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CASE REPORT

Case Report: Basal cell adenoma of esophagus, a rare tumor at an atypical site [version 1; peer review: 2 approved with reservations]

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Abstract

Basal cell adenomas are rare, salivary gland tumors. These are a class of non-malignant tumors that have glandular epithelial tissue origin. Often described to have a disputed recurrence rate after excision, the prognosis of basal cell adenomas is generally good. However, their rarity makes their continuous monitoring vital. Basal cell adenomas normally cause problems exerting a pressure effect on the surrounding organs or to the originating area. Often due to its location and effect, differentiation between other tumors, e.g. basal cell carcinoma, adenocarcinoma and cystic carcinoma, must be done. We were able to treat a patient with complaints of sensation of regurgitation that was diagnosed as an esophageal mass on esophagogastroduodenoscopy and later as basal cell adenoma on histopathological examination. The patient was treated with endoscopic submucosal dissection successfully and has had no fresh complaints since. Here, we present a case of basal cell adenoma in the esophagus, a rare entity in itself.

Keywords

basal cell adenoma, rare case, case report

Open Peer Review

Approval Status ? ?

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version 1	?	?
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1. Adriana Handra-Luca , APHP GHU Avicenne, Universite Paris Nord Sorbonne Cite, Bobigny, France		
2. Sadhna Dhingra , Baylor College of Medicine, Houston, USA		
Any reports and responses or comments on the article can be found at the end of the article.		

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Introduction

The National Cancer Institute, USA, describes adenoma as “a tumor that is not cancer. It starts in gland-like cells of the epithelial tissue (thin layer of tissue that covers organs, glands, and other structures within the body)”¹. Basal cell adenomas are a rare type of benign glandular tumors, salivary in origin. Accounting for less than 1–2% of salivary gland tumors², they are located usually superficial within the glandular body, and often a brownish appearance is observed³. First described by Kleinasser and Klein in 1967, they are termed as rare benign tumors with a high recurrence rate and, in general, good prognosis and recognized as an independent entity in the Second Edition of the Salivary Gland Tumors Classification of the World Health Organization⁴. Normally indigenous to (parotid) salivary glands, basal cell adenomas have also been reported in the buccal mucosa, palate and nasal septum. They are often seen in patients in their fifth-seventh decade of life, a contrast to other benign tumors⁵. Basal cell adenoma can be divided into four subtypes: solid, trabecular, tubular and membranous⁶. Histologically characterized by the presence of uniform and regular basaloid cells having two differentiated morphologies, the tumor cells are intermingled. Among the two groups, one has characteristic constitution of small cells with little cytoplasm and intense basaloid nuclei located near the tumoral nests while another is made up of large cells with abundant cytoplasm and pale nuclei in the centre of tumoral nests. The tumoral nests are separated from the surrounding connective tissues by a basal membrane like structure, which surrounds them⁵. Immunologically speaking, they strongly express PAN CK and CK 5/6, along with Calponin, P-63 and weak staining of Ki-67. Vimentin, actin also stain positive in basal cell adenomas, along with S-100 (alpha subunit) in ductal cells and S-100 (beta subunit) in basaloid cells.

Case report

A 63-year-old woman of Chinese (Han) origin was referred to Taihe Hospital, Department of Digestive Medicine after esophagogastroduodenoscopy (EGD) in her county hospital due to a protuberant mass in the esophagus, 35 cm from the incisors. The patient had initially complained of sensation of regurgitation and was a non-smoker and non-drinker. There was no history of

heartburn, chest pain, nausea, vomiting or diarrhea. She also had no complaints of hematemesis, hematochezia or melena. The patient was non-diabetic, non-hypertensive, with no history of renal disorders, cardiac diseases or tuberculosis. She had no remarkable family history (medically relevant) and no signs of genetic disorders. The patient had undergone appendectomy 13 years back and had been diagnosed and treated for intestinal obstruction 4 years previously. Laboratory investigation reports were normal. Upper abdominal and chest contrast-enhanced computer tomography (CECT) showed: lower esophagus and cardiac region of stomach wall hypertrophy (highly indicates presence of a tumor); right hepatic lobe diffuse (indicates small cyst present); B/L lung field showed some scattered fibrotic and calcified lesions.

EGD was repeated and showed a protuberant lesion in the esophagus, 3cm in size, 35 cm from the incisors (**Figure 1**). It also showed another protuberant lesion 0.7 cm in size in the gastric cardia. Multiple yellow spots (gastric xanthoma) were seen in the stomach. Pylorus, pyloric sphincter and duodenum were normal. Endoscopic ultrasound (EUS) of the protuberance in the esophagus showed that the lesion as hypoechoic, an originated from and limited to the submucosal layer with an area of more than 3 cm (**Figure 1**). No lymphadenopathy was found around the lesion. Radiofrequency ablation was done on the yellow spots.

With the working diagnosis of esophageal leiomyoma/polyp, the patient was scheduled for surgery. Endoscopic submucosal dissection (ESD) was done under general anesthesia (**Figure 2**). The surgery went well without complications. The protuberant mass was then sent for histopathological examination and the patient was kept for observation.

