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Case Report

A three dimensional printed endobronchial stent for the treatment of a broncho-esophageal fistula

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ABSTRACT

Broncho-esophageal fistulas (BEFs) are a rare but serious complication that can occur after esophagectomy, often resulting in aspiration, respiratory issues, and infection. Management depends on fistula size and location, with options including conservative treatments, surgical closure and stenting. Conventional treatment involves esophageal stents, which may be insufficient for larger or more complex fistulas. This case report describes the first use of a 3D-printed airway stent in combination with an esophageal stent to treat a broncho-esophageal fistula. A 70-year-old patient with distal esophageal adenocarcinoma, treated with neoadjuvant chemo-radiation, underwent robot-assisted minimally invasive esophagectomy. The procedure was complicated by a broncho-esophageal fistula, leading to multiple interventions. Despite dual stenting with a custom 3D airway stent, the fistula persisted, and the patient was transitioned to supportive care due to disease progression. This case, the first to use a 3D-printed airway stent for a broncho-esophageal fistula, demonstrates that the stent did not achieve closure, likely due to excessive pressure against the endobronchial wall. This underscores the need for improved 3D stent designs, offering important insights for interventional pulmonologists.

1. Introduction

A broncho-esophageal fistula is an uncommon yet significant complication that can occur after subtotal esophagectomy with gastric tube reconstruction [1]. Symptoms associated with broncho-esophageal fistulas include dyspnea, coughing and aspiration [2]. In case of small fistulas conservative treatment may be considered [3]. Treatment options for larger fistulas encompass surgical procedures or interventions, such as direct closure with well-vascularized tissue, closure utilizing local injection of biologic or chemical glue, as well as laser and argon plasma coagulation (APC) thermal ablation, or esophageal and/or airway stenting [4,5]. When opting for stenting, esophageal stents are mainly suitable for lower fistulas. The location and dimension of the fistula determine the stent length and diameter required. When fistulas are larger (>20 mm), or if an esophageal stent narrows the airway significantly, an esophageal stent is recommended to be combined with an airway stent [4,6]. If placing an esophageal stent is not feasible because there is a significant risk of stent migration, then opting for placement of an airway stent alone may be preferable [4]. Numerous cases

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have been documented involving patients with broncho-esophageal fistulas who underwent dual stenting of both the airway and esophagus, primarily for symptom relief [2,7]. Depending on the anatomy of the broncho-esophageal fistula and underlying disease, a variety of airway stents are available to choose from in terms of material (silicone, metallic and biodegradable) and shape (straight, L-shaped, Y-shaped). A new addition to the current arsenal of available stents is the three-dimensional (3D) printed silicon airways stent [8]. A 3D endobronchial stent is a custom silicone stent designed by an interventional pulmonologist for cases where a standard stent does not adequately fit the unique anatomy of a central airway obstruction [9,10]. These stents provide a personalized design achieving congruence to the airway, instead of the current limited choices in stent dimensions. The design of a 3D stent requires a CT scan, based on which a virtual 3D model is created [11,12]. For the final design, the interventional pulmonologist must provide input on various parameters, including the stent shape, maximal outer diameter and corresponding thickness (in mm), minimal outer diameter and corresponding thickness (in mm), and stent length [8]. Some cases have been reported of patients with a tracheo-bronchial stenosis treated with a 3D printed airway stent [8]. In the currently running phase II Treatment of Central Airway Stenoses Using Computer-Assisted Customized 3D Stents (TATUM) trial (www.clinicaltrials.gov:NCT04848025), the researchers investigate whether 3D printed airway stents will provide long-term safety and efficacy for all patients planned for airway stent placement. However, no cases have been reported of patients with broncho-esophageal fistulas being treated with a 3D printed airway stent. Here we present the first case of a patient with a broncho-esophageal fistula treated with a customized airway stent in combination with an esophageal stent.

