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Cementoblastoma in a red deer (*Cervus elaphus*) from the Late Pleistocene of Rochedane, France

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A B S T R A C T

Only relatively few cases of dental abnormalities in wild ruminants have thus far been described in the paleopathological literature. This study reports a case of cementoblastoma, a benign odontogenic tumor of ectomesenchymal origin, in a red deer (*Cervus elaphus*) from the Late Pleistocene of Rochedane, a prehistoric site in the French Jura. The tumor was attached to the root of a heavily worn loose left maxillary third molar. CT imaging revealed several radiolucent (former soft tissue) spaces of varying shape and size within the mineralized tumorous mass. Light microscopic analysis and backscattered electron imaging in the SEM showed that the process of dental wear had reached the tumor and that the tooth and the attached tumor had undergone considerable microbial diagenesis. This is the first case of cementoblastoma described in a prehistoric animal and also the first report of this type of odontogenic tumor in a deer.

Keywords:
Animal paleopathology
Dental pathology
Odontogenic tumor
Cementoblastoma
Late Pleistocene

1. Introduction

While there exist numerous reports on dental pathology in domestic animals from prehistoric sites, only relatively few cases of dental abnormalities in wild animals have been described in the paleopathological literature (Baker and Brothwell, 1980; Miles and Grigson, 1990; Bartosiewicz, 2013). Regarding odontogenic tumors and tumor-like lesions in prehistoric wild animals, thus far only a number of odontomas (tumor-like malformations or hamartomas; Praetorius and Piattelli, 2005a,b; Brown et al., 2007) in proboscideans (Van Essen, 2004) and a single case of odontoma in a red deer (*Cervus elaphus*) (Binois et al., 2014) have been reported. The present paper reports the case of an odontogenic tumor in a red deer from the Late Pleistocene of Rochedane, France.

2. Materials and methods

2.1. The site of Rochedane

The prehistoric site of Rochedane is located in the French Jura about 25 km south of the town of Montbeliard on the left bank of the Doubs river. It is a rock shelter dug into an

approximately 15 m high calcareous cliff at an altitude of about 355 m above sea level. Neighboring plateaus rise to about 700 m above sea level. The excavation (1968–1976) identified several archaeological layers covering the time span from the Late Pleistocene to the early Holocene (Thévenin and Sainty, 1998; Drucker et al., 2009). All layers contained remains of red deer (Bridault, 1990).

2.2. Stratigraphic position of the specimen

The specimen reported and discussed in this paper, a loose maxillary molar of a red deer (find number: Ro76, 402), originated from layer B of the site. Direct accelerator mass spectrometry (AMS) radiocarbon dating performed on collagen of a red deer metatarsal from this layer revealed an age of 13,650–13,282 cal BP (2σ), corresponding to the Early Alleröd pollen chronozone (Drucker et al., 2009).

2.3. Macroscopic, CT and microscopic analysis

The tooth was first inspected macroscopically and photographed using a digital camera (Canon EOS 300D; Canon, Tokyo, Japan). Computed tomography (CT) imaging of the specimen was performed with a Philips Brilliance CT 64-channel scanner (slice thickness 1 mm, 70 kV, 151 mAs). Subsequently, the tooth was embedded in epoxy resin (Biodur products, Heidelberg, Germany)

