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ORIGINAL ARTICLE

Clinical aspects of middle ear tympanokeratoma in dogs diagnosed through advanced imaging, otoendoscopy and histopathological evaluation

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Abstract

Background: Canine aural cholesteatoma (more appropriately named tympanokeratoma) is an epidermoid cyst whose aetiopathogenesis remains poorly recognised in veterinary medicine. There are a few reports published, possibly because it may be underdiagnosed.

Objectives: To characterise the clinical aspects of dogs with tympanokeratoma, to describe the otoendoscopic, advanced imaging and histopathological findings of tympanokeratoma and to report the best approach to diagnose canine auricular tympanokeratoma in a retrospective study.

Material and Methods: Of 890 dogs with suspected tympanokeratoma and otitis media, 100 animals underwent advanced imaging and otoendoscopy at radiology and dermatology reference centres in Brazil.

Results: Most affected dogs were male (71%) neutered (95%) with an average age of 6.8 years. Ninety-one of the 100 affected dogs were brachycephalic. Otitis externa (OE; 81%) was the main non-neurological manifestation observed. The main neurological clinical manifestations observed were: "head tilt" (66%), ataxia (31%) and nystagmus (25%). Advanced imaging findings could not propose a presumptive diagnosis of tympanokeratoma in 60 of 100 (60%) of the dogs. The absence of tympanic membrane and the presence of a dense pearly yellowish material resembling keratin in the tympanic bulla, after myringotomy, was the main otoendoscopic finding. The advanced imaging findings did not correlate with otoendoscopy and histopathological findings in more than half of the dogs.

Conclusions and Clinical Relevance: Tympanokeratoma should be suspected in brachycephalic dogs with OE and peripheral vestibular syndrome, and samples of keratinous material from the middle ear associated with histopathological results may be the best approach for the diagnosis.

KEYWORDS
computed tomography, histopathology, magnetic resonance, otoendoscopy

INTRODUCTION

Cholesteatoma or tympanokeratoma is a benign aural epidermoid cyst, generally located in the middle ear and capable of expansion and erosion of adjacent structures including the temporal bones. It is described as a rare disease in dogs, frequently associated with chronic otitis externa (OE) and otitis media (OM), and also may occur as a secondary manifestation to therapeutic, clinical or surgical approaches and ineffective OE treatments.[1]

Tympanokeratoma has been described in cocker spaniel, golden retriever, Doberman pinscher, Shetland sheepdog, English springer spaniel, Labrador retriever, English setter, pug, rat terrier, Chesapeake Bay retriever, Lhasa apso and mixed-breed.[1–3] However, according to Greci et al.,[4] no significant breed predilection has been reported in the literature and a higher incidence in male dogs has been found; however, this finding was not significant, probably owing to the small number of patients.[3,4] Authors point out that although tympanokeratoma

are more commonly found in middle-aged to elderly dogs, the ages reported in the scarce literature vary from 2 to 12 years.[3]

The actual occurrence of the disease is uncertain owing to the wide variety of clinical manifestations, as many animals are asymptomatic, and the diagnosis only occurs when there are external suppurative otitis and/or peripheral neurological manifestations.[5,6] Therefore, as the clinical manifestations in animals may be non-specific, some diagnostic exams need to be used in suspected cases.[7]

Imaging such as computed tomography (CT) and magnetic resonance imaging (MRI) are considered reliable methods to evaluate the middle ear and show lesions related to this anatomical region.[7] However, otoendoscopy has been used to diagnose suspected tympanokeratoma, as a consequence of the possibility of collecting characteristic material from the tympanic bulla for histopathological examination.[7,8]

The presence of keratin alone or keratin masses in the middle ear has been considered adequate to establish a diagnosis of aural tympanokeratoma; the detection of a lesion of pearly growth or yellowish or whitish material in the middle ear cavity that protrudes into the external auditory canal may suggest a possible presence of tympanokeratoma.[2,4,7,9,10]

However, tympanokeratoma is still an under-recognised condition and many practitioners may not be aware of the clinical features and findings leading to a diagnosis. Thus, the objectives of this retrospective study were to characterise the clinical aspects of dogs with tympanokeratoma, to describe the otoendoscopic findings, advanced imaging and histopathological findings and to report the best approach to diagnose canine auricular tympanokeratoma.

