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Original Article *Özgün Araştırma*

CLINICOPATHOLOGICAL ANALYSIS OF ODONTOGENIC KERATOCYSTS: 10 YEARS EXPERIENCE FROM A SINGLE CENTER

Odontojenik Keratokistlerin Klinikopatolojik Analizi: Tek Merkezden 10 Yıllık Deneyim

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ABSTRACT

Objective: The aim of the study is to update our understanding of the clinicopathological features of odontogenic keratocysts in a series of 43 cases from a single center.

Material and Methods: This retrospective study included 51 specimens from 43 patients diagnosed between 2010 and 2020. Microscopic findings, patients' age, gender, lesion location, and presence or absence of recurrence were noted.

Results: The study group included 22 men and 21 women with a mean age of 38.72 /year. In 37 patients, the lesion occurred in the mandible, while in 6 patients it was found in the maxilla. Three patients were associated with nevoid basal-cell carcinoma syndrome. Three patients had multiple odontogenic cysts. Satellite cysts were present on the cyst walls in 5 of the 43 cases. Epithelial islands were present in 4 of the cases. In 28 cases, moderate to severe inflammation was observed in the cyst wall. Among them, in 18 cases, classical odontogenic keratocyst features were lost in some areas in the cyst epithelium. Elongation of rete ridges and radicular cyst-like areas were observed in 12 cases. In only 41.66% of patients, the odontogenic keratocyst or keratocystic odontogenic tumor terms were given in the provisional diagnoses. Clinically, the most frequently confused lesions in the differential diagnosis were dentigerous cyst, radicular cyst, residual cyst, and ameloblastoma. No recurrence was observed in any of the patients.

Conclusion: Concomitant inflammation can cause changes in the epithelium and may hide the diagnostic features of odontogenic keratocysts. Therefore, careful examination is required for accurate diagnosis.

***Keywords**: Odontogenic cysts, pathology, oral, inflammation, basal cell nevus syndrome*

ÖZ

Amaç: Çalışmanın amacı, tek merkezden 43 olguluk bir seride odontojenik keratokistlerin klinikopatolojik özelliklerine dair anlayışımızı güncellemektir.

Gereç ve Yöntemler: Bu retrospektif çalışma 2010-2020 yılları arasında tanı almış 43 hastaya ait 51 spesmeni içermektedir. Mikroskopik bulgular, hasta yaşı, cinsiyeti, lezyonun yerleşim yeri ve nüks varlığı kaydedilmiştir.

Bulgular: Çalışma grubu 22 erkek, 21 kadın hastadan oluşmakta olup ortalama yaş 38.72 /yıl idi. Lezyon hastaların 37'sinde mandibulada, 6'sında maksillada yerleşim göstermekteydi. Üç hastada nevoid bazal-hücreli karsinom sendromu mevcuttu. Hastaların 3'ünde multipl odontojenik kist saptandı. Beş hastada kist duvarında satellit kistler mevcuttu. Epitelyal adalar 4 olguda görüldü. Hastaların 28'inde kist duvarında orta derecede veya şiddetli inflamasyon saptandı. Bunların 18'inde klasik odontojenik keratokist özellikleri kist epitelinin bazı alanlarında kaybolmuştu. Retelerde uzama ve radiküler kist-benzeri alanlar 12 olguda saptandı. Hastaların sadece %41.66'sında, odontojenik keratokist veya keratokistik odontojenik tümör terimleri klinik ön tanılar arasında yer almaktaydı. Klinik olarak, ayırıcı tanıda en sık karıştırıldığı lezyonlar dentijeröz kist, radiküler kist, rezidüel kist ve ameloblastom idi. Takipteki hastaların hiçbirinde nüks görülmedi.

Sonuç: Eşlik eden inflamasyon kist epitelinde değişikliklere yol açmakta ve odontojenik keratokistlerin tanısal özelliklerini gizleyebilmektedir. Bu nedenle, doğru tanı için dikkatli değerlendirme gerekmektedir.

