Unilateral Cholesteatoma in the First Millennium BC

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Objective: To analyze the bone lesions of the ear region from a late Bronze Age individual to establish the most probable diagnosis. **Background:** There has been evidence of diseases of the ear region since way back in history, but few human remains have been recognized. The case presented here corresponds to an ear lesion from a prehistoric skeleton found in the archeological site of *La Cova des Pas* (900–800 cal yr BC), located on Minorca island, in the western Mediterranean.

Methods: Macroscopic and radiologic (iCT) analysis had been performed.

Results: The remains belong to an elderly female subject who had a large cavity on the tympanic cavity as a result of the complete erosion of the outer wall of the attic and a large increase in the diameter of the outer ear canal. The cavity extends posterior to the mastoid.

Conclusion: The diagnosis suggests a probable cholesteatoma, being one of the oldest cases in Europe. **Key Words:** Bronze Age—Computed tomographic scan—Ear region disease—Minorca—Paleopathology.

Otol Neurotol 35:561-564, 2014.

Diseases of the ear region have been known since the Egyptians and the Assyrians (1). The Ebers Papyrus (764/770) and the Berlin Papyrus (70/71 and 200/203) dating from the New Kingdom, XIX dynasty, both described ear affections and their treatment (2). There is no high prevalence of aural region damage in archeological remains, and there are many difficulties with interpreting past ear diseases. Taphonomic changes to old bones due to external factors such as the chemical composition of the soil, changes in temperature, humidity, direct sunlight, or plant and animal modifications lead to the misdiagnosis of some of these lesions.

Otitis media (OM) and its complications, such as mastoiditis, chronic otitis, and cholesteatoma, may have had a major impact on ancient populations. The morphologic study of this region is often difficult, but there have been several previous case studies of ear diseases that have mainly focused on the radiologic examination of the mastoid. The

temporal bone is a complex 3-dimensional bone that makes it difficult to identify fine structures, whereas the absence of the ossicular chain is frequent given its fragility.

The oldest case of disease affecting the temporal bone is that of the Broken Hill skull dated between 300,000 and 125,000 years old discovered by A.S. Woodward in 1921 in the former Southern Rhodesia. Yearsley (3) was the first to describe the temporal pathology, and he thought that the presence of serious dental caries with alveolar abscesses plus a pneumatic anatomic variation of the mastoid and the loss of posterior wall of the attic and posterior tympanic spine could be diagnosed as Bezold's mastoiditis. Later, this was questioned because of a lack of antral involvement in this specimen (4). More recent and accurate diagnoses have been made of this case using medical endoscopes and radiographic images. For this lesion, the authors proposed, speculatively, a differential diagnosis such as an intradiploic dermoid or eosinophilic granuloma (5).

The case we present was found in the archeological site of *La Cova des Pas*, on Minorca (Balearic Islands) excavated from 2005 to 2006 (6). It can be considered an exceptional archeological discovery in the prehistoric Balearic and western Mediterranean region as it presents superb preservation and conservation of archeological and anthropologic records. The cave was of natural origin and was used as a necropolis by a pre-Talayotic culture community, in the late Bronze Age (900–800 cal yr BC) (7). Located in an inaccessible location of the wall of a ravine, had been

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This work was partially funded by the Spanish MEC: CGL2008-00800 and Generalitat de Catalunya SGR-2009-566.

The authors disclose no conflicts of interest.

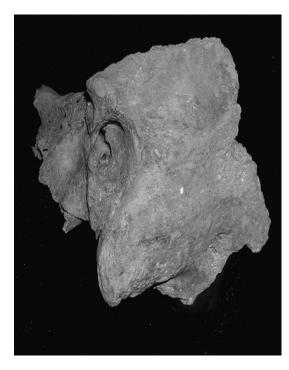


FIG. 1. IE. Left temporal bone with normal characteristics.

used to bury a minimum of 66 persons, representing both sexes and all age groups. During the funeral rite, the body was left on the cave floor and kept in a forced flexed position by the use of plant fiber ropes and shrouds. The specific environmental conditions of the cave also allowed the preservation of mummified soft tissue and hair, as well as remnants of wood and ropes (8,9).