MATERIALS AND METHODS

Medical records of dogs from the routine caseload of a veterinary centre specialised in dermatology, in the state of Rio de Janeiro, Brazil, with OM, suspected cases of tympanokeratoma, were retrospectively reviewed seeking data on clinical, including otoendoscopic, radiological imaging and histological findings.

The clinical records of dogs treated from November 2017 to March 2022 were reviewed and data including breed, age, sex, neuter status and clinical manifestations presented on the day of the otoendoscopic examination were collected, as well as diagnostic methods performed.

The inclusion criteria used in the selection of the medical records were: (1) fully completed records; (2) confirmed diagnosis of tympanokeratoma by anatomopathological examination, considering the following points: (i) origin of the tissue collected by biopsy being from the middle ear through otoendoscopy; (ii) histopathological presence of keratinised stratified squamous epithelium and laminar keratin.

Cases with incomplete medical records, with non-cholesteatomatous OM and those with inconclusive histopathological findings were excluded.

RESULTS

Medical records of 890 dogs diagnosed with OM, from a veterinary centre specialised in dermatology, in the state of Rio de Janeiro, Brazil, from November 2017 to March 2022, were reviewed. Among these 890 records, 100 dogs (11.23%) met the inclusion criteria for the diagnosis of tympanokeratoma, while 790 records did not meet the inclusion criteria, based on incomplete records, non-cholesteatomatous OM (bacterial OM, neoplasia, among others) or inconclusive histopathological findings.

There was a great predominance of breed dogs (98%). Pugs corresponded to 51% (51 of 100) of the dogs, followed by French bulldogs corresponding to 35% (35 of 100). Other breeds included English bulldogs (three of 100), and one of 100 each of cane Corso, Staffordshire bull terrier, shih tzu, American cocker, dogo Argentino, Maltese, Dalmatian and poodle (Table 1).

Most affected dogs were aged 6 years (21%, 21 of 100), followed by 5–6 years (14%), 10 years (13%), 8 years (10%), 4 years (9%), 3 years (7%), 9 years (4%), 12 years (3%), 13 years (2%) and 11 and 14 years (each 1%).

TABLE 1 Clinical aspects of middle ear tympanokeratoma in dogs.

Variable	Category	Number of cases (100)
Breed	Pugs	51
	French bulldogs	35
	English bulldogs	3
	Mixed-breed	2
	Other breeds	9
Age (years)	10	13
	8	10
	6	21
	5	14
	4	9
	3	7
	Others	11
Sex	Female	29
	Male	71
Ears affected	Bilateral	29
	Unilateral (right only)	43
	Unilateral (left only)	28
Clinical manifestations	Otitis externa	81
	Head tilt	66
	Ataxia	31
	Horizontal nystagmus	25
	Facial paralysis	18
	Horner syndrome	3
	Dysacusis	60
	Otalgia	18
	Difficult to open mouth	14
	Prostration	3

Regarding the advanced imaging analysis, most dogs 68% (68 of 100) underwent computed tomography exams. Magnetic resonance imaging was performed on 24/% (24 of 100) dogs and only eight of 100 (8%) dogs did not undergo imaging before otoendoscopy. Finally, 9% (nine of 100) dogs underwent two imaging exams at different times.

All dog's CT and MRI presented findings in the tympanic bulla compatible with a diagnosis of OM. Only 29% (29 of 100) of CT or MRI exams showed lysis and expansion of the tympanic bulla, which is a strong suggestion of tympanokeratoma.

The remaining 71% (71 of 100) of the imaging exams indicated changes compatible with suspected OM or incipient tympanokeratoma, with only 11% (11 of 100) of exams in addition to the soft tissue content, presenting bone lysis. Most imaging exams (60%, 60 of 100) only indicated soft tissue content, proposing a diagnosis of OM.

