Odontoameloblastoma in the posterior maxilla of a 6-years-old Ardi goat (Capra aegagrus hircus)

El-Sayed Ahmed El-Shafaey^{1*}, Mohamed Hamed² and Zeinab Dewidar²

¹Departement of Surgery, Anesthesiology & Radiology, Faculty of Veterinary Medicine, Mansoura University, Egypt. ²Departement of Pathology, Faculty of Veterinary Medicine, Mansoura University, Egypt.

^{*}Corresponding author at: Departement of Surgery, Anesthesiology & Radiology, Faculty of Veterinary Medicine, Mansoura University, Egypt. E-mail: sayedelshafaey@yahoo.com.

> Veterinaria Italiana 2021, **57** (3), 247-251. doi: 10.12834/Vetlt.1824.9678.2 Accepted: 04.11.2019 | Available on line: 31.12.2021

CASE REPORT

Keywords

Odontogenic tumors, Odontoameloblastoma, Maxilla, Histopathology, Goat.

Summary

A 6-year-old female Ardi goat was presented for evaluation of an expansile firm painless maxillary ovoid mass. Clinical and diagnostic imaging examination revealed a well-demarcated extensive mass occupying the left posterior maxilla and altering its anatomical features. Surgical excision was not deemed feasible with this apparently extensive infiltrative features of the tumor and the owner elected to euthanize the goat. Gross examination revealed plexiform and follicular arranged, ameloblast-like odontogenic epithelium. Follicular epithelium was disintegrated leaving spaces, identical to solid multicystic ameloblastoma, intermingled with primitive myxoid stroma resembling dental papilla, teeth like hard structure as enamel and dentine, and osteodentine matrix. Based on these findings, the tumor was diagnosed as odontoameloblastoma (OA) and to our knowledge this is the first report of OA in a goats.

Introduction

Odontoameloblastoma (OA) is an extreme rare matrix-producing mixed odontogenic tumor that has been described in both humans and animals (Barnes et al. 2005, Murphy et al. 2017). It is characterized by a mixed odontogenic epithelium and odontogenic ectomesenchyme with inductive changes leading to the formation of dentin and enamel in parts of the tumor (Dive et al. 2011). Although OAs are considered to be benign in nature, they might locally grow in expansile aggressive pattern with sequential destruction of adjacent bone and dental structures of the jaw (Mosqueda-Taylor et al. 2002). OA has been reported in humans (Dive et al. 2011), cattle (Lepri et al. 2013), horses (Murphy et al. 2017), sheep (Dubielzig and Griffith 1982), dogs (Kok et al. 2018), monkeys (Yanai et al. 1995) and rats (Burrough et al. 2010, Wong et al. 2018). While, to the best of our knowledge, OA has not been reported in goat. This report describes the clinical, radiographic, ultrasonographic and histopathological features of OA in a 6-year-old Ardi goat. To the best of our knowledge, OA has never been reported in goats.

Case description

A 6-year-old female Ardi goat (37 kg) was referred to the Veterinary Teaching Hospital, Faculty of Veterinary Medicine, Qassim University, Saudi Arabia, for investigation of a large firm painless ovoid mass overlying the left maxilla of undetermined duration (Figure 1A). The owner reported that the mass had been gradually increased in size during the last 6 months. The appetite, ruminal motility, rectal temperature, heart and respiratory rates of the goat were within normal range, except for difficulty in chewing with excessive drippling of salivation. However, in the last month with the progressive increase of the mass, there was apparent difficulty in mastication and swallowing and the goat starts to loss its appetite without any response to the traditional medicinal treatment.

Clinical examination

Oral examination revealed a well-demarcated, hard, sessile and dark red cauliflower-like mass of approximately 21 x 11 x 19 cm. It was located on the posterior maxilla extending from the left first premolar to the last molar, including the left



Figure 1. A. Left sided maxillary large ovoid mass in a 6-year-old Ardi goat. **B.** Left lateral radiograph of the soft tissue radiolucent-mass (white arrow) of the left maxilla of the aforementioned goat. **C.** Ultrasonographic view of the left maxillary mass showing irregular architecture with hyperechoic multiloculated core filled with heterogonous contents (black arrow).

maxillary sinuses, pteroid bone, overlying gingiva, left buccal mucosa, left masseter muscle and soft palate. The growth was almost parallel to the tooth and it was protruded in the oral cavity causing difficult prehension and swallowing. Surgical excision was not deemed feasible. The apparently extensive infiltration of the mass to the posterior maxilla and adjacent soft tissues indicated that the animal had a bad prognosis and the owner elected to euthanize the goat at this stage.