2. Case report/case presentation

A 70-year-old patient with adenocarcinoma of the distal esophagus was referred to our hospital for treatment as part of the Surgery

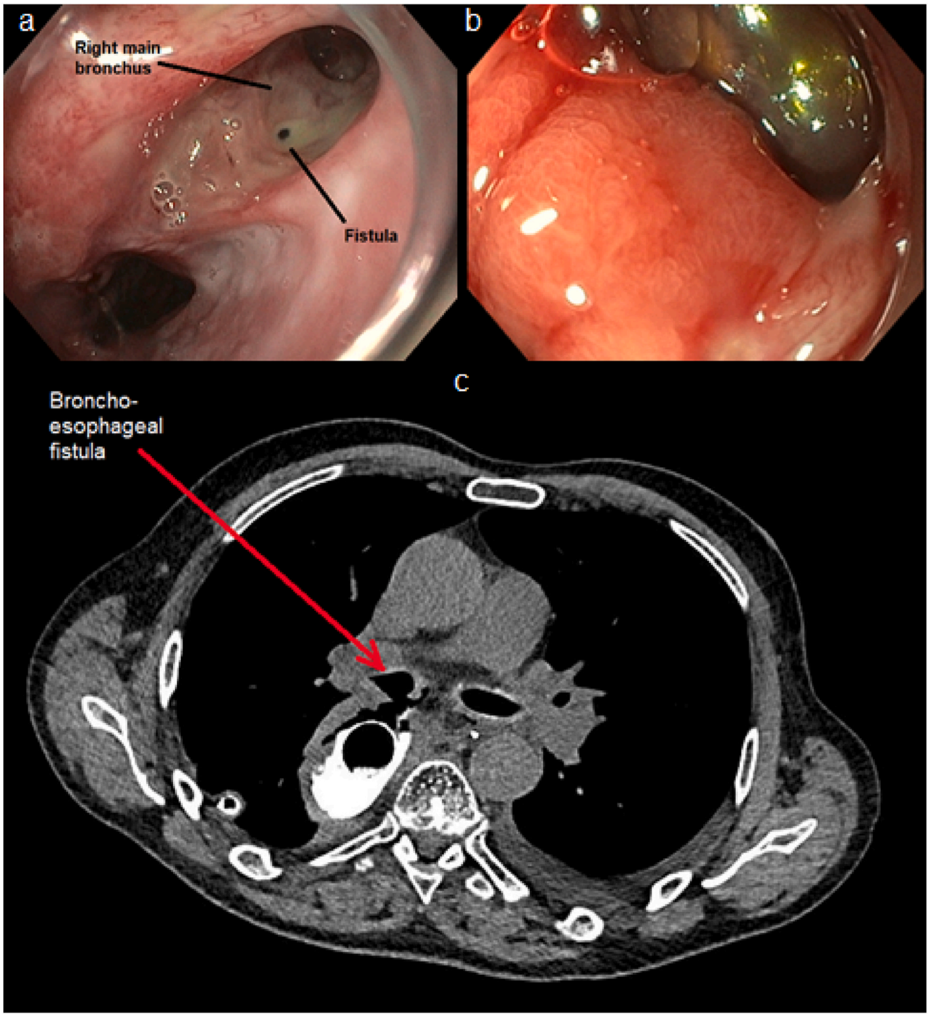

Fig. 1. | A. Anatomy of the broncho-esophageal fistula as seen with bronchoscopy. B. View of the endobronchial stent as seen with gastroscopy from the gastric conduit. C. Radiologic view of the broncho-esophageal fistula.