***Anahtar Kelimeler**: Odontojenik kistler, patoloji, oral, inflamasyon, bazal hücreli nevüs sendromu*

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INTRODUCTION

Odontogenic keratocysts (OKCs) are benign lesions of odontogenic origin with unique histological and clinical features. They were first described by Philipsen in 1951 (1). OKCs have been the subject of many studies due to their relatively high recurrence rate and aggressive behavior. OKCs are the third most common cysts of the jaw (after radicular cyst and dentigerous cyst, respectively). They constitute 10-20% of all odontogenic cysts (2). They occur in a wide age range and their incidence exhibits bi-modal distribution. The peak incidence is in the second to third decades and a second smaller peak in 50-70 years (3). Some of them (approximately 5%) occur as part of nevoid basal-cell carcinoma syndrome (NBCCS) and these cases tend to be multiple and in younger patients (3). In the past two decades, several changes have been made to the naming and classification of OKCs. This is due to the developments in the genetics, clinical follow-up, and treatment protocols of these lesions.

Our study aims to update our understanding of the clinicopathological features of OKCs in a series of 43 cases.

MATERIALS AND METHODS

This retrospective study included 51 specimens from 43 patients diagnosed with odontogenic keratocyst or keratocystic odontogenic tumors from 2010 to 2020 at the Pathology Department of Eskişehir Osmangazi University Medical Faculty.

The ethics approval was obtained from the "Non-Drug Clinical Research Ethics Committee of Eskişehir Osmangazi University Medical Faculty" (date: 02.03.2021, decision number: 26).

Eight patients were operated on after incisional biopsy, so there were two specimens of these patients. Hematoxylin & Eosin-stained slides were re-evaluated by two pathologists for the confirmation of diagnosis, and the presence of inflammation, epithelial changes, and additional histological findings. Also, patients' age at diagnosis, gender, lesion location, and presence or absence of recurrence was noted. Continuous variables were categorized to facilitate data analysis and presentation. The Chi-square test (Fisher's exact test) was used to compare categorical variables. IBM SPSS base system (SPSS, Version 23.0, USA) was used for statistical analysis. $p<0.05$ was considered statistically significant.

RESULTS

The clinicopathological characteristics of cases are shown in Table 1**.** The study group included 22 (51.16%) men and 21 (48.83%) women with a mean age of 38.72 /year. The male to female ratio was 1.04 and was close to equal distribution. The patients' ages ranged from 8 to 73 years. The lesion occurred most commonly in the 3rd and 4th decades, followed by the 7th decade (Table 2). In 37 patients (86%), the lesion occurred in the mandible, while in 6 patients (14%) it was found in the maxilla. Three patients (6.98%) were associated with NBCCS. Two of them were men and the other was a woman. The average age of the syndromic patients was 35 years. Three patients (6.97%) had multiple odontogenic cysts. Multiple cysts were seen in two out of three syndromic and one out of 40 non-syndromic patients. Therefore, the rate of multiple odontogenic cysts was 66.6% for syndromic patients and 2.5% for non-syndromic ones. One syndromic patient had multiple keratocysts and the other had keratocyst as well as a radicular cyst.

Since 8 patients were operated on after incisional biopsy, histopathological evaluation of both materials was performed together in these patients. Satellite cysts were present on the cyst walls in 5 (11.6%) of the 43 cases and one of them occurred in a syndromic patient.

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Table 1: Clinicopathological characteristics of the study population

	n	%
Total	43	100
Gender		
Male	22	51.16
Female	21	48.83
Localization		
Upper jaw	6	13.95
Lower jaw	37	86.04
NBCCS		
Present	3	6.97
Absent	40	93.02
Multiple cysts		
Present	2	4.65
Absent	41	95.34
Inflammation		
Present	28	65.11
Absent	15	34.88
Loss of classic OKC histopathological features		
Present	27	62.79
Absent	16	37.20
Epithelial hyperplasia		
Present	19	44.18
Absent	24	55.81
Radicular cyst-like areas		
Present	12	27.90
Absent	21	48.83
Satellite cysts		
Present	5	11.62
Absent	38	88.37
Squamoid or basaloid epithelial islands		
Present	4	9.30
Absent	39	90.69
Recurrence	0	0
Provisional diagnosis		
Present	36	83.72
Absent	7	16.27
Diagnosis compatibility		
Compatible	15	41.66
Incompatible	21	58.33

