

**Title:** An Unusual Exostotic Lesion of the Maxillary Sinus from Roman Lincoln.

**Authors:** Ross Kendall<sup>1</sup>, Ellen J. Kendall<sup>1\*</sup>, Iain Macleod<sup>2</sup>, Rebecca Gowland<sup>1</sup> and Julia Beaumont<sup>3</sup>

<sup>1</sup>Department of Archaeology, Durham University, Durham, DH1 3LE, UK

<sup>2</sup>School of Dental Sciences, Newcastle University, Newcastle upon Tyne, NE2 4BW, UK

<sup>3</sup>Archaeological and Environmental Sciences, University of Bradford, Bradford, West Yorkshire, BD7 1DP, UK

\*Correspondence to Ellen Kendall, Department of Archaeology, Durham University, Durham, DH1 3LE, UK. e.j.kendall@durham.ac.uk.

## **Abstract**

This report provides a differential diagnosis of an exostotic bony lesion within the left maxillary sinus of a Romano-British (3<sup>rd</sup> to 4<sup>th</sup> century AD) adult male from Newport, Lincoln. Macroscopic, radiographic, and cone beam computed tomography (CBCT) analyses suggest that the lesion is likely of odontogenic origin. The overall size of the lesion and areas of sclerosis and radiolucency, together with its hypothesised odontogenic origin, suggest that the lesion represents a chronic exostotic osteomyelitic reaction to the presence of odontogenic bacteria. While modern case studies of odontogenic maxillary sinus osteomyelitis are noteworthy, published cases of this condition are extremely rare in an archaeological context and may be underreported due to the enclosed nature of the sinuses. Such infections may have serious implications for individual and population health, and non-destructive investigation should be considered in cases where significant maxillary caries are present.

*Keywords:* Paranasal sinuses; osteomyelitis; odontogenic; sinusitis; Romano-British.

## **1. Introduction**

Diseases of the paranasal sinuses occur frequently in modern day populations, affecting both the young and old alike (Farman and Nortjé, 2002; Bell *et al.*, 2011). Chronic or recurrent low-grade infections of the maxillary sinus are aetiologically diverse, with triggers ranging from environmental irritants to allergy and respiratory tract infections (Spector, 1992; Corren and Rachelefsky, 1994; Osur, 2002). Around 10% of clinically reported cases have odontogenic aetiologies, such as periapical infections (Brook, 2006; Mehra and Jeong, 2008). The anatomical proximity of the maxillary dentition to the sinus is an important factor in the development of odontogenic sinusitis (Ugincius *et al.*, 2006).

Inflammatory osseous responses to pathogenic processes, including those affecting the maxillary sinuses, have been observed in the skeletal remains of many past populations (Boocock *et al.*, 1995; Merrett and Pfeiffer, 2000; Roberts, 2007). The often-chronic nature of mucosal infection increases the likelihood of changes to bone occurring within the sinus cavities (Ortner and Putschar, 1981; Roberts, 2007). However, it is generally assumed that the actual frequency of

past sinusitis is underestimated due to the enclosed nature of the sinuses when undamaged (Boocock *et al.*, 1995). An exostotic lesion of the type observed in this case is very uncommon in modern clinical literature (Tovi *et al.*, 1992; Moretti *et al.*, 2004). It is rarer still in archaeological literature, but, as with sinusitis, the potential for underreporting must be considered. We present this case to highlight the potential for maxillary caries to produce significant extra-oral lesions and to strongly urge future studies to consider non-destructive screening of individuals with extensive caries.

## 2. Materials and Methods

Excavations conducted in 2010 by Archaeological Project Services at Newport, Lincoln revealed a small assemblage of skeletal remains (n=11), with six individuals being securely dated to the early second to middle fourth centuries AD (Kendall, 2011). Within a few decades of the Roman conquest (by around AD 61), Lincoln had become established as a large military centre for the Second and Ninth Legions (Jones, 2003). Following the departure of the army in *c.* AD 78, Lincoln was converted into the military veteran settlement (*colonia*) of *Lindum* (Jones, 2003; Peachey, 2011). *Lindum* was later to develop into an important regional centre, becoming the capital of the province *Britannia Secunda* by the end of the third century (Mann, 1998).

