

# 3-D printouts of the tracheobronchial tree generated from CT images as an aid to management in a case of tracheobronchial chondromalacia caused by relapsing polychondritis

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## ABSTRACT

This report concerns a 67 year old male patient with known advanced relapsing polychondritis complicated by tracheobronchial chondromalacia who is increasingly symptomatic and therapeutic options such as tracheostomy and stenting procedures are being considered. The DICOM files from the patient's dynamic chest CT in its inspiratory and expiratory phases were used to generate stereolithography (STL) files and hence print out 3-D models of the patient's trachea and central airways. The 4 full-sized models allowed better understanding of the extent and location of any stenosis or malacic change and should aid any planned future stenting procedures. The future possibility of using the models as scaffolding to generate a new cartilaginous upper airway using regenerative medical techniques is also discussed.

## CASE REPORT

### CASE REPORT

In the two decades prior to diagnosis, the patient suffered several complaints which were dealt with in isolation - cardiac arrhythmias, vertigo and deafness (attributed to benign positional paroxysmal vertigo), intermittent painful ears. At the age of 63, he presented with an acute confusional state associated with a headache, complete lower motor neurone facial palsy, shortness of breath and pain and erythema of the pinna. On this admission, a clinical diagnosis of relapsing polychondritis (RP) was made with the mental status change

ascribed to limbic encephalopathy. A dynamic chest CT demonstrated tracheobronchial malacia (Figure 1 and Figure 2). Initial respiratory function tests were within normal limits. He received high dose intravenous steroids and cyclophosphamide with improvement. Multiple relapses occurred which were treated with immunosuppressants and immunomodulators including glucocorticoids variously in combination with cyclophosphamide, methotrexate, rituximab, infliximab, azathioprine, mycophenolate mofetil and leflunomide.

However, the symptoms of breathlessness have gradually worsened despite the lack of change in his respiratory function tests, and the patient was keen for intervention. Decision was made to use the DICOM files from the dynamic CT to produce inspiratory and expiratory 3-D models of the trachea and central airways as proof of principle to determine whether they could aid some form of tracheobronchial intervention in the future. The air was segmented to create negative models of the air within the airway using methods we have previously described [1] and plaster models printed (Figure 3, Figure 4) (on this occasion treating the air in the airway as a target) and the segmentation was then processed using Tomomask ([www.tomomask.com](http://www.tomomask.com)) to allow a cast of the airway to also be printed (Figure 5). The hollow versions were created by firstly making a mask to fill the interior of the trachea. The mask was then dilated in 3D using an oblate spheroidal structuring element with a radius of 5 pixels in the x & y plane and 3 pixels in the z plane. With the pixel spacing of 0.65625 mm and the slice spacing of 1 mm, this produced a dilatation (thickness) of approximately 3 mm (Figure 5). The interior mask was then subtracted from the dilated version to form a hollow shell with a 3 mm wall thickness.

## DISCUSSION

Relapsing polychondritis (RP) is a rare auto-immune disease with an incidence of less than 1 per million [2-6]. As the name suggests it tends to run a relapsing clinical course in which cartilage-containing structures (elastic, hyaline or fibrous) may be affected as indeed may structures containing mucopolysaccharides such as the eye and blood vessels [2-6]. Because of its rarity, relapsing nature and diverse clinical manifestations the diagnosis can be delayed by many years [2-6]. The commonest cause of death in RP relates to pulmonary complications and infection [2-6].

We report a case of RP in which the diagnosis was not made for many years that is now complicated by advanced tracheobronchial chondromalacia and endobronchial stenting is being considered.

The radiological literature concerning both normal and abnormal appearances of the trachea and central bronchi plus that specifically pertaining to RP is well-described. Airway involvement may be seen in 10% of cases of RP at presentation and can occur in up to 50% of patients and include fixed airway narrowing, wall thickening and calcifications. Smooth thickening of the anterior and lateral walls with sparing of the posterior membranous wall is characteristic and increased attenuation of the airway is the commonest sign [7-14]. There is now general consensus that patients with RP who have respiratory symptoms should have a chest CT in both full expiration as well as full inspiration as at least 50% of abnormalities are functional (particularly malacia and air trapping) and will be missed if only an inspiratory chest CT is carried out [11,12].

