A case of cholesteatoma in a medieval Hispano-Mudejar population (13th–14th centuries AD)

Enrique Dorado[1,2] | José Aso[3,4] | Cristina Sebastián[5] | Ildefonso Ramírez[6] | Manuel F. Carrillo[7] | Jorge Murillo-González[8]

[1]Forensic Anthropology and Odontology Department, Institute of Legal Medicine, Madrid, Spain

[2]Department of Legal Medicine, Psychiatry and Pathology, Faculty of Medicine, Complutense University, Madrid, Spain

[3]Neurosurgery Department, MAZ Hospital, Zaragoza, Spain

[4]Faculty of Health Sciences, San Jorge University, Zaragoza, Spain

[5]Radiology Department, MAZ Hospital, Zaragoza, Spain

[6]Grupo 365 Arqueología, Guadalajara, Spain

[7]Department of Surgery, Medical and Social Sciences, Faculty of Medicine, University of Alcalá, Madrid, Spain

[8]Department of Anatomy and Embryology, Faculty of Medicine, Complutense University of Madrid, Madrid, Spain

Correspondence
Jorge Murillo-González, Department of Anatomy and Embryology, Faculty of Medicine, Complutense University of Madrid, 28040 Madrid, Spain.
Email: jmurillo@med.ucm.es

Funding information
Santander Bank/Complutense University of Madrid, Grant/Award Number: PR108/20-18

Abstract

The case presented includes a left temporal bone from an individual exhumed from the Hispano-Mudejar necropolis in Uceda (Guadalajara, Spain) dated between the 13th and 14th centuries BC. External examination and computed tomography images show lesions in the external acoustic canal, in the form of diffuse widening, suggestive of a cholesteatoma originating in this canal, with invasion of the middle ear through the tympanic membrane. The difficulties with examining the internal elements of the ear are discussed, which causes the underestimation of these diseases in paleopathology studies.

KEYWORDS
bioarcheology, cholesteatoma, computed tomographic scan, ear disease, Islamic necropolis, paleopathology, temporal bone, Uceda

1 | INTRODUCTION

Paleopathological analysis constitutes an important source of the knowledge of diseases and health status of a population. Pathologies such as otitis, closely related to upper respiratory tract infections, are a reflection of the nutritional and hygienic conditions of the population, and ultimately their degree of development (Mann, 1992; Qvist & Grøntved, 2001).

However, the paleopathological study of otological diseases encounters important obstacles. In addition to the difficulty of accessing the examination of internal auditory structures, taphonomic phenomena can cause alterations that complicate their examination and facilitate diagnostic error (Armentano et al., 2014; Qvist & Grøntved, 2001).

All of this has led to scarce references to otological diseases in paleopathological registries and those of their differential diagnosis are very limited. However, these are diseases, such as cholesteatoma, which have presumably played a not inconsiderable role in the morbidity and mortality of ancient populations (Armentano et al., 2014; Mays & Holst, 2006).

Modern computed tomography (CT) techniques, applied to both bone remains and mummified bodies, have represented an extraordinary advance, allowing the noninvasive analysis of the complex region of the ear and its fine structures (Armentano et al., 2014).

Other techniques used in these studies include, in addition to direct examination with or without a magnifying operation microscope (Mann, 1992; Mays & Holst, 2006; Montgomery et al., 1994; Qvist & Grøntved, 2001), simple radiography (Drenhaus & Hilmann, 1991; Gregg et al., 1965), the optical endoscope (Qvist & Grøntved, 2001) and even histological and immunological techniques (Horne et al., 1976; Lynn & Benitez, 1974; Park et al., 2001; Soldati & Mudry, 2001).

In the present work, a case of cholesteatoma is exposed from an individual exhumed from a medieval cemetery and its possible etiology is discussed. The case highlights the difficulty in examining the internal elements of the ear, which causes an underestimation of these diseases in paleopathology studies.

2 | MATERIALS AND METHODS

The temporal bone of the skeleton analyzed came from one of the 116 exhumed bodies, all in individual burial, in the Hispano-Mudejar necropolis of Uceda (Guadalajara, Spain), dated between the 13th and 14th centuries AD. All burial pits showed a South–North orientation (Head–Feet), with the body in lateral or supine decubitus position and the head oriented to Mecca (Ramírez-González & Dorado, 2020) (Figure 1).