NBCCS, Nevoid basal-cell carcinoma syndrome

Thus, the rate of satellite cysts was 33.3% in syndromic patients and 10% in nonsyndromic ones. Basaloid or squamoid epithelial islands were present in 4 (9.3%) of the cases. Patients with special findings are presented in Table 3. Microscopically, in 28 (65.11%) cases moderate to severe inflammation (acute and chronic) was observed in the cyst wall.

Among them, in 18 cases, classical OKC features were lost in some areas in the cyst epithelium, and in 9 cases, classical OKC features were only focally observed. Two of these cases were not diagnosed as OKC at the first pathological examination. The epithelium was thickened in 19 cases. Elongation of rete ridges and radicular cyst-like areas were observed in 12 cases. In all cases where epithelial changes were observed, there was moderate to severe inflammation (active and chronic) in the cyst epithelium or connective tissue ($p<0.001$). There was no relationship between the presence of inflammation and the presence of epithelial islands and daughter cysts ($p>0.05$). Examples of histopathological features of OKC cases are shown in Figure 1a-1f.

A total of 49 provisional diagnoses were given to 36 patients. Some patients had more than one provisional diagnosis. In 15 (41.66%) of 36 patients, the odontogenic keratocyst or keratocystic odontogenic tumor terms were given in the provisional diagnoses and these cases were considered to be compatible with the pathologic diagnosis. In 21 (58.33%) patients, these terms were not presented in the provisional diagnoses so these cases were considered incompatible. There was no significance between the compatibility of provisional diagnoses and the presence of inflammation and epithelial changes ($p>0.05$). The provisional diagnoses and their frequencies are listed in Table 4. Clinically, the most frequently confused lesions in the differential diagnosis were dentigerous cyst, radicular cyst, residual cyst, and ameloblastoma, respectively.

Seventeen of 43 patients, had no re-examination visit. For the remaining 26 patients, the follow-up period varied from 1 month to 36 months, with a mean of 22.8 months. No recurrence was observed in any of these patients.

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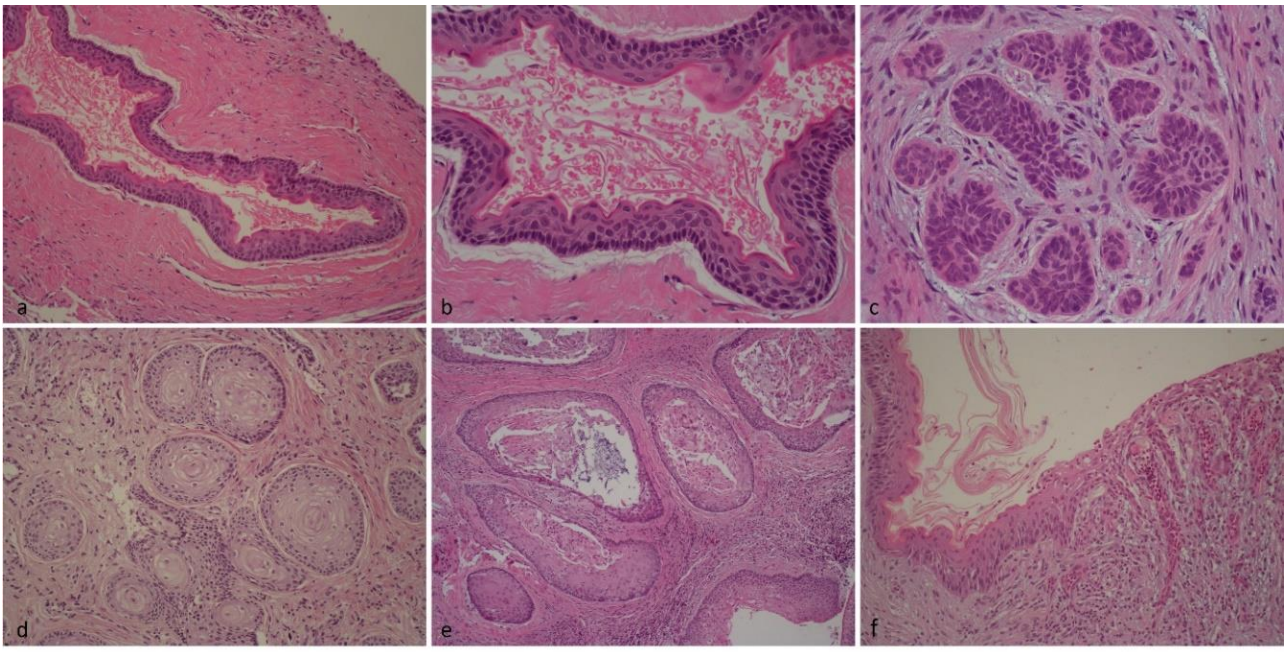

