Research Article

Adult Intussusception: Should Bowel Resection Always be Performed?

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Abstract

Introduction: Adult intussusception is considered a rare condition and it is commonly caused by an underlying lesion.

Material and methods: Retrospective review of 65 patients diagnosed at a tertiary centre and literature review.

Results: Median age was 52,7 years and 55,4% were male. 18 cases were incidentaly found

during CT exploration and had an asymptomatic course (27,7%). Most frequent symptoms were pain (70,2%) and nausea or vomiting (44,6%). In regards to their location, 33 intussusceptions were entero-enteric (50,8%), 14 entero-colic (21,4%) and 13 colo-colic (20%). Surgical procedure was carried out in 42 patientes. Bening lesion causing intussusception was found in 16 cases (mainly polips) and malignant lesion in 23 (mainly adenocarcinomas). 16 out of 18 entero-enteric intussusceptions had an asymptomatic course (88,8%) while those involving the colon caused symptoms in 92,6% of patients. Lesion causing instussusception was more likely to be malignant depending on its location: 35,7% in entero-enteric, 61,5% in entero-colic and 76,9% in colo-colic.

Conclusion: Symptoms and location of intussusceptions can be useful in predicting malignancy potential of underlying lesions. The more distal the more malignant. For intussusceptions involving the colon, oncologic surgical resection must be performed.

Keywords: Intussusception, Adult, Surgery

Introduction

Intussusception occurs when a bowel segment slides into the lumen of the adjacent. It is a rare condition in adults causing less than 5% of bowel obstructions [1]. Unlike in children, for whom most of cases are idiopathic and can be managed non-operatively by reduction, an underlying lesion is present up to 80-95% in adults and surgery is usually required [2].

Our objective is to analyse symptomatology, aetiology, diagnosis and treatment of these patients in our hospital and to summarize global management of intussusceptions in adults over the last decades.

Material and Methods

Retrospective study of intussusceptions diagnosed in adults from 2010 to 2017 in Universitary and Polytechnic Hospital La Fe from Valencia, Spain. Both asymptomatic intussusceptions, diagnosed as an incidental finding on imaging tests performed for other reasons, and symptomatic intussusception requiring hospital admission or surgery have been included.

Instussusceptions have been classified as follows: entero-enteric for those ones involving the small bowel (jejunal, jejuno-ileal or ileal), entero-colic for those ones involving terminal ileum and colon (ileo-cecal if the appendix is involved and ileo-colic if the appendix is not involved), colo-colic for those ones involving ascendent, transverse, descending or sigmoid colon and other instussusceptions (gastro-duodenal and rectal).

Length of symptoms has been divided in acute (onset less than 4 days before), subacute (between 4 and 14 days) and chronic (symptoms lasting more than 14 days).

Data about symptoms, imaging test, surgery, pathological examination and postoperative complications have been collected. Median follow-up was 4 years.

Percentage for discrete data and median for continuous data was use for descriptive analysis. SPSS® Statistics version 22.0 has been used for statistical analysis.

Studies including patients under the age of 18, case reports and all those cases not showing intussusceptions' aetiology have been excluded from literature review.

This research has been evaluated and approved by the investigation commission and the ethics committee from our hospital.

Results

Descriptive analysis

65 intussusceptions have been diagnosed in adults from 2010 to 2017 in our centre. Median age was 52.7 years (range from 18 to 95) and 55.4% were male. 18 intussusceptions had an asymptomatic course (27.7%) being diagnosed during imaging tests for other reasons (Table 1).

Among 47 symptomatic intussusceptions, 14 had an acute onset of symptoms (29.8%), 5 a subacute onset (10.6%) and 28 patients had chronic course (59.6%). Most frequent symptoms were pain in 33 patients (70.2%), nausea and vomiting in 21 (44.6%), gastrointestinal bleeding in 14 (29.8%), weight loss in 14 (29.8%) and abdominal distension in 8 (17%). Fever was only present in 3 patients, palpable mass in 1 and abdominal tenderness in 1. In univariate analysis the presence of symptoms (OR=17.7), weight loss (OR=17.5) and gastrointestinal bleeding (OR=6) were significantly related to the existence of an underlying malignant lesion.

