

peer reviewed **ARTICLE OF INTEREST**

## CT colonography in the visualisation of lymphangioma: a rare benign submucosal lesion

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### ABSTRACT

Lymphangioma of the colon is a rare benign lesion. CT colonography (CTC), compared to optical colonoscopy, does not entail compression or probing of submucosal lesions. It does routinely include different patient positions, measurement of lesions and their HU values. There seems to be a gap in the CTC literature in terms of the features of this benign submucosal lesion. The 3D and 2D images of colon lymphangioma of two cases at screening CTC are discussed to illustrate the difference between it and other submucosal lesions such as lipoma.

**Keywords** benign, cystic lesion, intramural, sessile submucosal, lipoma, leiomyoma

### LAY ABSTRACT

Based on the change of shape of a soft mass in the colon a diagnosis of lymphangioma was made for two cases that presented for screening computed tomography colonography.

### INTRODUCTION

For decades lymphangioma of the colon was thought to be a rare occurrence. Literature reports that lymphangioma is being increasingly diagnosed and reported with optical colonoscopy (OC), computed tomography (CT), magnetic resonance imaging (MRI) and endoscopic ultrasound.<sup>[1-7]</sup> Barium enema used to be the routine study to image the colon but has been replaced by CT colonography (CTC).<sup>[8]</sup> CTC has been used to visualise the colon for more than 20 years; it is an extremely safe and minimally invasive procedure.<sup>[9]</sup> In view of its increased acceptance as a screening procedure for polyps and colorectal carcinoma<sup>[9]</sup> radiologists would be the first to encounter a lymphangioma of the colon. CTC's routine technique of supine, prone, and decubitus views if necessary, makes it possible to observe that the soft tissue lesion seen initially on the supine view does change in appearance on subsequent views. Unlike OC, which involves pushing and compressing of an intramural lesion, CTC's range of views makes it easier to observe that liquid is within the lesion. CTC thus eliminates the possible danger of perforation by not compressing or pushing the lesion.

Most colon lymphangiomas are sessile in nature but may become pedunculated.<sup>[5]</sup> Lymphangiomas in the large intestine are benign submucosal lesions and are a

subset of nonneoplastic cystic lesions with an intramural origin.<sup>[10]</sup> They are covered by normal mucosa and are mass like protrusions.<sup>[10]</sup> OC only allows a visual evaluation of the surface of the colon lumen whereas CTC provides information on the origin and extent of non-neoplastic intramural lesions as well as their internal composition.<sup>[10]</sup> To my knowledge the CTC features of a lymphangioma have not previously been described. Two cases of colon lymphangioma diagnosed at screening CTC are presented and their features are discussed.

### CLASSIFICATION OF LYMPHANGIOMA

The majority (95%) of lymphangiomas occur in the neck or axillary regions. The remainder (5%) are located in the mesentery, retroperitoneum, abdominal viscera, lung and mediastinum.<sup>[2]</sup> There are three classifications of lymphangiomas.<sup>[4]</sup>

- Simple lymphangioma comprises small thin-walled lymphatic vessels.
- Cavernous lymphangioma has dilated lymphatic vessels and lymphoid stroma.
- Cystic lymphangioma consists of lymphatic spaces of various sizes. There is no communication with normal lymphatics.

Cavernous and cystic lymphangiomas have been found intra-abdominally and

the cystic type is usually situated under the skin.<sup>[11]</sup>

### HISTOLOGICAL FEATURES OF COLON LYMPHANGIOMA

Colon lymphangiomas are histologically dilated lymphatics and are found in the submucosa hence deep biopsy is required.<sup>[5]</sup> They are thin-walled, dilated and microcystic lymphatic spaces filled with fluid. The dilated lymph spaces are lined by attenuated endothelial cells.<sup>[2]</sup> Endothelium lines the spaces similar to normal lymphatics with smooth muscle bundles in the wall. The thin-walled cystic masses have smooth external surfaces and may contain chylous, serous, haemorrhagic or mixed fluid.<sup>[2]</sup>

### ENDOSCOPIC APPEARANCES OF COLON LYMPHANGIOMA

Lymphangioma of the intestinal tract is extremely rare and usually presents as a sessile or pedunculated polyp.<sup>[1]</sup> Endoscopically a lymphangioma is a transparent and fluctuating mass, which might be sessile or pedunculated, with the colour of normal mucosa. Dilated vessels may be found in the colonic mucosa and submucosa. A lymphangioma usually presents as a submucosal sessile polypoid lesion. The findings of 10 cases by Kim et al<sup>[1]</sup> were that a sessile or pedunculated lymphangioma at OC is a transparent mass with

normal mucosal colour. Its shape changes with endoscopic compression.<sup>[11]</sup>

### DIFFERENCE BETWEEN CT AND CTC TO VISUALISE THE COLON

Special software is used in a CT scanner to produce an accurate reconstruction of the colon. CTC produces two-dimensional (2D) images and three-dimensional (3D) endoluminal views. Attenuation values are obtained in CT and CTC studies. Electronic callipers are available to accurately measure lesions seen at CT or CTC. The main difference between an abdominal CT and a CTC examination is that the special CTC software includes a 3D fly-through of the colon. In other words this software pertains to a CTC study; vendors usually provide their own software.