Of the skeleton analyzed, from 14 stratigraphic units, most was recovered, with a moderate general degree of conservation (White, 2008). Age was estimated based on tooth enamel wear (Brothwell, 1987; Lovejoy, 1985), changes in the auricular surface of the ilium (Lovejoy et al., 1985), and changes in the pubic symphysis (Brooks & Suchey, 1990). Sex was established according to the morphological characters of the pelvis and cranium (Buikstra & Ubelaker, 1994), whereas height was estimated through the length of the femur (Mendonça, 2003).

The temporal bone was examined using noninvasive techniques, gross examination, and CT scans.

3 | RESULTS

The analyzed skeleton corresponds to a female individual, with an estimated age between 35 and 45 years and a height of approximately 150 cm. Both temporal bones were recovered in good condition, with partial bone loss of the petrous portion of the right temporal bone (Figure 2a,b). The external examination shows a broad widening of the external acoustic canal (EAC) of the left temporal bone, in clear contrast with the right temporal bone, which has a normal appearance. The rest of the skeleton shows no other pathological alterations, except for an osteochondritis dissecans in the trochlea of the right humerus.

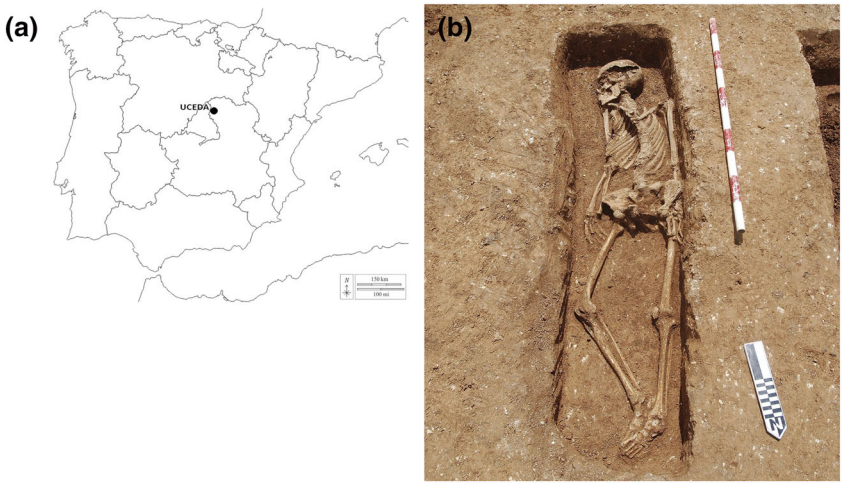

FIGURE 1 (a) Location of Uceda (Guadalajara, Spain). (b) Stratigraphic units 14 [Colour figure can be viewed at wileyonlinelibrary.com]

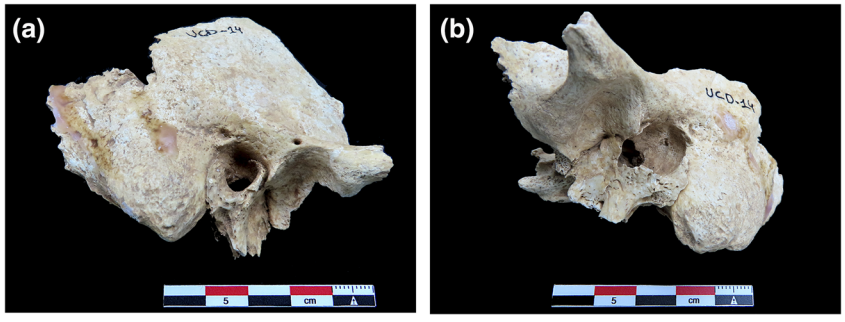

FIGURE 2 (a) Right temporal bone with normal characteristics. (b) Pathologic left temporal bone: macroscopically, great erosion affecting the external ear canal and tympanic cavity can be observed [Colour figure can be viewed at wileyonlinelibrary.com]

By CT, in the petrous portion of the left temporal bone, diffuse widening of the EAC, with scalloped and well-defined contours can be appreciated (Figure 3). No erosion of the scutum, Prussak's space, or widening or erosion of the tegmental wall was identified (Figure 4). The tympanic membrane (TM) was not identified in the study (Figures 3 and 4). The malleus head and the short process of the incus were displaced cranially (Figure 4), whereas the long process of the incus and the stapes were displaced medially (Figure 5). The vestible, semicircular canals and cochlea were unaltered (Figure 6). Mastoid involvement was not observed.