According to their location 33 were entero-enteric (21 jejunal, 2 jejuno-ileal, 10 ileal), 14 entero-colic (1 ileo-colic and 13 ileo-cecal), 13 colo-colic (9 colo-colic and 4 sigmoid) and 5 in other locations (2 gastro-duodenal, 2 duodenal and 1 rectal).

Computarized tomography scan (CT) was performed in 61 patients (93.8%) and was the most used diagnostic test followed by digestive endoscopy in 14, magnetic resonance imaging in 4, endoscopic capsule in 2 and contrast enema in 1 (Figure 1).

Of the 47 patients with symptomatic intussusceptions 42 required surgery (89.4%), 3 patients had a spontaneous resolution with conservative management and 2 patients were not operated due to their excessive comorbidity. Surgical interventions performed were right hemicolectomy in 16 patients, small bowel resection in 13, anterior rectal resection in 4, segmental colonic resection in 2, subtotal colectomy in 1, ileocecal resection in 1, enterotomy in 1, gastrostomy and polyp resection in 1, intussusception reduction in 1 and adhesiolysis in 1. In two cases, intussusception was not
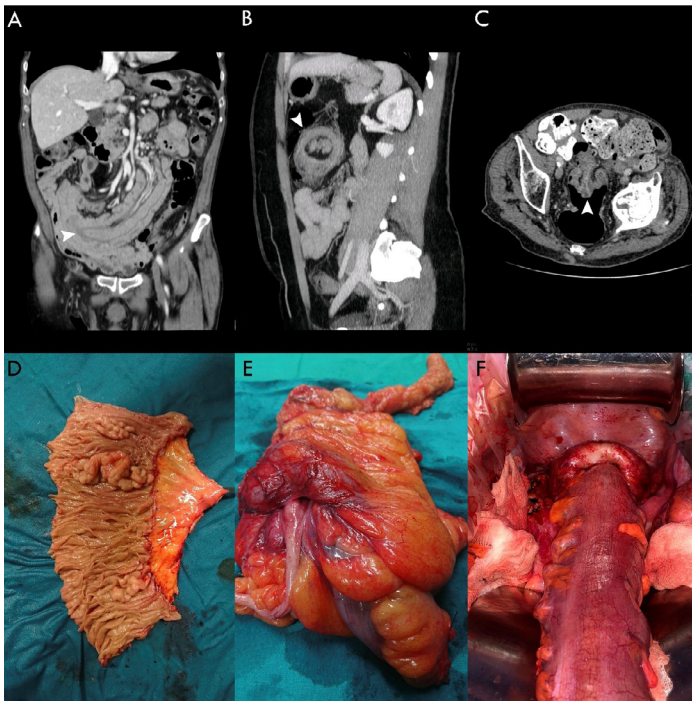
Figure 1: A. Coronal CT slice: long jejunal intussusceptions is shown; B. Satital CR slice: target sign can be observed on transverse colon intussusception; C. Axial CT slice: rectal intussusception with evident leading point; D. Ileal resection specimen showing underlying lesion causing intussusception; E. Right colectomy specimen with ileo-cecal intussusception; F. Intraoperative capture showing rectal intussusception.
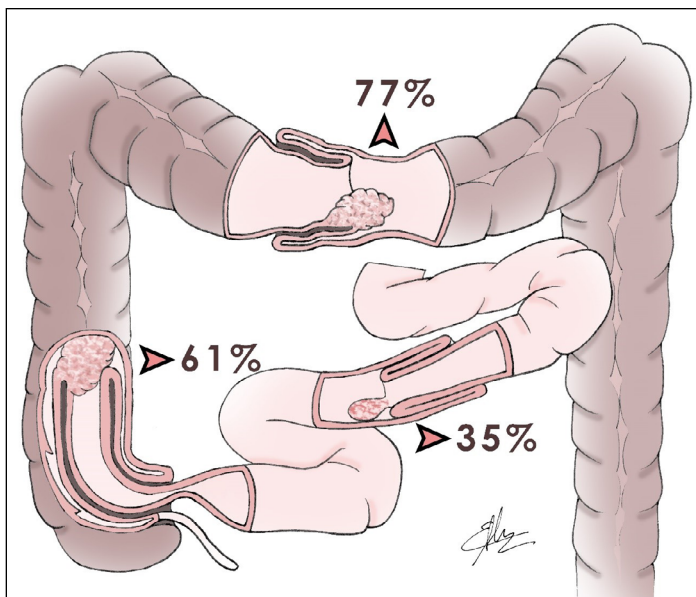

Figure 2: Illustration showing percentage of underlaying malignant lesions depending on intussusception's location. As noted, the more distal intussusceptions are more likely to be caused by malignant lesions

found during diagnostic laparoscopy (both entero-enteric intussusceptions diagnosed by CT scan).