### RIGOROUS TECHNIQUE FOR DETECTION OF SUBMUCOSAL LESIONS AT CTC

A 3D fly-through allows for viewing the inside of the colon from the rectum to the caecum and returning back to the rectum hence the entire colon is viewed twice. A fly-through is viewed as a video and the entire process takes two minutes to perform. One is able to stop the fly-through at any stage in order to 'eye-ball' any part of the colon that may have a lesion. Varying speeds may be selected hence one can 'travel' at a low or high speed. The use of a fly-through allows one to view the inside of the colon in both the supine and prone positions, and, if included in the decubitus views. In other words for a standard two view study comprising supine and prone positions the colon is viewed four times in its entirety: from rectum to caecum and back to rectum. If a decubitus study is added to an examination then the

inside of the colon will be viewed in its entirety six times. This will allow one to 'eye-ball' any change of shape of a lesion in the colon.

It is important to be skilled in being able to interpret the obtained 3D and 2D images.<sup>[12]</sup> Different window settings are used when assessing the 2D images. For example, for 2D polyp detection the window setting should be a window width of 2000 and centred at 10 to -200.<sup>[13]</sup> Both the supine and prone 2D images must be visualised to check for change in shape, for example. Particular attention should be paid to tagging of lesions and size of lesions. Electronic callipers are available to accurately measure lesions. The HU values of a region of interest (ROI) should be measured.

Viatronix software allows for an in-depth look at a lesion by means of a 3D translucent display. This provides a semi-transparent view of a lesion in a range of colours beneath the surface.<sup>[13,14]</sup> The different attenuation values of the software are: red indicates soft tissue; white indicates high attenuation values, such as barium; green indicates negative values in the fat attenuation range; and blue indicates negative values, such as air. For example, if the colour is red this indicates a high intensity, which is a feature of a polyp. If green is the predominant colour this indicates fat which means that a lipoma may be diagnosed. Electronic callipers are available to accurately measure lesions.

### LIPOMA AND LEIOMYOMA AT CTC

#### • Lipoma

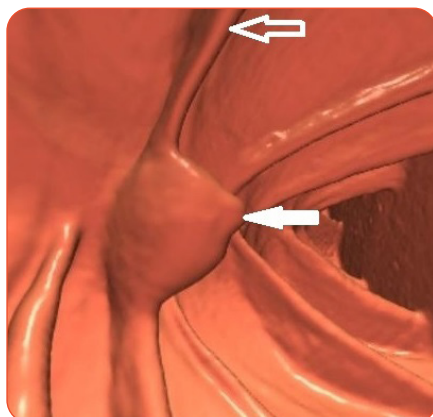
Lipoma is the most common of the non-epithelial tumors of the gastrointestinal

tract.<sup>[15]</sup> CTC is useful for detecting and demonstrating colonic lipomas on 2D and 3D views. On CT scan images the appearance of a lipoma is uniform, with a fat equivalent density range between -80 to -120 Hounsfield units.<sup>[16]</sup> They have a predilection for the right colon; especially the caecum and ascending colon, followed by the sigmoid and descending colon: they may be sessile or pedunculated. Colonic lipoma is usually clinically silent or mildly symptomatic.<sup>[17,18]</sup> Lipomas are almost always asymptomatic until their size becomes approximately 35mm (3.5cm).<sup>[19]</sup> Small lesions can be safely left in the colon, but as size increases > 30mm, symptoms may occur. There are two treatment options: endoscopic removal of lesions > 30mm, or surgical resection for benign larger tumors or those that result in intussusception.

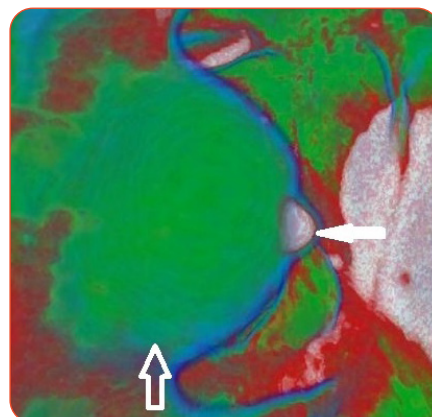
Lipomatosis of the ileocaecal valve may be present. This is easily diagnosed on CTC using translucent display (TD), which shows uniform green colour indicating fat. If a lipoma is present on the ileocaecal valve (ICV) it is usually visualised as a separate 'lump' and not part of a uniform fatty infiltration of the valve. Figures 1a to 1f are 2D and 3D views of a lipoma on haustral of ascending colon. Figures 2a and 2b are CTC views of a lipoma on the ileocaecal valve.