Figure 2. Gross appearance of the left maxillary mass of Ardi goat. **A.** The lateral aspect of the mass was smooth and lined by a whitish-gray thick capsule measured approximately $21 \times 11 \times 19$ cm in diameter (arrow). **B.** Grayish-brown, firm and gritty cut surface of the mass causing extensive destruction of the posterior maxilla and replacement by a dark red multiloculated tissue (arrow). 1 = Lower jaw, 2 = Maxillary sinus, Asterisk (*) = 1st premolar and last molar teeth.

Diagnostic imaging evaluation

Diagnostic imaging evaluation of the maxillary mass using skull radiograph (65 kVp, 2.0 mAs and a 60 cm focal film distance) and sector ultrasonography (5 MHz) have been acquired to assess the extent and characters of the maxillary mass. The radiographic picture revealed presence of a soft tissue radiolucent-mass originating from the posterior left maxilla. In addition, there was numerous radiopaque foci with osteolytic changes at the roots of the premolar and molar teeth and in adjoining area of the maxilla (Figure 1B). Ultrasonographically, the mass was irregular with hyperechoic multiloculated core filled with heterogonous contents (Figure 1C).

Gross examination

On gross postmortem examination, the outer surface of the mass from the oral view showed a number of purulent ulcerative nodules filled with a dark red clotted blood and viscid fluid. However, when the mass was viewed from the left fascial side, its outer surface was smooth and lined by a whitish-gray thick capsule (Figure 2A). The cut surface of the mass was grayish-brown, firm and gritty with its multiloculated core filled with irregular brownish trabeculae and granular matrix. There was extensive destruction of the posterior maxilla and replacement by a dark red multiloculated tissue including the 1st premolar and last molar teeth, while the other teeth were completely resorbed. When the mass was viewed from the maxillary sinus side, it was in the sinus causing effacement of the maxillary sinus (Figure 2B). No other abnormalities were seen in all other organs.

Histopathological examination

Serial sections from the mass, parotid, submandibular and retropharyngeal lymph nodes were taken and fixed in 10% neutral buffered formalin for 48 h, routinely processed, sectioned at 5 µm and stained with hematoxylin and eosin (HE) for microscopic examination. Histopathological examination showed solid ameloblastoma with follicular pattern, peripheral palisading and reversal of polarity, intermingled with primitive odontogenic ectomesenchyme resembling dental papilla, dysplastic eosinophilic matrix of osteodentine (osteoid matrix trapped odontoblasts) and basophilic enamel production in the form of crown with peripherally columnar-shaped palisaded ameloblasts due to inductive odontogenesis (Figure 3A). Plexiform ameloblastoma with cystic degeneration in the center, filled with eosinophilic



Figure 3. A. Follicular shaped ameloblastoma, basophilic enamel matrix in the form of crown with peripheral columnar shaped palisaded ameloblasts (arrow), dysplastic eosinophilic matrix (osteodentine) (thick arrow), primitive ectomesenchyme resembling dental papilla. HE. Bar, 200 μm. **B.** Plexiform ameloblastoma, cystic degeneration in the center filled with eosinophilic fibrin and neutrophils (thick arrow), primitive ectomesenchyme resembling dental papilla (thin arrow). HE. Bar, 200 μm. **C.** Squamous metaplasia of neoplastic odontogenic epithelium (arrow). HE. Bar, 100 μm.

fibrinous exudate and neutrophils, proliferation of odontogenic epithelium in the form of strands, intermingled with primitive ectomesenchyme was evident (Figure 3B). Nests of marked pleomorphism neoplastic cells with presence of ghost cells, mature fibrous tissue stroma were recognisable (Figure 3C). Necrosis, macrophagic and neutrophilic infiltrations, increased mitotic figures of neoplastic cells and squamous metaplasia were also seen (Figure 3C). Histopathological examination of parotid, submandibular, or retropharyngeal lymph nodes revealed no micro metastasis. Based on the clinical behavior, anatomic location, diagnostic imaging, histopathological characteristics and differential diagnosis for the maxillary odontogenic tumors, the mass was diagnosed as OA.