Bowel resection was performed in 39 patients. A benign lesion was identified in 16 surgical specimens. A malignant lesion was found in 23 patients (Table 2). No lesions were found in 3 patients during surgery although bowel resection was performed in one of them anyway (no lesion was observed in histological study).

Among 16 patients diagnosed with adenocarcinoma 1 had a T1, 4 a T2, 8 a T3 and 3 a T4a tumour. Metastatic adenophaties were found in 3 patients. Median harvested lymph nodes were 18,7. Of these patients 7 required an urgent surgery. Median harvested lymph nodes were significantly higher in patients who underwent an elective surgery, 23.6 vs. 13.4 (p<0.005).

Postoperative complications were registered in 9 patients (21.4%). Anastomotic leak occurred in 3 patients and all cases could be managed non-operatively until fistula resolution. 1 patient had an auto-limited lower gastrointestinal bleeding. Early postoperative deaths occurred in 5 patients: 2 nosocomial pneumonia in haematological immunosuppressed patients, 1 massive mesenteric ischemia caused by intussusception, 1 massive postoperative acute pulmonary embolism and 1 cardiac arrest following bronchoaspiration.

No intussusception recurrence was observed in operated patients. Ventral hernia was present in 3 patients after a 4-year median following. After a 12 to 84 months following time no recurrence or intussusception-related admissions were registered for patients diagnosed with an asymptomatic intussusception.

Clinical and location malignancy predictors

As noted above the presence of symptoms was related to the presence of an underlying malignant lesion (OR=17.7). Asymptomatic intussusceptions were mainly located at small bowel (88.9%), however, intussusceptions involving the colon (entero-colic and colo-colic) caused symptoms in 92.6% of patients.

Intussusceptions involving the colon were cause by a malignant lesion in 69.2% of cases. Moreover, intussusception location is a predictor of potential malignancy as the more distal it is the more proportion of malignant lesions are observed: 35.7% of entero-en-

Table 1: Demographic data

Age		52,7 (rango 18-95)
Sex (F/M)		29/36
Symptoms		47 (72,3%)
Pain		33
Nausea and vomiting		21
Weigh loss		14
GI bleeding		14
Diarrhea		13
Constipation		10
Anemia		9
Others		16
Symptoms onset	Acute	14 (29,8%)
	Subacute	5 (10,6%)
	Chronic	28 (59,6%)
Location	Entero-enteric	33 (50,8%)
	Entero-colic	14 (21,4%)
	Colo-colic	13 (20%)
	Other	5 (7,8%)
Surgery		42 (64,6%)
	Elective	17
	Urgent	25
Aetiology	Benign	18 (27,7%)
	Malignant	25 (38,5%)
	Idiopathic	22 (33,8%)

Table 1: Demographic data

	Benign		Malignant	
	Primary	Secondary	Primary	Secondary
Enteric	2 Peutz-Jeghers polyp	1 Bezoar	1 Ileal GIST	1 Peritoneal undifferentiated sarcoma
	1 Hamartomatous polyp	1 Meckel's diverticulum		1 Kaposi' sarcoma
	1 Submucous Lipoma	1 Jejunal diverticulum		1 Leukemia
	2 No lesion			1 MALT lymphoma
Entero-colic	2 Tubular adenoma	1 Fibrosing colonopathy	6 Adenocarcinoma	1 DLBC lymphoma
	1 Submucous lipoma	1 Adhessions		1 Pancreatic adenica. metastasis*
Colic	1 Submucous lipoma		10 Adenocarcinoma	
	1 Tubulo-villous adenoma			
	1 Villous adenoma			
Others	1 Gastric inflammatory polyp	1 Peptic duodenal stricture	1 Rectal adenocarcinoma	1 Gastric adenoca. carcinomatosis*
	1 Duodenal submucous tumor			
* non-resected lesions				

teric (15.1% if asymptomatic entero-enteric intussusceptions are considered), 61.5% of entero-colic and 76.9% of colo-colic intussusceptions were caused by a malignant lesion (Figure 2).