#### • Leiomyoma

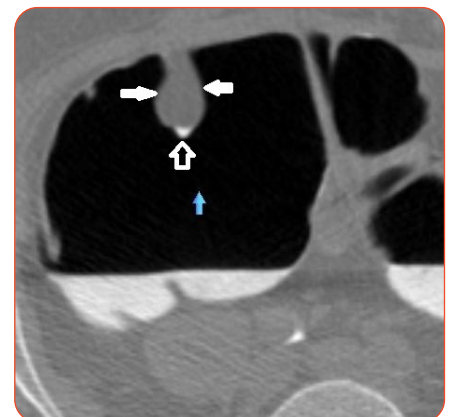
Only 3% of gastrointestinal leiomyomas occur in the colon; most occur in the oesophagus.<sup>[20]</sup> Leiomyoma of the colon (a.k.a. intestinal leiomyoma) is an uncommon, benign smooth muscle tumour that may arise from the muscularis mucosa or the muscle itself.<sup>[21]</sup> This tumour is usu-



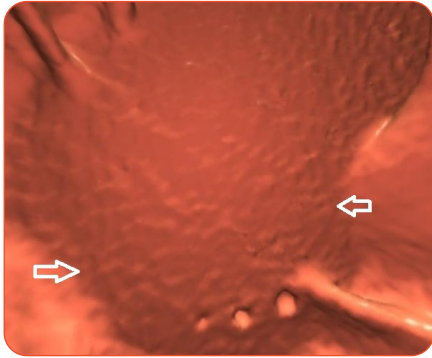
**Figure 1a.** 3D endoluminal view showing polypoidal lesion (closed white arrow) arising from haustral fold (open white arrow).



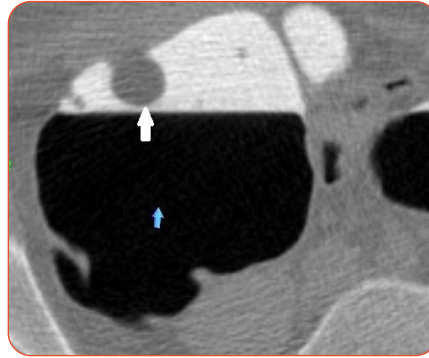
**Figure 1b.** TD showing lipoma: green = fat (open white arrow) and barium on tip (closed white arrow).



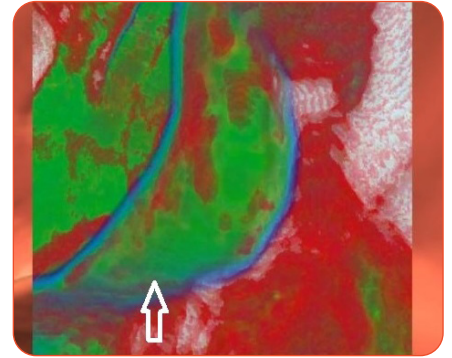
**Figure 1c.** Axial 2D soft tissue window showing tip of barium (open white arrow) on lipoma (closed white arrows).



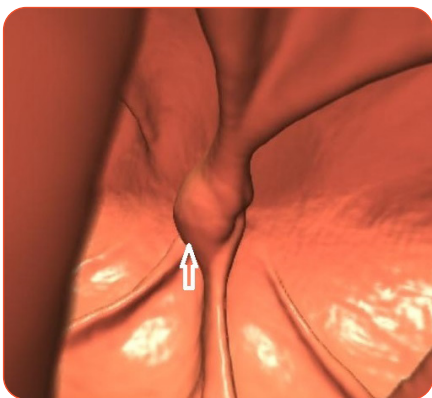
**Figure 1d.** 3D prone image showing barium covering lipoma (open white arrows).



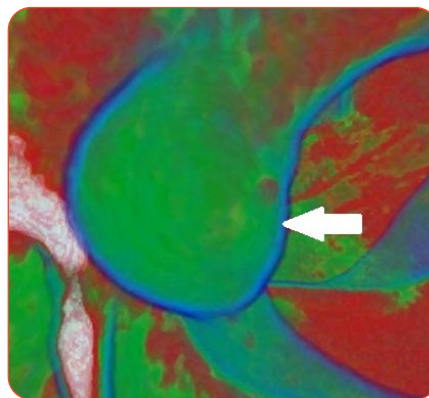
**Figure 1e.** Prone 2D axial view, soft tissue window showing filling defect (closed white arrow) in barium pool.



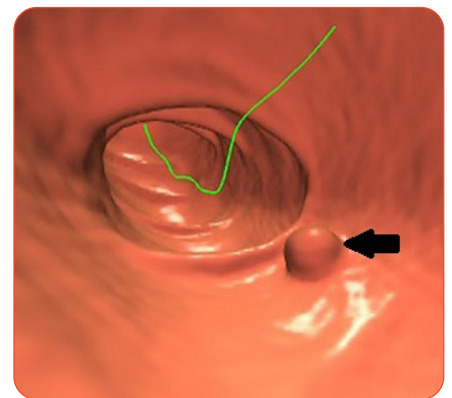
**Figure 1f.** TD showing diffuse infiltration of the ICV (open white arrow) indicating caecal lipomatosis with minimal high intensity (red).



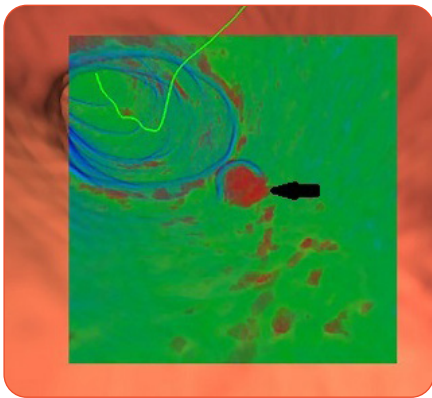
**Figure 2a.** 3D view shows a lipoma on ICV (open white arrow).



