## Middle ear metastasis from dormant breast cancer as the initial sign of disseminated disease 20 years after quadrantectomy

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## Abstract

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*We describe an unusual case of breast cancer metastatic* to the middle ear in a 71-year-old woman. The metastasis was the initial sign of disseminated disease 20 years after the patient had undergone a quadrantectomy for her pri*mary disease.* Computed tomography (CT) demonstrated the presence of an intratympanic mass with a soft-tissue density that was suggestive of chronic inflammation. The patient underwent a canal-wall-down tympanoplasty. When a brownish mass was found around the ossicles, a mastoidectomy with posterior tympanotomy was carried out. However, exposure of the tumor was insufficient, and therefore the posterior wall of the ear canal had to be removed en bloc. Some tumor was left on the round window membrane so that we would not leave the patient with a total hearing loss. Our case highlights the limitations of CT and magnetic resonance imaging in differentiating inflammatory and neoplastic lesions.

#### Introduction

Cancer metastases to the temporal bone, including the middle ear, are relatively uncommon. Affected patients are often asymptomatic for a long time. When signs and

Corresponding author: Ilaria Franceschetti, MD, Institute of Pathology, S. Maria del Carmine Hospital, Piazzale S. Maria 6, 38068 Rovereto (TN), Italy. Email: ilariafran@yahoo.com symptoms of middle ear metastasis do manifest, they may be misinterpreted as otitis media or mastoiditis. We describe a new case.

## Case report

A 71-year-old woman presented for evaluation of bilateral hearing loss. Audiometry revealed that the hearing loss was sensorineural on the left side and mixed on the right side. The patient also complained of severe problems with her balance.

Twenty years earlier, the patient had undergone a quadrantectomy and an axillary lymphadenectomy after she had been diagnosed with infiltrating ductal carcinoma with metastases in five lymph nodes (pT2N2a). The neoplasm had been negative for estrogen receptors, but it was characterized by moderate positivity for progesterone receptors and a medium proliferation index.

Postoperatively, the patient had undergone 6 weeks of adjuvant local radiotherapy with an average dose of 4,800 cGy (range: 4,600 to 5,000) followed by polychemotherapy. At the end of this multimodal treatment regimen, she experienced an apparently complete regression of her disease as documented by clinical and radiologic follow-up right up to the time of her presentation to us.

We obtained computed tomography (CT), which demonstrated the presence of an intratympanic mass with a soft-tissue density that was suggestive of chronic inflammation (figure 1). The mass had involved a thickened and calcific tympanic membrane, and the membrane was displaced toward the promontory. CT also showed infiltration of the ossicular chain, particularly the incus, and a diffuse increase in the density of the mastoid bone. Magnetic resonance imaging (MRI) showed hyperintense tissue in the right middle ear.

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The patient was taken to the operating room for a canal-wall-down tympanoplasty. The retroauricular area and the external auditory canal were infiltrated with a solution of 1:200,000 lidocaine and epinephrine. An incision was made along the retroauricular sulcus, and a periosteal flap was elevated. A vascular strip was prepared in the ear canal, and the middle ear was exposed.

A brownish mass was found around the ossicles. A mastoidectomy with posterior tympanotomy was carried out, but the exposure of the tumor was insufficient; therefore, the posterior wall of the ear canal had to be removed en bloc. The tumor was located primarily in the area of the promontory, but it did not extend to the hypotympanum, as CT had suggested.

The tumor was removed along with the ossicles and a section of the stapedial crura. A radical dissection was not pos-

sible, and a small piece of tumor was left on the round window membrane to avoid leaving the patient with a total hearing loss. The tumor had not come into contact with the facial nerve.

Histologic analysis of the excised specimen revealed the presence of ductal adenocarcinoma in the ossicle (figures 2 and 3). At 2 months postoperatively, follow-up revealed ongoing epithelialization. The patient subsequently experienced pulmonary metastases, and she died of the disease at 18 months of follow-up.