Discussion

Incidence

Intussusception in adults is extremely rare representing less than 5% of all intussusceptions. Based on historic series, intussusceptions are responsible for approximately 1-5% of hospital admissions for bowel obstruction in adults [1-5]. In most recent series, incidence of intussusceptions is less than 1% of all bowel obstructions and represent 0.003-0.02% of all hospital admissions [6-9].

Symptoms and clinical presentation

All series from the last century have reported pain as the most frequent symptom followed by nausea and vomiting. Chronic symptoms were more commonly reported in the first series but in the last two decades, intussusceptions are usually diagnosed early from the onset of symptoms [1,2,5,6,10]. Classical children's triad (pain, palpable mass and haematochezia) is only seen in 2.13-15.7% of intussusceptions in adults [11-14]. Complete bowel obstruction is present in less than 20% of patients [1,3,13] and abdominal mass in palpable in 3-15% of adults [7,11,13,15].

In the last twenty years, on account of CT scan increasing use, a new entity has been defined: transient intussusception. It refers to asymptomatic intussusceptions found as an incidental finding during imaging test for other reasons. They represent up to 50% of intussusceptions in some series and no extra inquiry is required as they trend to spontaneous resolution [9,13,14].

Some authors have related several symptoms to the existence of an underlying malignant lesion. Jong et al. found chronic symptoms to be related with malignancy [10]. Okendi et al. [13] found haematochezia (OR=14.41), occlusive symptoms (OR=9.13) and palpable mass (OR=4.56) to be related with malignancy on multivariate analysis.

Aetiology and location

Unlike childhood intussusception, which is idiopathic in 90% of cases, 80-90% of adult intussusceptions are caused by an organic lesion [1,5,9-12]. Mechanism causing intussusception is no well known but the most accepted theory is that normal peristalsis can be disrupted by any lesion or irritating factor leading to an intussusception.

In the fist reported series, up to 90% of intussusceptions were secondary to a lesion and 46-69% were malignant tumours with no differences between small and large bowel distribution [1-5]. From year 2000, transient asymptomatic intussusceptions have been increasingly diagnosed by CT scan representing up to 50% in some series [6-9,16]. They are mainly located at small bowel so intussusception's distribution has changed. In our literature review, including 1172 cases, entero-enteric intussusceptions represent 51% of cases, entero-colic 27% and colo-colic 21%. Malignant underlying lesions were found in 31%, 42% and 55% respectively (Table 3). As observed in our study, the more distal the intussusception is located the more likely to be caused by malignant lesions.

As general rule, most malignant lesions causing small bowel intussusceptions are metastatic (melanoma, sarcoma or lymphoma) and those causing large bowel intussusceptions are primary adenocarcinomas. In our literature review, intussusceptions were caused by benign lesions in 43% of cases, by malignant lesions in 37% and idiopathic in 20%.

Diagnosis

Classically, abdominal x-ray with or without contrast barium was the only available diagnostic tool for bowel obstructions. The characteristic coiled-spring image was only observed in 6.6-29.1% of intussusceptions [1-7,16-19].

Nowadays, CT scan has evolved into one of the primary diagnostic tools in abdominal pathology. In 1997, Azar et al reported a 78% accuracy for CT scan in detecting intussusceptions [5]. Babiera et al. [18] described 3 different radiologic patterns: target mass pattern, pseudokidney pattern and sausage-shaped pattern. The target pattern corresponds to an early intussusception with only minimal obstruction and no sign of ischaemia at pathology, the reniform pattern is described as a bilobed density with peripheral high attenuation and lower attenuation centrally, as a result of of thickening bowel wall surrounding the intussusceptum and the sausage shape results from alternating areas of low and high attenuation related to the bowel wall, mesenteric fat and fluid, intraluminal fluid, contrast material or air [14].

In the last two decades, intussusceptions have been more and more diagnosed due to increasing use of CT scan and several authors have tried to identify related factors to an underlaying lesion. Tabrizian et al found intussusception longer than 3,5cm (OR=25.8), radiologic bowel obstruction (OR=11.4) and colon involvement (OR=33,3) to be related with a secondary intussusception but only the two last factors were independently related on the multivariate analysis [9]. According to these finding they define a low risk group: unique entero-enteric no longer than 3,5cm intussusception in patients under age 50 without occlusive symptoms.