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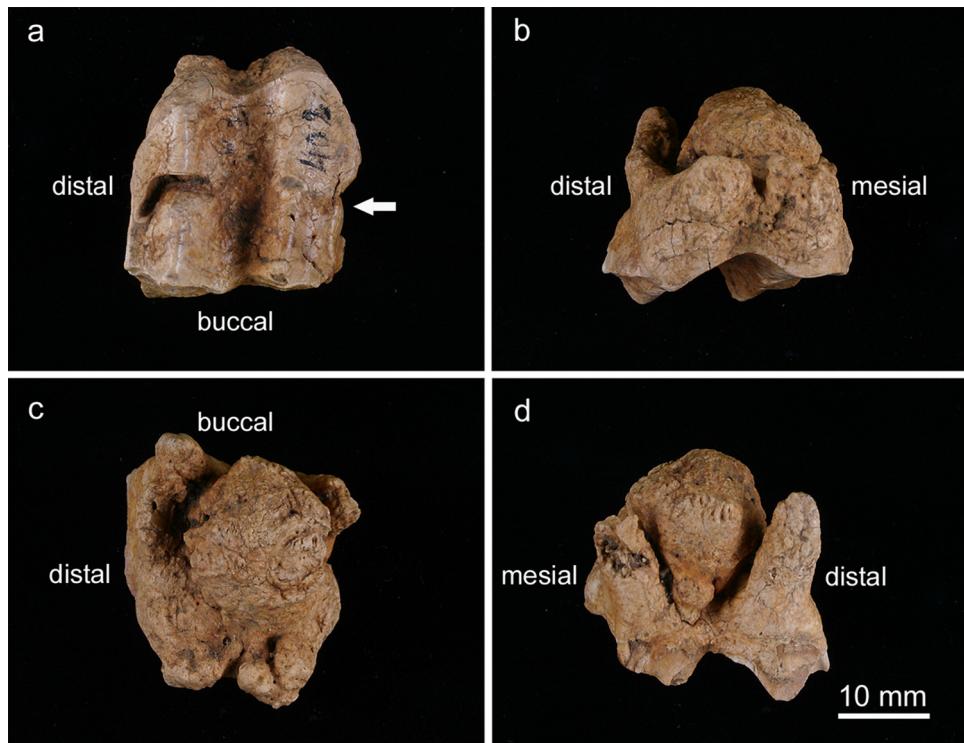


Fig. 1. Left maxillary third molar of a red deer from the Late Pleistocene of Rochedane (find number Ro76, 402) with cementoblastoma attached to the root; (a) occlusal view, (b) palatal view, (c) apical view, (d) buccal view. Arrow: approximal wear facet mesially.

and sectioned in a mesio-distal plane, using a rotary saw with a water-cooled, diamond-coated blade. The cut surfaces were smoothed and polished as described previously (Kierdorf et al., 2012). The polished, uncoated surfaces were then viewed in an environmental scanning electron microscope (ESEM; FEI Quanta 600 FEG, accelerating voltage 20 kV) operated in the backscattered electron (BSE) mode. Following capturing of the BSE-images, each tooth half was mounted with its polished side down on a glass slide, using the epoxy resin as glue. Two ground sections, each with a thickness of approximately 50 µm, were produced as described in Kierdorf et al. (2012). The cover-slipped sections were viewed and photographed in transmitted light in an Axioskop 2 Plus microscope (Zeiss, Jena, Germany) equipped with a digital camera (Canon PowerShot G2).

3. Case description

3.1. Macroscopic description

The specimen presented here was a loose left maxillary third molar of a red deer. The tooth was heavily worn, and its crown exhibited a facet from approximal wear mesially, whereas there was no wear facet distally, as is typical for a third molar. Only a very small remnant of the base of the infundibulum was present on the anterior (mesial) tooth lobe, while a more extended portion of the infundibular base had remained on the posterior (distal) lobe (Fig. 1a). The shape of the occlusal surface of the tooth was normal for a heavily worn red deer maxillary molar (*cf. e.g.* von Raesfeld, 1920, p. 160, figure 79). The molar's disto-buccal root was preserved completely, whereas the other roots were incomplete, as smaller or larger portions had been lost, probably due to post-mortem breakage (Fig. 1b–d).

A nodular mineralized mass was attached to the furcation area at the common base of the tooth roots (Fig. 1b–d). The nodular mass, which showed the same color and surface structure as the

adjacent root cementum, tapered in apical direction (*i.e.*, in the direction of the former root apices), forming a blunt tip. Its apical (tip) region exhibited a more irregular surface structure than the flanks, which could have resulted from the loss of a covering layer of unmineralized tissue and/or erosional processes.

