

Case Report

A Rare Subglottic Pleomorphic Adenoma: Magnetic Resonance Findings

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No previous study has published magnetic resonance imaging (MRI) findings for a subglottic pleomorphic adenoma. Here, we describe the case of a 62-year-old man with a subglottic pleomorphic adenoma. Endoscopic findings revealed a smooth-surfaced tumor arising from the subglottic posterior wall. MRI revealed the lesion as an isointense region on T1-weighted images, which was homogeneously enhanced. This lesion showed a heterogeneously hyperintense region on T2-weighted images. Diffusion-weighted imaging (DWI) showed slightly high intensity in the same area, with a normal or only slightly high apparent diffusion coefficient (ADC). Laryngomicrosurgery was performed for transoral excision of the subglottic tumor, resulting in a postsurgical diagnosis of pleomorphic adenoma.

Key words: subglottis, pleomorphic adenoma, MRI, transoral surgery

Pleomorphic adenomas are benign tumors that occur in the major or minor salivary glands. In the larynx, the glands are simple, tubular, and usually purely mucous, or large, branched, seromucous tubular-alveolar [1]. Pleomorphic adenomas are known to occur in the laryngeal regions; however, few reports of pleomorphic adenomas affecting the subglottic region have been published [2-6]. Moreover, no cases of subglottic pleomorphic adenomas evaluated using magnetic resonance imaging (MRI) have been reported. In this study, we report a rare case of a subglottic pleomorphic adenoma evaluated using MRI.

Case Report

Magnetic resonance scanning protocol. In this

case, we performed an MRI using a 3-T MR scanner (Achieva; Philips Medical Systems, Best, The Netherlands) with a head-neck coil. The imaging sequences were as follows:

(a) T2-weighted fast spin-echo imaging (T2WI) and fat-suppressed T2WI in the axial plane (repetition time [TR]/echo time [TE], 3,053/90 ms; echo train length, 19; matrix size, 269×352; field of view, 23 cm; slice thickness, 3 mm).

(b) T2WI and fat-suppressed T2WI in coronal plane (TR/TE, 3,053/90 ms; echo train length, 19; matrix size, 268×268; field of view, 23 cm; and slice thickness, 4 mm).

(c) T1-weighted fast spin-echo imaging (T1WI) in the axial plane (TR/TE, 452/5.9 ms; echo train length, 3; matrix size, 235×336; field of view, 23 cm; and slice thickness, 3 mm).

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(d) T1WI in coronal plane (TR/TE, 452/5.9ms; echo-train length, 3; matrix size, 245×352; field of view, 23 cm; slice thickness, 3 mm).

(e) Diffusion-weighted imaging (DWI) with *b*-values of 0 and 1,000 s/mm² in the axial plane using spin-echo type and single-shot echo planar imaging (EPI) sequences in parallel. (TR/TE, 6,000/76 ms; matrix size, 117×80; field of view, 23 cm; slice thickness, 3 mm).

(f) Post-contrast enhanced three-dimensional fat-suppressed T1WI (Gd-T1WI) in the sagittal plane (TR/TE, 4.7/2.1 ms; matrix size, 177×256; field of view, 23 cm; slice thickness, 0.9 mm). Gadobutrol was intravenously administered at a dose of 0.1 mmol per kilogram body weight. Axial and coronal images were obtained from sagittal images using multiplanar reconstruction (MPR).

A 62-year-old Japanese man underwent preoperative examination for surgical treatment of esophageal cancer at the Department of Surgery of our institution. The computed tomography (CT) scan incidentally revealed abnormal findings in the subglottic region, and the patient was referred to our department for further examination. The patient reported no subjective symptoms.

Endoscopic findings revealed a smooth-surfaced tumor arising from the subglottic posterior wall (Fig. 1A). The lesion was confined to the subglottic region, and mobility of the vocal folds was not affected. No other abnormalities were observed in the laryngopharyngeal region. Ultrasonography revealed no evidence of cervical lymphadenectomy. Moreover, CT

revealed an enhanced hemispherical mass at the subglottic posterior wall with no invasion of the surrounding tissue (Fig. 1B). MRI revealed a lesion 13 mm in diameter as an isointense region equal to the surrounding muscle tissue on T1WI, which was homogeneously enhanced. This lesion showed a heterogeneously hyperintense region on T2WI (Fig. 2). In this case, DWI showed slightly high intensity in the same area, and an apparent diffusion coefficient (ADC) that was normal to slightly high ($1.108 \times 10^{-3} \text{ mm}^2/\text{s}$).