From 2010, several studies have tried to identify malignant le-

Table 3: Literature review.

	Year	N	Most freq. symptoms	Location				Surgery	Histology		Idio-pathic	CT scan
				Enter-ic	Ente-ro-colic	Colic	Oth-er		Benign	Malig-nant		
Dean et al. [1]	1955	96	1. Pain; 2. Obstruction	29 (30%)	35 (37%)	32 (33%)	0	100%	26 (27%)	58 (60%)	12 (13%)	0
Stubenborn et al. [2]	1969	34	1. Pain; 2. Obstruction	12 (35%)	18 (53%)	4 (12%)	0	97%	17 (50%)	10 (29%)	7 (21%)	0
Nagorney et al. [3]	1980	48	1. Pain; 2. Obstruction	18 (38%)	6 (12%)	24 (50%)	0	96%	18 (38%)	22 (46%)	8 (16%)	0
Azar et al. [5]	1996	58	1. Pain; 2. Obstruction	44 (76%)	14 (24%)		0	100%	29(50%)	27 (47%)	2 (3%)	13 (22%)
Martín-Lo-renzo et al. [16]	2004	7	1. Pain; 2. Obstruction	4 (57%)	3 (43%)	0	0	100%	4 (57%)	3 (43%)	0	4 (57%)
Erkan et al. [6]	2005	13	1. Pain; 2. Obstruction	10 (77%)	1 (8%)	2 (15%)	0	100%	7 (54%)	5 (38%)	1 (8%)	7 (54%)
Zubaidi et al. [8]	2006	22	1. Pain; 2. Obstruction	14 (64%)	2 (9%)	6 (27%)	0	90%	11 (50%)	8 (36%)	3 (24%)	6 (27%)
Goh et al. [7]	2006	60	1. Pain; 2. Obstruction	16 (27%)	23 (38%)	18 (30%)	3 (5%)	100%	26 (43%)	28 (47%)	6 (10%)	30 (50%)
Ahn et al. [10]	2008	42	1. Pain; 2. Obstruction	22 (52%)	10 (24%)	10 (24%)	0	100%	23 (55%)	10 (24%)	9 (21%)	33 (79%)
Hanan et al. [25]	2009	16	1. Pain; 2. Obstruction	5 (31%)	6 (38%)	5 (31%)	0	100%	6 (38%)	8 (50%)	2 (12%)	3 (19%)
Yakan et al. [23]	2009	20	1. Pain; 2. Obstruction	13 (65%)	4 (20%	3 (15%)	0	100%	14 (70%)	4 (20%)	2 (10%)	12 (60%)
Morera et al. [24]	2009	30	1. Pain; 2. Obstruction	18 (60%)	6 (20%)	4 (13%)	2 (7%)	93%	16 (53%)	12 (40%)	2 (7%)	17 (57%)
Wang et al. [11]	2009	44	1. Pain; 2. Obstruction	20 (46%)	15 (34%)	9 (20%)	0	100%	28 (64%)	12 (27%)	4 (9%)	29 (66%)

Gupta et al. [12]	2011	38	1. Pain; 2. Obstruction	16 (42%)	12 (32%)	10 (26%)	0	100%	18 (47%)	18 (47%)	2 (6%)	20 (53%)
Onkendi et al. [13]	2011	196	1. Pain; 2. Obstruction	138 (70%)	28 (14%)	30 (16%)	0	61%	95 (48%)	43 (22%)	58 (30%)	117 (60%)
Cakir et al. [27]	2013	47	1. Pain; 2. Obstruction	5 (11%)	38 (81%)	4 (9%)	0	100%	23 (49%)	17 (36%)	7 (15%)	15 (32%)
Varban et al. [20]	2013	44	1. Pain; 2. Obstruction	35 (80%)	9 (20%)		0	100%	10 (23%)	13 (30%)	21 (47%)	64 (100%)
Honjo et al. [15]	2015	44	1. Pain; 2. Obstruction	12 (27%)	22 (50%)	10 (23%)	0	100%	9 (20%)	25 (57%)	10 (23%)	44 (100%)
Somma et al. [14]	2015	47	1. Pain; 2. Obstruction	35 (74%)	5 (11%)	7 (15%)	0	66%	20 (43%)	11 (24%)	16 (34%)	47 (100%)
Own Series*	2019	65	1. Pain; 2. Obstruction	33 (51%)	14 (22%)	13 (20%)	5 (7%)	65%	18 (28%)	25 (38%)	22 (34%)	61 (94%)
		1172		499 (51%)	259 (27%)	203 (21%)	10 (1%)		418 (43%)	359 (37%)	194 (20%)	

