



Case study

A suspected case of intranasal inverted Schneiderian papilloma in an adult male from post-Medieval Holland



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ABSTRACT

During the routine assessment of skeletal material unearthed from Middenbeemster, a post-Medieval (AD 17–19th century) cemetery in Northern Holland, an adult male with an unidentified choanal lesion was discovered. The affected individual was analysed macroscopically and via computer tomography. Based on the phenotypic and radiographic characteristics of the lesion, and after a comprehensive review of clinical literature, it was determined that the lesion was likely caused by an inverted Schneiderian papilloma (ISP), a benign but locally aggressive endophytic neoplasm histopathologically characterized by the inversion of the epithelium into the *lamina propria* (Schneiderian membrane) of the respiratory nasal mucosa. This study presents a detailed description of the pathophysiology and aetiology of ISPs, using both bioarchaeological and biomedical frameworks. Several differential diagnoses are discussed, with emphasis on the reasons for their rejection as the primary pathogenic mechanism(s). To the best of the authors' knowledge, this research is the first reported case of ISP within palaeopathology, which highlights the need to consider ISPs whenever slow-growing sinonasal neoplasms are suspected, as well as in cases that exhibit focal rhinitis.

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1. Introduction

The pathogenesis of osteological alterations to the sinonasal tract is often difficult to ascertain osteoarchaeologically, given the susceptibility of the region to malignancies, benign neoplasms and inflammation, as well as the lack of soft tissue masses upon which histological and radiographic diagnoses can be made. To date, *Mycobacterium leprae* and *Mycobacterium lepromatosis*, treponemal diseases, chronic (rhino) sinusitis, and nasopharyngeal carcinomas have been the most extensively studied causes of resorptive and reparative lesions within the sinonasal tract and contiguous structures. However, there is a comparative lack of literature detailing the occurrence and severity of other sinonasal conditions, such as inverted Schneiderian papilloma (ISP), a benign but locally aggressive endophytic neoplasm histopathologically characterized by the inversion of the epithelium into the Schneiderian membrane of

the nasal respiratory mucosa (Eggers et al., 2007; Ringertz, 1938). Diagnosing ISPs within archaeological samples is a challenging endeavor, as a variety of other disease processes produce phenotypically analogous lesions, have similar (if not exact) distribution patterns, and may co-exist secondary to, or independent of, ISPs (Anari and Carrie, 2010; Thapa, 2010; Verma et al., 2011; Wang et al., 2012; Wassef et al., 2012).

The aim of this research is to provide a macroscopic and radiographic description of a suspected case of ISP within the nasal cavity of an adult male from the predominately mid-19th century site of Middenbeemster, The Netherlands. To the best of the authors' knowledge, this is the first reported case of ISP within palaeopathological literature, suggesting the under-reported and under-considered nature of sinonasal ISPs in bioarchaeology. A more extensive understanding of the osteological and radiographic manifestations of ISPs may therefore enable the construction of more accurate morbidity profiles, and shed light on the range of sinonasal pathologies present within archaeological populations.

2. Materials and methods

2.1. The site

The individual in this study was excavated from Middenbeemster cemetery, located in the Beemster municipality of Northern

Abbreviations: ISP, inverted Schneiderian papilloma; ESP, exophytic Schneiderian papilloma; ANP, angiectatic nasal polyp; NBF, new bone formation.

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Fig. 1. Topographic map of The Netherlands, with the location of the Beemster municipality marked in black (after Jan-Willem van Aalst, 2015; Licensed under the Creative Commons Attribution 4.0 International License, 2015).

Holland (Fig. 1). The cemetery was in use between AD ~1613 and 1866, but archival documents suggest the site was cleared of most individuals in AD 1829 (Falger et al., 2012). The majority of the >450 excavated individuals are therefore considered to have died between AD 1829 and 1866.

The Beemster region was established in the early 17th century, after the draining of Lake Beemster in AD 1612. Throughout the 17th and early 18th centuries, the mainly Protestant population prospered, its economy based primarily on the trade of dairy products and cattle (de Jong, 1998). By the mid-18th and 19th centuries, however, a series of floods, rye infections and rodent infestations decimated crops and pastures, and two epidemics of rinderpest killed between one-half to two-thirds of Beemster cattle (Aten et al., 2012; Bergman, 1967). Famine (Waters-Rist and Hoogland, 2014), in addition to endemic malaria and outbreaks of paramyxoviridae, scarlet fever, diphtheria, smallpox, tuberculosis, whooping cough, typhoid fever, and cholera (Aten et al., 2012; Bergman, 1967; Wintle, 2000) left much of the Beemster population nutritionally and metabolically stressed (Lemmers et al., 2013; Vesekla et al., 2012), and, presumably, immuno-compromised.