3.2. CT imaging and microscopic analysis

CT-imaging of the tooth revealed that the nodular mineralized mass attached to the tooth root was not solid, but exhibited several radiolucent areas of different shape, some of which were of a large size (Fig. 2a).

Light-microscopic analysis of the ground sections confirmed the presence of several, partly relatively small, partly extended unmineralized (former soft tissue) spaces within the nodular mass (Fig. 2b). The more centrally located portion of the mass exhibited a pale-brown color, whereas the more peripheral areas showed a grayish/blackish staining. This dark staining was also present in parts of the tooth. A distinct boundary between the tooth and the nodular mineralized mass was not discernible.

Except for some parts bordering on the occlusal surface, the mineralized, cementum-like mass contained only few cementocyte lacunae, with larger portions showing none at all (Fig. 3a and b). In the central tooth portion this mineralized mass formed the occlusal surface (Fig. 3a), indicating that the process of tooth wear had eventually reached the nodular mass. Only in the portion of the mineralized mass that bordered on the occlusal surface, irregularly arranged patches or streaks of cementum containing numerous cementocyte lacunae were present.

Microscopic analysis further revealed that the tooth and the attached nodular mass had undergone considerable microbial diagenesis, the process having obscured much of the histological structure. Thus, numerous areas of tissue destruction were present both in the tooth (Fig. 4a and b) and the nodular mass (Fig. 4c and d). Infiltration with a filamentous (bacterial) material was

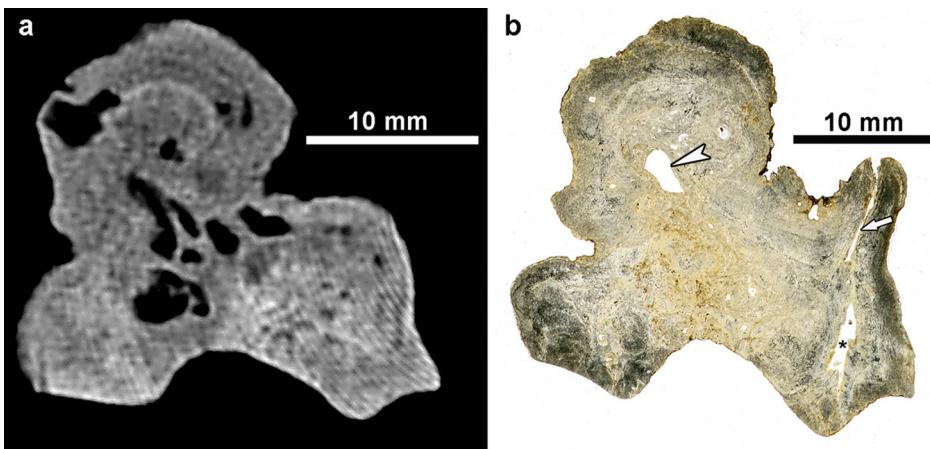


Fig. 2. CT scan (a) and unstained ground section (b) of the M^3 (find number Ro76, 402), both oriented in a mesiodistal plane; distal to the right of the images. (a) Note occurrence of several radiolucent spaces within the tumorous mass; (b) unmineralized (former soft tissue) spaces are also discernible in the section (arrowhead). Asterisk: pulp chamber; arrow: root canal.

