Case 16132

Eurorad

Odontogenic keratocyst of the mandible

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Section: Head & neck imaging
Area of Interest: Head and neck
Procedure: Diagnostic procedure
Procedure: Contrast agent-intravenous
Imaging Technique: CT
Imaging Technique: MR
Special Focus: Cysts Case Type: Clinical Cases
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Patient: 61 years, female

Clinical History:

A 61-year-old woman presented with episodes of paresthesia in the region of the left mental nerve and a slowly enlarging swelling of the left mandible for the past 2 years.

Imaging Findings:

A Cone Beam Computed Tomography (CBCT) showed a large radiolucent lesion in the left mandible with cortical breakthrough of the buccal cortex with an associated soft tissue component (Fig. 1a). The mesiodistal extent was larger than the buccolingual extension. (Fig. 1a) The roots of teeth 35 and 34 are located in the cyst (Fig. 1b, 2). The intimate relationship of the lesion with the mandibular canal may explain the paresthesia in the region of the left mental nerve (Fig. 2). Magnetic resonance imaging (MRI) showed an intramedullary lesion in the left mandible with scalloping of the lingual cortex and breakthrough of the buccal cortex (Fig. 4). The lesion was isointense to muscle on T1-WI (Fig. 3) and hyperintense on T2-WI with intralesional areas of intermediate signal (Fig. 4b). After gadolinium contrast administration, there was weak peripheral rim enhancement (Fig. 5a-b). Surgical enucleation revealed a cystic lesion containing keratin. On histopathological examination, the cyst wall consists of keratinised squamous epithelium.

Discussion:

Based on the imaging and histopathologic findings, the diagnosis of odontogenic keratocyst (OKC) was made. OKC is a developmental odontogenic cystic lesion arising from additional remnants of the dental lamina of the jaw bones or the basal cells from the oral epithelium. OKCs represent 2-11% of all odontogenic cysts, affect all ages with a peak from the 2nd–4th decade and have a male to female ratio of 2:1 [1, 2]. Clinically, most OKCs are asymptomatic but symptoms like pain, swelling and paresthesia of the lower lip can be seen in larger lesions [2, 3)]. In children, they are often associated with basal cell naevus syndrome (NBCCS or Gorlin’s syndrome), consisting of multiple OKCs, skin basal cell carcinomas, skeletal abnormalities and falx calcifications [2]. Radiologically, OKCs are difficult to differentiate from other radiolucent odontogenic lesions. A panoramic radiograph typically reveals a radiolucent

unilocular or multilocular lesion with well-demarcated or scalloped margins. Larger lesions tend to be more multilocular [2]. Most lesions (75%) are located in the posterior mandible with 50% occurring at the angle. Other lesions are located in maxilla with a possible expansion into the maxillary sinus. In 25%-40% an impacted tooth is seen and therefore it can be difficult to differentiate from a dentigerous cyst. Teeth displacement may occur but root resorption is very rare [1]. (CB)CT shows similar features as panoramic radiographs but (CB)CT is superior to determine lesion extension and cortical thinning or breakthrough [4]. OKC tend to grow in a mesiodistal direction while ameloblastomas usually show a predominant buccal-lingual expansion [2]. On MRI, the signal may vary, whereas ameloblastomas are hypointense on T1 and hyperintense on T2-WI [5-7]. There is a weak enhancement of the rim in OKCs whereas ameloblastomas display a nodular enhancement pattern [2, 8]. Diffusion-weighted imaging (DWI) can be used to differentiate between OKCs and ameloblastomas. OKCs contain desquamated keratin which increases the viscosity and therefore may cause diffusion restriction. Ameloblastomas containing proteinaceous fluid do not show restricted diffusion [6]. In case of NBCCS, (CB)CT and MRI can be used to detect multifocality. Because imaging features may be nonspecific, histological confirmation is mandatory [2]. The cyst wall is composed of a keratinised squamous epithelium. Treatment of OKCs depend on age, size and location of the cyst and soft tissue expansion. Treatment options are enucleation, marsupialisation, curettage and osteotomy [4]. Regular follow-up for 5 years is recommended as the recurrence rate is high [1].

Written informed patient consent for publication has been obtained.

