

“Ameloblastic fibrodentinitoma with a congenitally missing second premolar tooth:

A case report”

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## Abstract

Ameloblastic fibrodentinoma (AFD) is a rare benign mixed odontogenic tumor that occurs predominantly in children and young adults. AFD is usually associated with delayed eruption of the tooth and painless swelling. We present a case of an 8-year-old Japanese girl complaining of missing of the mandible second deciduous molar. Intraoral examination did not reveal any sign of pain and/or swelling in the left mandible. Panoramic radiography revealed impacted second deciduous molar and a radiolucent and well-circumscribed lesion, which contained radiopaque material at the premolar lesion of the left mandible.

The second deciduous molar tooth was unerupted and there was a congenitally missing second premolar tooth. Our provisional diagnoses were odontogenic tumor. Biopsy confirmed AFD. Patient treatment included total excision of the tumor and the second deciduous tooth. No recurrence has been noted during the two years of follow-up.

## **Introduction**

Although ameloblastic fibrodentinoma (AFD) has been defined as a neoplasm similar to ameloblastic fibroma (AF), it additionally exhibits inductive changes that lead to the formation of dentin<sup>1)</sup>. AFD is an exceedingly rare histological variant of AF in which there is the formation of dentin or dentinoid tissue. AFD arises mainly in the posterior mandible and is usually associated with unerupted molar teeth. Tumors related to the deciduous teeth usually arise in the incisor area. On the other hand, tumors related to the permanent teeth that develop in the molar region<sup>2)</sup>. AFD is more common in males than in females, and often grows slowly and is asymptomatic. Radiographically, it shows a fairly well delineated radiolucency with varying degrees of radiopacity<sup>3)</sup>. The recommended treatment is conservative surgical excision<sup>4)</sup>. At the present time, however, there is no agreement with regard to the preservation of teeth affected by the tumor.

The purpose of this article is to describe a case of AFD affecting the left mandible body included congenital missing of second premolar tooth with excisional surgery without recurrence after two years of follow-up.

## **Case Report**

On April 2010, an 8-year-old Japanese girl with missing of her left mandible second deciduous molar and her delayed eruption of her left mandible second premolar tooth were referred by her general dentist to the Department of Oral and Maxillofacial Surgery at Nagasaki University Hospital. The medical and family histories were unremarkable. Clinical examination did not reveal any sign of pain or swelling in her left mandible. There was no history of local trauma or infection. She had good oral hygiene with extra- and intra-oral inspection. There was no visible swelling or expansion and the overlying mucosa appeared normal. Radiographic examination revealed well-defined radiolucency with radiopacity noted above the tooth crown of the unerupted deciduous second molar, judging from its morphology, in the left mandible (Fig. 1). The impacted tooth was completely formed and located near the inferior border of her mandible. The patient also had a congenitally missing second premolar tooth in her left mandible. Computed tomography revealed radiopaque masses in the well-circumscribed lesion in her left mandible (Fig 2, 3). The clinical diagnosis was benign odontogenic tumor, with the tumor further classified as ameloblastoma.

After obtaining a biopsy under local anesthesia, the specimen's examination revealed that the tumor consisted of odontogenic ectomesenchyme tissue and odontogenic

epithelium arranged in islands, strands and cords. The ectomesenchymal component showed moderate density of oval or spindle cells around the epithelial nests. Hyalinous change was noticed in distant area from the epithelial nests (Fig 4). Some of the epithelial nests demonstrated enamel organ-like structure. In addition, premature dentine-like matrix with enclosed cells was followed by odontogenic ectomesenchyme tissues. Enamel organ-like epithelial follicles were found to be adjacent to and within the dysplastic dentine matrix (Fig. 5). At the time of the investigation, enamel formation was not seen, and there were no atypia in the tumor cells. After a histological diagnosis of AFD was made, the patient underwent surgical excision under general anesthesia. The tumor was encapsulated and there was no perforation seen in the buccal and lingual cortex bones. The dental sac of the second deciduous molar tooth adhered to the tumor, but was separated easily from the tumor. The completely formed second deciduous molar tooth was also removed with the tumor excision, because we deemed that it would not erupt. Since the excised specimens demonstrated similar features for the biopsy specimens, a final diagnosis of AFD was made. The 12-month radiographic follow-up showed there was new bone formation (Fig 6), and there was no recurrence observed at 24 months after the surgical excision.