**Figure 2b.** TD shows dense green coloration in keeping with fat (lipoma) (closed white arrow).



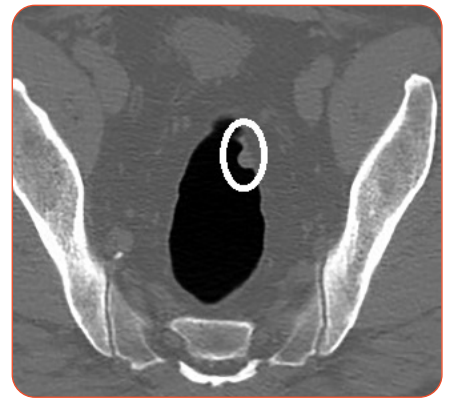
**Figure 3a.** 3D image showing a rectosigmoid polypoidal lesion (black arrow). (Courtesy of Prof Kim from University of Wisconsin).



**Figure 3b.** Translucent display showing high intensity as red (black arrow), which is usually typical feature of a polyp. (Courtesy of Prof Kim from University of Wisconsin).



**Figure 3c.** 2D supine coronal view showing soft tissue density (white circle) of the 3D polyp in Fig 3a. (Courtesy of Prof Kim from University of Wisconsin).



**Figure 3d.** 2D prone coronal view showing soft tissue density (white circle) of the polyp. (Courtesy of Prof Kim from University of Wisconsin).

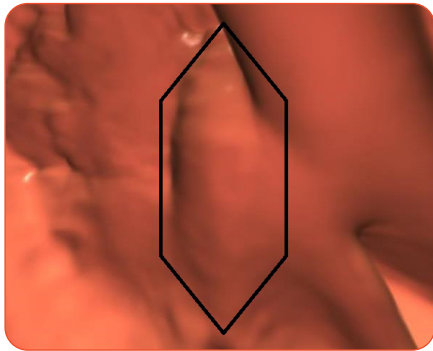
ally asymptomatic and frequently occurs in males over the age of 60 years. The location of these tumours is most often in the sigmoid and transverse colon. On colonoscopy they are identified as a sessile polyp<sup>[21]</sup> but occasionally may be pedunculated.<sup>[20]</sup> These lesions are removed for histology. Figures 3a to 3d show a rectosigmoid polyp at CTC screening; at polypectomy it was found to be a leiomyoma.

**CT APPEARANCE OF INTRA-ABDOMINAL LYMPHANGIOMA**

Intra-abdominal lymphangiomas account for less than 1% of all lymphangiomas for less than 1% of all lymphangiomas with an internal density approaching 15 HU.<sup>[22]</sup> They usually are well-demarcated, non-enhancing, homogeneous low attenuation cystic masses with an intact overlying mucosa; their margins are regular.<sup>[22,23]</sup>

**CTC APPEARANCES OF COLON LYMPHANGIOMA**

The CTC appearances of lymphangioma of two cases are presented. Screening CTC is a minimally invasive low-dose study that has been shown to be an accurate method of detecting submucosal lesions.<sup>[9,10]</sup> It is a two-view (supine and prone) examination to visualise the colon; a decubitus position may also be



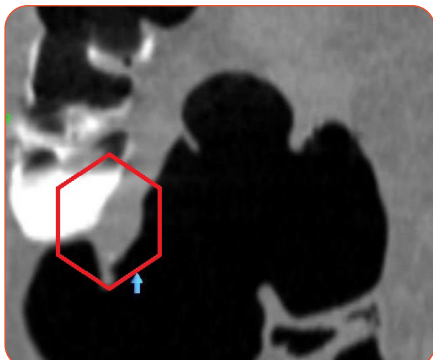
**Figure 4a.** 3D supine showing the soft tissue lesion resembling an extrinsic impression (black hexagon).



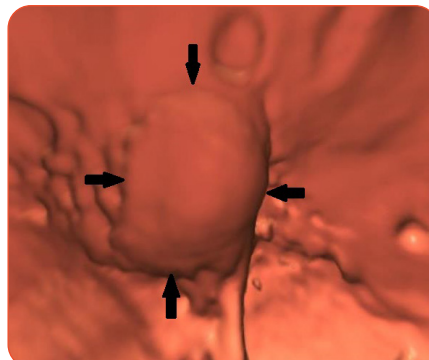
**Figure 4b.** 2D axial supine showing a polypoidal soft tissue density resembling a polyp.



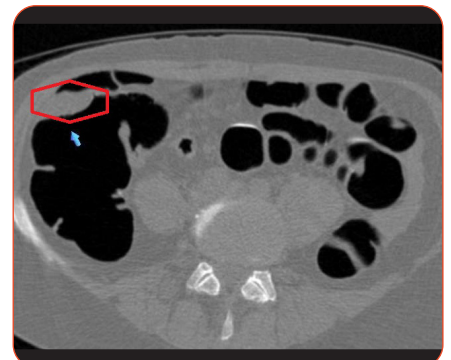
**Figure 4c.** 2D axial supine showing measurement of 30.8mm.