2.2. The skeleton

The skeleton is well-preserved; the cranium is intact and the post-cranium is >75% complete. The individual is estimated to be a male, ≥ 50 years old at the time of death. These estimates are predicated upon the accumulation of sexually dimorphic and age-related cranio-facial (Buikstra and Ubelaker, 1994; Meindl and Lovejoy, 1985; WEA, 1980) and pelvic traits (Brooks and Suchey, 1990; Buckberry and Chamberlain, 2002; Buikstra and Ubelaker, 1994; Phenice, 1969; WEA, 1980), as per disciplinary standards. Dental wear was not considered in the age estimate due to ante-mortem tooth loss and the presence of pipe-notches on the remaining dentition. Two outstanding pathological features are noted: unilateral osseous atresia of the hypoglossal canal, and the pars vascularis and

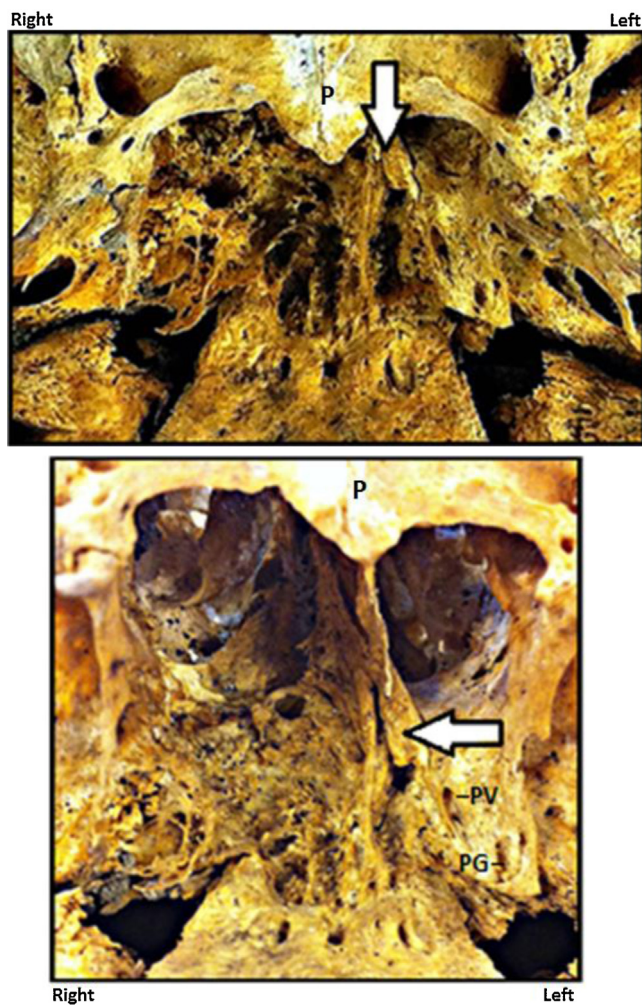


Fig. 2. Rear view of the localized erosive-reparative remodelling of the nasal choanae, and the hyperostosis of the nasal septum. Arrow: hyperostosis; P: palatine; PV: palatovaginal canal; PG: pterygoid canal (Carroll et al., 2015).

pars nervosa of the jugular foramen, and the presence of a localized erosive-reparative lesion within the nasal choanae. This research focuses primarily on the latter pathology.

2.3. Analytical methods

Radiographic assessment of the cranium was conducted at Leiden University Medical Center, using Computed Tomography (CT) and SPECTRA Diagnostic Imaging software (version 15.2.12.72; 2013).

Gross examination of the nasal vestibules, sinonasal ostia and basicranial foramina was assisted through the use of a Heine K180® fiber optic illuminated otoscope.

3. Nasal pathology

Examination of the right choana reveals focal osteolysis and erratic remodeling of the right half of the sphenoidal body, vomer, palatovaginal and pterygoid canals, perpendicular palatine, and medial pterygoid plate (Fig. 2). A circumscribed, ovate erosive-reparative lesion is visible on the right half of the sphenoidal body and vomer, anterior to the basiocciput (Figs. 2 and 3). Radiographically, the right greater wing of the sphenoid also exhibits demineralization and erosion. The lesions are multi-centric and their borders less clearly defined (Fig. 3). Apart from a small region

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