Discussion

Odontogenic tumors are histologically classified according to the presence of odontogenic epithelium, odontogenic mesenchyme, or both (Head et al. 2002). In animals, tumors with both components include OA, feline inductive odontogenic tumor, ameloblastic fibro-odontoma, and odontoma (Dubielzig 2003). These odontogenic tumors are seen as mass-like lesions interfering with mastication and even swallowing in affected animal (Gardner 1996). Regarding the frequency of occurrence of odontogenic tumor, the odontogenic tumors comprise 1% of all oral tumors in domestic animals (Munday et al. 2017). Among these various dental tissue tumors, OA has been reported in humans, cattle, horses, sheep, dogs, monkeys and rodents (Dubielzig and Griffith 1982, Yanai et al. 1995, Burrough et al. 2010, Dive et al. 2011, Lepri et al. 2013, Murphy et al. 2017, Kok et al. 2018, Wong et al. 2018). No reference information for incidence of OA in goats is available.

The etiopathogenesis of OAs, either in humans or animals, is not well known. Localized trauma, infection, and genetic predisposition have all been postulated as possible predisposing factors (Thompson *et al.* 1990, Mosqueda-Taylor *et al.* 2002). Despite the large size of maxillary mass and missing teeth, the goat involved in this report didn't have any known trauma or infection or known history of congenital inciting of OA.

OAs have similar microscopic features and often they are asymptomatic in both animal and human patients (Barnes *et al.* 2005, Lepri *et al.* 2013). Untreated tumors can become quite large, causing dysphagia, pain and destroying the affected jaw (Murphy *et al.* 2017). In humans, OAs are rarely associated with any symptoms. They usually are small and often incidentally identified on radiological examinations because of unerupted or missing teeth in the first 2 decades of life (Dive *et al.* 2011). However, in animals, clinical signs associated with OA include mandibular or maxillary swelling, missing teeth, or tooth-like structures erupted into the oral cavity (Yanai *et al.* 1995, Murphy *et al.* 2017, Kok *et al.* 2018). The difficulties in identifying missing teeth early in domestic species may contribute to this disparity, so the lesions in animals may progress further prior to identification and can require euthanasia due to adverse effects on quality of life specially in cattle and horses (Lepri *et al.* 2013, Murphy *et al.* 2017). Thus, this report describes the significant growth potential of maxillary OA in goat.

Differential diagnosis of OA from other mixed odontogenic tumors is of a great challenge for human and animal pathologists over the last decades (Mosqueda-Taylor et al. 2002). Histologically, OAs are closely related to the other matrix-producing odontogenic mixed tumors as odontomas and ameloblastic fibro-odontomas (Burrough et al. 2010). Odontomas have a limited growth potential (once they are fully formed they don't increase in size) with abundance of dental matrix, which creates the impression of dysplastic denticles (Head et al. 2002, Dubielzig 2003). In contrast, the ameloblastic fibromas have the greatest potential of unlimited growth of dental ectomesenchyme, untreated (Gardner 1996). OAs are primarily differentiated histologically by a relative abundance of irregular cords with follicles of odontogenic epithelium abutting a small cluster of odontoblasts resembling the enamel structure in the developing tooth germ with osteodentin formation (Mosqueda-Taylor et al. 2002, Kok et al. 2018). The present case showed relative abundance of neoplastic odontogenic epithelium, odontogenic ectomesenchyme, enamel formation, and mineralized dental tissues, including dentin, osteodentin, and bone-like tissue. Therefore, the histological characteristics and aggressive nature of the tumor reported here were consistent with the diagnosis of OA, suggesting a critical review of odontogenic tumors in animals that formerly diagnosed as ameloblastic fibro-odontoma. These findings were in agreement with Burrough and colleagues (Burrough *et al.* 2010), Lepri and colleagues (Lepri *et al.* 2013) and Murphy and colleagues (Murphy *et al.* 2017).

The surgical treatment of OA has proven to be a challenge, in ensuring adequate surgical excision of the tumor while maintaining function. The decision whether to surgically enucleate the tumor or to euthanize the affected animal was based on several factors as, tumor appearance, nature, size, invasion degree and the healthy condition of the affected animal (Lepri et al. 2013). Improper resection of the highly invasive tumor, resulting in high rates of tumor recurrence with extensive poor prognosis should also be considered (Murphy et al. 2017). The OA in the present case invaded the cancellous bone of the posterior maxilla, which would have indicated a higher likelihood of recurrence if surgical excision had been attempted. Thus, the surgical intervention was declined. This decision was in accordance with Mendez-Angulo and colleagues (Mendez-Angulo et al. 2014) and Kok and colleagues (Kok et al. 2018).