Among the clinical manifestations identified, 81% (81 of 100) of the dogs had a history of OE. "Head tilt" was the main neurological manifestation of peripheral vestibular syndrome observed in 66% (66 of 100) of dogs, followed by ataxia (31%), horizontal nystagmus (25%), facial paralysis (18%) and Horner syndrome (3%). Apart from the neurological clinical signs, dysacusis (reported by the owners) (60%, 60/100), otalgia (18%) and difficulty or pain in opening the mouth and/or chewing (14%) were the most reported manifestations. There also were reports of prostration in 3% (three of 100) patients.

Bilateral disease was seen in 29% (29 of 100) of the dogs. The 71% (71/100) dogs remaining, presented a unilateral condition, comprising 43 of 71 (60.6%) on the right side and 28 of 71 (39.4%) on the left. The gross appearance of material sampled by otoendoscopy from the middle ear was dense pearly white to yellowish (with shades of gold), firm to pasty, with a characteristic rancid odour. The histopathological analysis from all dogs was characterised by amorphous thick lamellar keratin debris lined by cornified epithelium (Figure 1). Furthermore, otoendoscopy showed a single polypoid mass in the distal portion of the horizontal ear canal in 15% (15 of 100) dogs. Histopathological examination revealed a hyperplastic epidermis and a core of collagenous connective tissue with bone metaplasia, confirming a diagnosis of fibroepithelial polyps. These polyps were only observed in pugs (27.4%, 14 of 51) and French bulldogs (8.6%, three of 35). After removing the polyps, the same yellowish material was sampled from the tympanic cavity for histopathological evaluation, which confirmed tympanokeratoma.

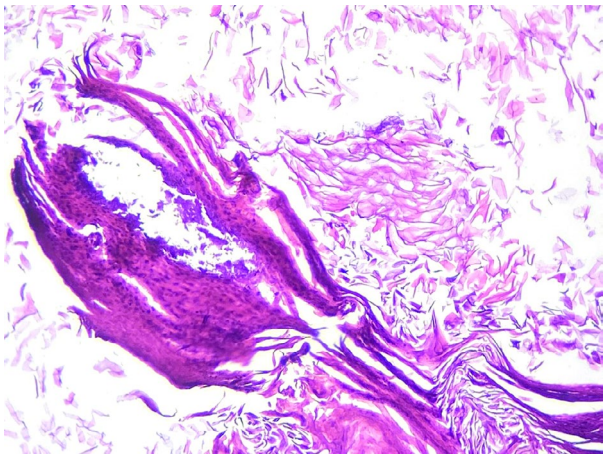

FIGURE 1 Tympanokeratoma (cholesteatoma). Cyst lined by keratinised stratified squamous epithelium and filled with keratin. Haematoxylin & eosin, ×200.

DISCUSSION

There are few reports of tympanokeratoma in the veterinary literature, which makes it difficult to make a comparison with the results reported here. Prior case series comprise 20 cases,[2] 13 cases,[11] 11 cases[12] and seven cases.[13] Authors of these case series reported little or no breed predilection, with Banco et al.[11] reporting mostly mixed-breed dogs. In a similar way to our study, Imai et al.[12] reported a high incidence of tympanokeratoma in brachycephalic breeds (63.6%). Of a total of 11 animals, five (45.4%) were French bulldogs and two animals (8.2%) were pugs. These data most closely align with the current study which included 51% pugs and 35% French bulldogs with tympanokeratoma.

Previous reports have documented that narrow nasopharynx and eustachian tube dysfunction may be predisposing factors for the development of primary acquired tympanokeratoma.[1,14] Furthermore, hypertrophic walls of the tympanic bulla and stenotic bone parts of the horizontal auditory canal may also predispose to tympanokeratoma.[14] This data may explain the great predominance of brachycephalic dogs in the present study. Yet, the aetiopathogenesis of tympanokeratoma in dogs, including in brachycephalic breeds, remains unknown. Furthermore, TECA- BO surgery has been suggested as a possible cause of tympanokeratoma development in dogs by other studies; however, none of the dogs in the present study had undergone surgery.

