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Ectopic thyroid tissue in the airway: a case report

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Abstract

Background Ectopic thyroid tissue (ETT) is a rare congenital anomaly caused by the abnormal embryonic migration of thyroid tissue, leading to its presence outside its usual pretracheal location. This condition can lead to diagnostic challenges, especially when located within the airway, as it mimics other respiratory disorders such as asthma.

Case presentation We report the case of a 69-year-old man with endotracheal ETT presenting with severe dyspnea, and the lesion was initially suspected to be malignant. The diagnosis of ETT was confirmed through bronchoscopy and histopathological examination. The patient underwent successful endoscopic interventional therapy, resulting in significant symptom improvement and complete resolution of the airway lesion.

Conclusion Although rare, ETT should be considered in the differential diagnosis of unexplained respiratory symptoms to prevent misdiagnosis and permit effective, minimally invasive treatment options such as endoscopic resection, which can significantly improve patient outcomes.

Keywords Ectopic thyroid tissue, Airway obstruction, Diagnosis, Treatment, Case report

Background

Ectopic thyroid tissue (ETT) describes the presence of thyroid tissue in locations other than its usual pretracheal position. This condition is caused by aberrant embryogenesis during the thyroid gland's descent from the foramen cecum to its final anatomical location. ETT is a rare event, potentially manifesting anywhere along the midline from the base of the tongue to the mediastinum. Its incidence is estimated as 1 in 300,000 individuals. Although ETT lesions are generally benign, they can sometimes present challenges in diagnosis and management, as histologically, ETT is indistinguishable from the main thyroid gland [1]. Malignancy occurs in approximately 1% of cases of ETT [2], and it should be considered in the differential diagnosis of pathological masses located in the central airway [3].

ETT accounts for approximately 48–61% of thyroid developmental anomalies [4], making it a common thyroid malformation. ETT typically manifests along the thyroglossal duct tract, which is the developmental pathway of the thyroid gland. Most cases of ETT involve the base of the tongue, whereas the laryngotracheal region is the least common location [5]. However, ETT in the laryngotracheal region holds clinical significance because of its potential to cause upper airway obstruction [6]. ETT can occur at any age, although it is most observed between the ages of 30 and 50 years. ETT exhibits female predominance with a ratio of approximately 3:1 [7].

Diagnosing ETT is especially difficult when it arises within the airway, often leading to a misdiagnosis of asthma and subsequent delays in appropriate

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management. Additionally, associated hypothyroidism can cause ETT enlargement, further worsening respiratory symptoms. Another key challenge in managing ETT is distinguishing it from malignancy. Therefore, routine follow-up imaging or invasive diagnostic procedures, such as surgical resection, are required to achieve a definitive diagnosis.

In this study, we discussed a case of ETT within the airway that was initially suspected to be malignant, and the lesion was treated successfully with endoscopic interventional therapy.

Case presentation

A 69-year-old man presented with intermittent dyspnea, cough, and sputum production for more than a year, and he experienced symptom exacerbation for the last two months. Despite the intermittent use of traditional Chinese medicine (unclear), there was no significant improvement. Two months before presentation, the patient's dyspnea worsened, and chest computed tomography (CT) identified a bronchial mass. He self-administered antibiotics, but his symptoms persisted. Additionally, he reported poor appetite, sleep disturbance, and the loss of approximately 10 kg in weight over the past year. His medical history included coronary artery disease for 5 years and thyroidectomy for benign thyroid nodules 5 years ago before presentation. He denied any history of smoking or alcohol consumption. He was currently on regular levothyroxine therapy (50 μg/day).