## **Discussion**

According to the World Health Organization, AFD is included along with AF and ameloblastic fibro-odontoma (AFO)<sup>5</sup>. All of the tumors in this group have odontogenic epithelium with odontogenic ectomesenchyme, with or without dental hard tissue formation. Thus, if there is dentin formation, the lesion should be diagnosed as AFD, while if there is also enamel formation, then it should be diagnosed as AFO<sup>5</sup>. In the present case, the tumors consisted of odontogenic epithelium with odontogenic ectomesenchyme cells. Additionally, there were dentin-like hard tissue formations without enamel formation in the tumor tissues. Based on these findings, a final diagnosis of AFD was made.

In the present case, we found that the AFD was accompanied with a congenitally missing second premolar tooth. However, we have not been able to find any reports in the literature of AFD accompanied by congenitally missing permanent teeth in past studies.

Additionally, in our case there was complete formation of the second deciduous molar, which was impacted near the border of the mandible. Lower second deciduous molar teeth normally complete their formation of the crown enamel at 10 months, with the emergence into oral cavity at 20 months and completion of the root formation at 30

months after birth<sup>6</sup>). The lower second premolar tooth germ has been shown to form at 7.5 to 8 months after birth<sup>6</sup>). Therefore, the AFD in this case may have already been present between 7.5 to 8 months after birth. These findings appear to indicate that the AFD interfered with the emergence of the second deciduous molar tooth into the oral cavity. We assume that this ectopic complete formation of the second deciduous molar caused the agenesis of the premolar tooth germ.

Although clinically AFD and AFO are similar to AF, there has been no definitive evidence that both tumors exhibit the same biologic behavior as AF<sup>7</sup>). Both AFD and AFO have a well-encapsulated lesion and there is no local invasion. In these types of cases, the recommended treatment is the conservative surgical excision<sup>4</sup>). However, there is as of yet no consensus regarding the preservation of teeth affected by the tumor. Okura et al<sup>8</sup>). have recommended unerupted teeth to be preserved. In contrast, other authors have recommended that teeth with an affected lesion to be removed<sup>9,10</sup>). In this case, the unerupted second deciduous tooth was extracted during the procedure that assisted the complete excision of the tumor tissues.

In the present case, there has been no recurrence observed in the patient at 2 years after the enucleation of the lesion and extraction of the deciduous second premolar.



Based on these findings, we recommend that the AFD treatment procedure include enucleation of the lesion along with the unerupted teeth.

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## Figure captions

Fig. 1 Panoramic radiography revealed well-defined radiolucency with a radiopaque area above the tooth crown of the unerupted deciduous second molar in the left mandible.

Fig. 2 Horizontal computed tomography showed calcified masses in the well-circumscribed lesion in the left mandible. Slight expansion and absorption were noticed in the lingual cortex in the left molar lesion

Fig. 3 Sagittal computed tomography showed calcified masses above the unerupted tooth in the well-circumscribed lesion.

Fig. 4 The tumor consisted of odontogenic ectomesenchyme tissues and odontogenic epithelium. Strands or cords of neoplastic epithelial components were seen in the cellular odontogenic ectomesenchyme tissues (hematoxylin and eosin  $\times 10$ ).

Fig. 5 Dysplastic dentine matrix embedded individual cells and enamel organ-like structures. Odontogenic ectomesenchymal cells adhered to dentinoid matrix and epithelial component (arrows). No induction of enamel formation was found (hematoxylin and eosin  $\times 20$ ).

Fig. 6 Panoramic radiography one year after surgery. Radiography showed ossification at original site of the lesion. Eruption of the left lower first and second premolar teeth was observed.