4 | DISCUSSION

The good state of preservation of the temporal bone exposed here has made it possible, in addition to its external examination, to obtain images by CT, which has allowed the precise examination of its structures. The diffuse widening of the EAC, with scalloped and well-defined contours, the displacement of part of the ossicle chain cranially and laterally (malleus head and short process of the incus) and part medially (long process of the incus and stapes), but remaining intact, are they indirect findings of benign and slow-growing lesions, ruling out infiltrating lesions such as squamous carcinoma of EAC. These findings are compatible with a primary cholesteatoma originating in the EAC with posterior invasion of the middle ear (ME) through the TM.

The first description of the cholesteatoma is attributed to Duverney (1648–1730), although previous references could be found described in the Hippocratic Corpus (Benmoussa et al., 2020). The medical term was proposed by Müller in 1838 (Soldati & Mudry, 2001).

Cholesteatoma consists of a nonneoplastic and ectopic proliferation of stratified squamous tissue, together with exfoliated keratin (Aswani et al., 2016).

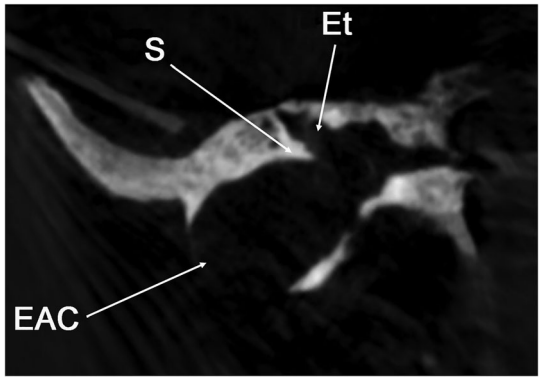

FIGURE 3 Coronal computed tomography (CT) of left temporal bone depict widening of the external acoustic canal (EAC), with scalloped and well-defined contours. Et, epitympanum; S, scutum

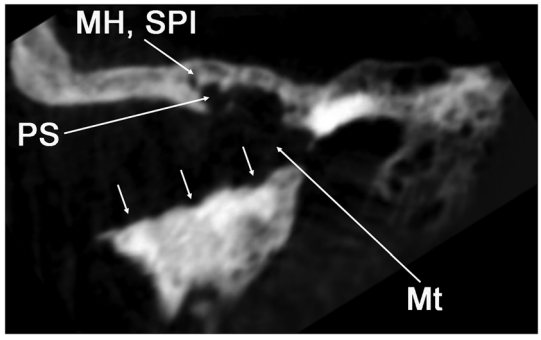

FIGURE 4 Coronal computed tomography (CT) of left temporal bone depict part of the ossicle chain in the epitympanum (MH, malleus head; SPI, short process of the incus), displaced cranially and laterally, near the tegmental wall. The middle ear and the external acoustic canal are widened (short arrows), although with scalloped contours. Mt, mesotympanus; PS, Prussak's space

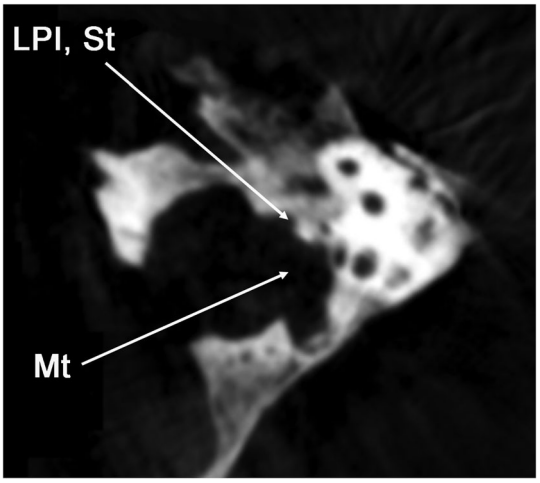

FIGURE 5 Axial oblique TC of left temporal bone depict part of the ossicle chain displaced medially but intact (LPI, long process of the incus; St, stapes). Mt, mesotympanus