OAs have been reported in a wide range of ages with various position in the mandible or maxilla (Dubielzig 2003, Burrough et al. 2010). However, most of the reported OA cases usually occur in animals younger than 5 years of age (Lepri et al. 2013), with high frequency mandibular involvement, particularly the incisor region (Murphy et al. 2017, Kok et al. 2018). However, in the present case, the OA was located in the posterior maxilla which confirmed by radiographic, gross and histopathological examination. This location strongly suggests that this neoplasm likely originated from the tooth germ tissues. This could be attributed to the relationship between tooth diseases and abnormalities in the animal, which might increase the risk of odontogenic tumors. Similar findings were reported by Thompson and colleagues (Thompson et al. 1990), Dive and colleagues (Dive et al. 2011) and Wong and colleagues (Wong et al. 2018).

To the best of our knowledge, the present report represents the first record of OA in goats. It should be included in the differential diagnosis of mixed odontogenic tumors in animals.

References

- Barnes L., Eveson J., Reichart P. & Sidransky D. 2005. Odontoameloblastoma. *In* Pathology and genetics of head and neck tumors (WHO Classification of Tumors series). Lyon, France, IARC Press, 312.
- Burrough E.R., Myers R.K. & Whitley E.M. 2010. Spontaneous odontoameloblastoma in a female Sprague Dawley rat. *J Vet Diagn Invest*, **22**, 998-1001.
- Dive A., Khandekar S., Bodhade A. & Dhobley A. 2011. Odontoameloblastoma. *J Oral Maxillofac Pathol*, **15**, 60-64.
- Dubielzig R.R. 2003. Histologic classification of tumors of odontogenic origin of domestic animals. *In* Histological classification of tumors of the alimentary system of domestic animals, vol. 10, 46-57. Washington, DC, Armed Forces Institute of Pathology.
- Dubielzig R.R.& Griffith J.W. 1982. An odonto a meloblastoma in an adult sheep. *Vet Pathol*, **19**, 318-320.
- Lepri E., Avallone G., Mandara M.T. & Vitellozzi G. 2013. Odontoameloblastoma in a calf. *J Vet Dent*, **30**, 248-250.
- Gardner D.G. 1996. Ameloblastic fibromas and related tumors in cattle. *J Oral Pathol Med*, **25**, 119-124.
- Head K.W., Else R.W. & Dubielzig R.R. 2002. Tumors of the Alimentary Tract. *In* Tumors in domestic animals (Meuten D.J., ed). 4th ed. Iowa State Press, Avenue, Ames, Iowa, 401-481.

KokM., Chambers J., Ushio N., Miwa Y., Nakayama H. & Uchida

K. 2018. Amyloid-producing odontoameloblastoma in a black-tailed prairie dog (*Cynomys ludovicianus*). *J Comp Path*, **159**, 26-30.

- Mendez-Angulo J.L., Tatarniuk D.M., Ruiz I. & Ernst N. 2014. Extensive rostral mandibulectomy for treatment of ameloblastoma in a horse. *Vet Surg*, **43**, 222-226.
- Mosqueda-Taylor A., Carlos-Bregni R., Ramirez-Amador V., Palma-Guzman J.M., Esquivel-Bonilla D. & Hernandez-Rojase L.A. 2002. Odontoameloblastoma. Clinic-pathologic study of three cases and critical review of the literature. *Oral Oncol*, **38**, 800-805.
- Murphy B., Bell C., Koehne A., Richard R. & Dubielzig R. 2017. Mandibular odontoameloblastoma in a rat and a horse. *J Vet Diagn Invest*, **29**, 536-540.
- Thompson I.O., Phillips V.M., Ferreira R. & Housego T.G. 1990. Odontoameloblastoma: a case report. Br J Oral Maxillofac Surg, 28, 347-349.
- Wong H., Hedley J., Stapleton N., Murphy B. & Priestnall S. 2018. Odontoameloblastoma with extensive chondroid matrix deposition in a guinea pig. J Vet Diagn Invest, **30**, 793-797.
- Yanai T., Masegi T., Tomita A., Kudo T., Yamazoe K., Iwasaki T., Kimura N., Katou A., Kotera S. & Ueda K. 1995. Odontoameloblastoma in a Japanese Monkey (*Macaca fuscata*). Vet Pathol, **32**, 57-59.