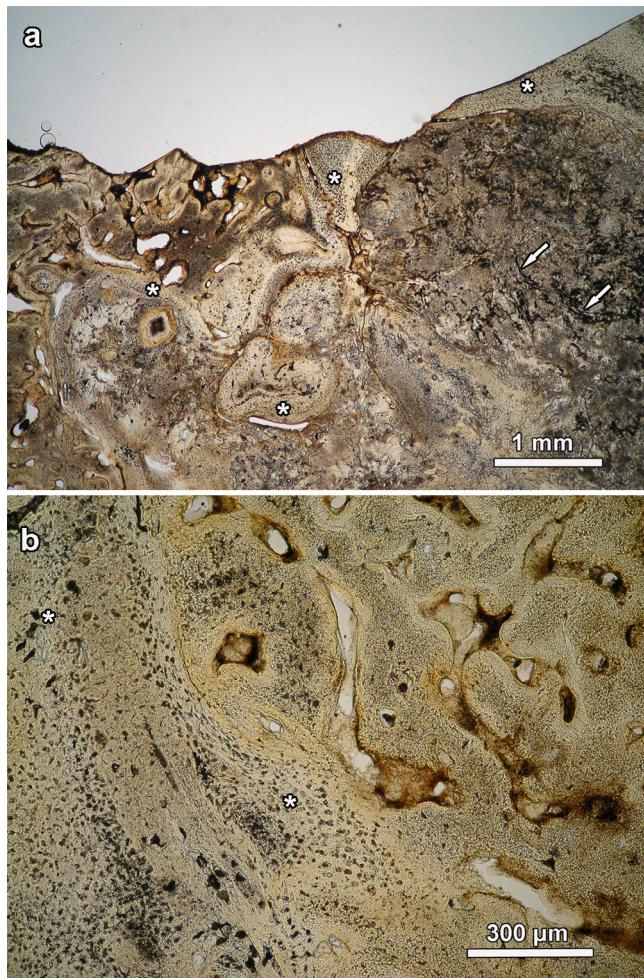


Fig. 3. Brightfield images of unstained mesiodistal ground section of the M^3 (find number Ro76, 402); occlusal direction to the top of the images. (a) The material beneath the occlusal surface consists partly of a tissue with only few cementocyte lacunae, partly of patches or streaks of a tissue with numerous small cementocyte lacunae (asterisks). Both tissue types are exposed at the occlusal surface. Arrows: filamentous material. (b) Higher magnification of the border between the material rich in cementocyte lacunae (asterisks) and that with few cementocyte lacunae.

especially prominent in the peripheral areas of the tooth and the nodular mass, causing the grayish/blackish staining of these regions in the ground sections. BSE-SEM imaging revealed the presence of numerous foci of bacterial invasion. These foci were of varying size and shape and characterized by numerous small holes. In some cases, the area around these small holes exhibited darker gray levels than their surroundings in the BSE-SEM images (Fig. 4a, c and d), indicating a lower mineral content due to mineral dissolution around routes of bacterial invasion. In some cases, the dark areas were lined by bright rims, or a complete focus of bacterial invasion appeared brighter (more highly mineralized) than its surroundings in the BSE-SEM images (Fig. 4c and d). These findings were indicative of a re-precipitation of dissolved hydroxyapatite in the respective regions (Turner-Walker, 2008).

4. Discussion

The nodular mineralized mass attached to the root of the red deer molar from Rochedane was diagnosed as a cementoblastoma. This is the term used in the current classification of odontogenic tumors by the World Health Organization (Van der Waal, 2005). Synonyms for this type of neoplasm that can be found in the literature are benign cementoblastoma, true cementoma, and benign cementoma (Cherrick et al., 1974; Kramer et al., 1992; Ulmansky et al., 1994; Kreutzer et al., 2007).

The cementoblastoma is a rare benign odontogenic tumor of ectomesenchymal origin that is histologically characterized as consisting of cementum or cementum-like tissue (Zachariades et al., 1985; Kramer et al., 1992; Ulmansky et al., 1994; Brannon et al., 2002; Kreutzer et al., 2007; Jundt and Reichart, 2008; Sankari and Ramakrishnan, 2011). In humans, just over 100 cases of this tumor were reported up to 2005 (Van der Waal, 2005). Most cementoblastomas reported in humans were located in the mandible, with a predilection for the mandibular permanent first molar (Brannon et al., 2002; Van der Waal, 2005; Huber and Folk, 2009). Association of this tumor with deciduous teeth has only rarely been observed (Van der Waal, 2005; Lemberg et al., 2007; De Noronha Santos Netto et al., 2012; Monti et al., 2013).