sions preoperatively. Okendi et al reported the presence of a head of intussuscipiens on CT scan as an independent factor for malignancy (OR=10.56) and surgery requirement (OR=10.08) [13]. Varban et al. [20] stablished personal history of malignant neoplasia (OR=3.7), mass present on CT scan (OR=2,9) and age over 60 (OR=2.2) as risk factors for malignant lesion causing small bowel intussusceptions. In 2018, a predicting score for determining the presence of an underlaying lesion was published by Tan et al. [21] from Singapore General Hospital. They listed 6 different items: female gender (1 point), abdominal pain (2 points), colon involvement (2 points), pathological lead point (2 points), distal diameter >27mm (1 point) and wall thickness >3mm (1 point). Leading point causing intussusception is present in less than 40% of patients scoring 3 points or less and in 70-100% of patients scoring 4 or more.

Treatment

Surgical resection was a constant in the first reported series form early twentieth century as most intussusceptions causing bowel obstruction were diagnosed intraoperatively [1]. From the first reported cases a high proportion of underlying lesions were observed so classical recommendation was to perform a surgical resection without intussusception reduction including locoregional lymph nodes [1-5]. In 2000, Sebbag et al published a paper titled "Intestinal intussusception in adults, treat it like cancer" where these recommendations are highlighted [17].

At the turn of the twenty-first century, due to increasing incidental intussusception being diagnosed, routinely resection for entero-enteric intussusceptions is queried. Evidence from published series show intussusceptions involving the colon (entero-colic and colo-colic) are more likely to be caused by malignant tumours. Pre or intraoperative colonoscopy is recommended when feasible for these patients by some authors [11]. However, entero-enteric intussusceptions are usually idiopathic or cause by benign lesions (except for patients with past history of primary neoplasia in whom metastatic lesion must be suspected). Therefore, unanimous recommendation for intussusceptions involving the colon is to perform an oncologic resection without reduction including locoregional lymph nodes [6-10].

At present, management of entero-enteric intussusceptions is controversial. As low percentage of entero-enteric intussusceptions are caused by malignant lesions some author advocate for intussusception reduction in order to evaluate the involved segment

viability and perform a limited bowel resection [10,15]. However, this is criticized by others arguing that intussusception reduction, when it is caused by a non-suspected malignant lesion, can cause intraluminal seeding and venous dissemination of malignant cells, possible perforation during manipulation and increased risk of anastomotic complications in the face of oedematous and inflamed bowel [11,12].

CT findings are crucial in management of asymptomatic intussusceptions. Several publications have identified risk factor for underlying malignant lesions. Those cases in which leading point is not identified for incidental asymptomatic entero-enteric intussusceptions (up to 50% in some series) no extra inquires are needed and surgical intervention must be avoided as an idiopathic origin is assumed [13,20-22]. In symptomatic patients with several short entero-enteric intussusceptions with no leading point identified on CT scan conservative management is a save option as transient intussusceptions trend to spontaneous resolution [21,22]. The 6-item score developed by Tan et al. could be useful in adult intussusception management [21]. For oncologic patients the presence of a bowel metastatic lesion must be kept in mind, especially for melanoma and lymphoma. In these cases, exploratory laparoscopy can be an appropriate alternative depending on the symptoms and patient status.

Conclusion

Intussusception is a rare cause of bowel obstruction in adults. Nowadays, it is more and more found as an incidental finding during a CT scan for other reason. For transient intussusceptions no additional inquiry is needed as they trend to spontaneous resolution. Intussusception's aetiology can be suspected depending on symptoms and location as symptomatic and more distal intussusceptions are more likely to be caused by malignant lesions. For intussusceptions involving the colon, regardless of symptoms, oncologic resection must be performed.

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