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CASE REPORT

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Oesophageal adenoid cystic carcinoma—a rare diagnosis on ultrasound guided percutaneous biopsy

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ABSTRACT

Introduction: Oesophageal adenoid cystic carcinoma (OACC) is a very rare form of malignant tumor in the oesophagus with estimated prevalence of around 0.04%–0.16% of oesophageal malignant tumors according to Japanese studies.

Case: We present a case of a 67-year-old man diagnosed with this cancer alongside all the imaging findings on Computerized tomography, magnetic resonance imaging (MRI), ultrasound, fluoroscopy, and nuclear imaging. Interestingly the mainstay of diagnosis of oesophageal malignancy—the endoscopic biopsy, was normal.

Conclusion: Ultrasound guided percutaneous biopsy is a useful adjunct in diagnosis for OACC.

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KEYWORDS

Adenoid cystic carcinoma; oesophagus; percutaneous biopsy

Introduction

Tumours of the oesophagus comprise <1% of all cancers. The most common histological types are squamous cell carcinoma and adenocarcinoma. Oesophageal adenoid cystic carcinoma (OACC) is a rare form of malignant tumour of the oesophagus, with only few cases documented in the literature so far with a prevalence of 0.04%–0.16% [1,2]. Despite their rarity in the oesophagus they are common in salivary glands. Histologically there are two main cell types: ductal and modified myoepithelial cells. There is not much data on the prognosis of OACC but overall distant metastasis were more frequent than with other oesophageal cancers. Average overall survival was approximately 7 months, with a 5-year survival rate of 35% [3,4].

Endoscopy is considered as method of choice of evaluation of oesophageal lesion and imaging is considered as mainstay for staging, assessment of prognosis and post treatment response assessment. In our case, diagnosis was established on ultrasounded guided percutaneous biopsy of esophageal lesion when preceding endoscopic results were negative for malignancy.

Case

67 year old male presented with occasional high dysphagia to solids. There was no history of dysphagia for liquids. He was an occasional smoker with only occasional alcohol intake. Physical examination didn't reveal any palpable lump or cervical lymphadenopathy. Laboratory examination findings were also unremarkable.

Endoscopy was performed to look for the cause of dysphagia, which suggested extrinsic compression of upper esophagus with smooth mucosal surface. However, histology from oesophago-gastro-duodenoscopy samples demonstrated chronic inflammatory changes only without any sign of dysplasia or malignancy.

Contrast enhanced computerized tomography (CT) scan was further requested which revealed a concentric stricturing soft tissue lesion in upper esophagus in subcricoid location which was inseparable from anterior wall of the trachea (Fig. 1). The lesion was causing anterior indentation in posterior wall of trachea. These findings were further confirmed by MRI where a T2 hyperintense concentric band was seen at the location of the CT abnormality (Fig. 2). There was no significant lymphadenopathy

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or distant metastasis identified on either of the investigation.

Barium swallow was also performed to evaluate the degree of dysphagia, which showed smooth circumferential narrowing in upper esophagus suggesting a submucosal location of the lesion (Fig. 3).

Ultrasound was performed to evaluate the accessibility of the lesion for percutaneous guided biopsy. It demonstrated a well-defined hypoechoic lesion posterior to right lobe of the thyroid gland and inferior thyroid artery in sub-cricoid location (Fig. 4). The lesion didn't demonstrate any vascularity on color Doppler. Ultrasound guided percutaneous core biopsy was performed on the right side of the neck using 14 gauge biopsy needle after obtaining informed consent. Local anaesthetic used was lignocaine 1%. 10 mls of the lignocaine was infiltrated to the skin and deeper tissues upto the oesophagus. Good visualization of the needle along its track to the thickened hypoechoic submucosal mass was obtained. On histopathoogical examination, the core of fibrocollagenous tissue showed infiltration by poorly differentiated basaloid tumour forming cohesive sheets and cords of uniform small cells with no clear squamous or glandular differentiation. The cells contained hyperchromatic round fairly uniform nuclei with scanty cytoplasm. No prominent nucleoli were seen. Some nuclear moulding was also observed. The obtained core also showed uniform positivity for cytokeratin, but patchy positivity for P63, CD117 and CK7. Occasional cells were also positive for P16. Proliferation activity seems to be variable, and focally up to 30%. The sample was negative for CK20, TTF1 and synaptophysis. Features collectively were consistent with intermediate grade adenoid cystic carcinoma.