Differential Diagnosis List: Odontogenic keratocyst, Dentigerous cyst, Radicular cyst, Ameloblastoma, Lateral periodontal cyst, Odontogenic fibroma, Odontogenic myxoma (myxofibroma), Aneurysmal bone cyst, Central giant cell granuloma

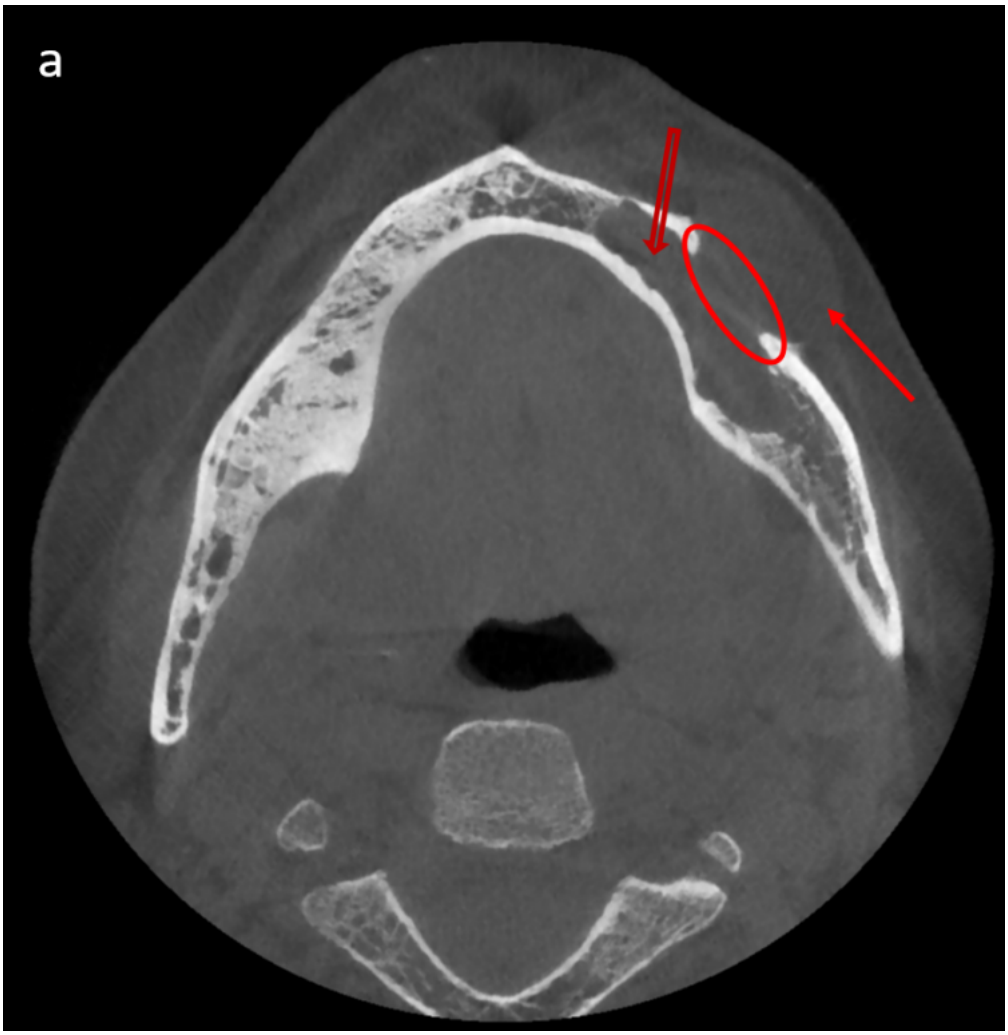
Final Diagnosis: Odontogenic keratocyst

References:

Chirapathomsakul D, et al. (2006) A review of odontogenic keratocysts and the behavior of recurrences. Oral Surg Oral Med Oral Pathol Oral Radiol Endod (PMID: 16360602)
Bernaerts A, Vanhoenacker FM, Hintjens J, Chapelle K, Salgado R, De Foer B, et al. (2006) Tumors and tumor-like lesions of the jaw: radiolucent lesions. JBR-BTR (PMID: 16729445)
Eryilmaz T, et al. (2009) Odontogenic keratocyst: an unusual location and review of the literature. Ann Plast Surg (PMID: 19158536)
Bhargava D, et al. (2012) Keratocystic odontogenic tumour (KCOT)--a cyst to a tumour. Oral Maxillofac Surg (PMID: 22072419)
Minami M, et al. (1996) Cystic lesions of the maxillomandibular region: MR imaging distinction of odontogenic keratocysts and ameloblastomas from other cysts. AJR Am J Roentgenol (PMID: 8610578)
Sumi M, et al. (2008) Diffusion-weighted MR imaging of ameloblastomas and keratocystic odontogenic tumors: differentiation by apparent diffusion coefficients of cystic lesions. AJNR Am J Neuroradiol (PMID: 18719033)
Fujita M, et al. (2013) Diagnostic value of MRI for odontogenic tumours. Dentomaxillofac Radiol (PMID: 23468124)
Konouchi H, et al. (2006) Usefulness of contrast enhanced-MRI in the diagnosis of unicystic ameloblastoma. Oral Oncol (PMID: 16488178)

Figure 1

a



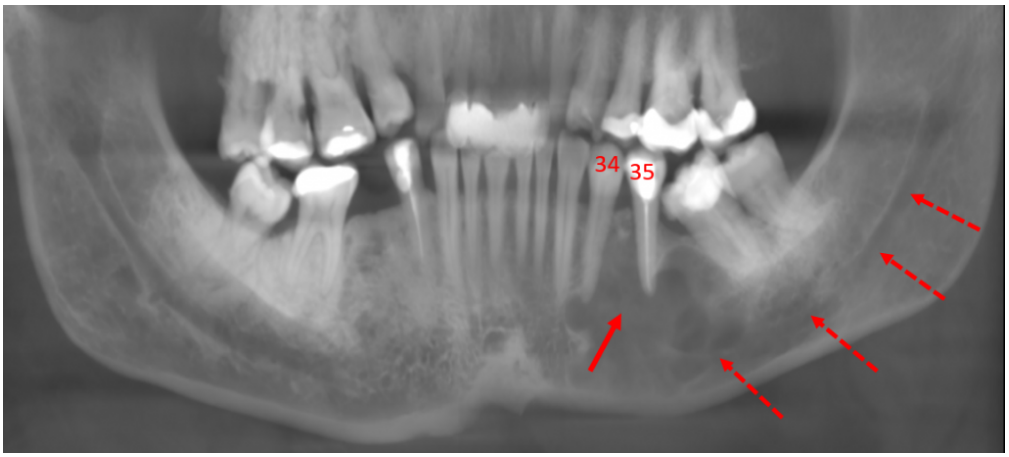
Description: Axial: Large radiolucent lesion (open arrow) with breakthrough of the buccal cortex (oval). Note a soft tissue component (arrow), the precise extent is difficult to evaluate on CBCT, due to lack of soft tissue contrast. **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint-Maarten, Mechelen, Belgium

b

Description: Sagittal reformatted image: the roots of teeth 35 (dotted arrow) and 34 (not shown on this image) are located in the lesion. Cortical breakthrough (oval). **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint-Maarten, Mechelen, Belgium