Early aggressive treatment may prevent irreversible cartilage destruction and there is a role for stenting in RP and

the site, severity and length of any strictures should be evaluated prior to intervention [15]. Imaging techniques are important to aid in the visualisation of anatomy and pathology and simple reformatted images may aid with 3-D visualization of focal stenosis. However, in complex cases where there is diffuse airway involvement or dynamic changes, our model may be particularly important in aiding procedure planning. Choice of stent size would be challenging, and the broadening and flattening of the airways in expiration is particularly striking when the physical models are held in the hand.

Rapid prototyping technology or, as it is now becoming better known, 3-D printing is becoming increasingly common place worldwide. A range of 3-D printing machines are available including the simplest 3-D printers costing around \$1000, desktop size printers capable of high quality relatively small prints and large complex commercial printers capable of printing models using multiple materials (including plastics and metals), which cost in excess of \$250,000. One might elect to have a 3-D printing company perform the actual printing of STL files sent to them or alternatively purchase a 3-D printer for one's own unit or hospital. Many 3-D printing online services impose restrictions on the types of models that can be printed such as a restriction on the overall file size. Other cost and technical considerations that will need to be considered in the future include demand, 'turn-around time', print size, the cost and type(s) of printing material used and the time required to perform the printing itself.

Some of the possible uses of 3-D models created from CT scans has recently been reviewed and includes roles in education, communication, procedure planning and medical device creation [16] but as far as we are aware, this technique has not been used to plan tracheal or bronchial stenting procedures in RP. Rapid prototyping has been used to print out models of normal airways which allowed assessment of flow patterns in the hollow cast [17]. The full-size 3-D models we have created will aid planning of stent location and size, of particular importance where stent migration is a well-recognized complication. The hollow cast could allow testing of stents and sealing zones. Related technology is used in a range of regenerative medicine applications. In particular Computer-Assisted Manufacture (CAM) can be used to produce scaffolds for tissue repair [18] and careful choice of materials can directly control cell identity and function [19]. The solid model could also be used as a scaffold for any potential cartilage regenerative treatments such as in-vitro cartilage engineering to produce novel and bespoke implants.