Figure 1. CT revealed non enhancing soft tissue density arising from the anterior wall of the upper oesophagus just below the level of cricoid causing sever luminal narrowing. No significant lymph node enlargement or distant metastasis present.

For cancer staging purposes a fusion positron emission tomography-computed tomography san was carried out which confirmed neither lymph nodal nor distant metastasis (Fig. 5).

Discussion

Adenocystic carcinoma is common tumor of salivary glands, respiratory tract, oropharynx, mammary glands and uterus however, its occurrence in esophagus is very rare [5]. Previous studies have concluded the incidence of OACC as 0.04% (1 of 2,370 cases) and 0.16% (4 of 2,521 cases) of all esophageal malignant tumors [1,2].

The clinical features of OACC is also not well studied due to small number of cases [6]. The mainstay of diagnosis of esophageal carcinoma is endoscopy guided biopsy, which turned out to be negative in

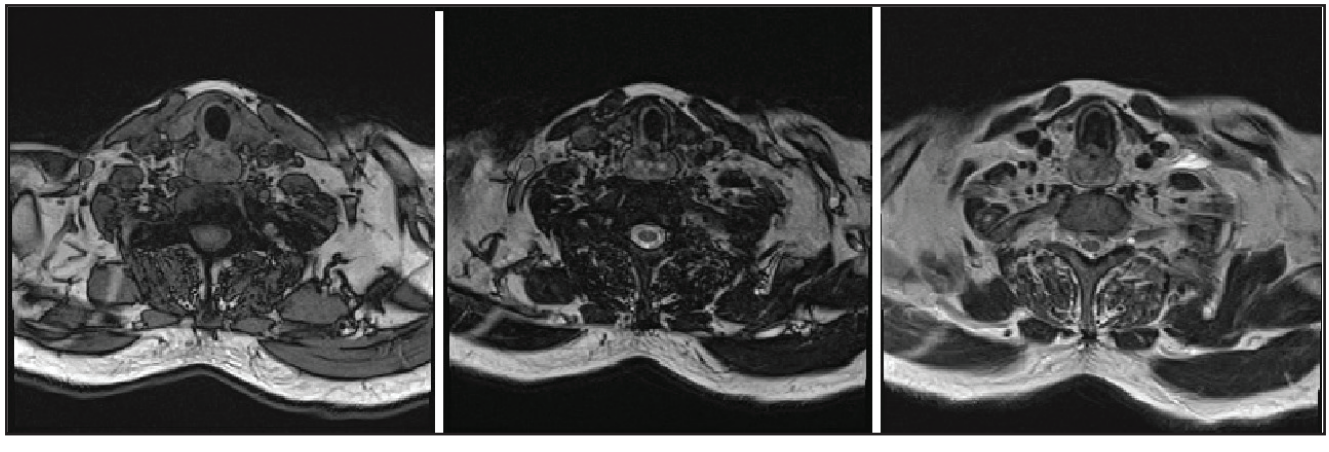


Figure 2. MRI shows an enhancing mass lesion arising from anterior wall of upper esophagus resulting into slit like lumen. It is also causing alight bulge in the posterior tracheal wall.