Fig. 1

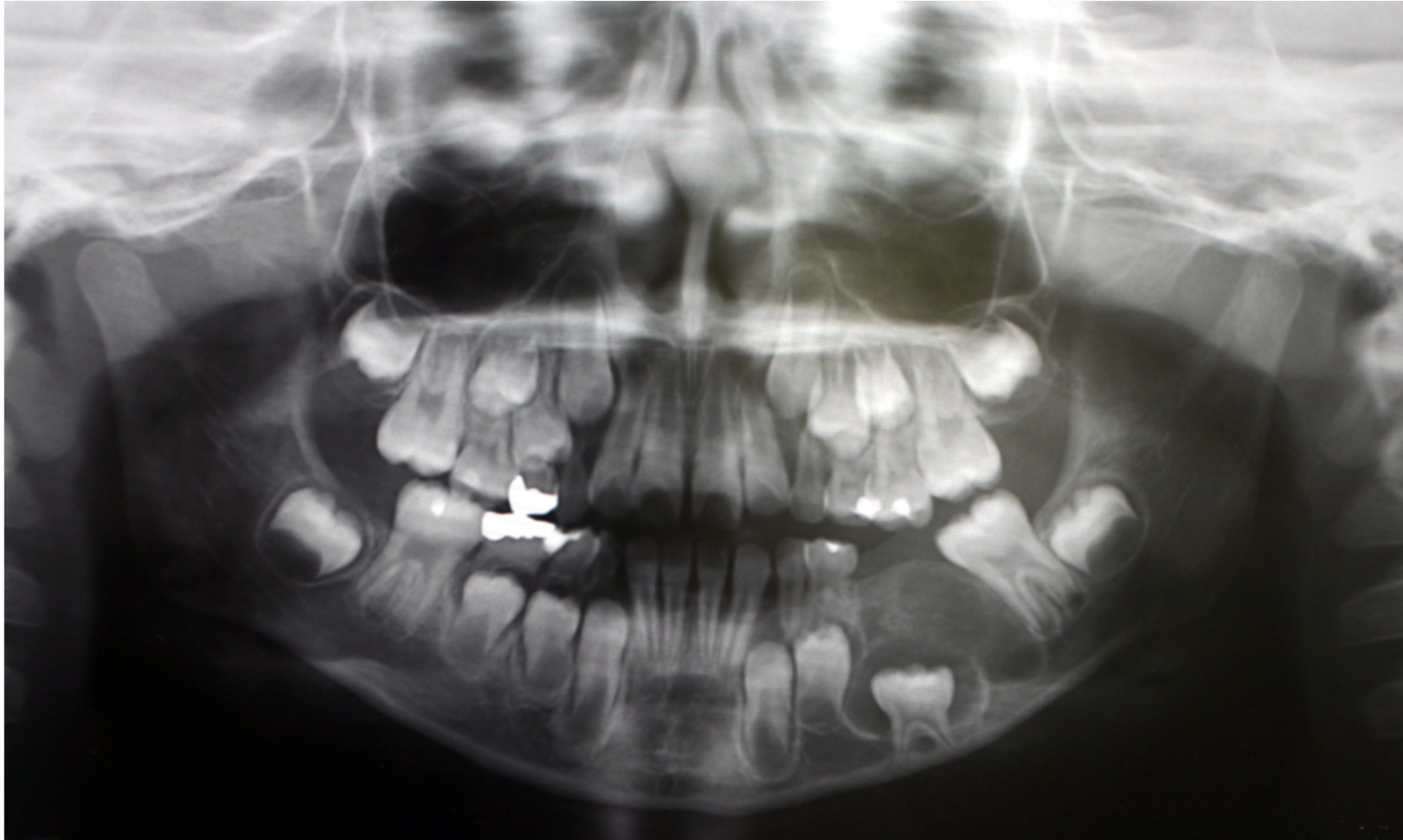


Fig. 2