In humans, cementoblastomas are characterized as slowly but steadily enlarging tumors that can eventually reach a considerable size (Zachariades et al., 1985; Brannon et al., 2002). While the cementoblastoma was originally believed to show no or only little tendency to recur (Kramer et al., 1992), a later study in humans reported a recurrence rate of 37% following surgical removal of the tumor (Brannon et al., 2002). Due to its unlimited growth

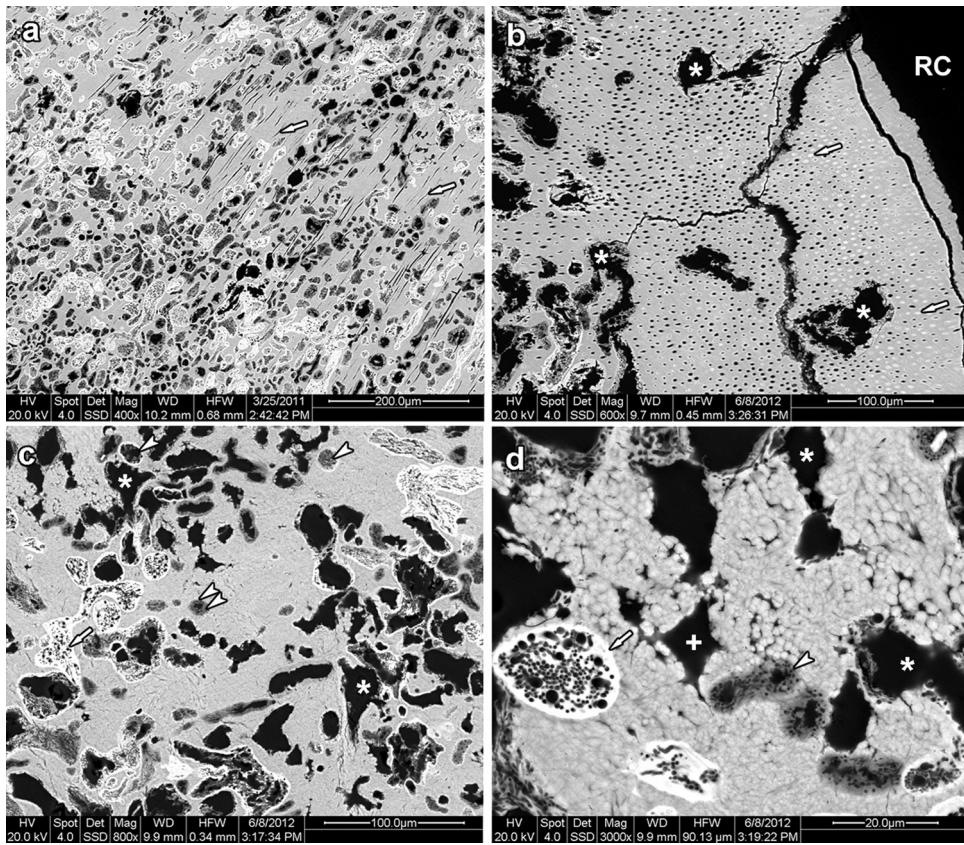


Fig. 4. BSE-SEM images of polished surface of a mesiodistally oriented section of the M^3 (find number Ro76, 402); occlusal direction to the top of the images. (a) Coronal dentin affected by diagenetic degradation. Arrows: dentinal tubules. (b) Dentin bordering on the root canal (RC); note numerous areas of tissue destruction (asterisks), and sclerosis of some of the dentinal tubules (arrows), especially in the dentin close to the root canal. (c) Diagenetic degradation of the tumor tissue; note numerous areas of tissue destruction (asterisks), dark areas, indicative of mineral dissolution, around foci of bacterial invasion, sometimes with (single arrowheads) sometimes without (double arrowhead) hypermineralized rims, and more extended hypermineralized areas (arrow). (d) Higher magnification of tumor mass with areas of tissue destruction (asterisks) and mineral dissolution (arrowhead) as well as hypermineralized areas (arrow) around foci of bacterial invasion. Cross: possible cementocyte lacuna.

potential, the expanding tumor can cause root resorption, obliteration of the periodontal ligament space and displacement of adjacent teeth, as well as resorption of the surrounding bone and facial bulging (Brannon et al., 2002; Van der Waal, 2005; Jundt and Reichart, 2008; Monti et al., 2013).

Histologically, human cementoblastomas consist of sheets or irregular trabeculae of a dense, acellular or paucicellular cementum-like tissue and numerous basophilic reversal lines in the more mature parts, interspersed with a fibrous vascular stroma (Abrams et al., 1974; Kramer et al., 1992; Ulmansky et al., 1994; Brannon et al., 2002; Van der Waal, 2005; Jundt and Reichart, 2008; Huber and Folk, 2009). The periphery of the tumor typically exhibits radially oriented trabeculae of unmineralized matrix (Abrams et al., 1974; Kramer et al., 1992; Ulmansky et al., 1994; Brannon et al., 2002; Van der Waal, 2005; Jundt and Reichart, 2008).