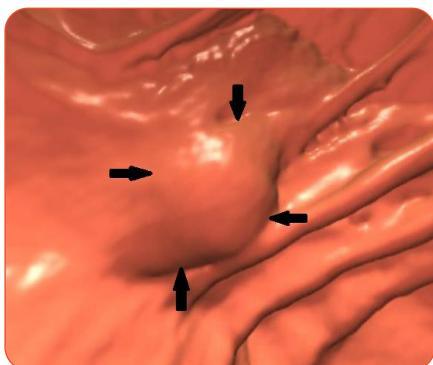
**Figure 4d.** 2D supine coronal view showing change of appearance of the shape of the soft tissue lesion (red hexagon).



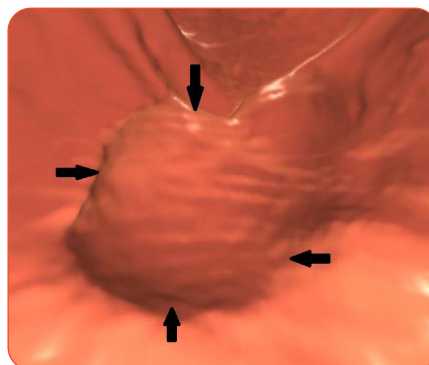
**Figure 4e.** 3D prone showing lesion resembling a polyp (black arrows).



**Figure 4f.** 2D prone view showing dramatic change of appearance compared to the supine view. The lesion (red hexagon) is lying horizontally on the anterior wall of the bowel.



**Figure 4g.** 3D right decubitus view showing the shape of the soft tissue lesion (black arrows).



**Figure 4h.** 3D left decubitus view showing how the lesion (black arrows) has changed in appearance between supine, prone and right decubitus views.

‘eye-balled’ eight times by means of the 3D fly-throughs.

As seen on Figure 4b the lesion was clearly visible. It was 5mm deep and 5cms long. The most striking CTC finding was that on the supine study the lesion resembled a sessile polyp (Figure 4b); its shape changed on the prone study (Figure 4f). Changes were noted between the two decubitus studies in keeping with the effect of gravity on the fluid (Figures 4g and 4h). In view of these findings a diagnosis of lymphangioma was made.

However, the referring physician was not satisfied with the diagnosis and referred her for an OC and endoscopic ultrasound examination. The finding of the OC was that of a “glistening submucosal polypoid structure, which on forceps pushing, showed what seemed to be fluid-filled structure”. The endoscopic findings were that of an “anechoic” structure which had a double layer to it. Its length was at least 4cms and its thickness was 5mm. It was not connected to any other structure. The lesion was punctured and approximately 3ml non-viscous slightly clear yellow fluid was aspirated. Multiple biopsies were obtained; analysis of the fluid showed no

used when in doubt.<sup>[24]</sup> It requires an adequately prepared colon. A ‘dry’ preparation of bisacodyl 5mg tablets and a saline non-ionic cathartic agent magnesium citrate is used on the day before the CTC examination.<sup>[24]</sup> Magnesium citrate, due to its osmotic action, reduces residual fluid in the colon. To maintain adequate hydration only clear liquids may be consumed on the day before the CTC examination, and patients are encouraged to drink two (2) to three (3) litres of liquid. Oral positive contrast media (250ml of 2% barium [Redi-Cat] and 50cc of non-ionic iohex-

ol [Omnipaque]) are also used the day before the CTC examination;<sup>[24]</sup> barium tags residual faeces and Omnipaque tags residual fluid.

Case 1 is a 64 year-old female who had undergone an OC and was told that a benign submucosal lesion was noted in the proximal ascending colon. She decided to have a five year follow-up examination and selected screening CTC as her choice. Supine, prone, right and left decubitus studies were obtained ( Figures 4a to 4h). In view of this her colon was virtually

epithelial or high grade malignancy. The biopsy finding was that of a cystic lymphangioma.

Case 2 is a 52 year-old healthy female who selected CTC as her first screening test for colorectal cancer. She did not have any gastro-intestinal symptoms. Supine, prone and right decubitus views were performed (Figures 5a to 5i). The inside of her colon by the use of the 3D fly-through was thus viewed six times.

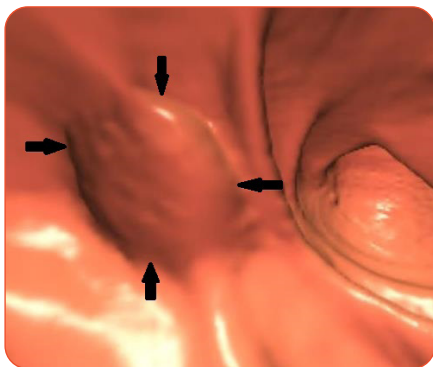
On the 3D fly-through study an extrinsic impression was noted at 76cms from the anal verge ( Figure 5a). This corresponded on the 2D image to a 1.5cm soft tissue polyp in the submucosa (Figures 5b and 5c). The appearance changed somewhat on the different views (Figures 5d to 5i) and a diagnosis of lymphangioma was made.