Figure 2

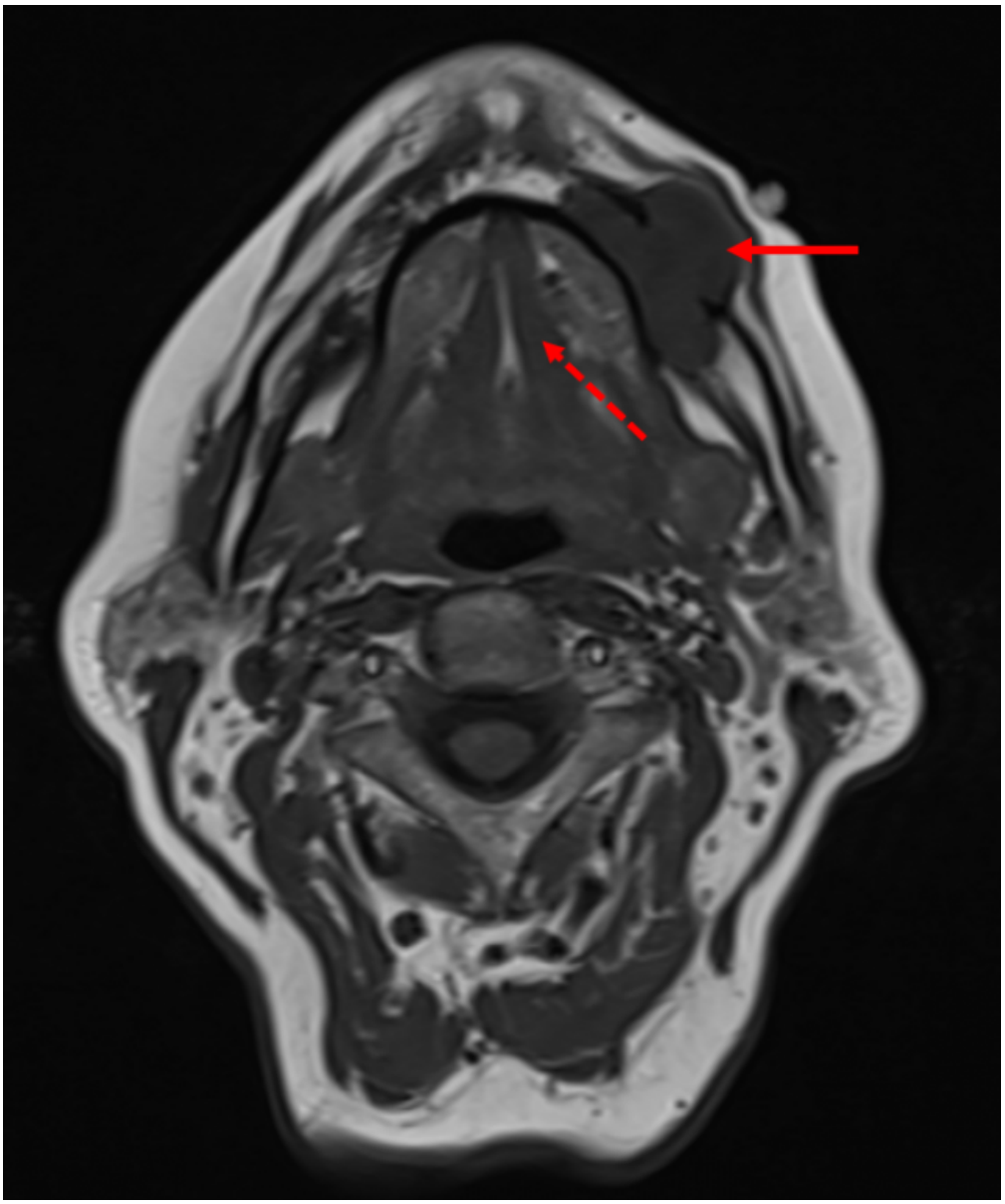
a



Description: Large multilocated radiolucent lesion (arrow) with an intimate relationship with teeth 34 and 35 and the left mandibular canal (dotted arrows). **Origin:** Vanhoenhacker FM, Department of Radiology, AZ Sint-Maarten, Mechelen, Belgium

