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Inflammatory Fibroid Polyp Presenting as Ileo-colic Intussusception: A Case Report and Review of Literature

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Abstract **AIM:** To provide a brief review of literature on intussusception due to inflammatory fibroid polyp (IFP). **METHODS:** A case report on inflammatory fibroid polyp presenting as ileo-colic intussusception made us review the literature on IFPs presenting as Intussusception, published in PubMed in English language. A comprehensive search of all case reports was done using keywords: intussusception due to IFP, intussusception and IFP, intussusception, IFP. The search covered all case reports from 2000 to 2020. **RESULTS**: 53 case reports with a total of 55 cases were analysed in this article, the range of age with such a presentation was from minimum10 years to maximum 79 yrs age, with median age of presentation 48years. 22 cases were males and 32 females, with one case gender not specified. Most common site of IFP presenting as intussusception was ileum (43cases), leading to ileo-colic intussusceptions, there were 11 case reports of jejunal intussusceptions also.

Keywords: *Inflammatory fibroid Polyp (IFP), Ileocolic Intussusception, Inflammatory myo-fibroblastic tumours (IMT)*

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1. Introduction

Intussusception is the prolapse of a portion of the intestine into an immediately adjacent portion [1]. It is the frequent cause of intestinal obstruction in infants and infrequent among adults with 2 to 3 cases occurring in a population of 1,000,000 per annum [2], accounting for less than 0.1% of all hospital admissions in adults [2]. The lead point associated with intussusception in small bowel could be a benign [70%] or a malignant lesion [30%], whereas in large bowel the lead point is usually a malignant lesion [66%] rather than benign [3]. Here we describe an adult female presenting to the emergency department with symptoms of sub-acute intestinal obstruction [SAIO] due to ileo-colic intussusception with a rare benign neoplastic lead point, an Inflammatory Fibroid Polyp (IFPs).

Inflammatory fibroid polyps are uncommon benign mesenchymal tumours of the gastrointestinal tract with histological resemblance with inflammatory myo-fibroblastic tumours (IMFTs) and consist of spindle shaped stromal elements with inflammatory infiltrate of eosinophils. Historically both were considered the same and IFPs were thought to be gastro-intestinal manifestations of IMFTs [4] Based on extensive studies, both the entities have been now distinguished and many authors consider them as separate entities under the classification of mesenchymal tumours. Because of slightly different biological properties their clinical outcomes vary. IMFTs are known to recur, unlike the IFPs. IFPs have overlapping features with the most common mesenchymal tumor of gastro-intestinal tract, the GIST [5]. IFPs tend to have activating PDGFRA mutations, having molecular overlap with GIST, but lack GIST specific tumor markers DOG-1 or c-KIT; however, both express CD34 [6]. Immunohistochemical staining of IFPs for PDGFRA supports the diagnosis of an inflammatory fibroid polyp [7].

2. Material and Methods

A case report of 70-year-old, female with inflammatory fibroid polyp presenting as ileo-colic intussusception is discussed. Review of literature on 'IFPs presenting as Intussusception', was conducted from database of PubMed. A comprehensive search of all case reports in English language was done using keywords: intussusception due to IFP, intussusception and IFP, intussusception, IFP. The search covered all case reports from January 2000 to March 2020. Articles with sufficient information on age, sex, duration of complaints, diagnostic tool, surgical intervention, site of tumor, and tumor size were taken for review, Inadequate articles with missing information were excluded from the study.

3. Case Report

A 70-year-old, female, presented with complaints of intermittent colicky abdominal pain in the umbilical region of 6 months duration, associated with recurrent episodes of nausea and vomiting. There was history of obstipation during the episodes of pain. Her past medical and family history was insignificant. The general physical examination and abdominal examination were otherwise normal, except for mild tenderness and a vague mass in the right iliac fossa.