An OC was performed and a soft-tissue lesion was seen. Biopsies of the lesion showed a leak of thin yellowish liquid

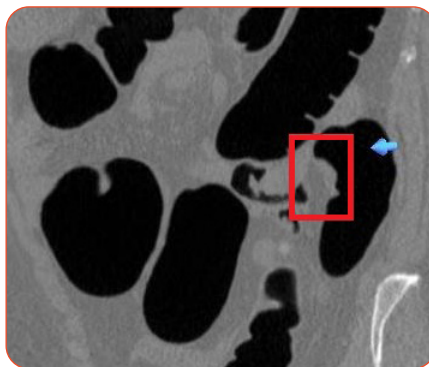
which was not collected for examination. The biopsy results confirmed the presence of a small cystic lymphangioma.

**DISCUSSION**

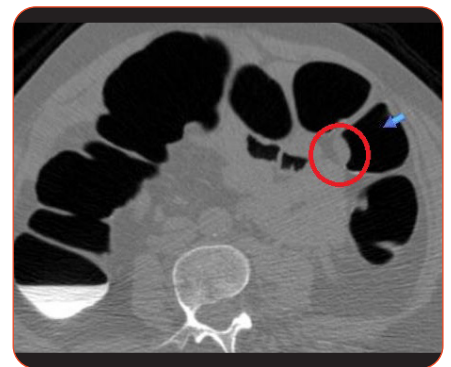
Literature reports that colon lymphangioma is a rare benign lesion.<sup>[2,5,10]</sup> Its treatment has changed over the years. Surgery, in the form of colectomy, was employed before the invention of endoscopic procedures.<sup>[25]</sup> Currently the latter are the mainstay of treatment. Surgery is



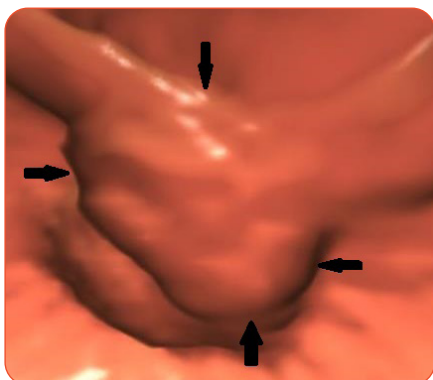
**Figure 5a.** 3D supine view showing soft tissue lesion (black arrows).



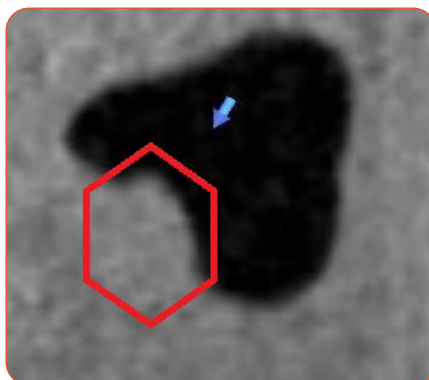
**Figure 5b.** 2D supine coronal view showing soft tissue lesion in the descending colon (red rectangle).



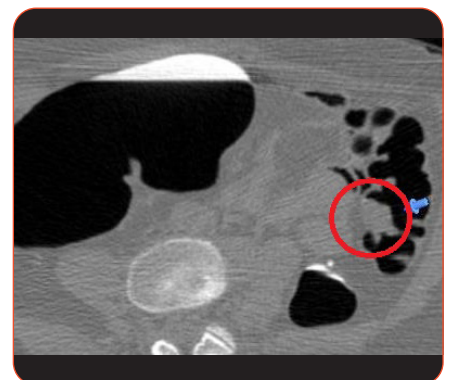
**Figure 5c.** 2D supine axial view showing a soft tissue lesion (red circle).



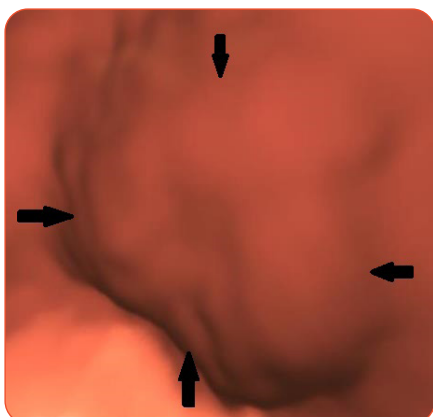
**Figure 5d.** Prone view showing difference in appearance of 3D lesion and appears as an extrinsic lesion (black arrows).



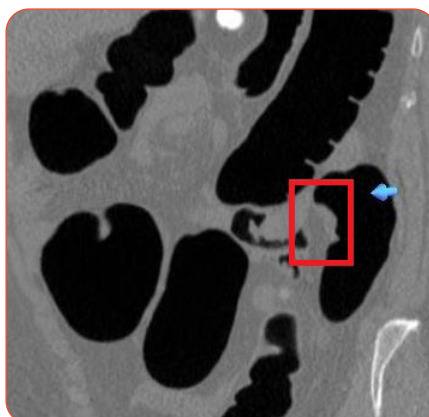
**Figure 5e.** 2D prone coronal view showing the soft tissue lesion (red hexagon).