The average age of the affected dogs in this study was 6.8 years, ranging from 2 to 12 years. This age variation reported in this study is similar to that reported by several authors, such as Little et al.,[13] Banco et al.,[11] Hardie et al.[2] and Imai et al.[12] The wide age variation may suggest different aetiologies, as tympanokeratoma can be primary or acquired, and this precise definition is not yet established in veterinary medicine. Male dogs were over-represented in different studies,[2,11–13] which was similar to the data from our study, with males representing 71% of the dogs. There is no known explanation in the literature for this prevalence.

In the present study, OE was the main clinical finding reported associated with tympanokeratoma, which in their entirety had a chronic history, agreeing with data from other authors. Indeed, chronic OE may represent an important clinical sign leading to the investigation of tympanokeratoma.[2–4] Surprisingly, 19% of the animals did not show any sign of OE, presenting only with variable neurological clinical signs, which reinforces the theory of acquired primary tympanokeratoma and

should be considered by veterinary surgeons, especially in brachycephalic dogs represented here as most cases.

Regarding involvement and laterality, only 29% of the dogs presented bilateral disease. This observation differs from some publications, where the minority of cases had unilateral disease (ranging from 0% to 18%).[4,12] Moreover, in another study,[2] 80% of the dogs had bilateral disease. Few authors comment on the correlation between laterality and neurological clinical signs. In the present study, neurological manifestations were always unilateral (probably the most severe or the most inflamed), even in dogs with bilateral diagnoses. This fact reinforces the need for an advanced imaging exam so that future therapeutic approaches can be assessed on both affected sides. Only two studies[11] proposed a more conservative approach with otoendoscopy and middle ear flushes, which should be considered even more in bilateral cases, as surgical management may not be curative and can result in the order of 50% recurrence.[2,15]

The main neurological clinical manifestations in the present study were head tilt (66% of the cases) and ataxia (31% of cases), which also were the main neurological signs reported by Hardie et al.,[2] Greci et al.[4] and Imai et al.[12] Although facial nerve paralysis was more frequently reported by Hardie et al.,[2] Greci et al.,[4] Imai et al.,[12] in the present study it was observed only in 18% of patients. Interestingly, nystagmus was rarely reported in other studies[2,4,12] and was an important observation found here in 25% of patients. In another study, it was reported that >50% of the dogs were affected by head tilt, facial nerve paralysis and ataxia, without, however, highlighting the frequency of occurrence of each clinical sign.[1]

Another clinical manifestation observed by different authors was pain when chewing, yawning or opening the mouth during physical examination, which was observed in 14% (14 of 100) dogs and reported as an important symptomatology by Hardie et al.[2] (20% of dogs), Greci et al.[4] (50% of dogs) and Little et al.[13] (50% of dogs). Hardie et al.[2] also defined oral pain as a marker of animal life expectancy, reporting that dogs with this manifestation presented lethal complications on average 40 months after diagnosis. Interestingly, there are authors who do not report oral pain.

Dysacusis, or anacusis, was reported by the owners via a questionnaire in 60% of the dogs, which also has been reported by several authors.[2,4,12] These are important data, as most dogs evaluated in this study no longer had their tympanic membrane present. The hearing impairment reported may be conductive as a result of the occlusion from the material in the middle ear in addition to the absent tympanic membrane. Observations of dysacusis, or anacusis, in this report may be related to the fact that the number of cases with tympanokeratoma presented here exceeds most published reports,[12,14] thus providing evidence of rarer or less frequent clinical signs.

Computed tomography plays an important role in the evaluation of dogs with middle ear diseases,[3,16] as well as MRI.[17] The main observation in imaging diagnostics that may suggest a tympanokeratoma is the expansion of the tympanic bulla, associated or not with osteolysis, observed in this study in 29% of the dogs and only lysis in another 11%; however, lysis can be observed in chronic cases of OM and is therefore not pathognomonic of tympanokeratoma.[16] Tympanic bulla expansion was not the most frequent observation in the cases of tympanokeratoma presented here. This change has been reported in 53.8%[12] and 55% of cases[2] by other authors. Travetti et al.[3] highlighted tympanic bulla expansion as common in cases of tympanokeratoma. Tympanic bulla lysis without expansion was observed in all dogs from several studies[2,3,12,16] and found here in 11% (11 of 100) patients.