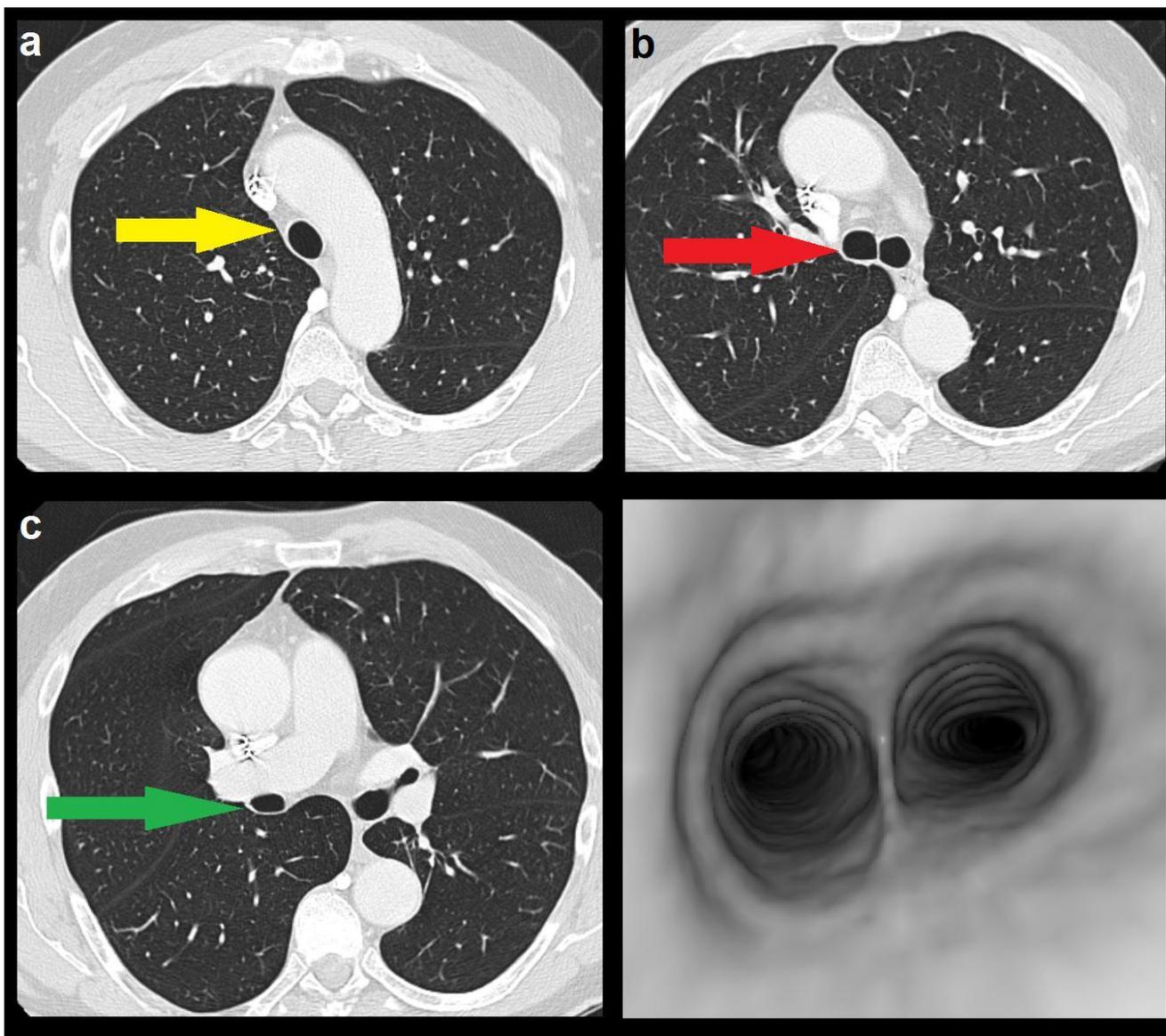
## TEACHING POINT

3D model of the tracheobronchial tree produced by rapid prototyping of CT images can aid surgical or interventional planning as well as facilitate education and patient understanding. They could also conceivably be used as a scaffold or a scaffold design aid for in vitro cartilage engineering in the preparation of novel and bespoke implants.