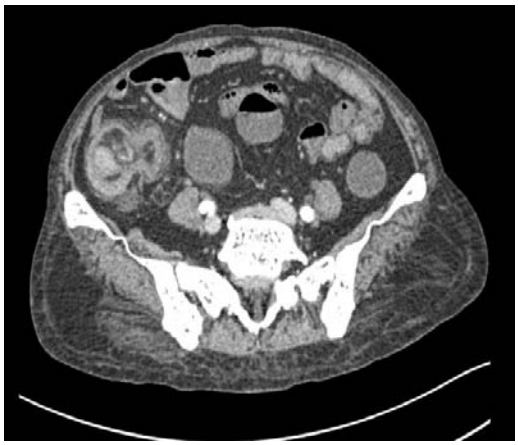

Figure 1. CECT Abdomen showing Ileocolic intussusception

On investigating, her routine investigations tested normal. The abdominal X-ray revealed non-specific air-fluid levels. Ultrasound abdomen revealed dilated small bowel loops with caecal thickening. The contrast-enhanced computed tomography (CECT) scan of the abdomen revealed an ileo-colic intussusception of 7cm length with heterogeneously enhancing mass of size 4.2 x 3.2 x 3.6cm arising from the lead point of intussusception and a few sub-centimetric homogenously enhancing lymph nodes around the ileo-caecal region (Figure 1). The appendix was distinctly visualized on the CT scan.

After an informed consent patient underwent a midline exploratory laparotomy and was found to have an ileo-colic intussusception (Figure 2), (Figure 3). The healthy appendix and the caecum were clearly seen before the reduction of the intussusception (Figure 4). Standard right hemicolectomy with side to side ileo-transverse anastomosis was performed in view of her age, the mass and enlarged mesenteric lymph-nodes on CT scan wherein the possibility of malignancy could not be ruled out.

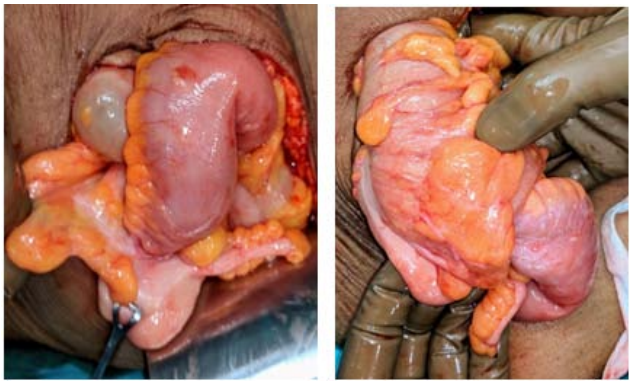

Figure 2. Intraoperative picture showing the ileocolic intussusception with normal appendix lying adjacent but not intussuscepting

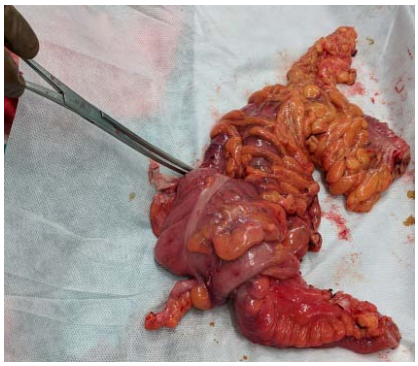

Figure 3. Resected specimen showing the ileo-colic intussusception

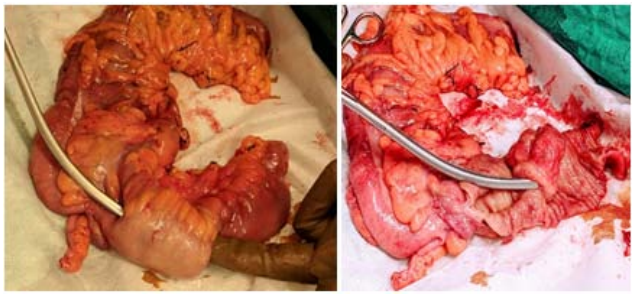

Figure 4. Resected specimen after reduction with the mass and after cutting open the ileum