Based on endoscopic and MR imaging findings, a benign submucosal tumor was suspected. Preoperatively, we explained to the patient the possibility of performing a tracheotomy or external incision if oral intubation or transoral resection of the subglottic tumor was difficult. However, an endotracheal tube with an internal diameter of 6.0 mm was inserted successfully. The tube was then shifted anteriorly using a Weerda laryngoscope to obtain a subglottic view, which allowed adequate visualization of the subglottic lesion. After the transoral excision, endoscopy confirmed the absence of residual lesions in the subglottic region. Histological examination of the excised specimen revealed a tumor consisting of a glandular structure of small round cells, including myoepithelial cells and small cysts (Fig. 3). The interstitium contained fibrous tissue with vitrification and myxoid stroma, resulting in a postsurgical diagnosis of pleomorphic adenoma. One year after surgical treatment, no signs of recurrence have been observed on imaging or upon physical examination (Fig. 4).

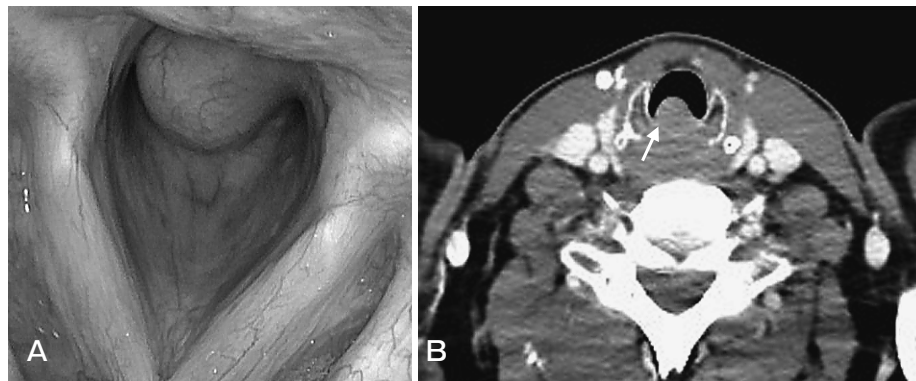


Fig. 1 Laryngeal and computed tomography (CT) findings at the initial visit. Endoscopic findings reveal a smooth-surface tumor arising from the subglottic posterior wall (A). Contrast-enhanced CT reveals an enhanced hemispherical mass in the subglottic posterior wall with no invasion of the surrounding tissue (B), as indicated by the arrow.