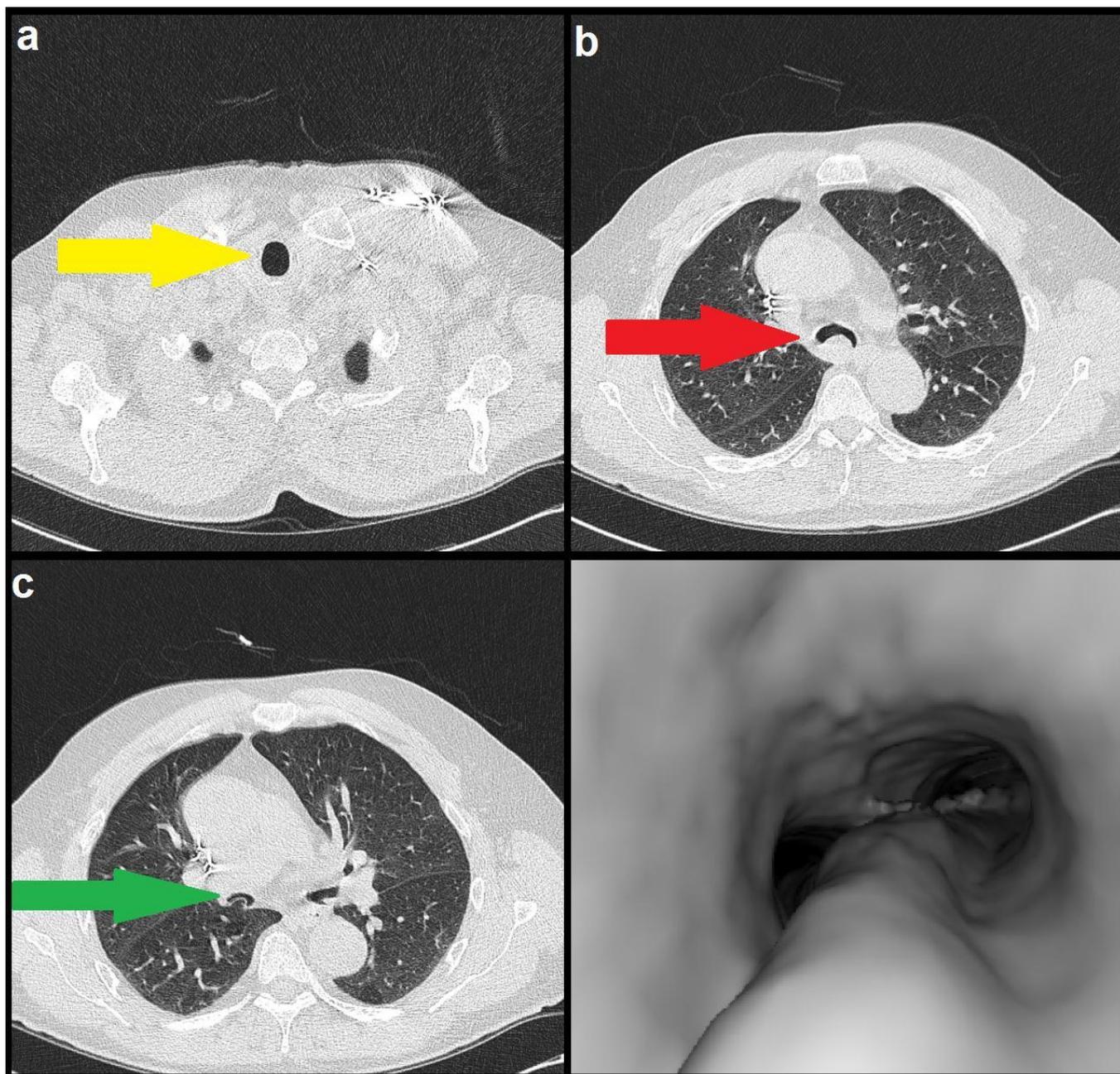
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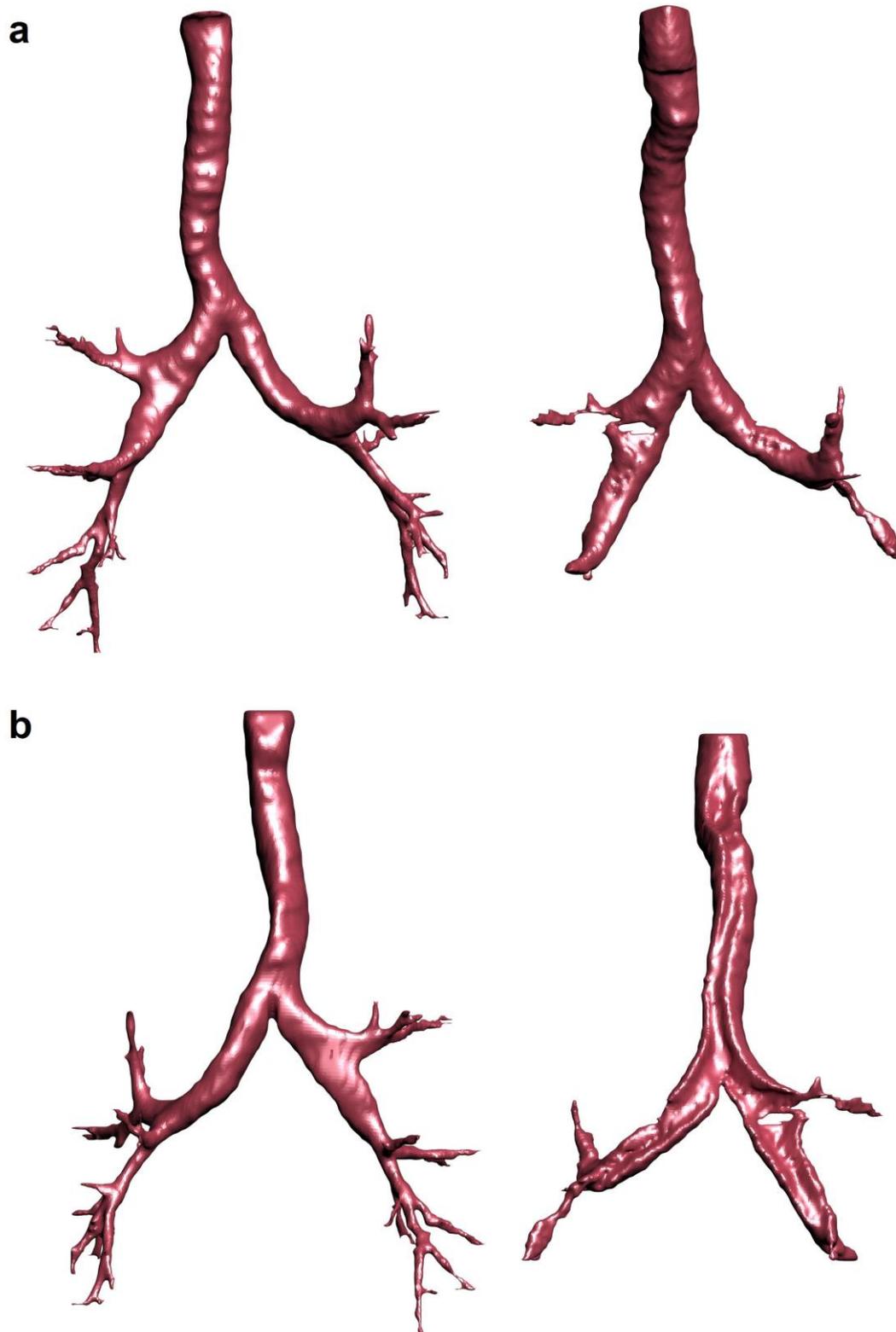
FIGURES