### Discussion

The most common malignant tumor in the middle ear is squamous cell carcinoma, which typically arises in the external ear canal and then extends from there into the tympanic cavity, the mastoid, or anteriorly. Reported metastases from primary tumors in other locations to the temporal bone, including the middle ear, are very unusual. When metastases do occur, they are most commonly found in the petrous portion (35% of cases), the internal auditory canal (17%), the mastoid (8%), and the external auditory canal (8%).<sup>1</sup> Facial nerve paresis is the most common symptom, followed by sudden and progressive hearing loss.<sup>1</sup> It has been suggested that temporal bone metastases are difficult to diagnose because symptoms and imaging findings mimic those of mastoiditis and chronic inflammation.<sup>2,3</sup>

Metastatic tumors involving the temporal bone usually



Figure 1. Axial (**A** and **B**) and multiplanar coronal (**C** and **D**) high-resolution CTs of the middle ear show the intratympanic soft-tissue density suggestive of chronic inflammation. Note the infiltration of the ossicular chain, particularly the incus, and the diffuse increase in the density of the mastoid bone.

spread from the breast (18% of cases), lung (12%), kidney (10%), stomach (8%), or larynx (5%).<sup>1,4-6</sup> The areas of the temporal bone that are most frequently involved are the petrous portion, particularly the osseous labyrinth; the middle ear is involved in only a minority of cases.<sup>4,7</sup>

Only a few reports have described in detail the precise site of metastatic involvement in the middle ear. Bates and Luscombe described a patient with a carcinoid tumor in the middle ear that arose 1 year after the patient had been diagnosed with a primary carcinoid of the ileum.<sup>8</sup> Merrick reported metastatic involvement of the left tympanic cavity in a patient who had undergone a hemicolectomy for an adenocarcinoma of the descending colon 6 years earlier.<sup>9</sup>

Porret et al reported a case of metastasis in the middle ear space and mastoid 1 year after a diagnosis of bladder cancer.<sup>10</sup> Wu et al described a case of metastatic hepatocellular carcinoma in the tympanic membrane 4 years after the initial diagnosis.<sup>11</sup> Yamaguchi et al reported a middle ear metastasis of a thymoma that had been diagnosed 6 years earlier.<sup>12</sup> Clamp and Jardine described a case of acute mastoiditis caused by metastatic lung cancer, and they reviewed the literature.<sup>3</sup>

The largest series was an autopsy study published in 2000 by Gloria-Cruz et al.<sup>2</sup> They examined the temporal bones of 212 patients with nonsystemic malignancies (excluding diseases such as lymphoma, leukemia, and multiple myeloma). Histologic examination of these

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Figure 2. Infiltration of islets of neoplastic cells is consistent with metastatic ductal carcinoma (H&E, original magnification ×50).

temporal bones demonstrated metastatic deposits in 47 patients (22.2%). Only 12 patients had an occult malignancy, and only 1 of them had experienced a metastasis to the temporal bone. Our patient presented with a metastasis 20 years after her initial diagnosis.

Breast cancer is known to metastasize to many organs.<sup>13,14</sup> Studies of patients with stage I or II breast cancer have indicated that only a few (<5%) exhibit signs of skeletal involvement at the time of diagnosis.<sup>15</sup> However, bone metastases have been reported in 28 to 75% of patients with stage III breast cancer<sup>8</sup> and in 50 to 80% of cases at autopsy.<sup>15,16</sup> Because metastases to the temporal bone are generally asymptomatic, most of those cases are not diagnosed until autopsy.<sup>2,17,18</sup>

Recurrence of breast cancer rarely occurs 20 years after the diagnosis of the primary, and the mortality rate for women 20 years past diagnosis of the primary is not significantly different from that of the rest of the



Figure 3. The neoplastic cells show positivity for estrogen receptors (original magnification  $\times 100$ ).

population.<sup>19</sup> The duration of breast cancer dormancy appears to be 20 to 25 years after the initial diagnosis.<sup>15-17</sup> Patients who survive that long without a recurrence or cancer in the contralateral breast are probably cured.<sup>19</sup>

To the best of our knowledge, our case represents the first published report of a middle ear metastasis from a dormant breast cancer in which the metastasis was the first sign of metastatic dissemination. Our case highlighted the limitations of CT and MRI in differentiating inflammatory and neoplastic lesions. Our first suspicion was a glomus tumor, although the location was unusual. Only histologic examination of the lesion can establish its malignant nature.

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