MATERIALS AND METHODS

Individual Description

The skeleton of the individual CP- $\overline{15}$ (6) was partially preserved. Sex and age were determined according to skeletal morphologic traits that suggest an older woman of 60 to 70 years of age (10–13).

She presents several diseases and bone abnormalities. A small trauma was observed on the frontal bone, but its remodeled aspect indicates that the injury occurred long before dying (14). The parietal bones show a significant bilateral thinning that is common in older women (15) and almost complete resorption of all observable alveoli of the mandible. Degenerative signs associated with osteoarthritis of the Atlas vertebra have also been observed. However, the most important injury affected the right temporal bone. The analysis had been performed in Phillips Brilliance iCT, cutting every 0.2 mm.

RESULTS

Macroscopic Description

The left temporal bone (Fig. 1) of the individual shows a well-preserved external auditory canal. The right temporal bone (Fig. 2) presents a broad widening of the external canal, a complete destruction of the posterior canal wall,

and erosion on an important part of the mastoid. The rest of the mastoid process shows sclerosal characteristics, without aerial cells that were possibly related with a chronic inflammatory process. The inspection under ×40 magnification using a low-power microscope shows the presence of several aligned spicular structures and low crests inside, which are residues of the edges of former connecting openings between the cells and the osseous walls between them. Also, undercut edges can be seen along the cavity because of plate-like proliferations that demonstrate the common interaction between bone proliferation and bone resorption in chronic mastoiditis. Furthermore, a few bony layers and a slight periostitis are present around the external auditory meatus. Periostitis is a nonspecific response to any aggressive disease. In this case, the presence of periostits around the external auditory meatus shows that inflammation is not acute, as occurs in cholesteatomas. It is therefore a reactive lesion coming from the cholesteatoma. Therefore, in this case, the resorption could be induced by a superinfected cholesteatoma.

Radiologic Description

The left ear TC shows normal structures (Figs. 3 and 4). The right ear (Figs. 5 and 6) shows a big cavity formed by the expansion of the middle ear, destruction of the attic wall, and an important increase in the external ear canal diameters. Part of the mastoid is eroded and the rest is completely sclerosed. *Tegmen tympani* is absolutely normal. The mastoid *antrum* is also normal, and the walls are not eroded, conserving little bone spicules similar to those noted in the normal ear. The structures of the inner ear, the inner wall of the tympanic cavity, and the oval and round foramina are normal. Inner ear canal is also normal.

This description is consistent with the characteristics of a cholesteatoma, which widely affected the middle ear (destruction of the wall of the attic and mastoid sclerosis)



FIG. 2. RE. Pathologic right temporal bone: macroscopically, great erosion affecting the external ear canal and tympanic cavity can be observed.

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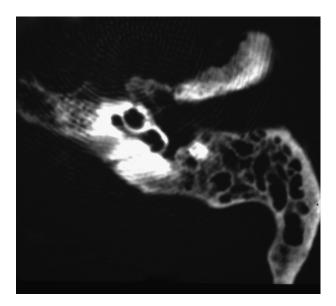


FIG. 3. IE. Axial CT of left temporal bone. Normal pneumatization of mastoid process and correct sizes of ear canal and tympanic cavity can be observed.

and was laterally externalized, destroying the superior and posterior walls of the ear canal. However, no signs of complications were found in the inner ear or in the endocraneal cavity. Therefore, after the macroscopic and radiologic analysis, the most accurate diagnosis is cholesteatoma.

DISCUSSION

Taking into account the age of the individual and the lesion patterns, the most probable diagnosis is an acquired unilateral cholesteatoma. There are very few published

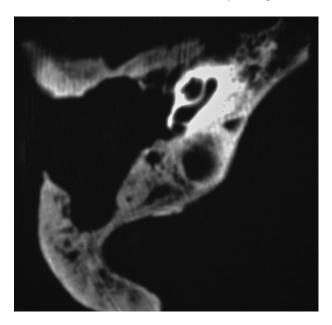


FIG. 4. IE. Coronal CT of the normal left ear where the wall of the attic is well differentiated (1).