On admission, the patient was lethargic with cyanotic lips and tachypnea. His vital signs were as follows: temperature, 36.5 °C; heart rate, 98 beats/min; respiratory rate, 24 breaths/min; and blood pressure, 142/81 mmHg. Physical examination revealed bilateral coarse breath sounds with inspiratory crackles and minimal moist rales in the left lower lung. Cardiac and abdominal examinations were unremarkable. The results of laboratory tests, including a complete blood count, coagulation profile, liver and renal function tests, thyroid function tests (TSH, 2.263 mIU/L; T4, 93.33 nmol/L; FT3, 15.90 pmol/L; T3, 1.37 nmol/L; FT3, 4.6 pmol/L), and assessments of markers for autoimmune and infectious conditions, were normal excluding a low blood potassium level of 2.82 mmol/L. Arterial blood gas analysis (with nasal oxygen at 4 L/min) revealed the following findings: pH, 7.46; PCO_2, 60 mmHg; PO_2, 65 mmHg; lactate, 0.8 mmol/L; HCO^{3-}, 42.7 mmol/L; base excess, 15.8 mmol/L; and oxygen saturation, 94%. Enhanced chest CT (Fig. 1) revealed an approximately 18-mm nodular soft tissue mass in the trachea with a density of 82 Hounsfield units. The mass exhibited heterogeneous enhancement and caused significant luminal narrowing, and it featured indistinct borders from the surrounding cartilage and the left thyroid lobe, raising concerns of malignancy.

Flexible bronchoscopy (BF-P290, Olympus Corporation, Tokyo, Japan) was utilized for both examination and

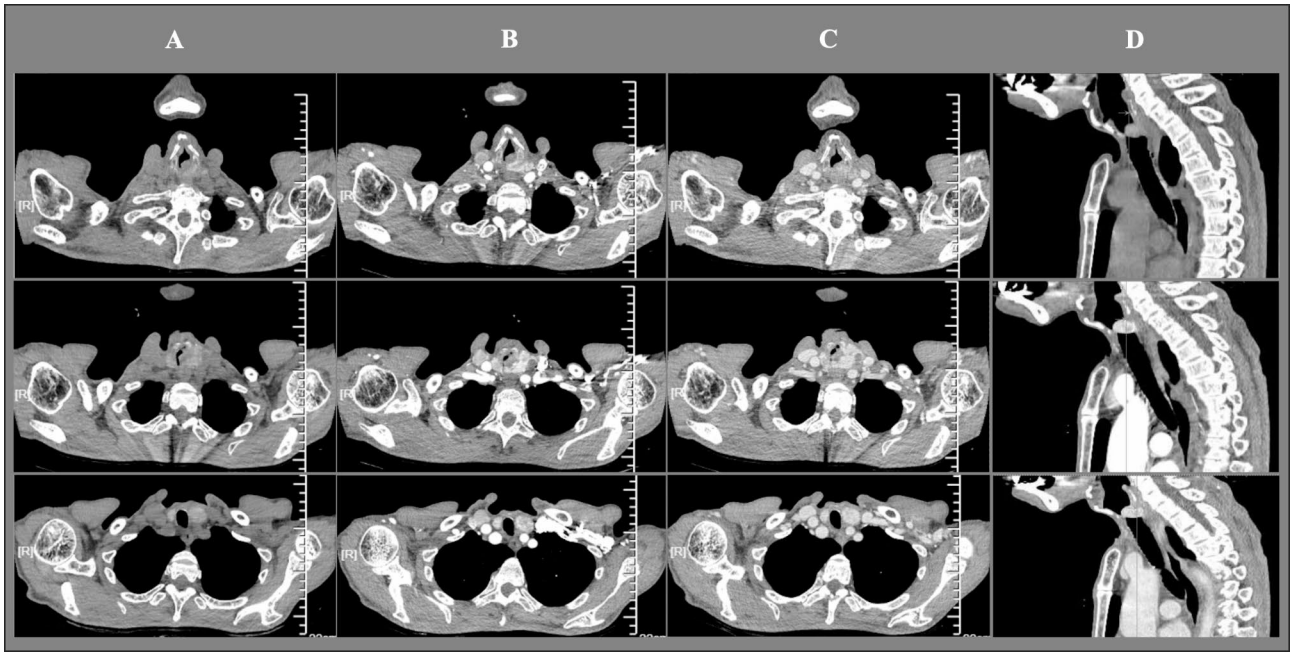

Fig. 1 Chest computed tomography (CT; **A**.**B**.**C**: axial view, **D**: sagittal view) revealed a nodular soft tissue mass in the trachea measuring approximately 18 mm in diameter with a CT attenuation value of 82 Hounsfield units. The mass exhibited heterogeneous enhancement and indistinct borders from surrounding structures and contributed to significant narrowing of the airway lumen