The site was located immediately west of Ermine Street, a major Roman thoroughfare linking London (*Londinium*) and York (*Eboracum*), some 650m to the north of the *colonia*'s north gate in the Newport suburb. In line with Roman burial law (Toynbee, 1971), this site likely represents one of a number of extramural burial grounds, and reflects a northward spread of settlement along Ermine Street beyond the *colonia* walls (Peachey, 2011). The concentration, stratigraphic sequence, and general north-south alignment of graves suggests that the cemetery may have evolved from an *ad hoc* burial site in the middle 2<sup>nd</sup> century to a small, organised roadside cemetery serving the 3<sup>rd</sup> and 4<sup>th</sup> century *colonia* (Kendall, 2011). Skeleton 611 (hereafter SK611) was interred supine on a northwest-southeast alignment in a coffin with a serrated iron object believed to be a saw.

Osteological analysis was performed using standard methodology (following Buikstra and Ubelaker, 1994, and Brickley and McKinley, 2004). Age-at-death and sex were estimated based on age-related degeneration of the auricular surface and pubic symphysis (Lovejoy *et al.*, 1985; Brooks and Suchey, 1990) and morphological features of the pelvis and skull (Phenice, 1969) respectively. Pathology was identified and described using standard macroscopic methods, with orbital lesions being recorded by the method of Stuart Macadam (1991). An exostotic lesion of the maxillary sinus, which forms the focus of this paper, was examined macroscopically and radiographically (GE Medical MPX 10; General Electric Wauwatosa, USA set to 70kVp at 0.500mAs, with a Carestream Point-of-Care digital CR reader; Rochester, USA) in the Department of Archaeology at Durham University, and CBCT (NewTom VGi; QR Verona, Italy) was carried out at the School of Dental Sciences, Newcastle University.

Destructive histological analysis of the lesion was not performed due to considerations of fragility and risk-benefit balance. Histological evidence may add confidence to a differential diagnosis where microscopic features are pathognomonic, but it is rarely possible to reach a definitive diagnosis based on histopathology alone, due to the similarity of histological features between distinct diagnostic entities (Eversole *et al.*, 2008; Silva and Wasterlain, 2010). With this

constraint in mind, a differential diagnosis was attempted based upon macroscopic, radiographic, and CBCT evidence.

### 3. Results

SK611 was estimated to be a male aged between 25 and 35 years at the time of death. His skeletal remains were largely complete, with good bone preservation (Figure 1). However, SK611's full remains were not excavated due to the diagonal truncation of his grave by the limit of excavation. Overall skeletal pathology for SK611 was low. Observed postcranial pathologies were limited to bilateral sacralisation and osteophytosis affecting the lower thoracic and lumbar vertebrae. Cranial and dental pathologies were observed in greater abundance. SK611 exhibited grade 2 bilateral cribra orbitalia and suffered from poor dental health. His mandibular teeth were slightly misaligned, with evident overcrowding. Table 1 presents a skeletal dental presence/absence chart for SK611. Two maxillary and eight mandibular teeth had enamel hypoplastic defects. All extant teeth displayed flecked to medium concretions of calculus. Large carious lesions were present on eight teeth (2, 5, 13, 15, 16, 18, 19, and 20), four of which were associated with periapical lesions and drainage sinuses (15, 16, 18, and 19). Six periapical lesions were observed in total (tooth numbers 3, 4, 15, 16, 18, and 19), all with accompanying drainage sinuses. All periapical lesions were associated with either carious or unobservable (lost antemortem or postmortem) teeth.