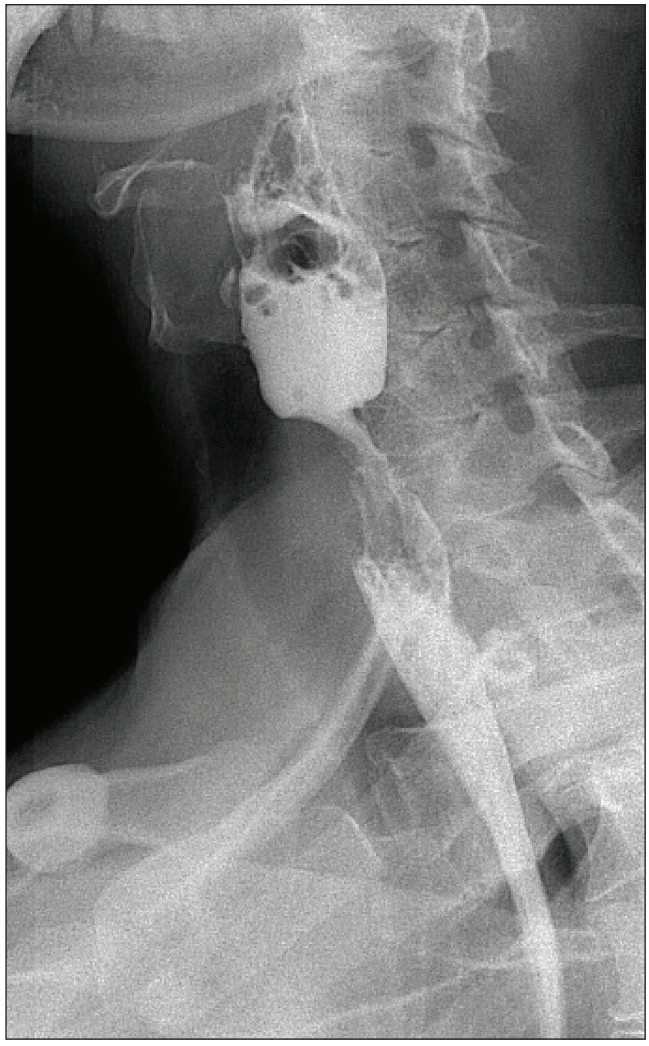


Figure 3. Barium study showed circumferential narrowing in upper esophagus with smooth mucosal surface consistent with the submucosal location of the lesion.

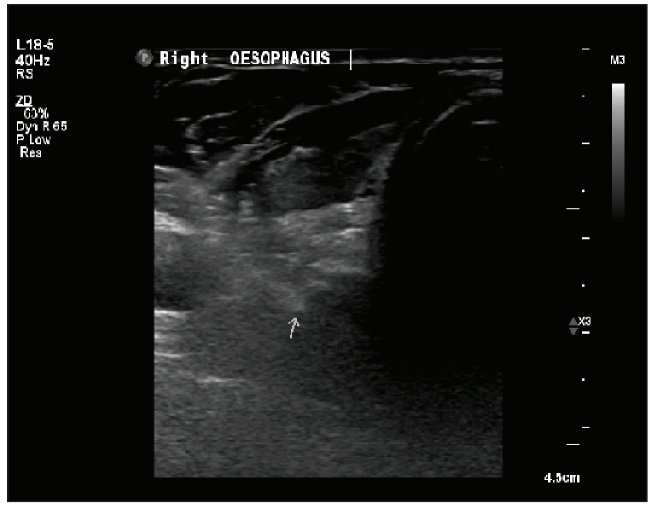


Figure 4. Ultrasound demonstrates a hypoechoic soft tissue lesion in the right side of the neck posterior to the right thyroid lobe and right inferior thyroid artery in the paratracheal region sowing no internal no definite vascularity. Guided core biopsy was performed, arrow indicates the tip of the needle.