Occurrence of cementoblastomas has also been reported from animals, with most of the cases observed in horses (Gorlin et al., 1963; Miles and Grigson, 1990; Kreutzer et al., 2007; Levine et al., 2008). Kreutzer et al. (2007) described three cases from three individuals, the affected teeth being a left maxillary first incisor, a left mandibular first incisor and a left maxillary second premolar. On clinical examination, moderate malocclusion and pain were recorded in the horses. In all three cases, the cementoblastoma was attached to the root of the affected tooth, and the tumorous masses consisted of a cementum-like material containing what the authors described as abnormal lacunae (Kreutzer et al., 2007). Levine et al. (2008) reported the occurrence of multiple cementoblastomas affecting the maxillary incisors as well as the mandibular canines and third incisors of a horse. Histological analysis of the

extracted incisors revealed the presence of atypical cementum with frequent reversal lines and cementoblasts embedded in lacunae. The tumorous masses abutted and had focally invaded the roots of the affected incisors (Levine et al., 2008).

Only few cases of cementoblastomas in animals other than the horse have been reported. Among bovids, a case of cementoblastoma affecting a mandibular incisor of an ox was described by Woods (1907). The case is also discussed and depicted by Miles and Grigson (1990, p. 598 and figure 25.32). The cementum, which had completely overgrown the tooth, showed a trabecular structure with marrow spaces. Andrews (1973) reported the case of a steer, in which a mass of cementum was attached to the crown of a right mandibular second molar that was not yet fully erupted. The mass, which also exhibited a trabecular structure, was apparently continuous with the coronal cementum. The fact that in this case the cementoblastoma was attached to the tooth crown shows that the definition of this tumor as a neoplasm consisting of “cementum-like tissue in connection with the root of a tooth” (Van der Waal, 2005) only holds for humans and other mammalian taxa that lack coronal cementum, including deer (Eidmann, 1939; Kierdorf and Kierdorf, 1992). In contrast, in species possessing coronal cementum, a cementoblastoma can, in principle, be attached to the root or the crown of a tooth.