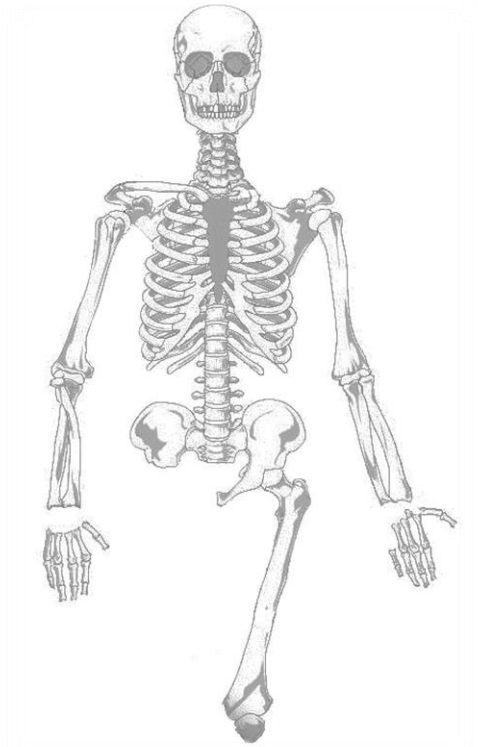


Figure 1 – Elements present in skeleton 611.

|       |    |    |    |    |    |    |    |    |  |    |    |    |    |   |    |    |      |
|-------|----|----|----|----|----|----|----|----|--|----|----|----|----|---|----|----|------|
| Right |    |    |    |    |    |    |    |    |  |    |    |    |    |   |    |    | Left |
|       | 1  | 2  | X  | \  | R  | \  | R  | 8  |  | \  | 10 | 11 | 12 | R | \  | 15 | 16   |
|       | 32 | 31 | 30 | 29 | 28 | 27 | 26 | 25 |  | 24 | 23 | 22 | 21 | R | 19 | 18 | 17   |
| Right |    |    |    |    |    |    |    |    |  |    |    |    |    |   |    |    | Left |

Table 1. Skeletal dental presence/absence chart for SK611, after Buikstra and Ubelaker (1994). Top line = maxillary teeth; bottom line = mandibular teeth. Key: Number = tooth present; X = lost antemortem; \ = lost postmortem; R = root only.

Pathological bone changes were observed in the region of the left maxillary molars. The zygomatic process of the left maxilla exhibited extensive porosity with remodeling deposits of woven and transitional new bone located inferior to the infra-orbital foramen and immediately superior to teeth 15 and 16, (Figure 2). This area of remodeling was concave, producing a notable asymmetry between the right and left sides of the face. A lingual drainage sinus, measuring approximately 30mm in diameter, was recorded in association with tooth 16 (Figure 3).

Directly superior to tooth 16, an amorphous, coral-like, exostotic bony lesion, measuring a maximum of 20mm in breadth and 30mm in height, was observed within the maxillary sinus (Figure 4) via post-depositional damage to the orbital plate of the maxilla. This mass, composed externally of compact lamellar bone, originated at the roots of the carious tooth 16 (Figure 3), and appears to have subtly elevated the orbital plate (Figure 4). Radiographic analysis showed sclerotic thickening of the lateral and anterior walls of the left sinus (Figure 5). CBCT of the lesion revealed peripheral sclerosis, and internal sclerotic and radiolucent areas, while also clearly showing the location of the maxillary molar root apices within the sinus cavity (Figure 6). Endoscopy of the right maxillary sinus revealed no pathological abnormality.



Figure 2 – Anterior view of SK611 skull with asymmetrical concave contour of maxilla (arrowed), and inset showing detail of contour.