As Needed for Esophageal cancer (SANO)-2 study (www.clinicaltrials.gov: NCT04886635), a prospective cohort study on active surveillance after neoadjuvant chemoradiation according to the ChemoRadiotherapy for Esophageal Cancer followed by Surgery Study (CROSS) regimen [13] (intravenous carboplatin [AUC 2 mg/mL per min] and intravenous paclitaxel [50 mg/m^2 of body surface area] for 23 days) with concurrent radiotherapy (41.4 Gy, given in 23 fractions of 1.8 Gy, 5 days per week) for esophageal cancer. Six months after active surveillance, a residual esophageal tumor was found, and the patient underwent robot-assisted minimally invasive esophagectomy (RAMIE) with intrathoracic anastomosis. The surgery was complicated by a defect at the thoracic anastomosis and right main bronchus. An esophageal stent (24 mm × 120 mm Niti-S) was placed to bridge the defect, followed by a right thoracotomy where an intercostal muscle flap was placed over the bronchial defect, and a latissimus dorsi flap was used to cover the gastric tube defect.

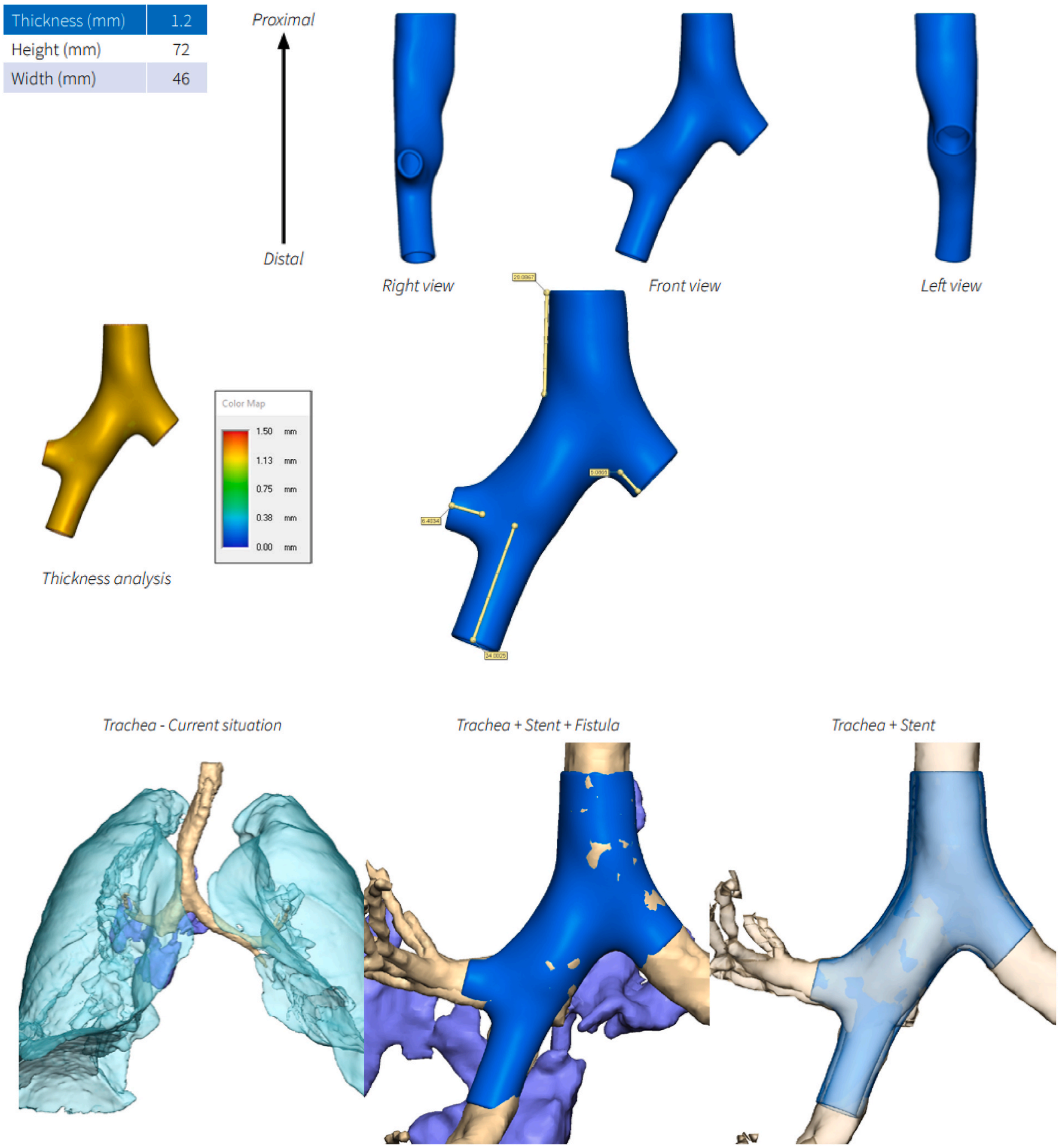

Fig. 2. | Three-dimensional printed endobronchial stent design for the treatment of a broncho-esophageal fistula (or fistula between the bronchus and gastric conduit).

One week later, the patient was readmitted with severe respiratory insufficiency from aspiration pneumonia due to a persistent esophago-bronchial fistula. Despite a second esophageal stent (24 mm × 60 mm Niti-S), contrast leakage was still visible. Subcutaneous emphysema suggested a possible fistula into the pleural cavity (see Fig. 1).

Given the complex fistula anatomy and lack of a suitable endobronchial stent, a custom 3D-printed stent was designed (see Figs. 2 and 3). After its placement and removal of the esophageal stent, follow-up CT-scan showed a reduced fistula opening towards the right main bronchus and bronchus intermedius. However, a small defect (16 mm × 24 mm) persisted, prompting reinsertion of an esophageal stent. Endoluminal vacuum therapy (endoVAC) was attempted but was unsuccessful (see Fig. 4).

2.1. Outcome and follow up

During a follow-up gastroscopy conducted two months later, a persistent broncho-esophageal fistula was observed, raising the suspicion that the defect could not close due to the bronchial 3D stent. The endobronchial stent was removed without complications.

