to the diagnosis of secondary adrenal insufficiency through endocrinological evaluation. Additionally, otologic examination and TBCT revealed previously unrecognized PET symptoms associated with ossified Eustachian tube cartilage.

Table 1 provides a comprehensive overview of petrified ear cases referenced in this manuscript, highlighting their diverse clinical manifestations and management approaches.<sup>2,5-13</sup> Endocrinopathies frequently accompany petrified ear, with one comprehensive review reporting endocrine abnormalities in 50 of 75 cases.<sup>4</sup> Various case reports have documented associations with primary or secondary adrenal insufficiency,<sup>2,7,8</sup> idiopathic adult-onset pituitary insufficiency,<sup>9</sup> primary hypothyroidism with adrenal insufficiency,<sup>10</sup> and isolated ACTH deficiency.<sup>11</sup>

Secondary adrenal insufficiency, characterized by inade-



**Fig. 3.** Histopathological findings of the left conchal cartilage biopsy. A: Low-power view (Hematoxylin and eosin [H&E] stain,  $\times 12.5$ ) of the whole decalcified tissue. B: High-power view (H&E stain,  $\times 200$ ) of the black-colored box area from image A demonstrates ossification of cartilage with only peripheral margins containing residual cartilage tissue (right to the dashed line). The transitional zone shows characteristic features including chondrocytes (a) within residual cartilage matrix, osteocytes (b) embedded in mineralized bone matrix, distinct cement lines (c) at the interface, newly formed osteoid tissue (d), and osteoblasts (e) along the mineralization front.

**Table 1.** Clinical features and management of petrified ear cases cited in this manuscript<sup>2,5-13)</sup>

| Study, year                                 | Patient | Side      | Petrified ear feature                                                                                                                                                                                                                                                                                                                             | Management                                                                                                                                                                     |
|---------------------------------------------|---------|-----------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Uthoff and Wiesli, <sup>2)</sup> 2011       | M/60    | Bilateral | <ul style="list-style-type: none"> <li>Stiffness in both ears discovered during emergency department visit for profound hypotension after minor hand laceration</li> <li>X-ray, CT: calcification</li> <li>Secondary adrenal insufficiency (autoimmune hypophysitis)</li> </ul>                                                                   | No specific treatment for petrified ear. Long-term glucocorticoid-replacement therapy which resulted in marked improvement of fatigue.                                         |
| Britton, et al., <sup>5)</sup> 2009         | M/28    | Right     | <ul style="list-style-type: none"> <li>Chronic swelling, tenderness, and firmness of his right ear</li> <li>Physical examination</li> <li>Chronic use of a bluetooth headset device for 5 months</li> </ul>                                                                                                                                       | No specific treatment for petrified ear.                                                                                                                                       |
| Thomas, et al., <sup>6)</sup> 2019          | M/52    | Bilateral | <ul style="list-style-type: none"> <li>12-month history of pain affecting the helix of his right ear</li> <li>X-ray: petrification, fracture at the lower pole of the right helix</li> <li>Frostbite affecting the ear 20 years earlier</li> </ul>                                                                                                | No specific treatment for petrified ear.                                                                                                                                       |
| James and Wermers, <sup>7)</sup> 2016       | M/63    | Bilateral | <ul style="list-style-type: none"> <li>Gradually progressive hardening of outer ears without pain or deformity</li> <li>Physical examination, X-ray</li> <li>Addison's disease (diagnosed at age 12 years)</li> </ul>                                                                                                                             | No specific treatment for petrified ear. Physiologic doses of hydrocortisone and fludrocortisone.                                                                              |
| Goswami, et al., <sup>8)</sup> 2016         | M/30    | Bilateral | <ul style="list-style-type: none"> <li>Marked rigidity of both ears without signs of inflammation or scarring, discovered during evaluation of generalized weakness</li> <li>X-ray, CT: calcification</li> <li>Secondary adrenal insufficiency with secondary hypothyroidism (possibly due to previous bacterial meningitis at age 16)</li> </ul> | Treated with prednisolone 5 mg in morning along with l-thyroxine. Patient recovered from symptoms, but ears remained unchanged.                                                |
| Gogate, et al., <sup>9)</sup> 2012          | M/53    | Bilateral | <ul style="list-style-type: none"> <li>Generally stiff ears discovered during evaluation of chronic diarrhea, general weakness, cold intolerance</li> <li>CT: calcification</li> <li>Panhypopituitarism possibly due to adult-onset pituitary insufficiency</li> </ul>                                                                            | Treated with intravenous hydrocortisone, l-thyroxine and testosterone replacement. Patient showed rapid recovery, but ears remained unchanged.                                 |
| Recalcatti and Fantini, <sup>10)</sup> 2021 | M/71    | Bilateral | <ul style="list-style-type: none"> <li>Progressive stiffening of both ears for 20 years with pain exacerbated in bed at night, hearing loss</li> <li>X-ray, CT: calcification</li> <li>Primary hypothyroidism and primary adrenal insufficiency for 18 years</li> </ul>                                                                           | Treated with hydrocortisone and l-thyroxine. Patient also underwent surgical excision and reconstruction with pre-auricular flap for squamous cell carcinoma on right auricle. |
| Taguchi, et al., <sup>11)</sup> 2017        | M/22    | Bilateral | <ul style="list-style-type: none"> <li>Inflexible stiffness in both ears discovered during evaluation for nausea and low sodium</li> <li>X-ray, CT: calcification</li> <li>Isolated ACTH deficiency with other pituitary hormones normal</li> </ul>                                                                                               | Hydrocortisone supplementation for 6 years led to improvement of all symptoms except ear stiffness.                                                                            |
| Shah, et al., <sup>12)</sup> 2019           | M/75    | Right     | <ul style="list-style-type: none"> <li>Clinical presentation: 6-month history of hardening and protrusion of right ear, thickened and hard pinna with mild overlying erythema and ear lobe sparing</li> <li>Image scans: X-ray</li> <li>Etiology: Idiopathic</li> </ul>                                                                           | No specific treatment due to mild symptoms.                                                                                                                                    |
| Weiss, et al., <sup>13)</sup> 2017          | M/69    | Bilateral | <ul style="list-style-type: none"> <li>Clinical presentation: 1-month history of non-painful rigidity of right ear progressing to involve left ear, mild discomfort while sleeping</li> <li>X-ray</li> <li>Idiopathic</li> </ul>                                                                                                                  | Orthotic pillow for sleeping comfort, no specific treatment due to mild symptoms.                                                                                              |