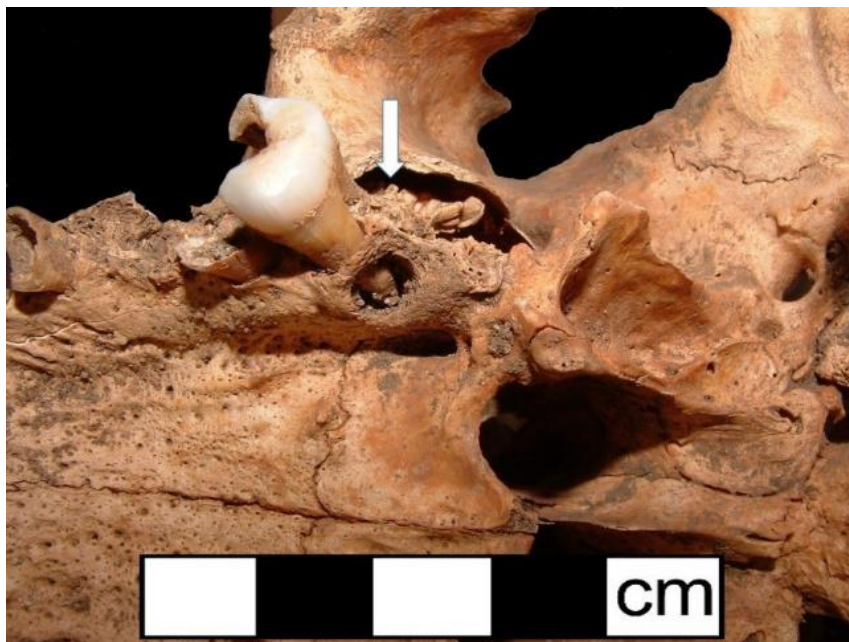


Figure 3 – Inferior view of left maxillary 3<sup>rd</sup> molar. Arrow indicates origin site of maxillary sinus lesion and close association with the molar roots. Note the drainage sinus exposing the lingual molar root.



Figure 4 – Irregular exostotic lesion in the left maxillary sinus of SK611.



Figure 5 - Radiograph with sclerotic thickening of the lateral and anterior walls of the left maxillary sinus.

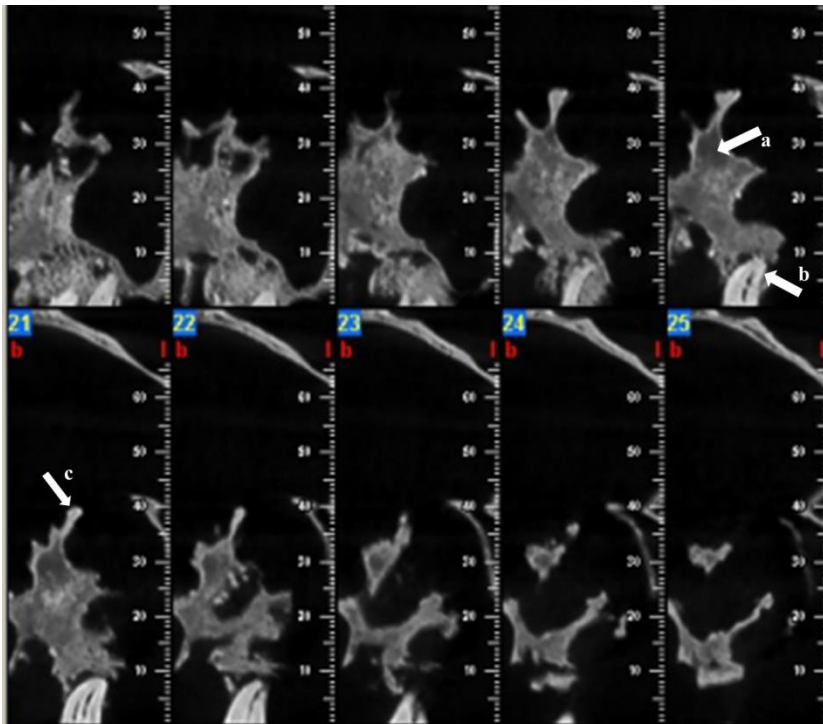


Figure 6 – CBCT scan of maxillary sinus lesion. Note areas of radiolucency (a) and sclerosis (c), and the intimate association between maxillary molar root apices (b) and the base of the lesion.