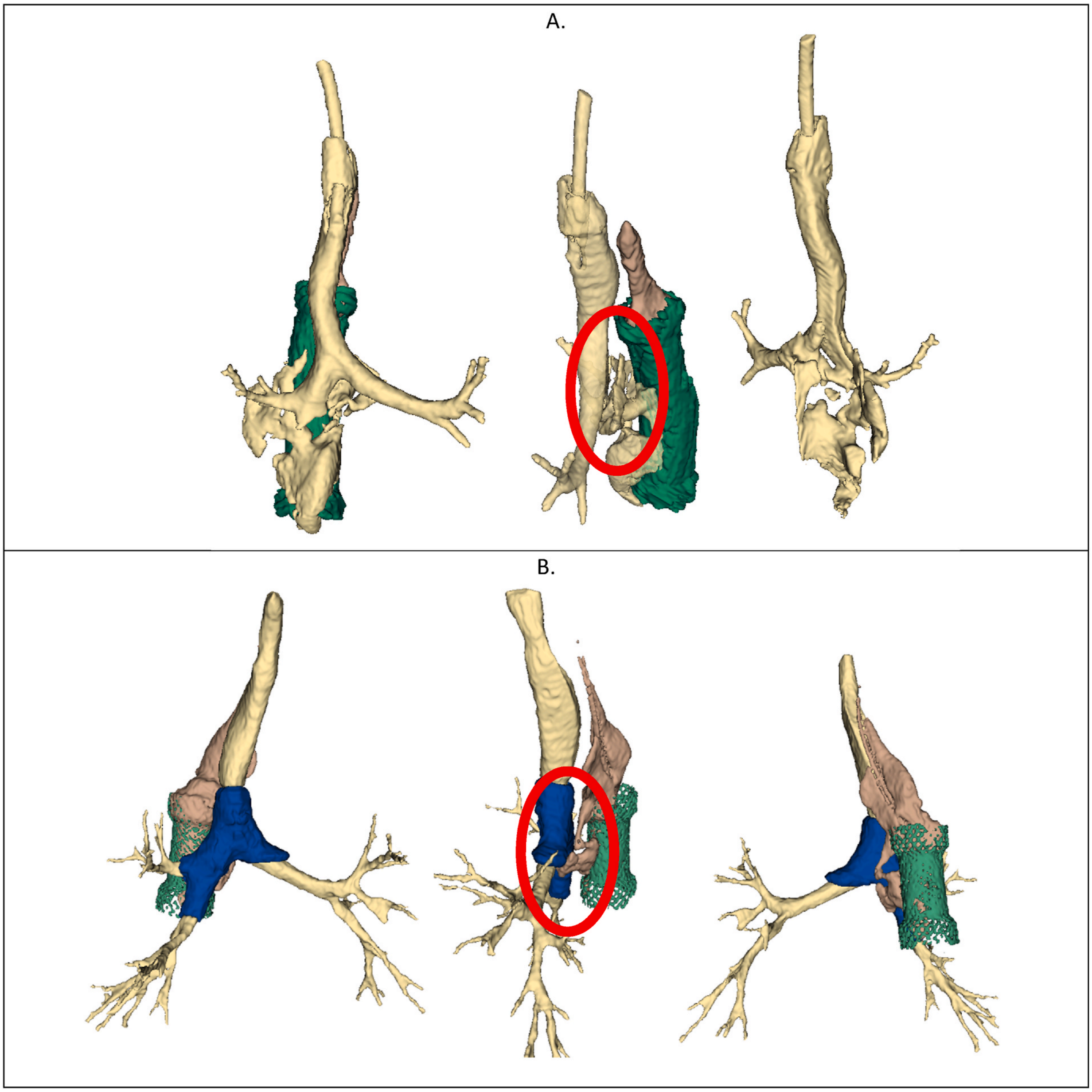

Fig. 3. | A) Segmentation of airways with broncho-esophageal fistula prior to stent placement. Yellow: airway and intrathoracic free air, brown: esophagus, and green: esophageal stent, red: location of fistula