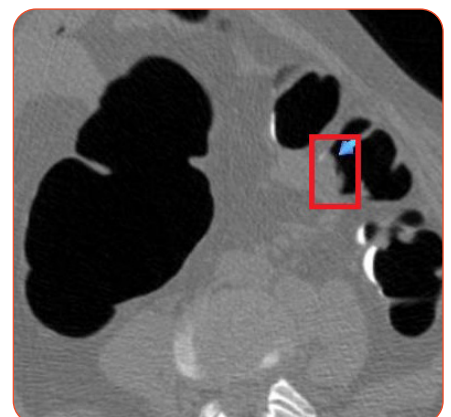
**Figure 5f.** 2D axial prone view showing the soft tissue lesion (red circle) has changed in appearance compared to the supine view.



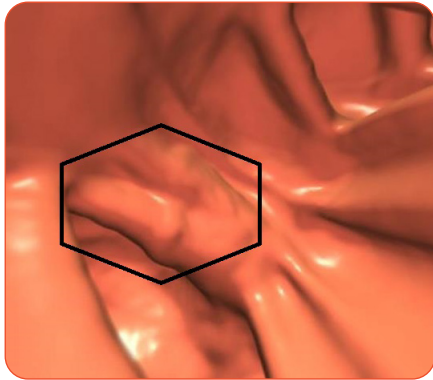
**Figure 5g.** 3D right decubitus view showing that the soft tissue lesion (black arrows) morphology is different from the supine and prone images.



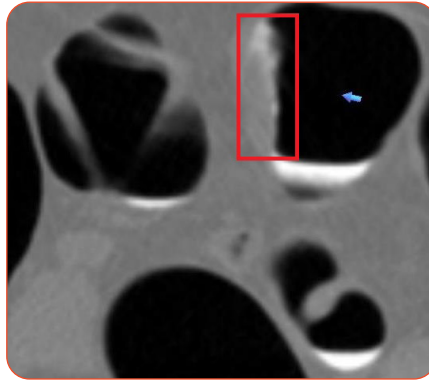
**Figure 5h.** 2D right decubitus coronal view showing the soft tissue lesion (red circle).



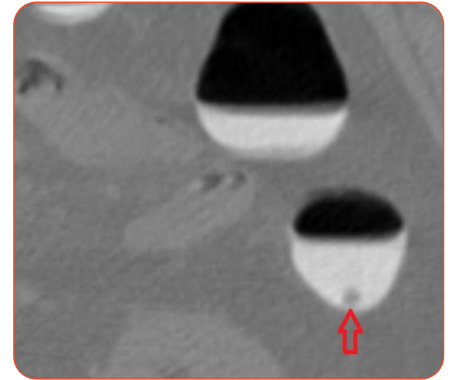
**Figure 5i.** 2D right decubitus sagittal view showing the soft tissue lesion (red rectangle).



**Figure 6a.** 3D of a patient with an irregular fold (black hexagon).



**Figure 6b.** 2D axial view of the 3D irregular fold. Note the barium coating the lesion (red rectangle) which is a feature of a polyp or carcinoma.



**Figure 7.** 2D supine axial view showing visualisation of a submerged polyp (open red arrow) in opacified residual fluid.

usually reserved for complications, such as intussusception.<sup>[25]</sup> Excessive use of invasive treatment for this benign lesion should be avoided; endoscopic resection should be the preferred treatment.<sup>[15]</sup> Although endoscopic resection is recommended for removal of 1-2cms lesions it has been successfully used for removal of 5cms lesions.<sup>[7,26]</sup>

In view of the described CTC features of colon lymphangiomas the treatment of non-pedunculated lesions or those smaller than 3cms could be managed conservatively with follow-up examination after 3-5 years. Endoscopically larger lesions can be punctured and the fluid sent for analysis, and several biopsies may also be taken at the same time. This was the procedure undertaken for the lymphangioma in the proximal ascending colon in case 1. In case 2 the lymphangioma was in the descending colon. Both patients had OC examinations and biopsies taken and the finding for both was lymphangioma.

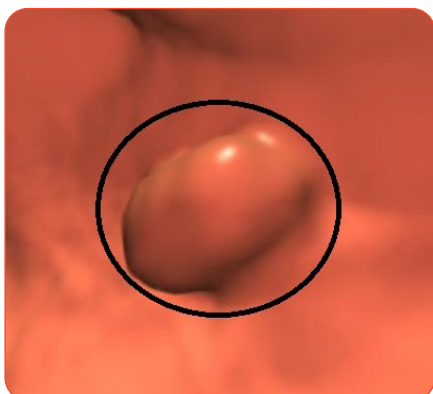
The 3D fly-through, when correlated with

2D findings, makes the diagnosis of colon lymphangioma a lot easier. The lesion in both cases was smooth and sessile and located within the submucosal area (Figures 4b, 4c, 4d, 1f, 5b, 5c, 5e, 5f, 5h, 5i). The 3D appearance was that of an extrinsic lesion (Figures 4a and 5a). Of significance, as shown in Figures 4d, 4f, 5c, 5e, and 5f, unlike what usually occurs with a polyp (Figures 6a and 6b), there is no 'tagging' of the lesion with barium.