Figure 1a: The typical histopathologic appearance of an odontogenic keratocyst; uninflamed fibrous wall with thin, regular, folded epithelial lining (H&E, x200). b. Uniform stratified squamous epithelium, hyperchromatic, palisading basal epithelial cells, and overlying wavy, corrugated parakeratin layer (H&E, x400). c. Basaloid islands on the cyst wall (H&E, x400). d. Squamoid islands on the cyst wall (H&E, x100). e. Satellite cysts on the wall of an odontogenic keratocyst (H&E, x100). f. Cyst epithelium exhibits characteristic histological features at the left and loss of the classic odontogenic keratocyst features in an area of inflammation at the right (H&E, x200).

Table 2: Age distribution of patients

Age	Male	Female	Total	%
0-10	2	-	2	4.6
11-20	2	3	5	11.6
21-30	7	2	9	20.9
31-40	3	6	9	20.9
41-50	2	3	5	11.6
51-60	1	4	5	11.6
61-70	4	3	7	16.2
71-80	1	-	1	2.3
Total (n %)	22 (51.16)	21 (48.83)	43 (100)	100

Table 3: Patients with special findings

	Age	Gender	NBCCS	Multiple cyst	Satellite cysts	Epithelial islands
Patient 1	32	Female	+	OKC+OKC	+	-
Patient 2	23	Male	+	-	-	-
Patient 3	17	Female	-	-	+	-
Patient 4	33	Male	-	-	+	-
Patient 5	36	Male	-	-	+	+
Patient 6	46	Female	-	OKC+OKC	-	-
Patient 7	13	Female	-	-	-	+
Patient 8	32	Female	-	-	+	+
Patient 9	9	Male	-	-	-	+
Patient 10	50	Male	+	OKC+Radicular Cyst	-	-
Total			3	3	5	4

NBCCS, Nevoid basal-cell carcinoma syndrome

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Table 4: The list of the provisional clinical diagnoses

Provisional clinical diagnoses	Number of patients
Odontogenic keratocyst	15
Dentigerous cyst	11
Radicular cyst	7
Residual cyst	6
Ameloblastoma	4
Odontogenic cyst	2
Squamous odontogenic tumor	1
Glandular odontogenic cyst	1
Incisive duct cyst	1
Sinus retention cyst	1