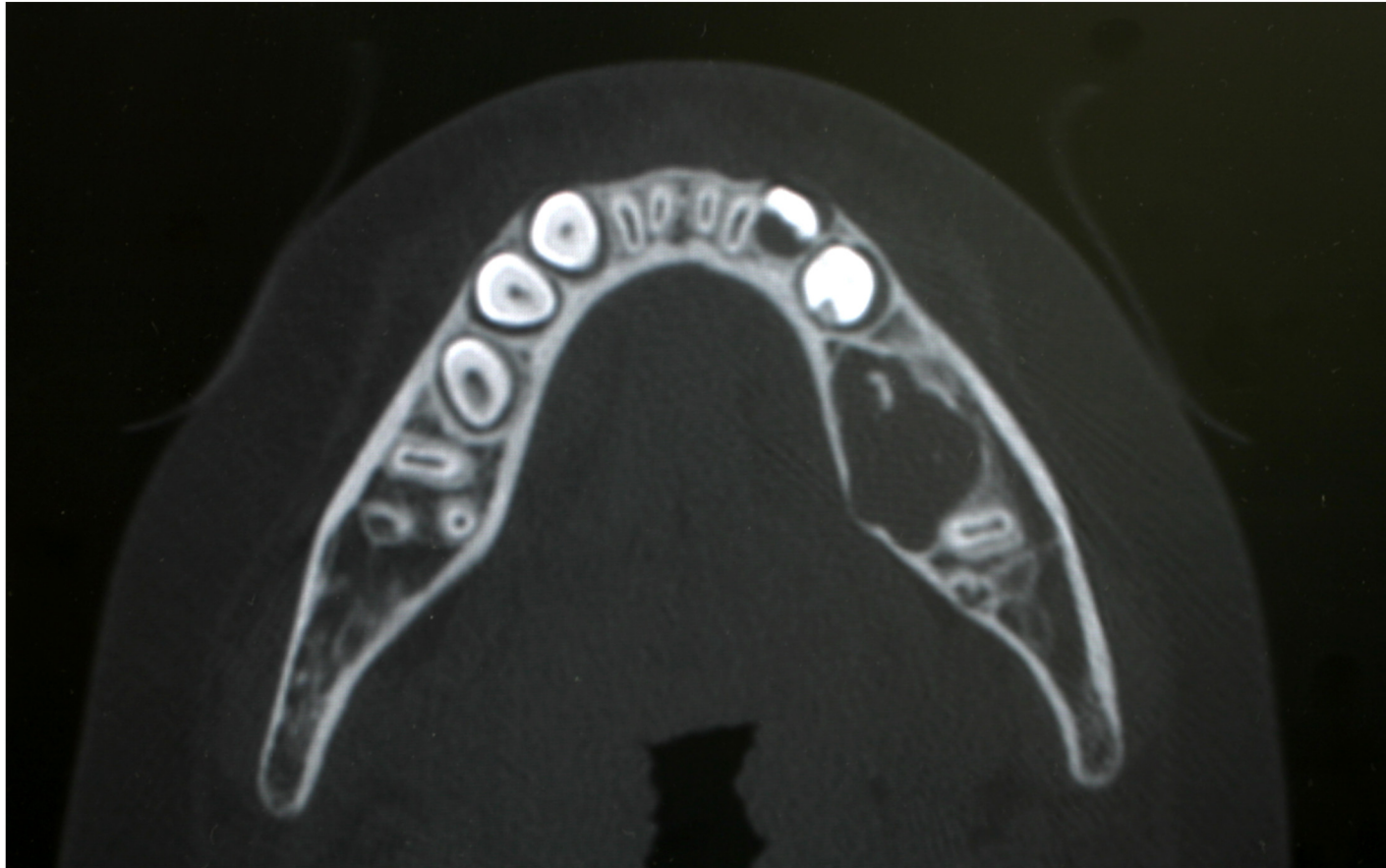


Fig. 3

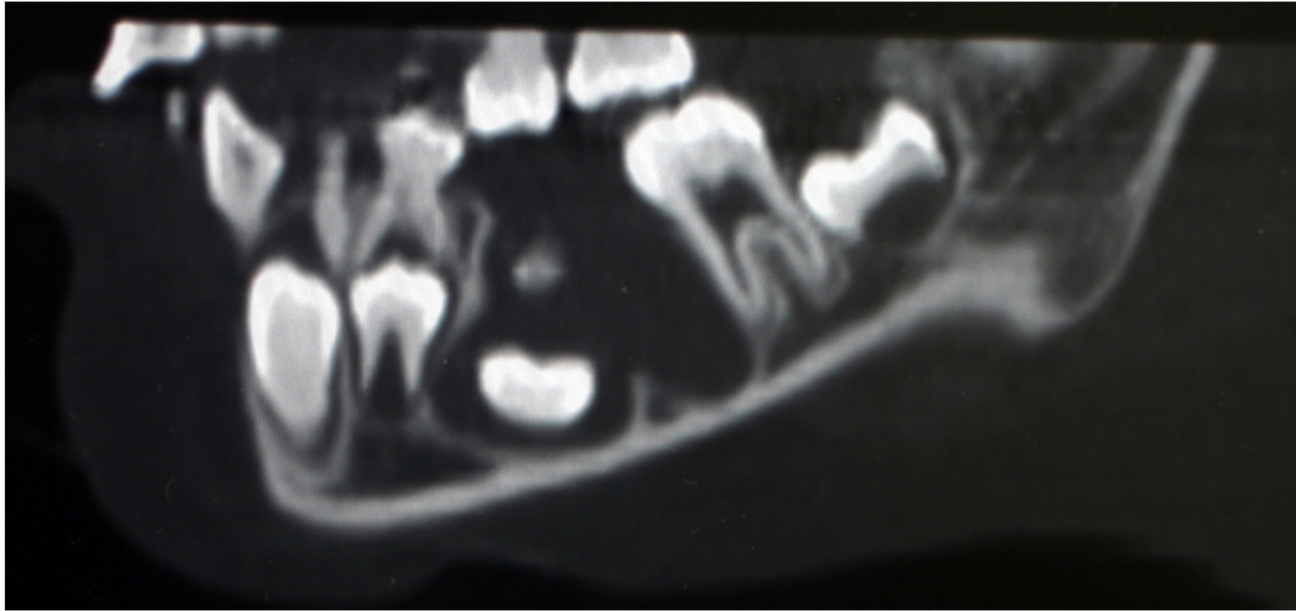