#### 4. Discussion

Numerous conditions may produce exostotic changes in bone, although it is rare for many of them to manifest within the maxillary sinus. Lesions affecting the paranasal sinuses may occur secondary to other conditions, such as osteopetrosis or Paget's disease (Seow, 1991; Labajian *et al.*, 2013); they may be formed as a response to bacterial infection (e.g. osteomyelitis) (Yoshino *et al.*, 2011), or associated with either malignant (e.g., osteosarcoma) or benign (e.g. osteoma, osteoblastoma, ossifying fibroma) neoplasms (Fu and Perzin, 1976; Jaswal *et al.*, 2007; Ortiz and Lin, 2012).

Osteopetrosis is a congenital condition affecting osteoclastic function, and which varies in the severity of phenotypic expression (Barry and Ryan, 2003; Stark and Savarirayan, 2009). Individuals affected by severe osteopetrosis often do not survive beyond infancy without intensive treatment (Stark and Savarirayan, 2009). Biological age and lack of gross morphological pathological markers (e.g., flaring long bone metaphyses and vertebral body sclerosis) rule out a diagnosis of severe osteopetrosis for SK611. Indicators of adult onset osteopetrosis, such as scoliosis and thickened long bone cortices (Barry and Ryan, 2003), are also absent.

Paget's disease of bone also involves abnormal osteoclastic function and affects one to two percent of adults over 55 years of age (Ralston *et al.*, 2008). Bone changes resulting from Paget's disease include cranial vault thickening, long bone bowing, and pathological fractures (Ralston *et al.*, 2008). Although paranasal sinus tumors can occur secondary to Paget's disease (Labajian *et*

*al.*, 2013), SK611's age and absence of commonly observed gross primary skeletal sequelae do not support a diagnosis of Paget's disease.

Given the extensive evidence for carious and periapical lesions and the close anatomical proximity between the posterior maxillary teeth and sinus lesion (Figure 5), infection of the paranasal sinuses was subsequently considered. The propinquity of maxillary tooth roots and sinuses is known to foster development of odontogenic maxillary sinusitis through compromise of the mucosa (Ugincius *et al.*, 2006). Such compromise may result from periapical infection, trauma, or following surgical intervention (Mehra and Jeong, 2008). For SK611, it is likely that periapical infection lead to bacterial transfer to the maxillary sinus. Roberts (2007:797) has argued that in dry bone, "while dental disease may lead to sinusitis, unless a direct connection is seen to the sinus from the dentition, then hypothesizing a dental origin for the sinusitis is not possible". SK611 presents an observable connection between the base of the exostotic lesion and the apices of the second maxillary molar buccal roots (Figures 3 and 5). The disease process may have been initiated by a carious lesion, followed by an infection of the pulp cavity spreading to the root apex, adjacent to the floor of the maxillary sinus. The drainage sinus clearly visible in Figure 3 suggests the subsequent formation of a dento-alveolar abscess (Dias and Tayles, 1997). The introduction of odontogenic bacteria into the maxillary mucosa would have led to sinusitis. Finally, chronic osteomyelitis of the left maxilla and formation of the reactive bone lesion within the sinus followed, as the body attempted to sequester the infection. Changes in contour and concavity to the zygomatic process of the maxilla, together with the mixed woven and transitional appearance to the exterior bone directly anterior to the exostotic lesion, (Figure 2) suggest that it may have been the site of a previous and considerable drainage sinus. This sinus had closed and was in the process of remodeling at the time of death, further signifying that this was an infection of significant duration.