DISCUSSION

OKC is an odontogenic cyst lined by thin (6-8 cell layers), regular lining of parakeratinized stratified squamous epithelium with little or no rete ridges (2-4). The parakeratin surface is typically wavy and corrugated. The basal-cell layer is well-defined and formed by cuboidal or columnar, often palisaded cells with hyperchromatic nuclei (3). Reverse polarization can be seen in the focal areas of the basal cells. Although basic histopathologic appearance includes these findings, some additional features may be present. OKCs can be seen over a wide patient age range (7 to 93 years) and show a bimodal distribution. While the highest incidence is seen in the 2nd and 3rd decades, the second small peak is between the ages of 50 to 70 years (2). In the present study, patients' ages ranged from 8 to 73 (mean 38.72) years and showed bimodal distribution. In many studies, the male to female ratio varies between 1.35 to 1.70 and exhibits slight male predilection (2, 5, 6). In the present study, this rate was 1.04 and was close to equal. OKCs are most frequently located in the mandible and approximately 80% of the cases arise from this area (3). Similar rates were found in the series of Brannon (65.4%), Myoung (76.5%), and Sung-II Yang et al. (69.5%) (5-7). In our series, this rate was found to be 86.04%, which was slightly higher than those in these series. It is well-known that the age of occurrence of OKCs in syndromic patients tends to be earlier and these cases tend to be multiple (3). Woolgar et al. showed that the mean age at the removal of the cyst was 40.4 (SD 19.2) years in the non-syndromic group, while it was 26.2 (SD 17.3) years in the syndromic group, and this difference was statistically significant ($p<0.001$) (8). In the present study, the mean age of non-syndromic patients was 39 (range, 8-73) years, while it was 35 (range, 23-50) years in the syndromic patients. OKCs are one of the most consistent features of NBCCS, occurring in 65-75% of cases (2). The mutation or inactivation of the PTCH1 gene activates the SHH signal pathway, resulting in abnormal cell proliferation of the OKC epithelium (3). Therefore, the frequency of satellite cysts, solid islands of epithelial proliferation, odontogenic rests within the capsule, and numbers of mitotic figures in the epithelium are statistically significantly higher in OKCs of syndromic patients (9). While the incidence of satellite cysts in syndromic cases varies between 36-78%, this rate varies between 2-18% in non-syndromic ones (8,10,11). In our series, this rate was 33.3% in syndromic patients and 10% in non-syndromic ones. Brannon found that epithelial islands and remnants were present in 29.8% of all cases (4). In the present study, epithelial islands (basaloid or squamoid) were present in 9.3% of all cases. Woolgar et al. reported that solid proliferation and odontogenic rests were present in 34% of syndromic cases, while 8% in non-syndromic ones (9). In the present study, basaloid or squamoid epithelial islands were present in 4 (9.3%) of the cases. While epithelial islands are expected to be seen more frequently in syndromic patients, these islands were not seen in syndromic patients in our study. Approximately 10% of patients have multiple OKCs, and half of them have NBCCS (3). In our series, 3 (%6.9) of 43 patients had multiple cysts, and two (%66) of them were syndromic. However, in a syndromic patient with multiple cysts, one of the cysts was diagnosed as OKC while the other was a radicular cyst.

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In cases accompanied by an intense inflammatory process, loss of specific features of OKC in the lining epithelium, epithelial thickening, the formation of rete ridges, and ulceration can be seen (2,3). Several publications report that 31.9 to 85% of cases are accompanied by varying degrees of inflammation in OKCs (4,5,9,11,12). Rodu et al. found that inflammation was present in 85 (76%) cases in their series and 75 (66%) of them had a transformation (nonkeratinized stratified squamous architecture) in the cyst epithelium (12). Inflammation without epithelial transformation was reported in 10 cases (8.9%) while no epithelial changes were observed in any case without inflammation. In the present study, 28 (65.11%) of 43 cases had moderate-to-severe inflammation in the cyst wall. In 27 (96.42%) of these 28 cases, classical OKC features were lost focally or widely in the cyst epithelium. Among them, in 18 cases, classical OKC features were lost in some areas in the cyst epithelium. Nineteen (44.18%) cases exhibited acanthosis and increasing epithelial thickness, 12 (27.9%) of them show rete ridges formation and radicular cyst-like areas. Typical epithelial areas could be observed only focally in 9 (20.9%) cases, and 2 of them were not diagnosed as OKC at the first pathological examination. There was no epithelial change in any of the cysts without inflammation.

Radiologically, OKCs do not have any reliable characteristic features of appearance to distinguish them from other radiolucent lesions of the jaw (6). It is usually in the form of a well-circumscribed radiolucent lesion with often a corticated margin. The lesions can be unilocular (sometimes scalloped) or multilocular (3). The multilocular variety can be misdiagnosed as ameloblastoma, occurrence in the periapical region of the vital standing teeth may give the appearance of a radicular cyst, and impeding the eruption of related teeth may result in a ‘dentigerous’ appearance radiologically (2).