To the best of our knowledge, thus far only a single case of cementoblastoma in a wild ruminant has been reported in the scientific literature. The case concerned a dama gazelle (*Nanger dama*, formerly *Gazella dama*) that exhibited a tumorous mass in the left maxillary region (Martin et al., 1985). While it is stated that the mass was located periapically, the report provides no information

to which tooth it was attached. The tumorous mass consisted of tangled trabecular arrays of cementum exhibiting irregular growth lines along the periphery, with some of the trabeculae described as being acellular (Martin et al., 1985).

In the case described here, the bulk of the mineralized mass attached to the tooth contained only few cementocyte lacunae, indicating that this cementum-like tissue had been of a paucicellular nature. However, in the portion of the tumor bordering on the occlusal surface, this material was interspersed with irregularly shaped areas exhibiting numerous cementocyte lacunae, i.e., with an originally cellular cementum. In red deer, a pad of cellular cementum is regularly laid down in the interradicular area of the cheek teeth (Eidmann, 1939). Formation of this cementum pad follows a seasonal pattern that enables the distinction between cell-rich "summer zones" and less cell-rich "winter zones" (Pike-Tay, 1991). It could be assumed that the development of the cementoblastoma had caused a disruption of the growth process of the interradicular cementum pad in the red deer from Rochedane, resulting in the formation of patches and streaks of cellular cementum intermingled with the paucicellular tumor tissue instead of regular layers of cellular cementum. In this case, the cellular cementum would not have been part of the tumor. Alternatively it might be assumed that the early-formed portion of the cementoblastoma had consisted of a mixture of cell-rich and paucicellular cementum/cermentum-like tissue. In this case it could be speculated that formation of the cementoblastoma in the red deer was caused by the neoplastic transformation of cells normally involved in the deposition of the interradicular cementum pad.

Judging from the normal shape of the occlusal surface (for a heavily worn M³ of *Cervus elaphus*), the growth of the cementoblastoma in the red deer from Rochedane had apparently not led to (significant) malocclusion. Whether the enlargement of the tumor had an effect on the wear rate of the molar by lifting it from the alveolus, must remain open to speculation.

Regarding differential diagnosis, the distinction between cementoblastoma and osseous tumors (osteoblastoma, osteoid osteoma, osteosarcoma) is problematic based on histological examination only (Kramer et al., 1992; Ullmansky et al., 1994; Brannon et al., 2002; Bilodeau et al., 2010). It has been stated that a diagnosis cannot be made on a biopsy alone (Van der Waal, 2005). In human dental pathology, the distinctive feature for the diagnosis of cementoblastoma is the relationship of the tumor with the root of a tooth (Kramer et al., 1992; Ullmansky et al., 1994; Brannon et al., 2002; Van der Waal, 2005). This relationship was also given in the specimen presented here.

Hypercementosis, a non-neoplastic condition, must also be considered in the differential diagnosis (Napier Souza et al., 2004; Kreutzer et al., 2007). Hypercementosis is characterized by the deposition of excessive cementum in continuation with the normal root cementum. Contrary to the cementoblastoma, no or only few and small soft tissue spaces are present in the calcified mass of cementum in the case of hypercementosis, and the cementum is more regularly mineralized (Euler and Meyer, 1927; Napier Souza et al., 2004; Kreutzer et al., 2007). In humans, hypercementosis often occurs as an age-related phenomenon, but can also develop in response to overloading of teeth or infection (Pindborg, 1970; Napier Souza et al., 2004). A hypertrophic growth of the interradicular cementum pad in the red deer molar from Rochedane can be excluded, since in this case the complete mass would be expected to consist of a dense cellular cementum with a layered appearance.

In conclusion, the case described here is the first report of a cementoblastoma, a benign odontogenic tumor of ectomesenchymal origin, in a (prehistoric or modern) deer. We are also not aware of any previously published case of this type of tumor in the archeozoological literature. As has been stated recently (Binois et al., 2014), rare and unique pathological findings in prehistoric

animals should always be reported, as they are of interest both for the archeozoologist who may encounter similar cases in his material as well as for the pathologist studying corresponding cases in recent animals.

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