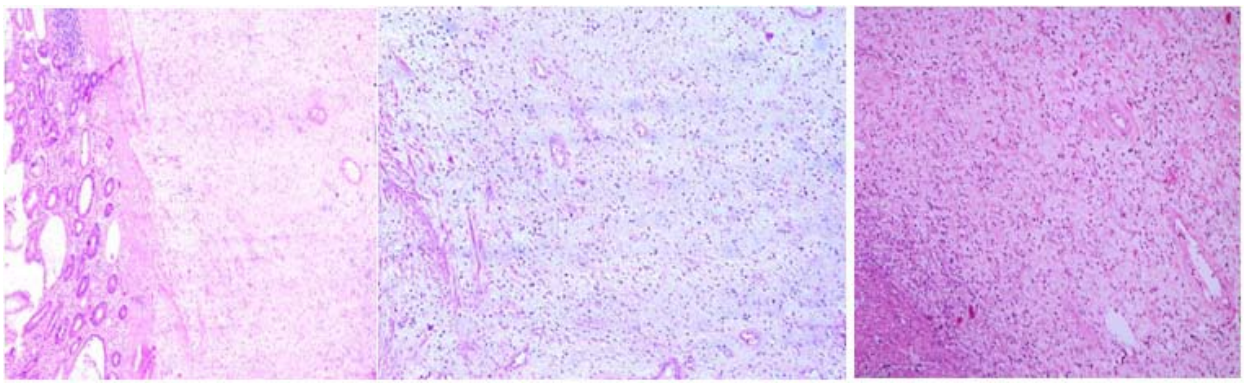

Figure 5. Focal surface ulceration and submucosal growth is seen. The underlying stroma is composed of loosely dispersed fibroblastic proliferation having spindle to stellate shape, imperceptible cytoplasm, bland nuclear features and focal reactive atypia. Interspersed are numerous plasma cells, mast cells, lymphocytes, and eosinophils. Presence of thin and thick-walled vessels are seen within the lesion. No necrosis, atypia, or increase in mitosis is noted. Histopathology is suggestive of IFP

On macroscopic examination of the gross excised specimen, the serosal surface was unremarkable; on cutting it open, the lumen of small intestine showed a protruding polypoidal mass of size 4x3.5x2.5cm. Microscopic examination showed a polypoidal growth with focal surface ulceration arising from the submucosal location. Underlying stroma showed loosely dispersed fibroblastic proliferation having spindle to stellate shaped cells, interspersed with numerous inflammatory cells suggestive of inflammatory fibroid polyp (Figure 5). The IHC markers were supportive of the diagnosis. Patient had an uneventful postoperative recovery. She was kept under follow-up. There has been no recurrence in the last one year.

4. Literature Review

Extensive search of all published articles in PubMed from year 2000 to March 2020 was done. A total of 53 case reports with a total of 55 cases were included for the review. On descriptive analysis of the data, the range of age with such presentation was from 10 years minimum to maximum of 79 years, with median age of 48 years. 22 were males and 32 females, in one case gender was not specified. Most common site of IFP presenting as intussusception was at ileum (43cases), leading to ileo-colic intussusception, there were few, only 11 case reports of jejunal intussusceptions. Individualities of all cases is briefed in Table 1.

Table 1.