Even if OKCs show certain typical histological features and have been confirmed in numerous publications (4,13,14), in the past, different names and histological criteria have been used to identify them. OKCs were described as a dermoid cyst and cholesteatoma in the earlier literature (2). In 1956, Philipsen first suggests the term odontogenic keratocyst for all odontogenic cysts that show keratinization of epithelium regardless of type (1). Histologic criteria were established by Shear in 1960 and Pindborg, Philipsen, and Henriksen, in 1962 (13,14). In 1992 WHO classification, it was categorized under the heading of “epithelial, developmental, and odontogenic cysts” and was used as synonymous with the primordial cyst (15). In 2005, the WHO consensus panel acknowledged some features of OKCs as evidence of neoplastic nature and renamed them as keratocystic odontogenic tumor (KOT) (16). Although its aggressive growth, the rare occurrence of its “solid” variant, recurrence after treatment, and most importantly mutations in the PTCH gene were considered justifications for reclassification, some authors suggest that these justifications are not sufficient to classify these lesions as neoplasms (17). In the 2017 classification, the keratocystic odontogenic tumor was moved back into the cyst category and it was called odontogenic keratocyst again (3,17). While neoplasms should not regress spontaneously, it has been documented that OKCs regress completely after decompression, and the epithelium of decompressed cysts resembles the oral mucosa rather than OKC. Loss of heterozygosity on chromosome 9q22.3 (where the PTCH1 gene has been mapped) is not specific for OKC and has also been shown in other non-neoplastic odontogenic cysts (3,18-20). In light of all these findings, it is believed that there is not enough evidence to support a neoplastic origin, and therefore OKC remains the most appropriate name for this lesion (3,17).

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There are many benign and malignant lesions in the differential diagnosis of OKCs, and it can be difficult to make a correct diagnosis clinically, radiologically, and even pathologically. Myoung et al. reported that concordance between the radiological impressions with the pathological diagnosis was 25.2% and the most common radiologic impressions were dentigerous cyst, OKC, primordial cyst, ameloblastoma, residual cyst, radicular cyst, and postoperative maxillary cyst respectively (5). In the present study, the most common clinical provisional diagnoses were OKC, dentigerous cyst, radicular cyst, residual cyst, and ameloblastoma, respectively. Although OKCs are often confused with benign lesions (mostly dentigerous cysts), they can also be confused with malignant lesions. Large size, multilocular radiological appearance, and clinical behavior can be misinterpreted clinically as ameloblastoma (6). To distinguish from other benign and malignant lesions, it is very important to sample the entire specimen for pathological examination and carefully evaluate all histopathological findings. Quite different rates of recurrence in OKCs have been reported in the literature, ranging from 0% to 62%. The majority of recurrences arise within the first 5 years following initial treatment. There are different treatment options for OKCs. With improvements in treatment modalities, recurrence rates have reduced dramatically compared to the past. The recurrence rate after enucleation with Carnoy solution is 8% and recurrence after resection is rare (in <2% of cases) (3). The presence of persistent satellite cysts or incomplete removal of cysts may be responsible for recurrences. In our series, recurrence was not observed in any patient. But, in our series, follow-up information was available in only 26 of the patients, and the short follow-up period (ranged from 1 to 36 months, with a mean of 22.8 months). The absence of recurrence in any of our patients may be related to short follow-up times and/or improvements in treatment modalities.

Looking at the history of OKCs, it is thought to have a neoplastic character but is currently classified in the cyst category. Recurrence was reduced with improvements in treatment modalities. Treatment effect and concomitant inflammation can cause changes in the epithelium and may hide the diagnostic features of OKC. Therefore, careful examination is required for accurate diagnosis, especially in cases where inflammation is accompanied.

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Ethics Committee Aproval: Non-Drug Clinical Research Ethics Committee of Eskişehir Osmangazi University Medical Faculty, date: 02.03.2021, decision number: 26.

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