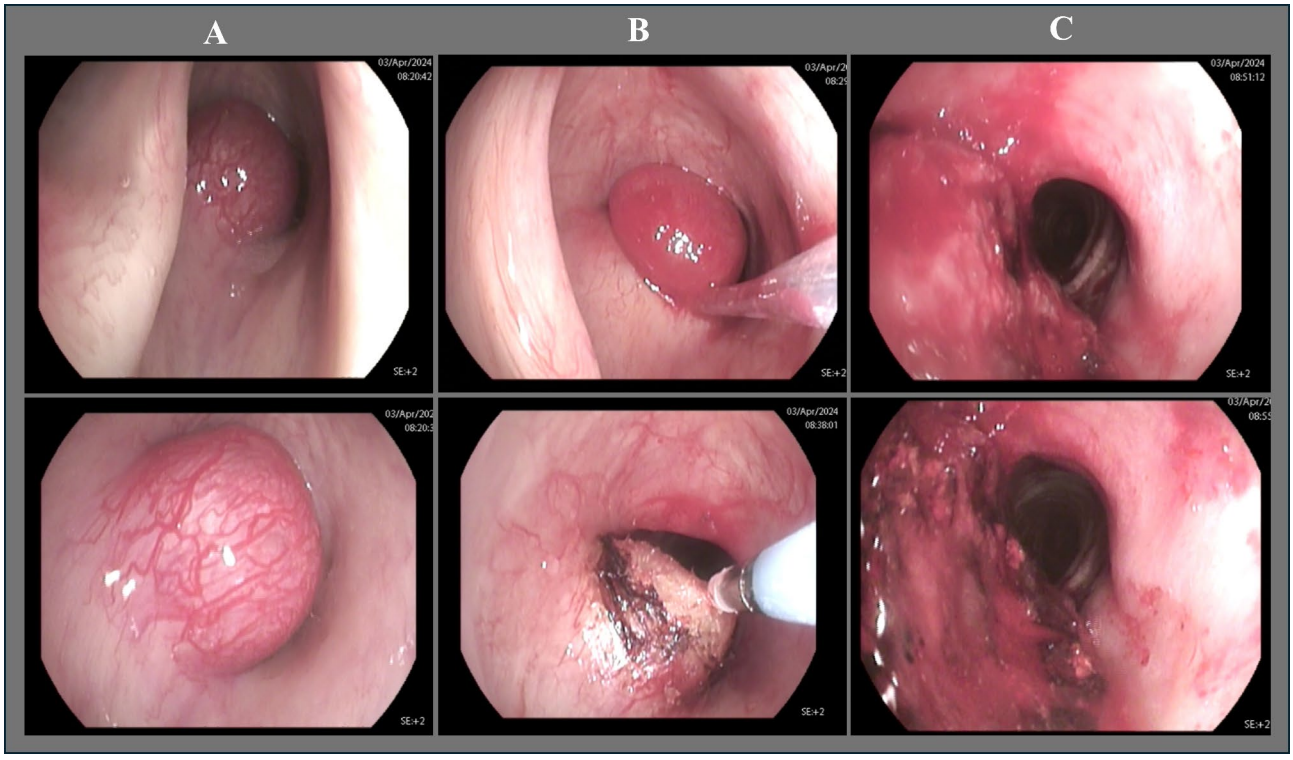

Fig. 2 **(A)** Flexible bronchoscopy revealed a smooth-surfaced lesion obstructing the upper segment of the main airway located approximately 1.0 cm below the vocal cords. **(B)** The lesion was treated using snare resection, argon plasma coagulation, and cryotherapy. **(C)** Post-procedure, the main airway stenosis was significantly improved