Cases of osteomyelitis of the maxilla are rare in both the clinical and archaeological literature. In clinical settings, osteomyelitis more commonly affects the mandible, seldom manifesting in the maxilla (Suma *et al.*, 2007). The reasons for this disparity are not fully established, but are believed to be due to the richness of blood supply and thinner cortical bone present in the maxilla (Baltensperger and Eyrich, 2009). Whether maxillary lesions were equally rare in the past, particularly in periods where oral hygiene appears to have been poorer, is less certain. As osteomyelitis is highly challenging to treat in modern cases, requiring a combination of antibiotic and surgical therapies (Singh *et al.*, 2010), it is difficult to estimate the impact or prevalence of such infections in the absence of modern treatments. Likewise, it should be assumed that the true frequency of sinus lesions in past populations is underreported in bioarchaeology. Archaeological human skulls are frequently damaged in the post-depositional environment, leading to incomplete skeletal representation in many cases (Roberts and Lewis, 2002). Alternately, an intact skull may preclude visual examination of enclosed sinuses (Roberts and Lewis, 2002). Routine population-level application of methodologies such as radiography or endoscopy would greatly improve the accuracy of bioarchaeological prevalence estimates, but may not be practical in environments such as cost-competitive commercial archaeology. In the present case it is certainly unlikely that the lesion would have been observed without the serendipitous breakage of the left orbit. The distinction between true infrequency in modern populations and infrequent *representation* in skeletal series must therefore be made, in keeping with the "osteological paradox" (Wood *et al.*, 1992).



Having established the likelihood of an infective odontogenic origin for the maxillary sinus lesion, both malignant and benign neoplasms were considered as potential secondary manifestations. Malignant neoplasms of bone which affect the paranasal sinuses are relatively rare in the present. For instance, although osteosarcoma accounts for 20% of all bone cancers currently (Ottaviani and Jaffe, 2010), it is responsible for less than 2% of tumours of the paranasal sinuses, and is rarer still in patients under the age of 30 years (Park *et al.*, 2003). Additionally, malignant neoplasms of the paranasal sinuses follow characteristic patterns: while metastatic disease more commonly results in destructive lesions of bone, where growth does occur it is nearly always rapid and invasive (Park *et al.*, 2002; Smith, 2010). As the lesion under discussion here is composed externally of lamellar bone, suggesting a chronic and constrained growth, we feel that metastatic disease is unlikely. Furthermore, the absence of diffuse or multifocal osteolytic or osteoblastic lesions does not support metastasis.

In contrast, benign, slow-growing neoplastic lesions such as osteomas may produce paranasal sinus lesions similar to that observed in SK611. While osteomas are the most commonly-observed benign lesions affecting the paranasal sinuses, in absolute terms sinus osteomas are very uncommon, with a modern incidence of 0.43% (Viswanatha, 2012). While the most common age of onset for osteoma, usually occurring in young individuals in their teens or twenties (Moretti *et al.*, 2004), is consistent with the maturity of the bone lesion and estimated age for SK611, we can definitively rule out osteoma based upon the size of his lesion, which exceeds the one to two centimetre diameter specified in diagnostic criteria (Jones *et al.*, 2006; Nielsen and Rosenberg, 2007). This larger size may instead indicate a giant osteoid osteoma, or osteoblastoma, another uncommon lesion which accounts for 1% of all primary tumours of bone, 10-15% of which may be found craniofacially (Nielsen and Rosenberg, 2007). Though the lesion seen in SK611 shares some of the radiographic characteristics of osteoblastoma, such as the presence of radiopaque and radiolucent features, and must be considered in a differential diagnosis, it lacks the well-defined, circumscribed, and rounded appearance common to osteoblastomas (Chatterji *et al.*, 1978; Ueno *et al.*, 1994; Jones *et al.*, 2006; Nielsen and Rosenberg, 2007). Cementoblastoma, a rare benign tumor which represents less than 1% of odontogenic lesions (Pogrel *et al.*, 2005), was also considered. This lesion develops most commonly in association with the molar or premolar tooth roots, with the affected tooth/teeth retaining pulp vitality (Souza *et al.*, 2013). Although extremely difficult to distinguish from osteoblastoma and focal sclerosing osteomyelitis radiographically and histologically (Pogrel *et al.*, 2005), cementoblastoma is distinctive in that it connects directly to the tooth root surface, often with the loss of root contour due to fusion with the tumor (Sankari and Ramakrishnan, 2011; Souza *et al.*, 2013). Macroscopic and CBCT observations demonstrate that SK611's affected molar retains its contour. Furthermore, it is unlikely that the molar retained pulp vitality in this instance.