Figure 3

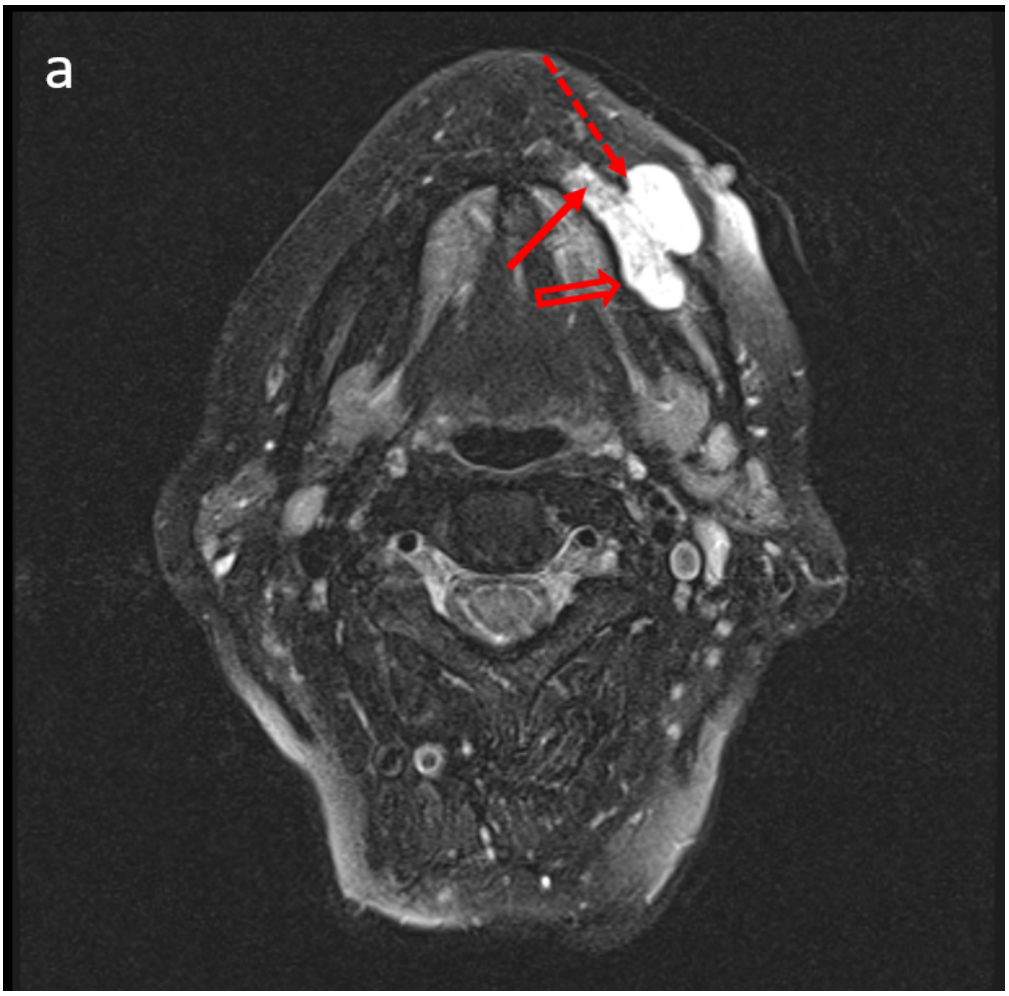
a



Description: The lesion (arrow) is isointense to muscle (dotted arrow). **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint Maarten, Mechelen, Belgium

Figure 4

a



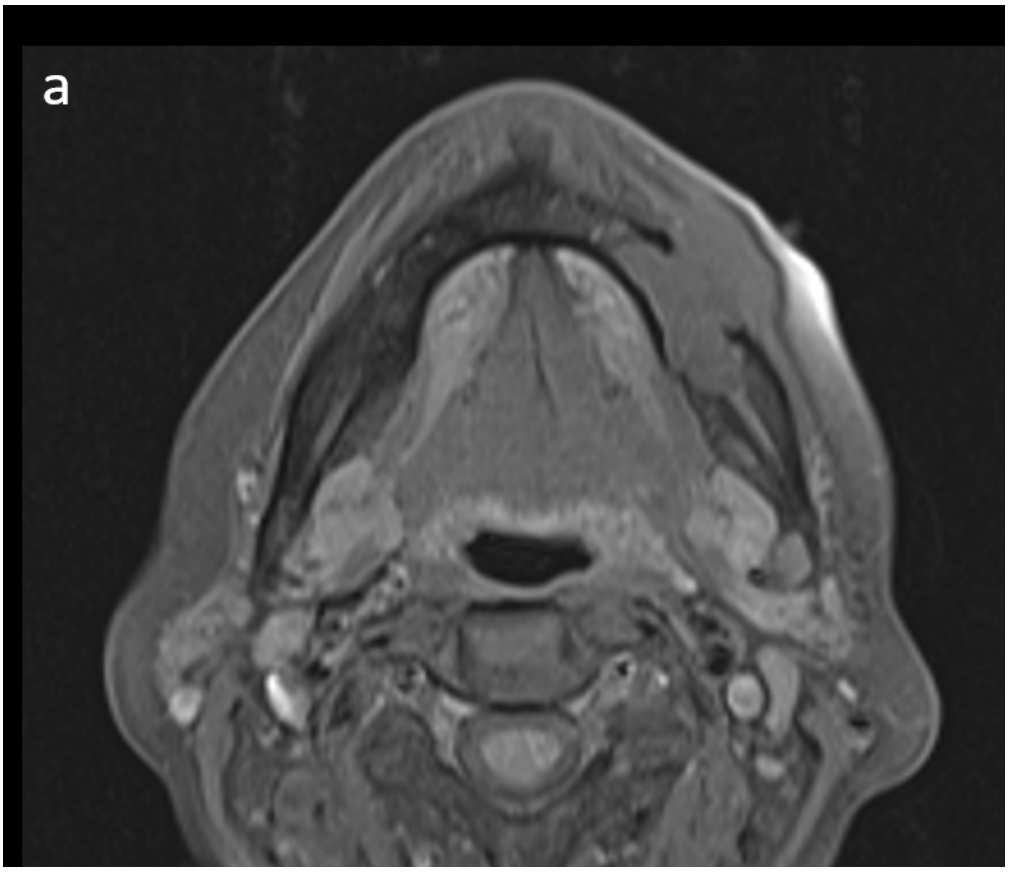
Description: Axial T2 with fat suppression (FS): Intramedullary lesion causing slight thinning of the lingual cortex (open arrow) and cortical breakthrough of the buccal cortex (dotted arrow). **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint Maarten, Mechelen, Belgium.

