Tracheal granular cell tumour presenting with throat discomfort

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Abstract
Granular cell tumours (GCTs) are a rare form of neoplasm found throughout the body. Tracheobronchial involvement is less common. We describe a case of tracheal GCT in a 37-year-old Japanese woman presenting with throat discomfort. A tracheal tumour was found during laryngoscopy for undefined throat discomfort. Bronchoscopy demonstrated a white sub-epithelial solitary nodule on the tracheal wall, and pathological examination of the biopsy samples confirmed GCT. No therapeutic procedures were performed, and the tumour is currently under strict observation. Throat discomfort is a rare presentation of tracheal tumours, but an early inspection using laryngoscopy and bronchoscopy may be helpful in determining an accurate diagnosis.

Keywords
Bronchoscopy, granular cell tumour, throat discomfort, tracheal tumour.

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Introduction
Granular cell tumours (GCTs) are a rare form of mostly benign soft tissue neoplasm. They may be found anywhere in the body, and more than half of primary lesions are found in the head and neck. GCTs in the tracheobronchial tree are less common. Tracheal GCTs usually present with symptoms such as respiratory distress, stridor, and cough. Throat discomfort is a rare complaint in tracheal GCT cases. Here, we describe a case of tracheal GCT presenting with throat discomfort.

Case Report
A 37-year-old Japanese woman presented to the otorhinolaryngology clinic with a 1-month history of throat discomfort. She had undergone a surgery for pharyngeal squamous cell papilloma 4 years ago and had a previous smoking history of 10 pack-years. Laryngoscopy did not indicate a pharyngeal papilloma recurrence or abnormal findings in the upper airway but detected an intraluminal tumour in the proximal trachea. She was referred to our hospital for further investigation. Physical examination and routine laboratory tests were unremarkable. A chest X-ray did not show any abnormal findings. Neck and chest computed tomography (CT) showed a solitary intraluminal tumour of the trachea at the level of the thyroid gland (Fig. 1A). The lesion measured 6 × 6 × 3 mm, and there was no apparent extra-tracheal extension on the CT scan. Following radiological examinations, a diagnostic bronchoscopy demonstrated a white sub-epithelial solitary nodule on the left side of the cartilage portion of the trachea (Fig. 1B), which corresponded to the CT findings. The tumour was well defined from surrounding normal epithelium. No other tumours were found in the distal tracheobronchial tree. Microscopic examination of the bronchoscopic biopsy specimens showed a dense proliferation of large tumour cells under the epithelial cell layer on haematoxylin and eosin (H&E) staining (Fig. 2A). The cytoplasm of the tumour cells was abundant with fine and eosinophilic granules, a characteristic finding in GCTs. Immunostaining for S-100 protein and CD68 were positive (Fig. 2B). The nuclei were pyknotic but did not show atypia, and Ki-67-positive tumour cells accounted for <1% on immunostaining. These findings indicate a pathological diagnosis of a benign tracheal GCT.

Treatment options, including tracheal sleeve resection, endoscopic tumour resection, and endoscopic laser...
ablation, were explained to the patient. However, she opted to observe the tumour over follow-up appointments. A repeat CT after 1 month did not show interval changes in tumour size.

Discussion

We describe a rare tracheal GCT in a patient who had throat discomfort as the initial symptom. The patient’s complaint was initially suspected to be associated with a metachronous recurrence of pharyngeal papilloma; however, laryngoscopy showed a solitary tracheal tumour.

GCTs are believed to arise from neural cells and are characterized pathologically by abundant cytoplasmic granules visible on H&E staining and positive staining for neural cell markers such as S-100 protein. While tumours can be found in the whole body, tracheobronchial GCT cases are less common. A registry survey of the Dutch Network and National Database for Pathology conducted between 1990 and 1999 reported 11 tracheal and 19 bronchial GCT cases in a population of about 15 million (0.07 cases of tracheal GCT annually per a million individuals) [1]. A study by Guarnieri et al. in 2014 found only 40 cases of tracheal GCTs in the English literature [2], including the cases in the Dutch registry survey. Initial symptoms were described in 25 of 40 cases of tracheal GCTs [2], including stridor/wheeze (n = 8), respiratory distress (n = 6), and cough (n = 5). Haemosputum/haemoptysis (n = 2), chest pain (n = 1), or dysphagia (n = 1) was less common. Asymptomatic GCT cases were also common (n = 7), in which tracheal tumours were accidentally found during examinations for other purposes. There have been

![Figure 1](image1.png)

**Figure 1.** Computed tomography showed a solitary intraluminal tumour of the trachea at the level of the thyroid gland (arrow mark) and no apparent extra-tracheal extension (A). Bronchoscopy showed a white sub-epithelial solitary nodule on the left side of the cartilage portion of the trachea (B).

![Figure 2](image2.png)

**Figure 2.** Pathological features of the tracheal tumour (400x magnification) on haematoxylin and eosin staining (A) and on immunostaining for S-100 protein (B). A dense proliferation of large tumour cells is seen under the epithelial mucosa. Fine and eosinophilic granules are abundant in the cytoplasm of the tumour cells (A). The tumour cells are positive for S-100 protein (B).
no previous reports of throat discomfort as an initial manifestation. An association between the patient’s complaint and the existence of tracheal GCT was not definitely determined in this study. However, the complaint persisted for several months, and no other cause was found.

Regarding the treatment options for tracheal GCTs, surgical resection is highly curative, but the procedure is substantially invasive, with a perioperative mortality of 14% [3]. In contrast, endoscopic tumour resection is less invasive and safer than surgical resection. However, local recurrence is observed in up to 50% of treated cases [3]. Endoscopic laser ablation is an alternative option to be considered, but not curative. In the present case, the patient hesitated to receive any invasive procedures for the less symptomatic benign tumour. Hence, we agreed to keep the tumour under observation. While a CT scan is less invasive and sensitive enough for routine checking of the tracheal tumour, bronchoscopy should be performed to yield detailed information when needed. Malignant behaviour in GCTs is a little concerning during long-term observation, reportedly accounting for 1–2% of the cases [3]. In the present case, pathological examination did not show any malignant features such as necrosis, spindle-formed cells, high nuclear/cytoplasmic ratio, or high Ki-67-positive tumour cells.

In summary, we report a rare case of tracheal GCT presenting with throat discomfort as an initial symptom. While the symptom was atypical for a tracheal tumour, early laryngoscopy and bronchoscopy led to the correct diagnosis. Clinicians should be made aware that throat discomfort can be an unusual manifestation of tracheal GCT.

Disclosure Statement

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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