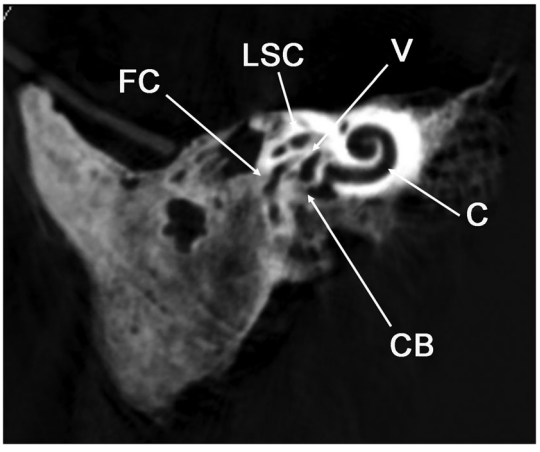

FIGURE 6 Coronal computed tomography (CT) of left temporal bone. The normal structures of the inner ear are observed. C, cochlea; CB, cochlea base; FC, facial canal; LSC, lateral semicircular canal; V, vestible

It is an otological pathology that generally presents with hearing loss and a purulent and fetid otorrhea that requires frequent cleaning. The hearing loss is of conductive or mixed type, caused by perforation of the TM and involvement of the ossicles chain. Otalgia, earfullness, and itching sensation may also be present (Kim et al., 2014). Its natural evolution is progressive growth invading the ME, with destruction of the bone and the ossicles chain. Possible complications may compromise the patient's life, especially in times prior to antibiotic therapy such as the present case. Complications include retroauricular abscess, facial nerve palsy, cerebrospinal fluid fistula, meningitis, brain abscess, or lateral sinus thrombosis (Muñoz et al., 2007).

From various studies, the incidence of cholesteatoma was 3–15 per 100,000 person-years (Im et al., 2020). Its usual location is the ME, being up to 60 times less frequent in the EAC (Dubach & Häusler, 2008; Kim et al., 2014; Owen et al., 2006).

ME cholesteatomas are produced by a perforation or retraction pocket in the flaccid pars or the tense pars of the TM (Phillips et al., 2012).

EAC cholesteatomas can be primary or secondary. Primary cholesteatoma is produced by a slowing of the expulsion to the exterior of the keratin debris from the external surface of the TM and EAC, producing erosions of soft contours, which are frequent in the lower wall of the canal. In secondary cholesteatoma, the retention of keratin debris can be due to different causes, such as trauma (Bhagat et al., 2013; Heilbrun et al., 2003; Shin et al., 2010).

EAC cholesteatomas can invade the ME, mastoid cells, or temporomandibular joint (Chawla et al., 2015). When also invading the ME, it is very difficult to distinguish them from the cholesteatoma originating in the ME, as in the case at hand. The absence of erosion of the scutum, Prussak's space, or widening or erosion of the tegmental wall, makes the diagnosis of a cholesteatoma originating in the ME unlikely.

Otological conditions have received little attention in the paleopathological literature, mainly due to the difficulties associated with their study (Armentano et al., 2014; Mann, 1992; Mays & Holst, 2006).

Actually, CT has represented an unquestionable advance in the study of the temporal bone. In addition to avoiding the overlap of images, it provides a scale of densities much higher than conventional radiology and identifies remains of earth, frequent in archeological simples (Villanueva et al., 1997). In the application of this technique, it is important that the bone has a good state of conservation, although the structures of the ME are usually well protected (Qvist & Grøntved, 2001).

Paleopathological studies have mainly collected cases of mastoiditis (Drenhaus & Hilmann, 1991; Gregg et al., 1965; Loveland et al., 1990; Rathbun & Mallin, 1977; Wells, 1962). Next, we will briefly refer to the few specific cases of colesteatoma found after the literature review.

The oldest case described corresponds to the Broken Hill skull, dated 250–130,000 years old, although subsequent analyses propose another diagnosis distinguishing, together with postmortem traumatic lesions, the possible existence of an intradiploic dermoid or an eosinophilic granuloma in the squamous portion (Montgomery et al., 1994).

Corresponding to the late Bronze, pre-Talayoticin culture, on the island of Menorca (Spain), a case of cholesteatoma has been described in the skeleton of an elderly woman, used for this, in addition to external examination, images obtained by CT (Armentano et al., 2014). Likewise, several cases of possible cholesteatoma have been described in mummies of ancient Egypt, observing, in an examination of 453 Egyptian skulls from Nagada, evidence of this disease in 22 of them (Mann, 1992).