Cases are arranged according to their citation order in the manuscript. M, male; CT, computed tomography; ACTH, adrenocorticotropic hormone

quate cortisol production due to ACTH deficiency and consequent adrenal atrophy, presents distinct features from primary adrenal insufficiency. While common etiologies include exogenous glucocorticoid therapy, pituitary tumors, cranio-pharyngiomas, pituitary surgery, head trauma, and radiation therapy, patients typically present with generalized weakness, fatigue, anorexia, weight loss, and gastrointestinal symptoms. Notably, unlike primary adrenal insufficiency, these patients do not develop hyperpigmentation or significant electrolyte disturbances.<sup>15)</sup>

Cartilaginous calcification and ossification are distinguished by their microscopic tissue findings. Pathologic calcification refers to the deposition of amorphous calcium within the cartilage matrix, where the cartilaginous tissue remains intact but becomes hardened. In contrast, ectopic ossification represents a more advanced process where cartilage is actually replaced by bone tissue. Calcification can be induced by endocrine disorders. In cortisol-deficient states, hypercalcemia may develop through various mechanisms. Indeed, clinically significant hypercalcemia occurs in approximately 6%–8% of patients with adrenal insufficiency, including Addison's disease, with proposed causes including increased bone resorption, enhanced renal calcium reabsorption, and hemoconcentration due to hypovolemia.<sup>16)</sup> However, hypercalcemia in adrenal insufficiency patients is often transient rather than persistent, typically occurring during dehydration or acute adrenal crisis, leading to the hypothesis that cumulative episodes of transient hypercalcemia may result in calcium deposition in cartilage.<sup>4)</sup> Ultimately, we can postulate that hormonal imbalance affects cartilage initially by altering chondrocyte metabolism and matrix composition to create an environment conducive to calcium deposition, and potentially progressing to ectopic bone formation if osteoblast recruitment and differentiation are stimulated.<sup>4)</sup>

To our knowledge, this represents the first reported case of petrified ear accompanied by PET. This report describes not only the association between petrified ear and secondary adrenal insufficiency but also demonstrates a direct pathophysiological connection between adrenal insufficiency and PET through cartilage ossification. Notably, TBCT of patient revealed ossification of both auricular and Eustachian tube cartilage, providing visible evidence of this connection. CT image showed persistently patent Eustachian tubes even in the resting state, suggesting that cartilage ossification had structurally altered the Eustachian tube's normal closing mechanism. This finding strongly suggests that the same underlying

endocrine pathology affecting auricular cartilage also impacts Eustachian tube cartilage, leading to PET symptoms. PET typically develops due to decreased peritubal fat or altered mucosal lubrication. Primary and secondary adrenal insufficiency, which cause chronic dehydration and weight loss, can result in peritubal fat reduction leading to persistent Eustachian tube patency. Additionally, hormonal imbalances in endocrine disorders including hyperthyroidism, diabetes, and pregnancy can affect mucosal surface tension and alter the pressure dynamics required for normal Eustachian tube function.<sup>17)</sup> Despite steroid replacement therapy, the ossification itself did not resolve in our case, consistent with previous reports indicating that ossification remains irreversible even with treatment. Consequently, the PET symptoms persisted due to the unchanged structural alteration of the ossified cartilage. Should PET symptoms exacerbate, surgical management options, including ventilation tube insertion or transtympanic catheter insertion, may be considered for symptomatic relief.