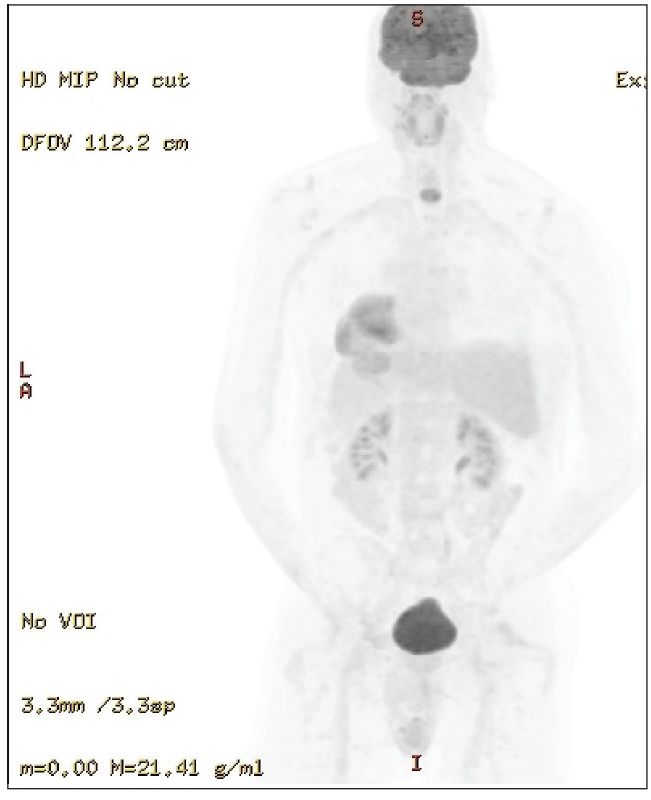


Figure 5. PETCT showed intense (SUVmax 8.8) FDG uptake by esophageal lesion.

our case. Similar incidents of diagnostic inaccuracy of endoscopy were documented in literature, which were diagnosed after surgical resection or postmortal autopsy [7,8]. The reasons are multifactorial and user dependent, one of the main reason is likely due to the layer of esophagus involved. As the adenocystic carcinoma usually involve submucosal layer, hence sometimes it might lead to sampling from inaccurate location due absence of direct visibility of tumor.

As endoscopy has limited role in OACC which most commonly shows fungating or polypoidal mass, imaging can play an important role in diagnosis and management.

Due to lack of enough cases, imaging findings are not available in literature. However, in our case, the imaging appearances were not of typical malignant lesion. It might be due to several reasons; firstly, submucosal location of the lesion, secondly, early detection before destruction of mucosal layer and lack of lymph nodal or distant metastasis at the time of detection.

Incidence of lymph nodal metastasis is less frequent in OACC as compared to other types like squamous cell carcinoma, however, nodal metastasis

indicate poorer prognosis. Despite being aggressive in nature, early detection have shown better prognosis [9].

Ultrasound guided percutaneous biopsy aided in diagnosis of the lesion. Fistula formation is a potential complication of this procedure in this location which did not occur in our case.

Conclusion

In our case, the patient was not very symptomatic and endoscopic findings were of reactive changes. Ultrasound guided percutaneous biopsy clinched the diagnosis. It is a safe procedure in expert hands where the visualization of the lesion using high frequency transducers is good. In our case the patient had a contrast swallow immediately post procedure to exclude oesophageal perforation almost immediately after the biopsy. In our report we have emphasised the importance of imaging for diagnosis of OAAC and highlighted the imaging appearances which is not available in literature.

References

1. Hiroyasu I, Hoichi K. Non-squamous cell carcinoma of the esophagus: the National Cancer Center Hospital, Tokyo, Experience. I to Cho 2005; 40(3):354–62.
2. Masao H, Sachiko K, Takaya K, Yasunori N, Akira K, Masahiro F. Non-squamous cell carcinoma of the esophagus. I to Cho 2005; 40(3):363–70.
3. Petursson SR. Adenoid cystic carcinoma of the esophagus. Complete response to combination chemotherapy. Cancer 1986; 57:1464–7.
4. Mafune K, Takubo K, Tanaka Y, Fujita K, Primary adenoid cystic carcinoma of the esophagus. Gan No Rinsho 1986; 32:513–9.
5. Khan AJ, DiGiovanna MP, Ross DA, Sasaki CT, Carter D, Son YH, et al. Adenoid cystic carcinoma: a retrospective clinical review. Int J Cancer 2001; 96:149–58.
6. Cerar A, Jutersek A, Vidmar S. Adenoid cystic carcinoma of the esophagus. A clinicopathologic study of three cases. Cancer 1991; 67:2159–64.
7. Shou C, Li Z, Yang W, Tjhoi W, Zhao Z Yu J. Adenoid cystic carcinoma of the gastroesophageal junction. Medicine 2019; 98(35):e16999.
8. Morisaki Y, Yoshizumi Y, Hiroyasu S, Shibata H, Terahata S, Tamai S, et al. Adenoid cystic carcinoma of the esophagus: report of a case and review of the Japanese literature. Surg Today 1996; 26:1006–9.
9. Kabuto T, Taniguchi K, Iwanaga T, Terasawa T, Sano M, Tateishi R, et al. Primary adenoid cystic carcinoma of esophagus. Cancer 1979; 43:2452–6.