Similar cases have been described in later times. The skeleton of a 25- to 30-year-old woman, from a burial in the 4th century BC, in San Fernando (Cádiz), examined with the aid of CT, showed signs of a cholesteatomatous lesion, in addition to chronic otomastoiditis (Macías et al., 2001; Villanueva et al., 1997).

Other cases of cholesteatoma in ancient remains are the ME cholesteatoma observed in a skeleton from Quarrington (Lincolnshire), 5th–6th century AD (Mays & Holst, 2006); calcified cholesteatoma in a 40- to 50-year-old individual from the Frankish-Alemannic cemetery, dated between 500 and 725 AD (Schultz, 1979); or the case of cholesteatoma-like destructions of the foramen ovale area and the labyrinth capsule, exposing the vestibulum, from a medieval Danish cemetery (Qvist & Grøntved, 2001). Two cases dated from the 19th century, one corresponding to a cholesteatoma of the external canal, in a skeleton of Wharram Percy North Yorkshire (Mays & Holst, 2006) and another from Uummaannaq (Greenland) (Homøe et al., 1992).

5 | CONCLUSIONS

CT represents an advance in the paleopathological study of otological diseases contributing to the knowledge of the health and disease of the population, in the present case of a medieval Mudejar population.

ACKNOWLEDGMENTS

This work is part of the doctoral thesis that the first author is completing at the University of Granada (Spain). We would like to thank Dr Miguel Botella, Full Professor, for his collaboration. We also thank Proof-Reading-Service.com for the proofreading and English editing of this manuscript. This work was partially supported by Grant PR108/20-18 from Santander Bank/Complutense University of Madrid.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data available on request from the authors.

ORCID

Jorge Murillo-González https://orcid.org/0000-0002-3455-2378

REFERENCES

Armentano, N., Malgosa, A., Martínez, B., Abelló, P., de Juan Delago, M., Prats-Muñoz, G., & Isidro, A. (2014). Unilateral cholesteatoma in the

first millennium BC. *Otology and Neurotology*, *35*(3), 561–564. https://doi.org/10.1097/MAO.0b013e3182a03542 PMID: 24136313

Aswani, Y., Varma, R., & Achuthan, G. (2016). Spontaneous external auditory canal cholesteatoma in a young male: Imaging findings and differential diagnoses. *The Indian Journal of Radiology & Imaging*, *26*(2), 237–240. https://doi.org/10.4103/0971-3026.184419 PMID: 27413272

Benmoussa, N., Fabre, C., Deo, S., Prêtre, C., & Charlier, P. (2020). The first description of cholesteatoma by Hippocrate. *European Archives of Oto-rhinolaryngology*, *277*(6), 1651–1653. https://doi.org/10.1007/s00405-020-05899-8 PMID: 32162060

Bhagat, S., Varshney, S., Bist, S. S., Mishra, S., Kabdwal, N., & Kumar, R. (2013). Primary external auditory canal cholesteatoma presenting as cerebellar abscess. *Indian Journal of Otology*, *19*(2), 88–91. https://doi.org/10.4103/0971-7749.113515

Brooks, S., & Suchey, J. M. (1990). Skeletal age determination based on the os pubis: A comparison of the Acsádi-Nemeskéri and Suchey-Brooks methods. *Human Evolution*, *5*, 227–238. https://doi.org/10.1007/BF02437238

Brothwell, D. (1987). *Desenterrando huesos: la excavación, tratamiento y estudio de restos del esqueleto humano*. Fondo de Cultura Económica. ISBN: 84-375-0367-1.

Buikstra, J. E., & Ubelaker, D. H. (1994). Standards for data collection from human skeletal remains. Proceedings of a Seminar at the Field Museum of Natural History, Arkansas Archaeological Sur Res Ser No. 44, Fayetteville. AR.

Chawla, A., Ezhil Bosco, J. I., Lim, T. C., Shenoy, J. N., & Krishnan, V. (2015). Computed tomography features of external auditory canal cholesteatoma: A pictorial review. *Current Problems in Diagnostic Radiology*, *44*(6), 511–516. https://doi.org/10.1067/j.cpradiol.2015.05.001 PMID: 26050022

Drenhaus, U., & Hilmann, H. (1991). Mastoiditis in a skull of the middle pre-Roman Iron Age. *Journal of Palepopathology*, *4*(1), 15–23.