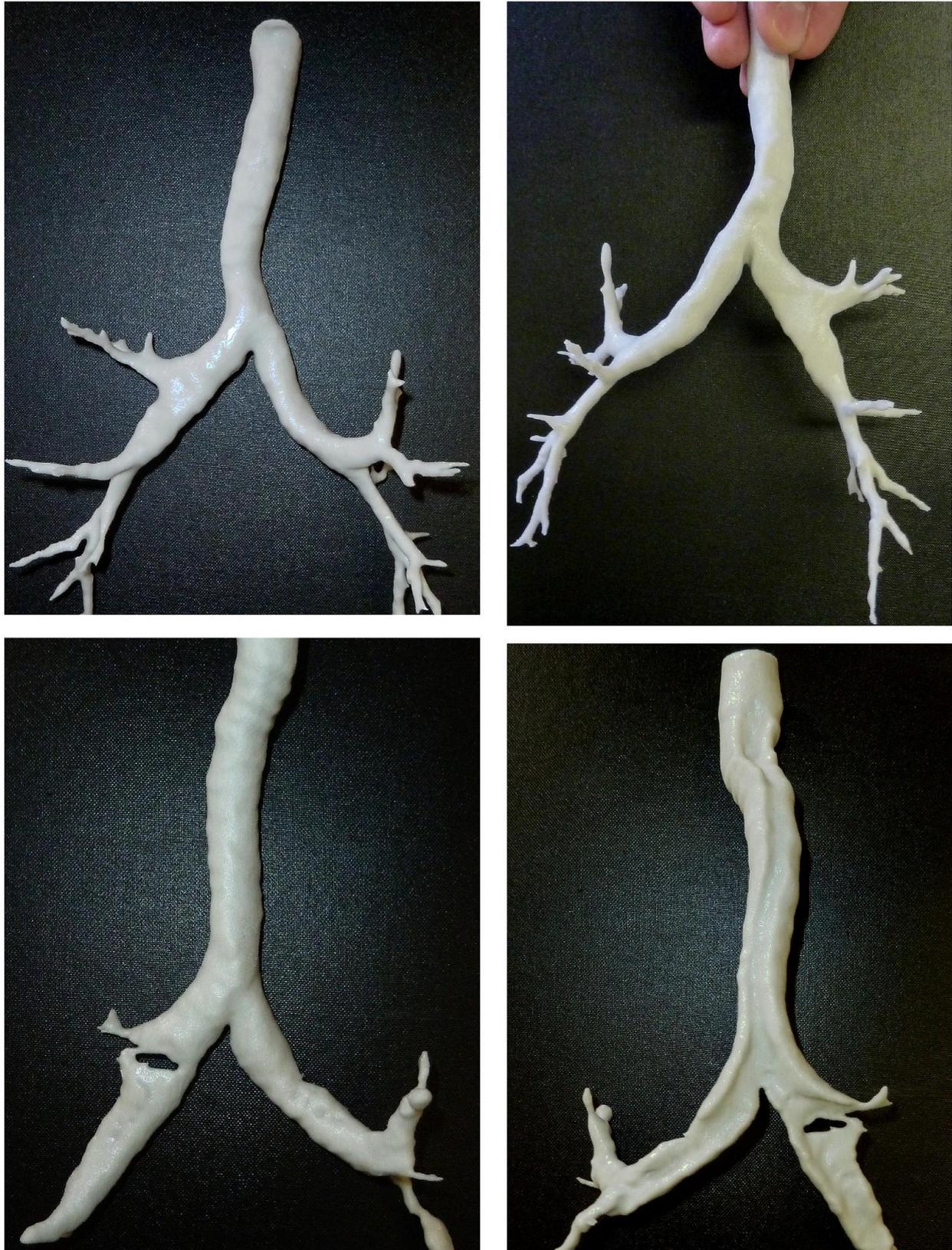
**Figure 1:** 67 year old man with known relapsing polychondritis. Images demonstrate an apparently normal CT chest. Axial contrast enhanced CT of the chest in arterial phase taken in inspiration. The trachea (yellow arrow), carina (red arrow) and bronchus intermedius (green arrow) are demonstrated in (a), (b) and (c) respectively, with a virtual bronchoscopy rendered image (looking down onto the carina) demonstrating normal airway appearances (d). (Protocol: Siemens 64 slice CT, slice thickness 1 mm, kVp 120, mAs 110).