Published year	Author	Age	Sex	Duration of complaint	Diagnostic tool	Surgical intervention	Site of tumor	Tumor size
2020	Park *et al* [22]	23	M	3d	CT	laparoscopic S.Resection (Segmental Resection)	2 polyps in Jejunum	5cm,2.5cm
2020	Karuhanga *et al* [23]	48	M	2w	AXR	NS (Not Specified)	Jejunum	4cm
2019	Kang et al [24]	48	F	NS	CT	Enterotomy and excision	ileum	2.5cm
2019	Gupta *et al* [25]	79	M	2d	CT	S.Resection	Ileum	6cm
2019	Paramythiotis *et al* [26]	42	F	7d	CT	Right hemicolectomy	Ileum	NS
2019	Mader *et al* [27]	38	F	3d	CT	laparoscopic right hemicolectomy	Ileum	3.5cm
2018	Jan *et al* [28]	60	NS	NS	CT	S.Resection	Ileum	NS
2018	Al *et al* [29]	47	F	4d	CT	S.Resection	Ileum	16cm
2018	Tajima et al [30]	70	F	NS	USG	S.Resection	Ileum	4cm
2017	Carvalho et al [31]	41	F	6h	USG,CT	S.Resection	Ileum	4.8cm
		51	F	2m	CT	S.Resection	Ileum	5.5cm
2017	Mochizuki *et al* [32]	35	F	2d	CT	laparoscopic S.Resection	Ileum	4.5cm
2017	Grover et al [33]	45	M	10d	NS	S.Resection	NS	NS
2017	Adams *et al* [34]	61	F	2w	CT	S.Resection	Ileum	7.5cm
2017	Rais *et al* [35]	22	M	1Y	CT	S.Resection	Ileum	3cm
2016	Basara *et al* [36]	42	F	NS	CT	S.Resection	Ileum	NS
		51	F	NS	CT	S.Resection	Ileum	5cm
2015	Hiremath *et al* [37]	46	M	4d	CT	S.Resection	Ileum	4cm
2015	Sakran et al [38]	40	M	NS	CT	laparoscopic S.Resection	Ileum	NS
2015	Kimura *et al* [39]	30	M	2w	CT, Diagnostic Laparoscopy	S.Resection	Ileum	4cm
2015	Kang et al	51	F	1m	CT	S.Resection	Jejunum	4cm
2014	Bae et al [40]	48	F	7days	CT	S.Resection	Ileum	3.5 cm
2014	Joyce et al [41]	62	M	12hr	CT	S.Resection	Jejunum	42mm
2014	Sulu *et al* [42]	41	F	-	Colonoscopy	Right hemicolectomy	Ileum	12cm
2013	Teli *et al* [43]	45	F	3d	US,CT	S.Resection	Ileum	2.5cm
2013	Neishaboori *et al* [44]	40	F	3days	US	S.Resection	Jejunum	18cm
2013	Siminas *et al* [17]	10	M	10d	US,CT	Right hemicolectomy	Ileum	3.5cm
2013	Jacobs *et al* [45]	41	F	3m	CT	Right hemi-colectom	Ileum	2.5cm
2012	Akbulut *et al* [18]	38	F	10d	US	S.Resection	Ileum	4cm
2012	Lasithiotakis K, et al [46]	58	M	8hrs post colonoscopy	CT	Right hemicolectomy	Ileum	-
2012	Rabbani et al [47]	39	F	3m	CT	S.Resection	Terminal ileum	2 cm
2011	Morales-Fu *et al* [48]	42	M	8 d	CT	S.Resection	Ileum	3
2011	Nonose *et al* [20]	56	F	45 d	CT	S.Resection	Ileum	4.5
2010	Toydemir *et al* [49]	54	M	2 mo	CT	W.Resection	Ileum	4
2009	Gara *et al* [50]	76	F	NS	CT	S.Resection	Ileum	5.5
2009	Akbulut *et al* [18]	73	F	5 d	US	S.Resection	Ileum	11
2009	Ruffolo *et al* [51]	44	F	3 d	CT	Laproscopic S.Resection,	Ileum	3.7
2008	Mohamud *et al* [52]	70	M	6 yr	CT	Laproscopic R.Hemicol,	Ileum	3

2008	O'Kane *et al* [53]	65	F	1 mo	CT	S.Resection	Ileum	3.5
2008	Deschamps *et al* [54]	22	M	NS	CT	S.Resection	Ileum	3
2008	Coulier *et al* [55]	58	M	Several weeks	CT	Ileocecal resec.	Ileum	6
2008	Costamagna *et al* [56]	62	M	3 wk	NS	S.Resection	Ileum	4
2007	El Hajj *et al* [57]	52	F	10 d	CT	Laproscopic S.Resection,	Jejenum	3.5
2006	Karamercan *et al* [58]	56	M	1 d	US, CT	S.Resection	Ileum	4
2005	Parasi *et al* [59]	35	F	NA	NA	S.Resection	Ileum	4
2005	Jabar *et al* [60]	34	M	2 d	US	S.Resection	Ileo-jejunal	3
2004	Bays *et al* [61]	54	F	8 mo	US, CT, Ecl	S.Resection	Ileum	3.5
2004	Vijayaragh *et al* [62]	20	F	3 wk	DL	S.Resection	Jejenum	6
2004	Miyata *et al* [63]	64	F	6 wk	US, CT, Enteroscopy	S.Resection	Jejenum	4.5
2004	Gonul *et al* [64]	48	M	1 d	Urgent	S.Resection	Ileum	5
2003	Topaloglu *et al* [65]	56	M	10 d	US, CT	S.Resection	Jejenum	5
2004	Martin-Lor *et al* [21]	28	F	NS	Urgent	R.Hemicolectomy	Ileum	NS
2003	Savargaon *et al* [66]	52	F	7 mo	CT	S.Resection	Ileum	3
2002	Sah *et al* [67]	45	M	2 wk	NA	NA	Jejenum	NA
2000	Balci *et al* [68]	71	F	NS	MRI	S.Resection	Ileum	1.5