B) Segmentation of the airways 1 month post-3D stent placement. Yellow: airways, brown: gastric conduit, green: esophageal stent, red: location of fistula and blue: 3D endobronchial stent.

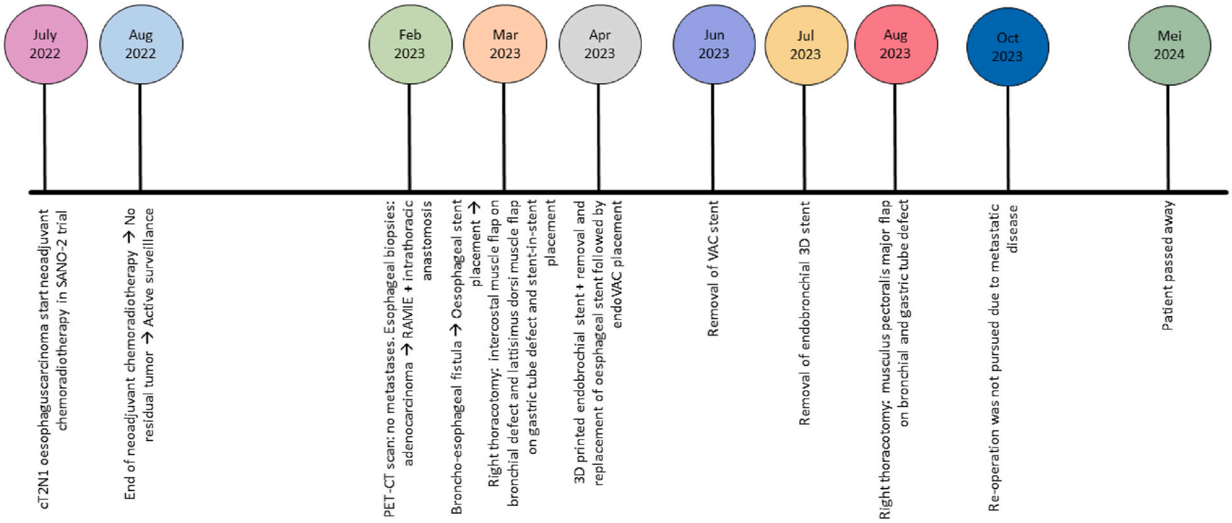

Fig. 4. | Timeline of events.