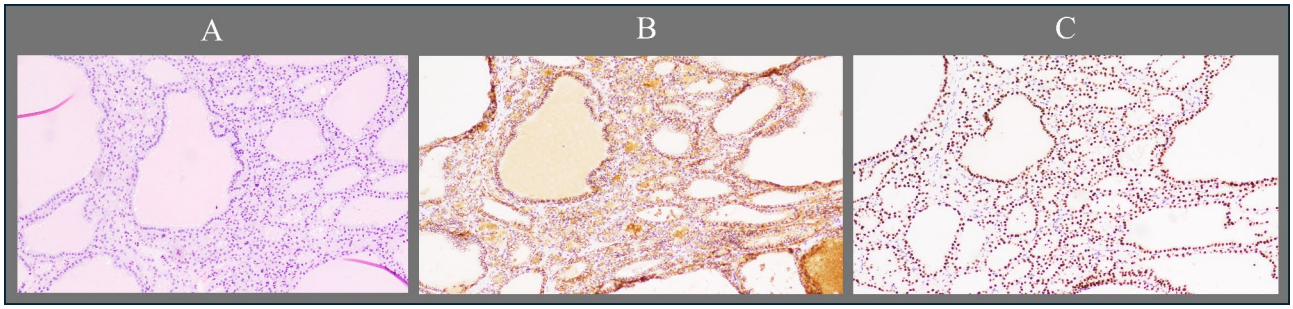

Fig. 3 Histopathological examination of ectopic thyroid tissue. **(A)** Hematoxylin and eosin staining revealed follicles of varying sizes lined by a single layer of follicular cells with either flattened or short columnar morphology and eosinophilic cytoplasm. **(B)** Immunohistochemical analysis revealed positive staining for CD56. **(C)** Immunohistochemistry demonstrates negative staining for thyroid transcription factor-1. All images were taken at ×100 magnification

bronchoscopic interventions. Prior to these interventions, general anesthesia was induced, and a laryngeal mask airway was used for ventilation, with nasal oxygen supplemented as needed. It revealed a smooth-surfaced lesion obstructing the upper segment of the main airway approximately 1.0 cm below the vocal cords, with severe luminal narrowing (Fig. 2). Tumor resection was performed with a snare, followed by hemostasis using lyophilized thrombin powder locally and snake venom-derived thrombin systemically. Residual neoplastic tissue was ablated with argon plasma coagulation and cryotherapy. After the procedure, the main airway stenosis was significantly improved. Histopathological examination of the resected tissue (Fig. 3) revealed thyroid tissue in the bronchial submucosa without atypical cellular features. Immunohistochemical staining of the biopsy indicated the presence of ETT. Staining disclosed positivity for cyclin D1 (30%), CD56, paired box gene 8 (PAX-8), p63, and thyroglobulin and negativity for BRAFV600E, P40, and thyroid transcription factor-1 (TTF-1).

After treatment, the patient experienced significant improvement in dyspnea, and he was discharged. One month later, follow-up chest CT revealed complete resolution of the airway lesion. (Fig. 4). Meanwhile, the