Since systematic studies directly comparing site-specific cartilage ossification patterns in endocrine disorders have not been reported, it is difficult to definitively explain why only auricular and Eustachian tube cartilage showed ossification in our case. However, CT-based data indicate that calcification frequency of external ear elastic cartilage is 19.5%,<sup>3)</sup> while Eustachian tube torus tubarius calcification occurs in only 0.6% of cases,<sup>18)</sup> suggesting biological and mechanical differences between these sites. Both cartilages are avascular elastic (fibro)cartilage that rely on diffusion from adjacent tissues for nutrient and oxygen supply. Endocrine abnormalities such as cortisol and thyroid hormone deficiency can lower the mineralization threshold of cartilage matrix through alterations in calcium-phosphate metabolism<sup>4)</sup> and increased alkaline phosphatase activity,<sup>19)</sup> while repetitive mechanical stress during mastication and swallowing may form calcification nuclei.<sup>20)</sup> We hypothesize that when endocrine-induced matrix susceptibility combines with local mechanical stress, the relatively vulnerable auricular and Eustachian tube cartilage are more likely to undergo calcification first.

Currently, no established treatment exists to reverse auricular ossification. While most asymptomatic cases are managed conservatively, surgical interventions such as wedge resection may be considered for severe symptomatic cases.<sup>4)</sup> Our patient requires ongoing monitoring to prevent further progression through avoidance of local trauma and cold exposure, while maintaining regular endocrinological follow-up

for steroid and thyroid hormone replacement therapy. Regular monitoring of auricular rigidity, tympanic membrane status, and PET symptoms is essential. Notably, the patient's initial mild hearing loss showed unexpected improvement during post-treatment follow-up. Although the mechanism of this audiometric improvement remains unclear, continued audiological monitoring is warranted.

Regarding the mechanism of low-frequency hearing loss and its recovery, we hypothesize that steroid therapy may have restored microcirculation in the cochlear apex and normalized aquaporin and ion channel expression in the endolymph, partially recovering the function of hair cells in the apical turn responsible for low-frequency hearing.<sup>21)</sup> However, this remains hypothetical and requires further validation. Previous reports have documented partial improvement in low-frequency hearing following levothyroxine supplementation in patient with hypothyroidism,<sup>22)</sup> which has been attributed to thyroid hormone normalizing outer hair cell electrophysiology and endolymphatic ion homeostasis. Therefore, the low-frequency hearing recovery observed in our case may result from the synergistic effects of steroid-induced stabilization of inner ear electrolyte metabolism and thyroid hormone-mediated electrophysiological normalization.

In conclusion, this case highlights two important clinical implications in the management of petrified ear. First, it emphasizes the critical importance of a comprehensive medical history and multidisciplinary approach, particularly regarding endocrinological evaluation, as the early detection of underlying conditions such as adrenal insufficiency can significantly impact patient outcomes. Second, this case demonstrates a novel pathophysiological connection between adrenal insufficiency and PET through cartilage ossification, which was confirmed by TBCT findings showing concurrent involvement of both auricular and Eustachian tube cartilage. These findings suggest that clinicians should actively screen for and evaluate symptoms of PET in patients with auricular ossification, as these symptoms may be subtle or underreported by patients. Furthermore, the co-occurrence of these conditions may affect the patient's quality of life and require additional therapeutic considerations.

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the content as needed and take full responsibility for the content of the publication.

### Author Contribution

Conceptualization: Dongwon Yi, Hyun Min Lee. Data curation: Ji Hyun Bae, Hyun Min Lee. Investigation: Hyun Min Lee. Methodology: Hyun Min Lee. Project administration: Hyun Min Lee. Resource: Dongwon Yi, Hyun Min Lee. Visualization: Ji Hyun Bae, Jung Hee Lee, Hyun Min Lee. Writing—original draft: Hyun Min Lee. Writing—review & editing: Dongwon Yi, Jung Hee Lee, Hyun Min Lee.

### ORCIDs

|              |                                                                                           |
|--------------|-------------------------------------------------------------------------------------------|
| Ji Hyun Bae  | <a href="https://orcid.org/0009-0000-8669-1356">https://orcid.org/0009-0000-8669-1356</a> |
| Dongwon Yi   | <a href="https://orcid.org/0000-0003-3574-036X">https://orcid.org/0000-0003-3574-036X</a> |
| Jung Hee Lee | <a href="https://orcid.org/0000-0003-3003-2217">https://orcid.org/0000-0003-3003-2217</a> |
| Hyun Min Lee | <a href="https://orcid.org/0000-0001-6306-5256">https://orcid.org/0000-0001-6306-5256</a> |

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