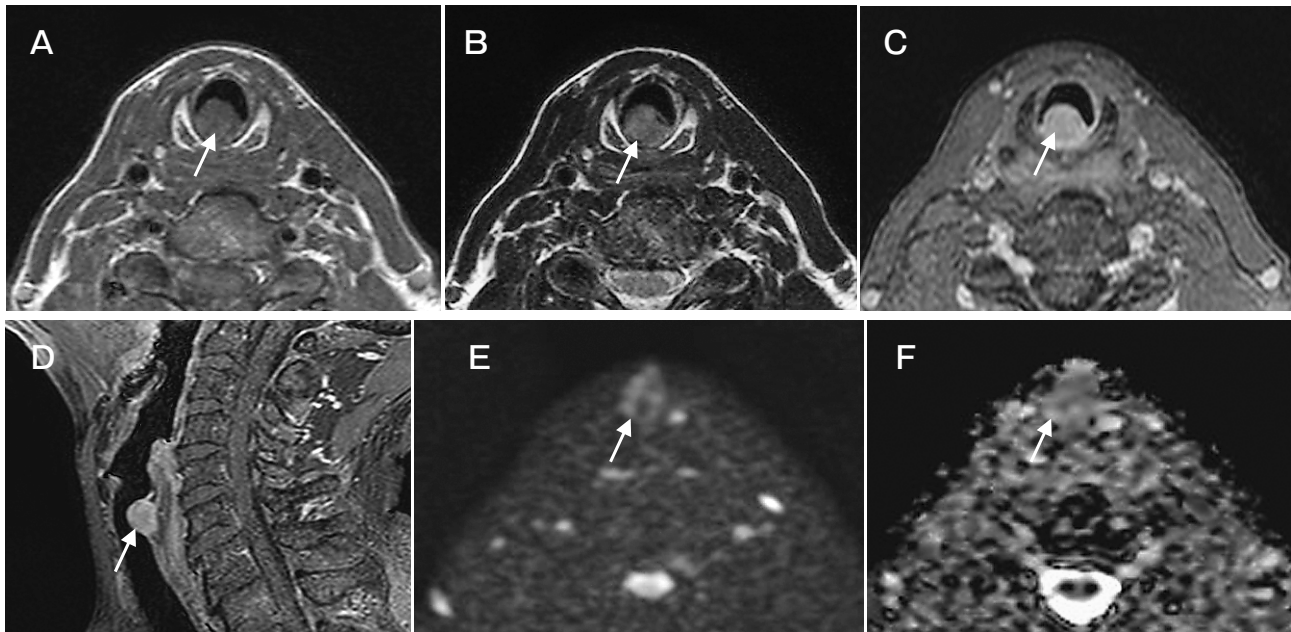


Fig. 2 Magnetic resonance imaging (MRI) findings of subglottic tumor. MRI displays a lesion 13 mm in diameter as an isointense region equal to the surrounding muscle tissue on T1-weighted imaging (**A**) and a hyperintense region on T2-weighted imaging (**B**). This region is homogeneously enhanced on T1WI (**C**, **D**), as indicated by arrows. DWI shows slightly high intensity (**E**), and ADC is normal or slightly high (**F**).

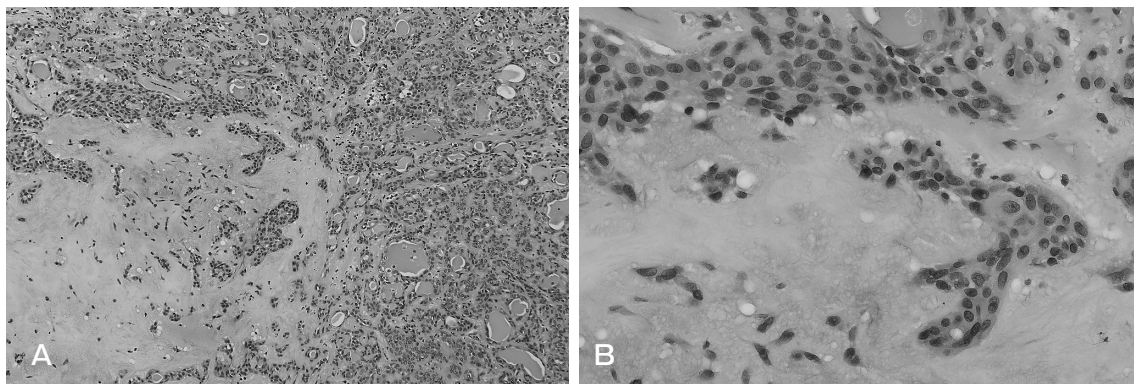


Fig. 3 Histological findings of resected specimen (hematoxylin and eosin stain). The tumor consists of a glandular structure of small round cells, indicating the presence of myoepithelial cells and small cysts. The interstitium contains fibrous tissue with vitrification and myxoid stroma (**A**, $\times 10$; **B**, $\times 40$).

Discussion

Only five cases of subglottic pleomorphic adenoma have been reported in the English literature [2-6]. In all previous cases, lesions were discovered after the onset of dyspnea, and three patients with subglottic pleomorphic adenoma underwent emergent tracheostomy [3,4,6]. In this study, a subglottic pleomorphic ade-

noma was discovered incidentally during a preoperative examination for esophageal cancer and was diagnosed without subjective symptoms. However, in any patient with chronic respiratory symptoms, the possibility of stenosis in the subglottic region should be considered. Observing the subglottic region during laryngoscopy is important for routine medical care.