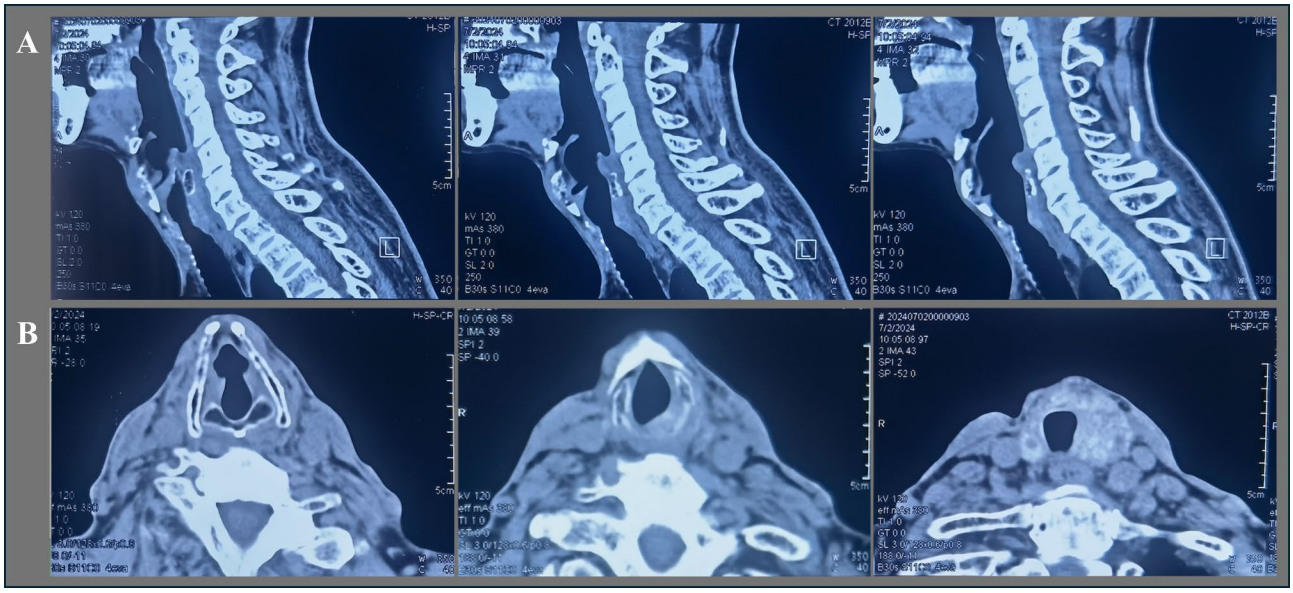

Fig. 4 After 1 month of follow-up, the cross-sectional and sagittal planes of computed tomography revealed no obvious abnormal tissue in the airway, and no stenosis was observed in the trachea

patient's thyroid function was normal. During 5 months of follow-up after surgery, the patient did not experience any respiratory distress symptoms, and his quality of life improved. Additionally, he continued to take 50 μg levothyroxine daily.

Discussion and conclusions

ETT is attributable to the failure of normal thyroid migration along the thyroglossal duct during early embryonic development, and it can occur at any age [8, 9]. However, the exact pathogenesis of endotracheal thyroid tissue remains unclear. Two main theories have been proposed to explain this pathological finding. The first theory suggests that the thyroid gland is divided by the forming trachea and its cartilaginous rings during fetal development, resulting in endotracheal thyroid tissue [10]. The second theory posits the existence of an ingrowth of thyroid tissue into the tracheal lumen [11, 12]. Other potential causes of ETT include trauma and invasive procedures that can lead to the implantation of thyroid tissue, as well as the locoregional spread of thyroid malignancies from the thyroid gland or nearby structures [13]. Additionally, hormonal imbalances can contribute to the development of ETT. However, these occurrences are much rarer and less well understood. Recent genetic research has identified several key genes, including TTF-1, TTF-2, and PAX-8, with critical roles in thyroid gland migration and development [14, 15]. Mutations in these genes might contribute to the abnormal migration of thyroid tissue, leading to ectopic locations such as the trachea.

ETT is most frequently located at the base of the tongue, but it can also arise in the head and neck, chest, digestive system, urinary system, and reproductive system [16]. Its manifestations vary depending on the location, with ETT within the airway leading to symptoms such as progressive dyspnea, stridor, cough, difficulty swallowing, and hemoptysis [17–19]. Dyspnea induced by ETT is often misdiagnosed as asthma, and distinguishing stridor from wheezing during a physical examination can be challenging [10]. However, once ETT is considered, the diagnosis can be straightforward, and it is often confirmed by bronchoscopic examination [10, 20]. ETT typically occurs alongside a normally functioning orthotopic thyroid gland, resulting in a euthyroid state. Many patients remain asymptomatic until a change in the endocrine status occurs, and this change can be triggered by puberty, menarche, pregnancy, or menopause. ETT growth can be stimulated by factors such as TSH, epidermal growth factor, and human chorionic gonadotropin [21]. In this case, the presence of ETT in the airway caused severe respiratory distress. Because of the rarity of this presentation, initial imaging and presumptive diagnoses could result in the misdiagnosis of an endotracheal tumor, posing life-threatening risks such as suffocation. Therefore, a thorough bronchoscopic examination was essential to establish a definitive pathological diagnosis.

Airway stenosis caused by ETT presents significant challenges, particularly in older adults with comorbidities. The normal trachea diameter ranges 13–25 mm (averages 19.5 mm) in men [22]. In this case, an 18-mm mass led to severe luminal narrowing, resulting in intense dyspnea and respiratory distress. The severity of stenosis is correlated with symptoms; specifically, mild cases might mimic asthma with chronic cough [23],

whereas moderate-to-severe cases can cause progressive dyspnea, stridor, and even acute respiratory failure, necessitating urgent intervention [24]. Management of ETT-induced airway stenosis depends on the obstruction's severity and the patient's overall health. Endoscopic interventions, such as snare resection, argon plasma coagulation, and cryotherapy, are preferred because of their minimally invasive nature, shorter recovery times, and lower complication risks. In this patient, these procedures effectively alleviated the airway obstruction and ablated the lesion. However, surgical excision is reserved for cases where endoscopic methods are insufficient or when malignancy is suspected, although it increases the risk of morbidity and requires specialized surgical expertise [25].