Fig. 4

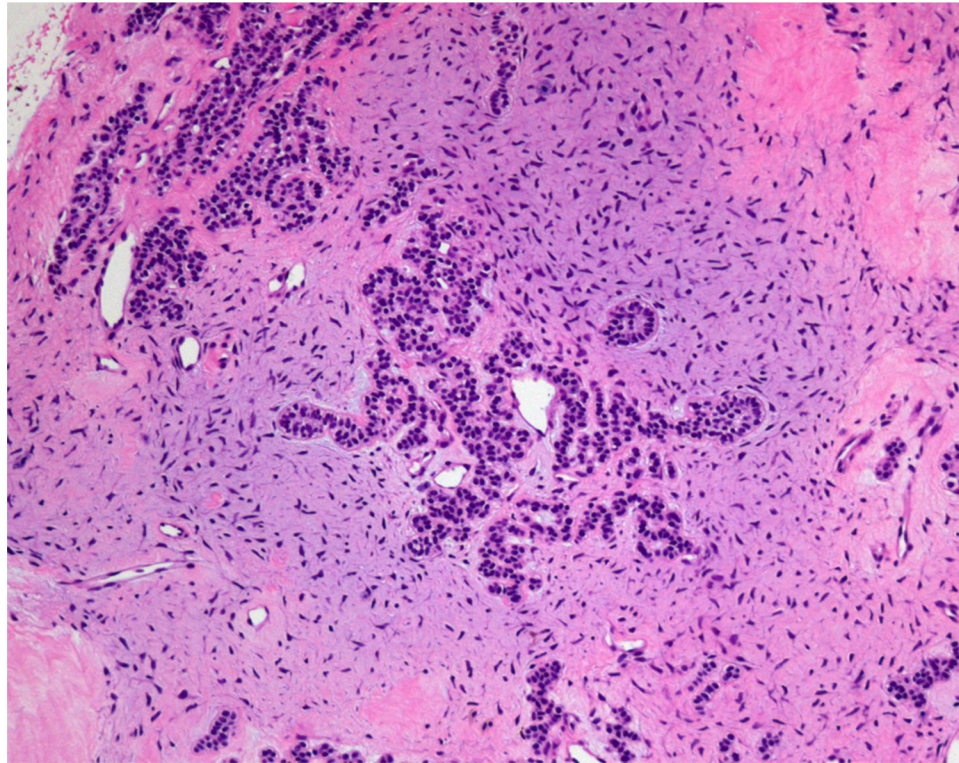