In all dogs in this study, with or without bullae expansion and with or without lysis, the tympanic bulla was filled with a dense opaque material, revealing itself to be nonvascularised and noncontrast enhancing. However, the initial stages of tympanokeratoma are indistinguishable from those of OM, a fact also pointed out by Imai et al.[12] Thus, the early stages of tympanokeratoma may remain unidentified by CT or MRI.

Considering the data from this retrospective study, in 60% of the dogs, OM was considered the most likely presumptive diagnosis based on the advanced imaging tests. However, when performing otoendoscopy followed by histopathological examination, the diagnosis of tympanokeratoma was confirmed, revealing that, in early cases, the combination of these two methods should be considered the gold standard for diagnosis. This fact also was pointed out by another author.[12]

In the present study, the main changes visualised by otoendoscopy were the absence of tympanic membrane, also reported by Imai et al.[12] and the presence of brownish pearly material filling the tympanic bulla, with firm or pasty characteristics on palpation, and with a strong rancid odour. Unfortunately, these findings cannot be compared with other studies, as there have been no descriptions of the otic changes assessed by otoendoscopy.

Furthermore, otoendoscopy showed a mass in the distal region of the horizontal ear canal in 15 dogs where the histopathological examination revealed a fibroepithelial polyp represented by hyperplastic epidermis and a core of collagenous connective tissue with bone metaplasia, which was only observed in pugs (14 of 51) and French bulldogs (three of 35). Interestingly, only one study[12] reports the presence of ear polyp, in seven of 11 (63%) dogs.

Histopathologically, the lesions in dogs appear to be different in character from typical aural polyps in cats. The presence of ciliated columnar epithelial cells is a prerequisite for the histopathological definition of feline polyps,[18] yet polyps and fibroepithelial polyps can be defined as small papular–nodular lesions in human and veterinary dermatology. In dogs, the association between these polyps and tympanokeratoma has not yet been fully elucidated. Another author[12] observed that the lesion may be related to

tympanokeratoma, which corresponds to what is proposed in the present study.

Aside from polyps, histological findings proved to be the main diagnostic test for confirming tympanokeratoma. Histologically, the tympanokeratoma was represented by laminar keratin enclosed in a cystic structure covered by keratinised stratified squamous epithelium, as described by several authors,[2,4,9,12,18] which provides the definitive diagnosis of the disease.

CONCLUSIONS

In this study, brachycephalic dogs, especially pugs and French bulldogs, were most commonly affected by tympanokeratoma. The main neurological clinical manifestations were: head tilt, ataxia, nystagmus and facial paralysis. The main non-neurological clinical manifestations were dysacusis and pain when opening the mouth. Presumptive diagnoses for tympanokeratoma made by advanced imaging methods, including CT scan, may be inconclusive in more than half of the patients and, based on our findings, they should not be considered definitive diagnostic methods. The main otoendoscopic observations were the absence of a tympanic membrane and the presence of dense pearly material inside the bulla. The presence of a polyp in the horizontal ear canal may be an indicator of tympanokeratoma. In conclusion, otoendoscopy with the collection of material from the middle ear associated with histopathological investigation, following advanced imaging, maybe the best approach for the diagnosis of canine tympanokeratoma.

AUTHOR CONTRIBUTIONS

Cristiane Bazaga Botelho: Conceptualization; writing – original draft; methodology; validation; visualization; writing – review and editing. **Rafaella Tortoriello:** Writing – original draft; visualization; writing – review and editing; conceptualization. **Sandra Nogueira Koch:** Writing – original draft; writing – review and editing; conceptualization. **Julio Israel Fernandes:** Writing – original draft; writing – review and editing; supervision; project administration.