The treatment options for ETT include thyroid suppression therapy, radioiodine ablation, and surgical excision. Surgical excision is typically prioritized in cases of airway obstruction and recommended for ETT, as indicated by a systematic review of benign intratracheal thyroid cases [26, 27]. Surgical approaches vary, ranging from open cricoid procedures to endoscopic laser-assisted techniques [28]. For asymptomatic patients with ETT and normal thyroid function, clinical intervention is generally not advised. Due to the fact that ectopic thyroid tissue is usually nonfunctional, some scholars suggest that patients with asymptomatic ETT take exogenous thyroid hormone to prevent the eventual occurrence of hypothyroidism in most patients [29]. Treatment decisions for symptomatic patients should consider the ectopic gland's location, size, clinical presentation, and thyroid function status. Radioiodine ablation is less desirable because of the risks of radiation thyroiditis and radiation-induced tracheitis and the potential for endotracheal ETT swelling, which can lead to upper airway obstruction. Additionally, its potential ineffectiveness is a concern, particularly because ETT might not efficiently accumulate iodine. Additionally, radioiodine treatment could destroy a coexisting orthotopic thyroid gland, which is usually present with ETT. In cases where biopsy is required for histological diagnosis, fiberoptic laryngoscopy is recommended to visualize the mass, which typically appears as a broad-based submucosal lesion on the lateral subglottic or upper tracheal wall. Additionally, integrating imaging tools such as CT, magnetic resonance imaging, and ultrasound can significantly enhance the effectiveness of fiberoptic laryngoscopy. These imaging modalities provide detailed anatomical information, enabling the precise localization and characterization of lesions. Consequently, clinicians can achieve more accurate diagnoses and optimize the management of submucosal masses within the airway. Caution is needed during biopsy because of the risk of significant bleeding from ETT [10]. In this case, flexible bronchoscopy with interventional therapy was chosen, leading to marked symptom improvement and rapid recovery. Nonetheless, endobronchial interventions near the vocal cords entail several risks, including bleeding from nearby major vessels, airway obstruction attributable to swelling or bleeding, and potential damage to delicate structures such as the vocal cords. Additionally, there is an increased risk of infection. An additional follow-up demonstrated excellent outcomes. This report is limited by the absence of detailed information regarding the patient's previous thyroidectomy and thyroid function tests, which restricts our understanding of the ectopic tissue's origin and the potential impact of prior thyroid conditions.

In conclusion, although ETT is a rare cause of upper airway obstruction, it might be more common than previously believed, and it should be considered in the differential diagnosis, especially when patients present with unexplained respiratory symptoms. ETT is often unrecognized until the ectopic tissue hypertrophies to a degree that causes clinical symptoms. Importantly, ETT can be effectively managed with endoscopic resection, offering a minimally invasive treatment option that can significantly relieve symptoms and improve patient outcomes.

Abbreviations

ETT	Ectopic thyroid tissue
PAX-8	Paired box gene 8
TTF-1	Thyroid transcription factor-1

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Author contributions

Qun Zhang and Yanling Xu: Conception and design of study; Lin Zhou, Wei Li and Yanling Xu: Data analysis and/or interpretation; All authors: Drafting of manuscript and/or critical revision, Approval of final version of manuscript.

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Data availability

All data generated or analyzed during this study are included in this published article. Further enquiries can be directed to the corresponding author.

Declarations

Ethics approval and consent to participate

Ethics approval was not required because this is a case report. This study was conducted in compliance with the Declaration of Helsinki. Written informed consent was obtained from the individual participant included in the study.

Consent for publication

Written informed consent was obtained from the individual for the publication of any potentially identifiable images or data included in this article.

Competing interests

The authors declare no competing interests.

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