Fig. 5

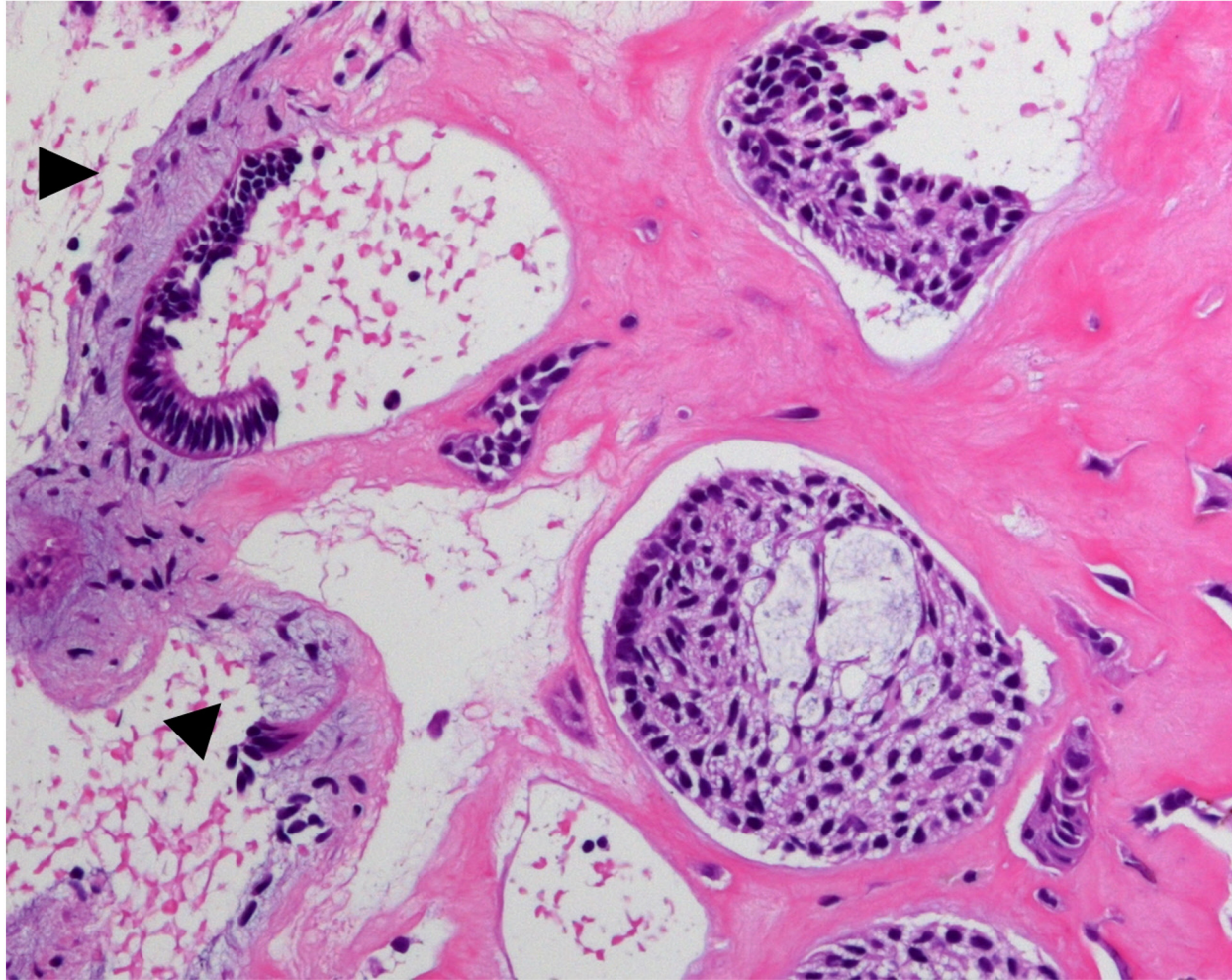


Fig. 6