Dubach, P., & Häusler, R. (2008). External auditory canal cholesteatoma: Reassessment of and amendments to its categorization, pathogenesis, and treatment in 34 patients. *Otology and Neurotology*, *29*(7), 941–948. https://doi.org/10.1097/MAo.0b013e318185fb20 PMID: 18758389

Gregg, J. B., Steele, J. P., & Holzhuetter, A. (1965). Roentgenographic evaluation of temporal bones from South Dakota Indian burials. *American Journal of Physical Anthropology*, *23*(1), 51–61. https://doi.org/10.1002/ajpa.1330230120 PMID: 14326955

Heilbrun, M. E., Salzman, K. L., Glastonbury, C. M., Harnsberger, H. R., Kennedy, R. J., & Shelton, C. (2003). External auditory canal cholesteatoma: Clinical and imaging spectrum. *American Journal of Neuroradiology*, *24*(4), 751–756. PMID: 12695217

Homøe, P., Lynnerup, N., & Videbaek, H. (1992). CT-scanning of ancient Greenlandic Inuit temporal bones. *Acta Otolaryngologica*, *112*(4), 674–679. https://doi.org/10.3109/00016489209137458 PMID: 1442014

Horne, P. D., MacKay, A., Jahn, A. F., & Hawke, M. (1976). Histologic processing and examination of a 4,000-year-old human temporal bone. *Archives of Otolaryngology*, *102*(12), 713–715. https://doi.org/10.1001/archotol.1976.00780170031001 PMID: 803176

Im, G. J., do Han, K., Park, K. H., Cho, C. H., Jang, H., Lee, J. H., & Lee, S. H. (2020). Rate of chronic otitis media operations and cholesteatoma surgeries in South Korea: A nationwide population-based study (2006–2018). *Scientific Reports*, *10*(1), 11356. https://doi.org/10.1038/s41598-020-67799-5 PMID: 32647201

Kim, C. W., Baek, S. H., Lee, S. H., Kim, G. W., & Cho, B. K. (2014). Clinical characteristics of spontaneous cholesteatoma of the external auditory canal in children comparing with cholesteatoma in adults. *European Archives of Otorhinolaryngology*, *271*(12), 3179–3185. https://doi.org/10.1007/s00405-013-2820-6 PMID: 24258852

Lovejoy, C. O. (1985). Dental wear in the Libben population: Its functional pattern and role in the determination of adult skeletal age at death. *American Journal of Physical Anthropology*, *68*(1), 47–56. https://doi.org/10.1002/ajpa.1330680105 PMID: 4061601

Lovejoy, C. O., Meindl, R. S., Pryzbeck, T. R., & Mensforth, R. P. (1985). Chronological metamorphosis of the auricular surface of the ilium: A new method for the determination of adult skeletal age at death. *American Journal of Physical Anthropology*, *68*(1), 15–28. https://doi.org/10.1002/ajpa.1330680103 PMID: 4061599

Loveland, C. J., Pierce, L. C., & Gregg, J. B. (1990). Ancient temporal bone osteopathology. *Annals of Otology, Rhinology and Laryngology*, *99*(2 Pt 1), 146–154. https://doi.org/10.1177/000348949009900214 PMID: 2405762

Lynn, G. E., & Benitez, J. T. (1974). Temporal bone preservation in a 2600-year-old Egyptian mummy. *Science*, *183*(4121), 200–202. https://doi.org/10.1126/science.183.4121.200 PMID: 4587441

Macías, M., Villanueva, A., Matero, A., & Ruza, M. (2001). Enfermedades otológicas halladas en una muestra de población púnica y romana. https://webs.ucm.es/info/aep/boletin/actas/14.pdf [update 16 July 2021].

Mann, G. E. (1992). The identification of chronic ear disease in the dried skull. *International Journal of Osteroarchaeology*, *2*(1), 19–22. https://doi.org/10.1002/oa.1390020104

Mays, S., & Holst, M. (2006). Palaeo-otology of cholesteatoma. *International Journal of Osteroarchaeology*, *16*(1), 1–15. https://doi.org/10.1002/oa.801

Mendonça, M. C. N. (2003). Contribución para la identificación humana a partir del estudio de las estructuras óseas: determinación de la talla a través de la longitud de los huesos largos. https://eprints.ucm.es/2917/1/T22911.pdf [update 1 July 2021].