5. Discussion

Adult intussusception is an infrequent finding and a rare cause of bowel obstruction accounting for only 5% of bowel obstructions [8]. Intussusception, itself being pathological is associated with a lead point. It could be primary or idiopathic without any lead point. Idiopathic intussusception is common in children. In adults, 20% of intussusception is idiopathic [2]. Intussusception is also classified based on the location of its occurrence as intussusception within small bowel [entero-enteric], terminal ileum telescoping into colon [ileocolic], ileocecal valve as lead point telescoping into colon [ileocecal] and in large bowel [colo-colic]. In a large case series described by Brayton et al of 745 adult patients operated for intussusception, small bowel was the most common site, 52% small intestinal origin (95 patients ileocolic + 294 entero-enteric), 38% colonic origin (124 ileo-caecal + 31 appendico-caecal + 130 colo-colic), 4% stomal (in 30 patients), 6% gastric and duodenal (in 45 patients) [9].

Intussusception can be also classified based on its cause as benign, malignant, or idiopathic (primary). In adults, the precise mechanism of bowel intussusception is ill-defined (primary or idiopathic) in 8%-20% of cases and in such, is more likely to be entero-enteric [8,10]. Patho-physiological mechanism of secondary intussusception is understood to be initiated from irritant within the lumen that changes normal peristaltic activity leading to intussusception or any structural lesion in the bowel wall that acts as a lead point, leading to telescoping of one segment of the bowel into another along with the mesentery of the proximal segment, causing bowel obstruction and compromised blood supply to the proximal segment, which initiates inflammatory bowel changes causing thickening of the bowel wall to ischemic necrosis [11]. 70% of lead points in the small bowel are benign lesions that are mostly intraluminal like adenomatous polyp, submucosal lipomas, leiomyomas, hamartomas, diverticulum, rarely inflammatory lesions like inflammatory fibroid polyp [12,13]. Benign etiology, without lead points can also lead to transient intussusception in conditions like celiac sprue and Crohn's disease [13]. Malignant lead points could be lymphomas, Adenocarcinomas(most common in colon), metastases (most common in small bowel] [14]. In large bowel the lead point is often malignant (66%) [3].

Inflammatory fibroid polyps (IFPs) are uncommon benign lesions. It was first described in stomach as gastric submucosal granuloma with eosinophilic infiltrate by Vanek in 1949 [15]. Known by terminologies like Vanek's tumor, polypoid myo-endothelioma, eosinophilic granuloma, fibroma with eosinophilic infiltration [15], it involves both sexes equally and the peak age of incidence is 5th to 7th decade of life [16]. Many theories have been proposed to explain the inflammatory growth, the most accepted one being the benign reactive growth in response to an unidentified irritant [16]. Suspected irritants that need yet to be proved are H. pylori, parasite, and foreign body [16]. The latest theory includes mutations in PDGFRA as reports of familial occurrence have also been reported [17]. The commonest site of IFPs is gastric antrum (60-70%), followed by small bowel (18-20%) and colo-rectum (4-7%), and least common (1%) is oesophagus, duodenum, gallbladder, and appendix [18]. Meta-chronicity in IFPS is rare and is seen in familial cases [17].Gross Size of IFPs at the time of diagnosis usually is 3 - 4cm, with a size ranging from 0.4 - 30cm [7].

Depending on the site of Inflammatory fibroid polyps, the clinical presentation varies. The majority of IFPs are incidental findings detected endoscopically and during laparotomy [19]. They are asymptomatic and rarely present with anaemia, chronic pain abdomen and intussusception [20]. Intussusception and obstruction are the most common presentation of IFPs of small bowel origin [9]. The Intussusception triad of palpable abdominal mass with pain abdomen and bloody diarrhoea is rare in adults but more common in children as an acute presentation. Adult intussusceptions are usually chronic with signs and symptoms of partial to complete bowel obstruction with or without GI bleeding [21]. IFPs are benign but can present with a lower GI bleed. Malignant lead points are more likely to be hemoccult-positive stools.