Immunohistology of the sample showed Calponin (+), CD117 (+), CD 56 partially (+), CKP (+), GATA-3(-), CK5/6 (+), CK7 (+), SOX 10(+), P63 (+), INI-1 (+), and Vimentin (+) (**Figure 3**). It also showed P63 (+), S-100(+), KI-67 about 2%. Light microscopy revealed the submucosal layer of esophagus showing glandular duct like structure also with double layer epithelium. Around the gland duct there was transparent

Figure 1. Left panel: Esophagus, 35mm from the incisors; middle panel: the base of the protuberance, closer view; right panel: endoscopic ultrasound (EUS) of the protuberance. (EGD by OLYMPUS EVIS LUCERA CV-260SL; EUS by OLYMPUS MAJ-935.)

Figure 2. Left and middle panels: Endoscopic submucosal dissection (ESD) removal of the mass under evaluation. Right panel: Closure of the defect by hemoclips. (ESD by single-use Olympus Dualknife™, tip thickness-0.3mm.)

Figure 3. Immunohistological staining of the tumor by different stains (1)HE Stain (2)SOX-10 (3)CD56 (4) CD117 (5)Calponin (6)Vimentin (7)S100 (8) P63 (9) IN1 (10)Ki67.

substance partially solid in nature. The sample also showed two types of epithelium-muscular epithelium and glandular epithelium, which corresponds with the diagnosis of basal cell adenoma, a type of salivary gland tumor. The patient was then discharged seven days after surgery with PPI (Cap. Lansoprazole 30mg BD for 45 days), Itopride (50mg TDS for 45 days) and Hydrotalcite (1gm TDS for 7 days) as discharge medication. Follow up was scheduled two months later.

Follow-up EGD of the patient was performed two months later and showed no recurrence of the tumor. The patient has had no fresh complaints since the ESD procedure (Figure 4).

Discussion

Basal cell adenomas are uncommon variants of benign salivary gland tumor with varying recurrence rates. There have been very

few reported cases of basal cell adenomas in ectopic sites. Its rarity makes comparison between two similar cases very difficult⁷. Basal cell adenomas are generally characterized by no regional node involvement, no calcification and no cystic component within the tumor⁸.

This case posed a challenge to our team since ectopic occurrences of basal cell adenoma have been reported very rarely in literature and is not a very common occurrence in itself. The prognosis of basal cell adenoma, when found at ectopic sites is not clear and thus poses a challenge to predict the outcome even after intervention.

Differentiation between benign and malignant basal cell adenoma is vital since they share the same site and characteristics. However, the mechanism and rate of progression is vastly

Figure 4. Site of endoscopic submucosal dissection (ESD) in follow-up after 2 months. The scar from ESD procedure is indicated by the red arrow.

different. Histology is not alone enough to differentiate or predict the progression, hence immunohistology plays a vital role in differentiation. Immunohistological staining with CD5/6, Keratin, Alpha-1 antichymotrypsin, CEA, S-100, Vimentin, CD117, Actin must be done to differentiate it from other benign and malignant tumors.

Whenever encountered and suspected, differentials must be established in favor of basal cell adenoma. Pleomorphic adenoma, adenoid cystic carcinoma, basaloid squamous carcinoma must be excluded first with the help of gross microscopic examination and immunohistological staining.

Surgical excision remains the first and foremost approach of choice for treatment of basal cell adenoma. However, during removal, it is mandatory not to disturb the tumor capsule to

prevent recurrence. Moreover, the patient must be kept under regular follow-up to monitor malignant transformation as well as recurrence.

Consent

Written informed consent was obtained from the patient for the publication of this case report and any associated images.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Grant information

The author(s) declared that no grants were involved in supporting this work.

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Version 1

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The authors describe an interesting and unique case of submucosal basal cell adenoma in the esophagus presenting as a symptomatic esophageal nodule, and diagnosed when resected by endoscopic submucosal dissection.

Endoscopic submucosal dissection is an innovative minimally invasive technique for resection of early cancers of gastrointestinal tract. It became popular in Japan in early 2000 for treatment of superficial gastric cancers and esophageal squamous cell carcinoma. It has gained momentum in other parts of the world for treatment of early superficial neoplastic lesions of the gastrointestinal tract. A byproduct is incidental detection of rare and unique submucosal lesions at unusual sites that present as a nodule and are amenable to endoscopic resection, such as this case.

The diagnosis rests on the histopathological description and supportive images. The histopathological description and the images provided in the case report to support the diagnosis are not very clear. Good quality microscopic description accompanied with quality images will enhance the quality of the submission. Only one hematoxylin and eosin (HE) stained image is provided. Addition of a couple of more HE stained images that depict clear unequivocal pathology of this lesion as basal cell adenoma will help improve the case report.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Partly

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

Yes

Is the case presented with sufficient detail to be useful for other practitioners?

Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Gastrointestinal and Liver Pathology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 20 June 2019

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Adriana Handra-Luca

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The authors report a case of basal cell adenoma of the esophagus.

- The authors could focus on benign tumors of the esophagus, as well rare epithelial tumors.
- The authors could add information on the 2 other lesions observed. (Biopsied or not biopsied?)
- The standard microscopy features should be noted before the IHC features.
- What do the authors mean by “muscular epithelium”?
- Indicators (arrows) could be added to photos.

Is the background of the case's history and progression described in sufficient detail?

Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?

Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?

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I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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