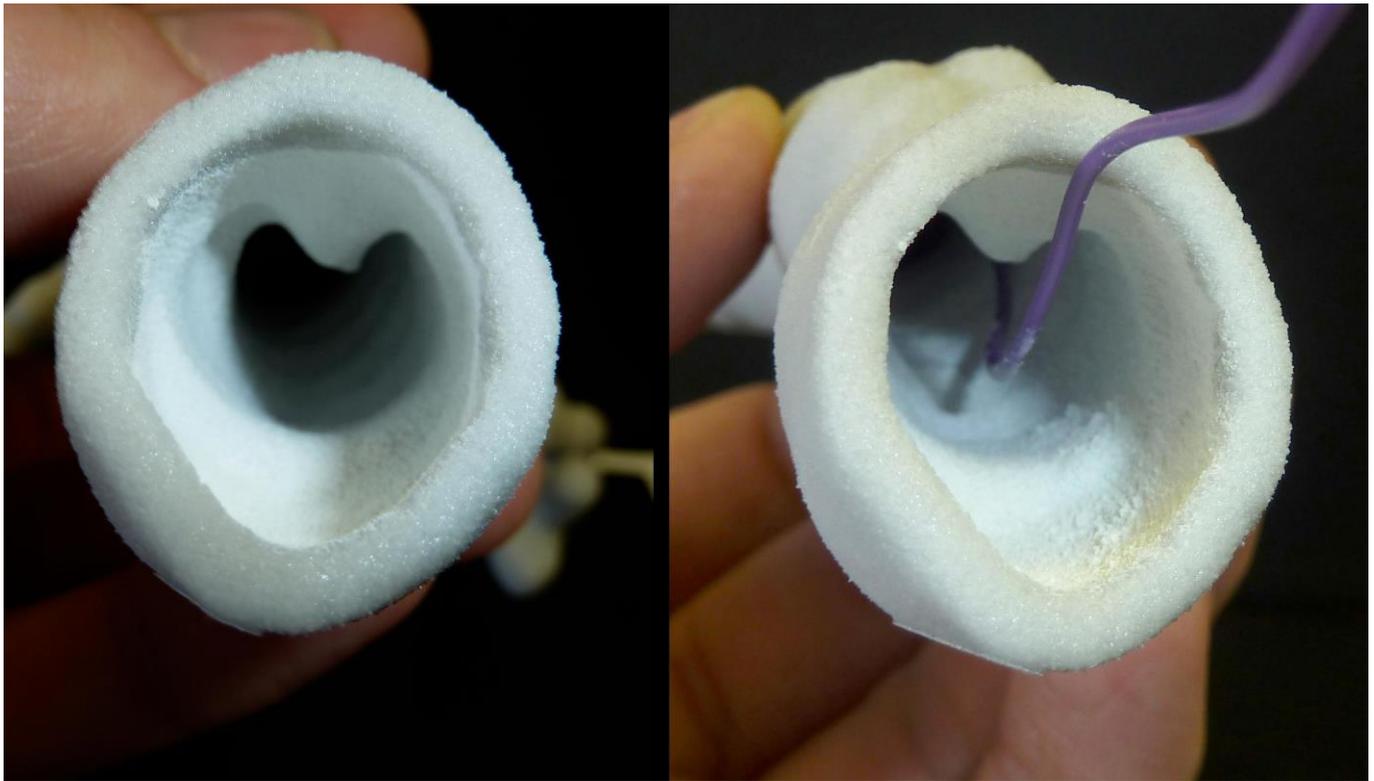
**Figure 2:** 67 year old man with extensive collapse of the airways due to chondromalacia secondary to relapsing polychondritis on unenhanced dynamic expiratory CT. The proximal trachea is normal (yellow arrow) but there is marked bowing of the posterior wall of the lower trachea (also known as the "frown" sign) and carina (red arrow) and further significant collapse of the bronchus intermedius (green arrow) is demonstrated in panes (a), (b) and (c), with a virtual bronchoscopic view looking down toward the carina showing significant airway collapse (d). (Protocol: 64 slice CT, slice thickness 1 mm, kVp 120, mAs 106).