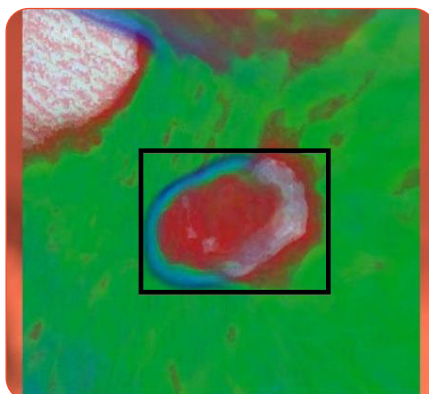
When one interprets CTC images, including each 3D fly-through, it is important to always consider all possible pathology based on the patterns and shape of a lesion as well as its HU values. In view of this the lesion in the two cases above did not resemble a polyp, lipoma, and leiomyoma. The shape of the lesion changed, as shown in Figures 4b, 4f, 5c, and 5f, on the axial, sagittal and coronal plane 2D images. The fluid in the lymphangioma causes a change in shape in different postural positions. The change in appearance between supine and prone images may be quite striking.

Stool and polyp may look identical, therefore tagging is an essential part of bowel preparation. On 2D viewing small air bubbles in a lesion indicates this is stool. The most important differentiation between stool and polyp rests on whether there is movement of the polypoidal lesion between supine and prone views.<sup>[24]</sup> If a lesion moves from the posterior to the anterior wall when a patient's position is changed from supine to prone then this is indicative of stool. Occasionally stool may be adherent to the wall of the colon and this is when tagging plays an important role. In the majority of polyps barium has been shown to stick to the surface of a polyp. A black filling defect in tagged residual fluid is a feature of a submerged polyp; in untagged fluid the black filling defect will not be seen (Figure 7).

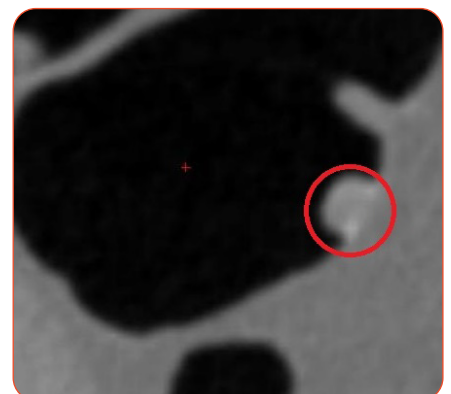
As discussed above CTC is a useful clinical tool in the diagnosis of colonic lymphangioma. CTC routinely includes supine and prone studies; right and/or left decubitus views may be performed if needed. Measuring of the HU is a simple



**Figure 8a.** 3D view showing a polypoid lesion (black circle).



**Figure 8b.** Translucent display (TD) of the polypoid lesion in Fig 5a. The red intensity in the black rectangle indicates this is a soft tissue lesion.



**Figure 8c.** 2D view showing the lesion (red circle) with thin barium layer.



Figure 8d. 2D axial view HU value of 147.

way of deciding whether a lesion is cystic or solid. The fluid in cystic lymphangiomas is often homogeneous with varying attenuation; negative attenuation occurs when the fluid is chylous and there is high attenuation when the fluid is haemorrhagic.<sup>[13]</sup> A polyp is solid and its HU value is usually > 40 HU (Figures 8a to 8d).

With all the different views being available, together with HU measurements, means that a diagnosis of a lymphangioma may be made radiologically.

## CONCLUSION

Colonic lymphangioma is a very uncommon

finding. This state of affairs as reported in the literature is changing because more of these lesions are being identified by OC and treated by puncturing the lesion and by taking multiple biopsies.<sup>[1,7]</sup> Literature refers to barium, ultrasound, CT and MRI for making a diagnosis<sup>[2]</sup> but there is no mention of CTC. In view of screening CTC being recognised as a colorectal cancer test<sup>[9,27,28]</sup> it is postulated that in the years ahead lymphangiomas will be diagnosed more frequently at CTC.

The cystic lesion in both CTC cases presented and discussed changed shape in the various positions. If a colon lesion changes when compressed during either OC or endoscopic ultrasound then a diagnosis of lymphangioma is made.<sup>[1,10]</sup> The complications and risks of OC are well described in the literature.<sup>[29,30]</sup> CTC is the least invasive of all the tests available hence radiologists may be the first to come across this uncommon condition.

It is important to check the shape of lesions in the colon in the different positions of patients. Small 1-2cms soft tissue lesions that change shape and dimensions in the different CTC positions could safely be followed-up in 3-5 years; this would be a less aggressive management of such

lesions. Pedunculated soft tissue lesions, and those larger than 3cms could be referred directly to OC for treatment by cyst aspiration and endoscopic biopsy.

Collaboration between radiologists, gastroenterologists, and referring physicians, should lead to optimal evaluation and management of intramural submucosal lesions of the colon.

## CONFLICT OF INTEREST

None.

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## PATIENT PERMISSION

The patients were informed that their images would not include their names and that their identities would not be mentioned in the paper. Verbal consent was obtained.

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