There were no signs of granuloma formation at the edges of the stent. The patient was scheduled for surgical closure of the fistula with a (musculus pectoralis major) muscle flap on the bronchial and gastric tube defect and the esophageal stent was also exchanged during this procedure. A re-thoracotomy was discussed with the patient for another attempt at closing the broncho-esophageal defect using an anterior serratus muscle flap. However, before proceeding, a PET-scan was performed to check for metastatic disease, which revealed new bone metastases. Due to the progressive nature of the disease, it was decided to refrain from further interventions to treat the broncho-esophageal fistula, and a best supportive care approach was discussed. To prevent fistula-related aspiration symptoms, the patient was fed via a jejunostomy. Unfortunately, the patient passed away 5 months later.

3. Discussion

We report a case of a broncho-esophageal fistula in a patient with distal esophageal carcinoma, previously treated with esophagostomy and gastric tube reconstruction following neoadjuvant chemoradiation. Management involved an esophageal stent and a 3D-printed airway stent. However, the stent did not effectively close the fistula, likely due to high pressure exerted on the endobronchial wall, which prevented proper sealing. This is the first reported use of a 3D-printed airway stent instead of a conventional stent for such a case, presenting a novel approach that may broaden treatment options in complex cases.

Silicone stents offer certain benefits over metal ones, including reduced risk of esophageal secretions entering the airway and granulation tissue formation. Due to the complex anatomy of the fistula, a standard stent was not suitable, necessitating a custom-designed stent. However, despite the potential for shape customization to fit complex anatomies, limitations were encountered: the stent's pressure on the airway wall likely contributed to inadequate sealing, requiring its removal.

The process of designing a 3D airway stent also has notable drawbacks, such as the time-intensive manufacturing and the expertise required in stent customization. While 3D-printed stents enable customized solutions for challenging anatomies, further research is essential to optimize stent designs. Enhancing retention, improving sealing effectiveness, and reducing complications, such as granulation tissue and mucus plugging, will be crucial in advancing the success of 3D stents for broncho-esophageal fistulas.

3.1. Take home message

Broncho-esophageal fistula treatment aims to prevent infection by sealing the fistula and stopping airway contamination. The efficacy of 3D-printed airway stents over conventional ones remains unclear. This case, the first to use a 3D-printed airway stent for a broncho-esophageal fistula, demonstrates that the stent did not achieve closure, likely due to excessive pressure against the endobronchial wall. This underscores the need for improved 3D stent designs, offering important insights for interventional pulmonologists.

CRediT authorship contribution statement

Illaa Smesseim: Writing – review & editing, Writing – original draft, Visualization, Validation, Software, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Sophia van Beelen:** Writing – review & editing, Writing – original draft, Formal analysis, Data curation, Conceptualization. **Jolanda M. van Dieren:** Writing – review & editing, Supervision, Data curation. **Koen J. Hartemink:** Writing – review & editing, Supervision, Data curation. **Johanna van Sandick:** Writing – review & editing, Writing – original draft, Supervision, Data curation, Conceptualization. **Jacobus A. Burgers:** Writing – review & editing, Writing – original draft, Validation, Supervision, Formal analysis, Data curation, Conceptualization.

Statement of ethics

This material is the authors' own original work, which has not been previously published elsewhere. This study protocol was reviewed and granted an exemption from requiring ethics approval by the Medical Ethics Committee of the Netherlands Cancer Institute. Written informed consent was obtained from the patient for publication of the details of their medical case and any accompanying images.

Data availability statement

The data that support the findings of this study are not publicly available due to their containing information that could compromise the privacy of research participant but are available from the corresponding author. Email: i.smesseim@nki.nl.

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Written informed consent was obtained from the patient.

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