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Are there specific endosonographic features in Crohn's patients with perianal fistulae?

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Abstract

Both 2-dimensional and 3-dimensional endoanal ultrasounds have been shown to be accurate in the definition of the anatomy of complex fistulae-in-ano in patients with perianal Crohn's disease. Recently, a Crohn's Ultrasound Fistula Sign (CUFS) has been suggested as a discriminating feature of perianal Crohn's disease as has the presence of fistulous debris and fistular bifurcation. We blindly assessed 197 patients (39 Crohn's fistulae and 158 cryptogenic fistulae) to determine if these signs differentiated fistula types. The incidence of CUFS in Crohn's cases was 17/39 (43.6%) and in cryptogenic cases was 4/158 (2.5%) (P<0.0001). The sensitivity, specificity, positive and negative predictive values and accuracy for CUFS were 43.6%, 97.5%, 80.9%, 87.5% and 86.8%, respectively. The presence of debris and fistula bifurcation in evaluable cases had a high specificity (87.2% and 81.8%, respectively) but poor sensitivity. The kappa values for or against CUFS, debris and bifurcation in Crohn's cases between 2 observers blinded to the diagnosis were 0.85, 0.72 and 0.93, respectively and in cryptogenic fistulae were 0.89, 0.85 and 0.80, respectively. The kappa values of an agreed consensus for CUFS in Crohn's disease, cryptogenic fistulae and overall with a third observer with no ultrasound experience were 0.62, 0.85 and 0.77, respectively. The presence of CUFS differentiates Crohn's-related from cryptogenic fistulae-in-ano with a high level of agreement for this sign between experienced and inexperienced observers blinded to the underlying diagnosis. © 2012 European Crohn's and Colitis Organisation. Published by Elsevier B.V. All rights reserved.

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1. Introduction

In a number of cases of Crohn's disease, the first manifestation may be in the form of perianal fistulae.^{1,2} A variety of clinical findings are indicative of perianal Crohn's disease including fleshy skin tags, cavitating ulcers and complex abscesses and fistulae, with a range of specific disease activity and classification systems. 3-6 The natural history of perianal Crohn's disease differs from that of anal fistulae of cryptoglandular origin where the accuracy of preoperative imaging,⁷ in particular, enhanced magnetic resonance (MR) imaging, provides in some complicated and recurrent cases, a gold standard for outcome following surgery.⁸ MR imaging has also been shown to be discriminating of underlying fistula type.⁹ Three-dimensional endoanal sonography has shown a high accuracy for the determination of the anatomy of complex perianal fistulae¹⁰ and for the delineation of the site of the internal fistula opening.¹¹ Recently, some have used specific endoanal ultrasonographic features to separate Crohn's-related from cryptogenic anal fistulae with Zawadzki et al. suggesting that the presence of a hypoechogenic rim with a surrounding hyperechoic region around abscesses and fistulae is a specific ultrasonographic sign of Crohn's disease.¹² This sign has been designated the Crohn's Ultrasound Fistula Sign (or CUFS) and may represent a peri-fistula inflammatory process specific to Crohn's disease. Further, Blom et al. have suggested that on 3-dimensional endosonography, that Crohn's fistulae are more often bifurcating in structure, are wider than cryptogenic fistulae and tend to contain more hyperchoic debris.¹³ They have postulated that these fistula-specific features correlate with overall disease activity.¹⁴ This study aims to determine if 3-dimensional ultrasonographic criteria previously described are pathognomonic of Crohn's fistulain-ano.

2. Patients and methods

Between January 2010 and March 2012, 214 patients with perianal sepsis were assessed with 3-dimensional endosonography at the Chaim Sheba Medical Center Israel. This center acts as a tertiary referral coloproctology and inflammatory bowel disease service. All cases of Crohn's disease were confirmed histologically with perianal fistula biopsy, enteric or colonic biopsies at endoscopy or from prior resectional specimens. All cases were examined with a ProFocus ultrasound machine (Bruel-Kjaer Medical Herlev, DK) equipped with a 2050 13 MHz rotating probe. Patients were examined by one of 3 clinicians who had a minimum of 10 years' experience performing 3-dimensional endosongraphy and who performed >100 ultrasounds per annum. All 3-dimensional volume datasets were all stored on computer and were blindly analyzed retrospectively by 2 of the ultrasonographers (AZ and DC) without knowledge of the diagnosis. Discordant results were re-analyzed for a final agreed consensus on the presence or absence of CUFS. To assess whether an untrained observer without endoanal ultrasound experience could detect CUFS, a further blinding was created against the accepted diagnosis using one our colorectal residents (NH). Fig. 1 shows a 3-dimensional image of a CUFS fistula and a cryptogenic fistula without evidence of the CUFS sign,



Figure 1 *Top image* 3-dimensional axial image of the Crohn's Ultrasound Fistula Sign (CUFS) showing an abscess collection (A) crossing the external anal sphincter (EAS) flanked by an hypoechoic edge (*arrow*) and a surrounding hyperechoic rim (*block arrow*). This is distinct from the appearance of a cryptogenic fistula (*bottom image*) where the fistula track (*arrows*) shows no hypo- or hyperchoic rims, appearing as a simple hypoechoic track traversing the external anal sphincter (EAS).

highlighting the ultrasonographic distinguishing features between the two fistula types.

As a secondary study, a blind assessment of the presence of fistula bifurcation or secondary extension and determination as to whether the fistula was filled with hyperechogenic debris was made between the 2 experienced observers on post-processing of the 3-dimensional datasets. An example of each ultrasound feature is shown in Figs. 2 and 3, respectively. All patients undergoing ultrasound with perianal Crohn's disease were incorporated onto an electronic database permitting retrieval of the basic clinical information including, age, gender, duration of perianal Crohn's disease, the presence of associated enteric Crohn's disease and its duration, the history of prior anal and/or abdominal procedures, the history of prior biologic therapy and a calculation of a simplified perianal Crohn's Disease Activity Index (PCDAI) as reported by Hughes.³ The duration of fistula symptoms and the history of prior anal surgery were recorded for the cryptogenic fistula group.

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