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CONFLICT OF INTEREST STATEMENT

No conflicts of interest have been declared.

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REFERENCES

1. Risselada M. Diagnosis and management of cholesteatomas in dogs. Vet Clin North Am Small Anim Pract. 2016;46:623–34.
2. Hardie EM, Linder KE, Pease AP. Aural cholesteatoma in twenty dogs. Vet Surg. 2008;37:763–70.
3. Travetti O, Giudice C, Greci V, Lombardo R, Mortellaro CM, Di Giancamillo M. Computed tomography features of middle ear cholesteatoma in dogs. Vet Radiol Ultrasound. 2010;51:374–9.
4. Greci V, Travetti O, Di Giancamillo M, Lombardo R, Giudice C, Banco B, et al. Middle ear cholesteatoma in 11 dogs. Can Vet J. 2011;52:631–6.
5. Rivera CM, Fernandez LAM, Meza TRFC. Colesteatoma de oído. Anales de Radiología México, Ciudad de México. 2007;6:71–7.
6. Fossum TW. Cirurgia de ouvido. In: Fossum TW, editor. Cirurgia de pequenos animais. 4th ed. Rio de Janeiro: Elsevier; 2015. p. 341–5.
7. Newman AW, Estey CM, McDonough S, Cerda-Gonzalez S, Larsen M, Stokol T. Cholesteatoma and meningoencephatilis in a dog with **chronic** otitis externa. Vet Clin Pathol. 2014;44:157–63.
8. Miller WH, Griffin CE, Campbell KL, Miller WH, Griffin CE, Campbell KL. Muller and Kirk's small animal dermatology. 7th ed. St Louis, MO: Elsevier Mosby; 2013. p. 741–73.
9. Ferlito A, Devaney KO, Rinaldo A, Milroy CM, Wenig BM, Iurato S, et al. Clinicopathological consultation: ear cholesteatoma versus cholesterol granuloma. Ann Otol Rhinol Laryngol. 1997;106:79–85.
10. Sturges BK, Dickinson PJ, Kortz GD, Berry WL, Vernau KM, Wisner ER, et al. Clinical signs, magnetic resonance imaging features, and outcome after surgical and medical treatment of otogenic intracranial infection in 11 cats and 4 dogs. J Vet Intern Med. 2006;20:648–56.
11. Banco B, Grieco V, Di Giancamillo M, Greci V, Travetti O, Martino P, et al. Canine aural cholesteatoma: a histological and immunohistochemical study. Vet J. 2014;200:440–5.
12. Imai A, Kondo H, Suganuma T, Nagata M. Clinical analysis and nonsurgical management of 11 dogs with aural cholesteatoma. Vet Dermatol. 2019;30:42-e12.
13. Little CJ, Lane JG, Gibbs C, Pearson GR. Inflammatory middle ear disease of the dog: the clinical and pathological features of cholesteatoma, a complication of otitis media. Vet Rec. 1991;128:319–22.
14. Schurenemann RM, Oechtering G. Cholesteatoma after lateral bulla osteotomy in two brachycephalic dogs. J Am Anim Hosp Assoc. 2012;48:261–8.
15. Vanhamäki L, Seppänen R, Candido M, Anttila M, Reunanen V. Long-term non-surgical management of canine aural tympanokeratoma with repeated removal of middle ear keratinous debris and CT follow-up. Vet Rec Case Rep. 2023;11:e561.
16. Garosi LS, Dennis R, Schwarz T. Review of diagnostic imaging of ear diseases in the dog and cat. Vet Radiol Ultrasound. 2003;44:137–46.
17. Harran NX, Bradley KJ, Hetzel N, Bowlt KL, Day MJ, Barr F. MRI findings of a middle ear cholesteatoma in a dog. J Am Anim Hosp Assoc. 2012;48:339–43.
18. Gotthelf LN. Doenças do ouvido em pequenos animais: guia ilustrado. 2nd ed. Sao Paulo: Ed. Roca; 2007.

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Résumé
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