Ossifying fibromas are rare, benign lesions representing a variety of pathognomonic ossifying or calcifying processes that most often affect the craniofacial bones of middle aged adults, with a tendency towards malignancy (Pogrel *et al.*, 2005; Eversole *et al.*, 2008). Although they may contain a varying quantity of calcified, bone-resembling tissue, like osteomas and osteoblastomas, ossifying fibromas are generally well-defined in radiolucency and are regularly organised (Fu and Perzin, 1976; Liu *et al.*, 2010; Pogrel *et al.*, 2005). A proliferative lesion within the maxillary sinus of a Late Neolithic adult has been recently described and proposed to be ossifying fibroma (Silva and Wasterlain, 2010), a rare case within the published

archaeological literature. The morphological and radiographic dissimilarities of SK611's lesion to this, or any of the potential benign or malignant neoplasms known to affect the maxilla or paranasal sinuses, make them an unlikely primary cause. However, we are aware that without histological analysis, neoplastic growth secondary to infection (ossifying fibroma or cementoblastoma being the most likely) cannot be definitively ruled out.

The potential impacts of this lesion for SK611 are significant and variable, ranging from physical impairment to social disability. We can conservatively suggest that in addition to his existing dental issues, he would have suffered from the common symptoms of maxillary osteomyelitis: purulent discharges from the nose and mouth resulting in severe halitosis, facial swelling, pain, intermittent fever, and loss of appetite (Barry and Ryan, 2003; Baltensperger and Eyrich, 2009). In addition, the fractured remnants of the left orbital plate of the maxilla appear slightly convex, which may suggest that the lesion and its associated soft tissue created some degree of proptosis. While this must remain a tentative suggestion, due to the poor preservation of the orbital plate, unilateral proptosis is a known complication of orbital cellulitis, which commonly stems from infection of the paranasal sinuses and carried a significant risk of death or blindness before the advent of antibiotics (Bergin and Wright, 1986). Although odontogenic infections are believed to be caused by pathogens of low virulence (Perdikaris et al., 2007), it is known that "osteomyelitis of the jaws is a serious and potentially lethal lesion because of its frequently-associated bacteremia and surely must have been responsible for many deaths in antiquity..." (Aufderheide and Rodriguez-Martin, 1998:409). The impact of this lesion upon the quality of life for SK611 may therefore have been considerable. Social stigma may have resulted from an abnormal eye appearance and foetid breath, while more physically detrimental effects would have consisted of potential visual impairment, pain, and risk of premature death.

## **5. Conclusions**

We have presented here a maxillary sinus exostotic lesion, which is highly unusual in the archaeological literature. A direct connection observed between the roots of a carious molar and the exostotic bone lesion strongly suggests an infective odontogenic origin, resulting in maxillary sinusitis and triggering the formation of a reactive bony mass within the sinus cavity. While a secondary neoplastic reaction (e.g., ossifying fibroma or cementoblastoma) cannot be ruled out without histological analysis, morphological and radiographic dissimilarity to clinical cases leads us to propose a differential diagnosis of chronic suppurating osteomyelitis of the maxillary sinus secondary to periapical infection. Maxillary sinus osteomyelitis of odontogenic origin is a rarely-reported condition, clinically or archaeologically, and would have represented a significant threat to the life and wellbeing of affected individuals in a pre-antibiotic era. This study demonstrates the importance of routine maxillary sinus investigation during palaeopathological analysis, particularly in cases evidencing poor maxillary dental health. In such cases, endoscopic or radiographic analysis of sinuses is strongly recommended.

## **Acknowledgements**

The authors thank Dr. Jo Buckberry (University of Bradford) and Julie Peacock (Durham University) for their help with radiography, and Jeff Veitch (Durham University) for assistance with photography. Thanks also to Prof. Charlotte Roberts (Durham University), Dr. Willard Fee Jr. (Stanford School of Medicine), and Drs. Sarah and Douglas Watt for their useful comments.

Thanks are also due to Gary Taylor, Mark Peachey, and Mark Dymond (Archaeological Project Services) for their assistance, and to CgMs Consulting Limited for excavation and post-excavation funding.

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