Since the patient presents with features of intestinal obstruction the initial diagnostic modality is plain

abdominal X-ray and USG whole abdomen. Plain X-ray features suggest intestinal obstruction and the probable site of obstruction [69]. Contrast studies demonstrate 'coil spring' appearance in upper GI study and 'cup-shaped' filling defects in patients with colic intussusceptions on barium enema examinations [70,71]. Ultrasonography is a useful imaging tool in diagnosing intussusception in both adults and children [72] especially in emergency. USG features in intussusception are doughnut or target sign in transverse view and pseudo-kidney sign in longitudinal view [73]. The most sensitive imaging modality to diagnose intussusception however, is CECT with diagnostic accuracy as high as 58% to 100% [74]. Imaging signs include target sign, mesenteric vessels within the bowel lumen [74]. Nevertheless, the visibility of mass within intussusception is poor [75].

The management of IFPs is based on its location and presentation. Asymptomatic IFPs located in the stomach, duodenum, and colon can even be resected endoscopically [76]. If IFPs are complicated with intussusception it warrants surgical intervention to avoid the fear of bowel ischemia and necrosis. If intraoperative features are suggestive of a benign lesion, reduction and limited resection of small bowel intussusception is advised instead of en-bloc resection of the intussuscepted segment of bowel as in colonic intussusception [77]. Since colonic intussusception is more commonly associated with malignant lead points, which on bowel handling leads to perforation and seeding of tumor cells into the abdominal cavity, hence a wide resection is preferred [77].

Histologically, an inflammatory fibroid polyp has a characteristic localized proliferation of mononuclear spindle-shaped cells with an inflammatory infiltrate [16]. IFPs arise from submucosal layer and extend outside to muscularis propria and inner mucosa [7]. IFPs are very well demarcated from surrounding normal tissue and are unencapsulated tumours [7]. They predominantly have eosinophils infiltrate and more fibrosis when compared to IMFTs. Spindle shaped cells are loosely dispersed in a fibro-myxoid stroma with a prominent regular vascular network [4]. Whereas IMFTs occurs mostly in young age, and have a tendency to infiltrate, more cellular with a fascicular architecture and a poor vascular network [4]. Both appear to be benign processes.

Immunohistochemically, IFPs show inconsistent positivity for CD34 and negativity for CD117 and S100 protein [16], whereas IMFTs are always CD34 negative. Positive CD34 suggests primitive perivascular or vascular cell origin [20]. IFPs are differentiated from GIST as IFPs is CD117 negative and they have no risk of recurrence or metastasis after removal. Other differentials of IFPs include plexiform fibro-myxoma and smooth muscle tumours. Plexiform fibro-myxoma contains spindle cells in fibro-myxoid stroma but lack the characteristic eosinophilic inflammatory infiltrate and are CD34 negative [7]. Smooth muscle tumours exhibit desmin, SMA and caldesmon, and are CD34 negative [7]. Most of gastric and small bowel IFPs have expression of PDGFRA (100% of gastric IFP and 95% of small bowel IFP) [5]. Immunohistochemical staining for PDGFRA may be useful to support a diagnosis of inflammatory fibroid polyp.

6. Conclusion

In conclusion, IFPs are rare benign mesenchymal tumours of the GI tract with resemblance to inflammatory myo-fibroblastic tumours IMFTs. Immunohistochemical staining may be useful to support a diagnosis of inflammatory fibroid polyps in case of diagnostic dilemma. Being a benign tumor management guidelines have not been standardized. In case of small bowel IFPs where diagnosis is not possible with preoperative biopsy, or when the presentation is in emergency, management is always limited resection and anastomosis. In other sites if preoperative diagnosis of IFPs is made and growth is small and asymptomatic, management of vigilant wait vs early resection is yet to be studied. IFPs are known to cause lower GI bleed and further complicate with intussusception and obstruction. Therefore, early resection is a better option than vigilant wait. No reports of recurrence of IFPs have been documented till date. Hence, no follow-up protocol guidelines have been established.

Declarations

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Ethical approval: Not required.

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