



Who Originally Described Pindborg Tumor?

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To the Editors,

The well-written and elegantly illustrated paper on calcifying epithelial odontogenic tumor (CEOT) by Sánchez-Romero et al. [1] aroused our interest and concern simultaneously. Here we describe how the disease concept of CEOT has changed and developed into the present one by a thorough review of its historical background.

The medical and dental literature usually cites the 1958 article by Pindborg [2] in Denmark as the first full documentation of CEOT. He summarized three cases, one of which was described in a Danish dental journal by Husted and Pindborg [3] 5 years earlier. They named it “adenoid type of ameloblastoma” because of its similarity to an early American case of adeno adamantoblastoma reported by Thoma and Goldman [4, 5] in 1946. At that time, Pindborg [2, 6] studied 24 cases of ameloblastoma and was fortunate enough to encounter three examples of CEOT during 5 years (1953–1958). In 1955, he initially introduced the concept of CEOT at the 11th Scandinavian Congress of Pathology and Bacteriology [6], and this transaction was followed by the first of many English-language reports on the topic [2]. In the same year (1958), Pindborg and Clausen [7] authored another important paper on a new classification of odontogenic tumors in which CEOT was separately categorized. Of interest is that 1958 was a notable year for CEOT because of the publication of two other papers, one by Langer [8, 9] in Germany and the other by Bradfield and Broadway [10] in England.

In his survey, Pindborg [2] referred to the previously published four cases of CEOT described by Thoma and Goldman [4, 5] in 1946, Ivy [11] in 1948, Wunderer [12] in 1953 and Stoopack [13] in 1957, respectively. Nevertheless, in a search of the extensive world literature, we were able to retrieve four references to CEOT that predated the above reports. The first of these was provided in 1932 by the German pathologist Heinz [14] under the term “odontogenes Psammocarcinom.” This German-language paper included one radiograph, two gross images and three photomicrographs of pericoronal CEOT. Until the late 1950s, the scientific literature traditionally adopted a general nomenclature “adamantinoma or ameloblastoma” for all epithelial odontogenic tumors. As early as 1934, Thoma [15], a pioneer of oral pathology, used the name “adamantinoma” to describe CEOT. This American case was included in the initial edition of his textbook “Oral Pathology” in 1941 [16] and retained in the 4th edition in 1954 [17].

In 1935, the Swedish pathologist McGregor [18] reported a series of adamantinoma in which a single example of CEOT was clearly described. He observed hyaline masses and calcified whorls within the tumor nests of polygonal epithelial cells and concluded that the lesion resembled craniopharyngioma. Hertz [19, 20] in the same institution was correct in excluding this Scandinavian case from the 1952 literature review of adamantinoma. Another earlier European paper on CEOT was a French contribution by Lattès [21] in 1937. He coined the designation “tumeur dysembryoplasique.” In the British literature, Kramer [22] in 1963 was likely the first to refer to CEOT.

In the United States, a somewhat different approach had been employed for CEOT classification under the influence of Thoma’s earlier works [4, 5, 15–17]. Although there is little microscopic similarity, CEOT and ameloblastoma had been grouped together until the early 1960s. A mostly American group of oral pathologists drew attention to peculiar calcifications rather than a unique epithelial morphology. In his textbook “The Management of Oral Disease”, Bernier [23] in 1955 illustrated CEOT as a type of ameloblastoma with a legend “partially calcified bodies in a nest of odontogenic

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epithelial cells which have undergone squamous metaplasia.” Despite the 1958 publication of Pindborg’s original report in an international peer-reviewed medical journal [2], the descriptive terminology “calcifying ameloblastoma [24] or ameloblastoma with enamel droplets [25]” still haunted the American literature thereafter. Likewise, a widely used atlas of tumor pathology published in 1960 by the Armed Forces Institute of Pathology mentioned Pindborg’s CEOT, but provided only a brief comment “a variant of ameloblastoma with calcified structures” [26]. Soon after the publication of the first American report with a title of CEOT in 1962 [27], an old set of terms was obsolete. It is interesting that the now famous eponym “Pindborg tumor” had its origin in the 2nd edition of “A Textbook of Oral Pathology” published in 1963, and Shafer [28], author in chief of this authoritative book, was an internationally known, influential oral pathologists in the United States. Also of note is that in CEOT, the term “amyloid” was initially used by Vickers et al. [29] in 1965. Vickers [27] is remembered as co-author of one of the earliest report of CEOT in the American literature.

In summary, our brief review provides a historical timeline that indicates how knowledge of CEOT pathology developed. Although Pindborg [2] published the best-known, first English-language paper on CEOT in 1958, Heinz [14] originally described Pindborg tumor in a German dental journal 26 years before that. Between early 1960 and the middle of 1970, the American oral pathologists did much to popularize Pindborg tumor.

Compliance with Ethical Standards

Conflict of interest The authors declare no conflict of interest.

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