b

Description: Coronal T2 without FS: The lesion is predominantly of high signal with intralesional areas of intermediate signal (arrows in a and b). **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint Maarten, Mechelen, Belgium.

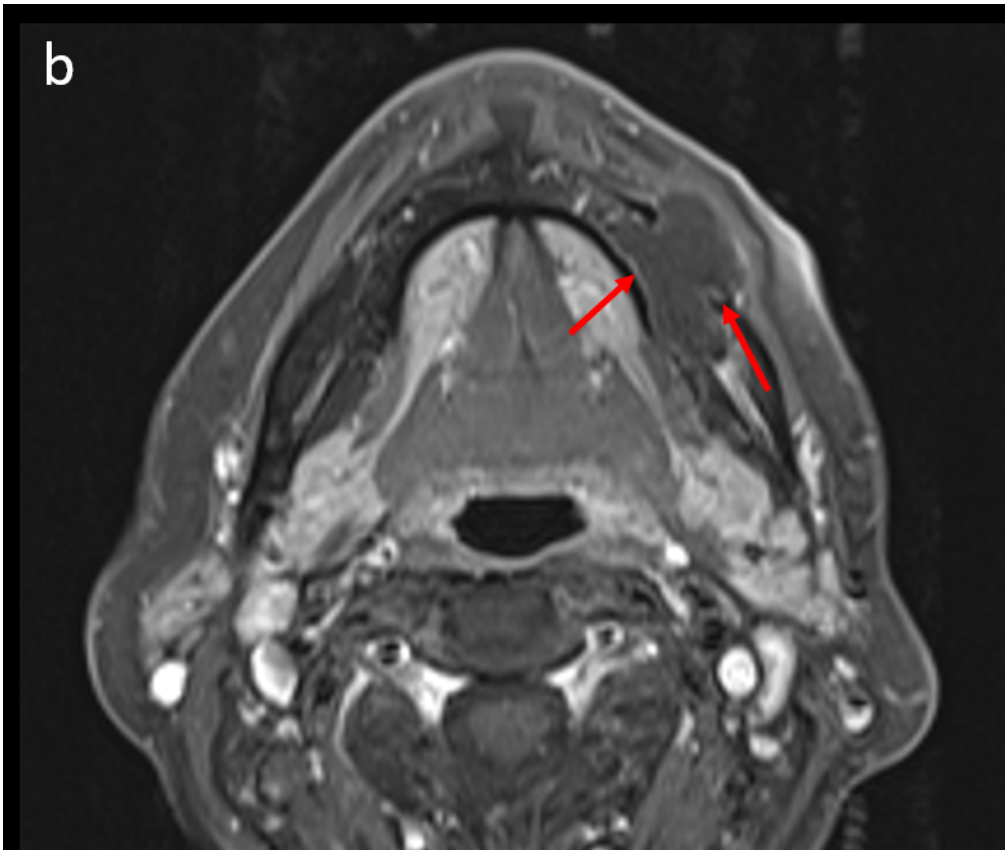
Figure 5

a



Description: Axial T1 FS before gadolinium contrast. **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint Maarten, Mechelen, Belgium.

b



Description: Axial T1 FS after gadolinium contrast: faint rim enhancement (arrows). **Origin:** Vanhoenacker FM, Department of Radiology, AZ Sint Maarten, Mechelen, Belgium.