**Figure 3:** 67 year old man with known relapsing polychondritis. Presentation of STL files rendered from DICOM CT. Views of the tracheobronchial tree are presented as follows -anterior inspiratory (top left), anterior expiratory (top right), posterior expiratory (bottom left) and posterior inspiratory (bottom right).



**Figure 4:** Photographs of 3-D printed models of the tracheobronchial tree of a 67 year old man with known relapsing polychondritis. Top row: anterior and posterior views of the inspiratory phase; bottom row: anterior and posterior views in expiration. Again, note the expiratory airway collapse.



**Figure 5:** 67 year old man with known relapsing polychondritis. Photographs of the hollow 3d printed models. The hollow cast of the expiratory airway is demonstrated and internal access with a wire is shown.

<b>Etiology:</b>	Recurrent inflammation and destruction of cartilage? autoimmune
<b>Incidence:</b>	1 in 3.5 million
<b>Gender ratio:</b>	1:1
<b>Age predilection:</b>	Average age at diagnosis is 47 years, peak prevalence is 40 to 60 years, but non-specific symptoms can be present for years prior to diagnosis
<b>Risk factors:</b>	N/A
<b>Treatment:</b>	Immunosuppressants, tracheobronchial stenting
<b>Prognosis:</b>	Relapsing course, death from pulmonary complications or infective complications

**Imaging findings:**

**CT head** Thickened pinna and cartilage of the external auditory meatus

**CT chest** Can be normal, so inspiratory and expiratory phases are warranted

Increased attenuation of tracheal wall (compared to mediastinal soft tissue)

Airway wall calcifications

Focal airway strictures

Thickening of airway wall \*posterior membranous wall often spared

50% of patients with RP will develop respiratory complications

In cases of known RP, 70% may have tracheal wall thickening

Diffuse wall thickening occurs more commonly than nodular/focal changes

Tracheal stenosis is present in up to 15%

Air trapping on expiratory scans in 50%

Associated cylindrical bronchiectasis

Can also affect peripheral joints, nasal cartilage, laryngeal cartilage, uveal inflammation and aortitis and aortic root dilatation

**Table 1:** Summary table for Relapsing Polychondritis (RP)

- ❖ Amyloidosis (often circumferential)
- ❖ Sarcoidosis (often circumferential)
- ❖ Wegener's granulomatosis (often circumferential)
- ❖ Tuberculosis, histoplasmosis,
- ❖ Tracheopathia osteochondroplastica (idiopathic multiple submucosal calcified nodules)
- ❖ Rhinoscleroma (granulomatous infection)
- ❖ Laryngeal papillomatosis (viral) [20]

**Table 2:** Differential diagnosis table for airway thickening

<b>Malignant</b>	<ul style="list-style-type: none"> <li>• Primary airway tumour – scc, adenoid cystic carcinoma</li> <li>• Bronchogenic carcinoma</li> <li>• Oesophageal or thyroid carcinoma</li> <li>• Airway metastases (e.g. renal cell carcinoma or colon cancer)</li> </ul>
<b>Benign</b>	<ul style="list-style-type: none"> <li>• Post-intubation / post-tracheostomy</li> <li>• Inflammatory – TB, post-infectious</li> <li>• Anastomotic – lung transplantation, sleeve resection</li> <li>• Tracheobronchomalacia</li> <li>• Congenital tracheal stenosis</li> <li>• Compression by oesophageal stent</li> </ul>

**Table 3:** Differential diagnosis table for airway stenosis

#### ABBREVIATIONS

DICOM = Digital Communications in Medicine  
 RP = relapsing polychondritis

#### KEYWORDS

Rapid prototyping; DICOM; STL; 3D printing; 3D modeling; relapsing polychondritis; tracheobronchomalacia

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