In the present case, the lesion was resected as a

benign subglottic tumor, and histopathological examination revealed a pleomorphic adenoma. Benign tumors occurring in the subglottic submucosal region include hemangiomas and neurogenic tumors such as schwannomas, paragangliomas, neurofibromas, and solitary fibromas. Hemangiomas and paragangliomas are vascular lesions that can be easily differentiated from pleomorphic adenomas based on the color of the mass [7-9]. In contrast, other neurogenic tumors may present with a surface-smooth, spherical mass as an endoscopic finding, similar to those of the present case [10-12]. To our knowledge, only five cases of schwannoma, one case of neurofibroma, and one case of solitary fibrous tumor have been reported in the subglottic region in the English literature [10-16]. Details of our MRI findings are shown in Table 1. Similar MRI find-

ings have been reported for these diseases, suggesting that subglottic pleomorphic adenomas may mimic neurogenic tumors in terms of both local and MRI findings.

MRI findings for the imaging diagnosis of subglottic pleomorphic adenomas have not previously been reported. Parotid pleomorphic adenomas commonly show an isointense or hypointense signal on T1WI, a characteristic bright hyperintense signal on T2WI, and a high signal on both DWI and ADC [17-19]. In this case, the lesion was detected before the patient developed respiratory symptoms, allowing for an MRI evaluation. The lesion had findings characteristic of a parotid pleomorphic adenoma, except for those on T2WI, which showed a heterogeneously hyperintense signal. On T2WI, hyperintense areas correspond to an

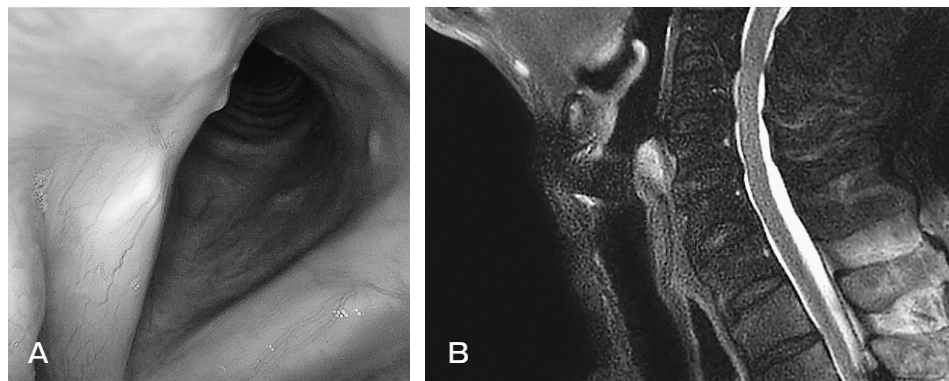


Fig. 4 Laryngeal and magnetic resonance imaging findings 1 year after the surgical treatment. No signs of recurrence are observed on imaging or endoscopy.

Table 1 MRI findings of subglottic tumors

	Author	MRI			
		T1WI	T2WI	Contrast-Enhanced	DWI
Schwannoma	Murata, T ⁽¹¹⁾ 2013	iso	high	homogeneously enhanced	–
	Takeda, K ⁽¹⁴⁾ 2003	low	high	–	–
	Pulli RS ⁽¹⁵⁾ 1997	iso	high	homogeneously enhanced	–
Neurofibroma	Tanaka ⁽¹³⁾ 1997	iso	high	homogeneously enhanced	–
Pleomorphic adenoma	Present case	iso	high	homogeneously enhanced	high

abundance of myxoid components, whereas hypointense areas indicate fibrosis [19]. We speculate that the mixture of fibrous tissue and myxoid stroma in the present case caused a heterogeneously hyperintense signal on T2WI. In a previous report on pleomorphic adenoma originating in the epiglottis, the tumor had high signal intensity on T2WI, low signal intensity on T1WI, and low signal intensity on the ADC map [20].

In the context of postoperative management of esophageal cancer, we decided to excise the subglottic pleomorphic adenoma transorally, without a neck incision. One patient diagnosed with malignant disease on preoperative biopsy underwent total laryngectomy, whereas the other underwent transoral resection or laryngofissure removal [2-6]. According to previous reports, tumors with a diameter of 18 mm or less can be removed transorally [3]. In our case, the laryngoscope provided an adequate operative field except for the lower edge of the tumor, and endoscopy confirmed the absence of residual lesions. No apparent recurrence was observed 1 year after surgery. Because sufficient margins were not obtained, continuous monitoring of the patient for recurrence has been necessary.

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