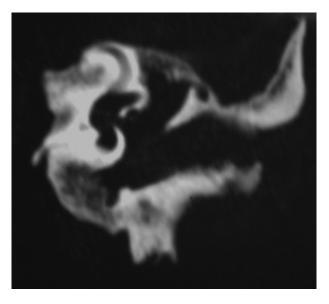


FIG. 5. RE. Axial CT scan of the right ear: (1) great erosion causing a large cavity formed by the tympanic cavity and part of the mastoid process, (2) disappearance of mastoid pneumatization, and (3) good preservation of the structures of the inner ear.

cases of this kind of lesion originating in prehistoric times. However, attribution of these ancient lesions to cholesteatoma is not always confident as are based exclusively in macroscopic exam.

The oldest case of probable cholesteatoma belongs to a Late Stone Age skull from Boskof (Transvaal / RSA) discovered in 1913 and described by Singer (16). In Ancient Egypt the presence of otologic diseases are not uncommon. Both in Predynastic and Protodynastic times, some examples of lesions diagnosed as possible cholesteatomas are

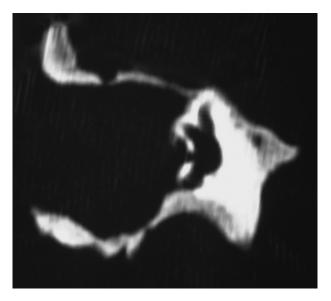


FIG. 6. RE. Coronal cut of the right ear. The image shows the following: (1) destruction of the attic external wall and erosion of the tympanic cavity roof, (2) tegmen tympani preserved, (3) normal inner ear structures, and (4) major extension of the tympanic cavity and the CAE.

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found in literature (17), some of them involving temporomandibular joints. From this period, a feasible case of cholesteatoma was found in a Predynastic skull from Nubia, now stored at the British Museum, the right temporal bone of which shows a considerable destruction of the mastoid (18). The cranium shows no sign of healing, and the individual probably died from the extensive inflammation in the ear region. Otherwise, it resembles the early dynastic skull from the Tarkhan discovered by Fitzsimons in Nubia that shows a well-defined breach in the meatal wall, possibly because of a mastoid abscess (19). Thus, the most impressive find of this piece is the presence of a fairly well-healed trephine hole at some distance on the inferior parietal bone that could be an ancient curative procedure. In America and in Asia, there have been few studies of the prevalence of mastoid infection in prehistoric times (20,21).

In Europe, few cases have been described of ear/mastoid disease in Prehistoric and Classical times. The oldest could be a Neolithic left mastoidal fistula belonging to a 20-year-old female temporal bone from Nerja (Malaga, Spain) (22). From the beginnings of the Bronze Age (Catalan Megalithic Culture), a 12-year-old child from the Dolmen at the *Cementiri dels Moros* (Torrent, Girona / Spain) shows a hole in the postero-superior wall of the left temporal bone that affects the sigmoid sinus in addition to a pneumatic mastoid process (15). However, both cases of cholesteatoma are doubtful. Also dating from the Bronze Age is the Irish skull from Knockast (23) with a lesion involving the temporomandibular joint.

More recent specimens described to experience cholesteatoma come from the Punic–Roman Necropolis of Cadiz (24). A possible case of cholesteatoma was found in a computer-assisted tomographic study of 33 temporal bones; the case is that of a female subject dating from the 4th century AD. Two other possible cases of cholesteatoma were described from Sedgeford and Red Castle–Thetford/Norfolk burials, both dating from the Late Saxon times (25) and another from Quarrington II (Lincolnshire) dating back to the 5th to 6th centuries AD (26). Finally, cholesteatomas were mentioned in four Merovingian skulls dating from the 5th to 7th century (27) in Germany.

After this revision of ancient cases of diagnosed cholesteatomas from prehistoric and historic times, the case of *La Cova des Pas* could be one of the most ancient cases of cholesteatoma in Europe ever to be published, if not the most ancient case. Also, it could be one of the most true as demonstrated by the morphologic and radiologic characteristics of these very well conserved ancient bones.

Acknowledgments: The authors thank the team and sponsors (Consell Insular de Menorca and Caixa de Catalunya) of the excavation of *La Cova des Pas*.

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