Montgomery, P. Q., Williams, H. O. L., Reading, N., & Stringer, C. B. (1994). An assessment of the temporal bone lesions of the Broken Hill cranium. *Journal of Archaeological Science*, *21*(3), 331–337. https://doi.org/10.1006/jasc.1994.1032

Muñoz, C., Macías, L. A., & Cruz, T. R. (2007). Colesteatoma de oído. *Anales de Radiología México*, *1*, 71–77.

Owen, H. H., Rosborg, J., & Gaihede, M. (2006). Cholesteatoma of the external ear canal: etiological factors, symptoms and clinical findings in a series of 48 cases. *BMC Ear, Nose, and Throat Disorders*, *6*, 16. https://doi.org/10.1186/1472-6815-6-16 PMID: 17187684

Park, K., Park, H. J., & Chun, Y. M. (2001). Immunohistochemical study on proliferative activity of experimental cholesteatoma. *European Archives of Otorhinolaryngology*, *258*(3), 101–105. https://doi.org/10.1007/s004050100315 PMID: 11374247

Phillips, G. S., LoGerfo, S. E., Richardson, M. L., & Anzai, Y. (2012). Interactive web-based learning module on CT of the temporal bone: Anatomy and pathology. *Radiographics*, *32*(3), E85–E105. https://doi.org/10.1148/rg.323115117 PMID: 22582369

Qvist, M., & Grøntved, A. M. (2001). Chronic otitis media sequelae in skeletal material from medieval Denmark. *Laryngoscope*, *111*(1), 114–118. https://doi.org/10.1097/00005537-200101000-00020 PMID: 11192878

Ramírez-González, I., & Dorado, E. (2020). Memoria de la excavación en las parcelas 9H, 9I y 9J en el Camino Cañada Araguz de Uceda (Guadalajara). Depositada en la Junta de Comunidades de Castilla-La Mancha.

Rathbun, T. A., & Mallin, R. (1977). Middle ear disease in a prehistoric Iranian population. *Bulletin of the New York Academy of Medicine*, *53*(10), 901–905. PMID: 338065

Schultz, M. (1979). Diseases in the ear region in early and prehistoric populations. *Journal of Human Evolution*, *8*(6), 575–580. https://doi.org/10.1016/0047-2484(79)90109-X

Shin, S. H., Shim, J. H., & Lee, H. K. (2010). Classification of external auditory canal cholesteatoma by computed tomography. *Clinical and Experimental Otorhinolaryngology*, *3*(1), 24–26. https://doi.org/10.3342/ceo.2010.3.1.24 PMID: 20379398

Soldati, D., & Mudry, A. (2001). Knowledge about cholesteatoma, from the first description to the modern histopathology. *Otology and Neurotology*, *22*(6), 723–730. https://doi.org/10.1097/00129492-200111000-00003 PMID: 11698787

Villanueva, A., Macías, M., Ruza, M., & Mateo, A. (1997). Utilidad de la tomografía computarizada de oído en paleopatología. https://www.academia.edu/43062695/Utilidad_de_la_Tomograf%C3%ADa_computadorizada_de_o%C3%ADdo_en_Paleopatolog%C3%ADa [update 16 July 2021].

Wells, C. (1962). Three cases of aural pathology of Anglo-Saxon date. *The Journal of Laryngology and Otology*, *76*(11), 931–933. https://doi.org/10.1017/s002221510006014x PMID: 13999874

White, W. (2008). The human skeletal remains from the burial ground of St Benet Sherehog. In A. Miles, W. White, & D. Tamnkard (Eds.), *Burial at the site of the parish church of St Benet Sherehog before and after the Great Fire*. MoLAS Monograph Series 39. (pp. 70–92). Museum of London Archaeological Service Publications. ISBN: 978-1-901992-75-5.

How to cite this article: Dorado, E., Aso, J., Sebastián, C., Ramírez, I., Carrillo, M. F., & Murillo-González, J. (2022). A case of cholesteatoma in a medieval Hispano-Mudejar population (13th–14th centuries AD). *International Journal of Osteoarchaeology*, *32*(1), 294–299. https://doi.org/10.1002/oa.3062