

Review Article

# Idiopathic Chronic Intussusception in an 11th Months Old boy. Is The Freely Caecum One of Risk Factor?

N Leonidas<sup>1\*</sup>, M Omid<sup>2</sup>, M Helle<sup>2</sup>, E Ravel<sup>1</sup>, S Mansour<sup>1</sup>, Y Issoufou<sup>1</sup>, A Diori<sup>1</sup> and A habibou<sup>2</sup>.

<sup>1</sup>National Hospital Amirou Boubabar Diallo of Niamey, France

<sup>2</sup>Abdou moumouni University of Niamey, France.

**Corresponding Author:** N. Léonidas, National Hospital Amirou Boubabar Diallo of Niamey, France.

Received: 📅 2023 Oct 20

Accepted: 📅 2023 Nov 17

Published: 📅 2023 Nov 30

## Abstract

Chronic intussusception is rare in childhood, symptoms are variable and persist more than 2 weeks. The classical triad of abdominal pain, vomiting, and bloody stool is absent. Here we present an 11-month-old boy with recurrent abdominal pain, vomiting and weight loss, with an evolution of more than 2 months. Ultrasound confirm an intussusception. A laparotomy was performed, which showed an irreducible intussusception by Hutchinson reduction maneuver. Due to laborious tight fibrous adhesions. Strictures resection was performed to complete reduction. There was no intestinal perforation. The final diagnosis was an ileocolic chronic intussusception due probably to the freely mobile ileocecal junction. The patient had an uneventfully recovery and gained 3 kg three months post operatively. In the literature we found that malignancy is the main cause of chronic intussusception in children as proven in adults.

**Keywords:** Intermittent abdominal pain, Chronic intussusception, Freely cecum, Childhood, Surgery, Gastric disorders.

## Abbreviation

CI: Chronic intussusception.

## 1. Introduction

Chronic intussusception (CI) is a rare condition defined as intussusception persisting for over 14 days. However, CI is an uncommon disease that is more accompanied by leading points, such as polyps or neoplasms, and typically requires surgical treatment. In adults, it is frequently associated with leading points like neoplasm and other pathologies. CI is characterized by being non-strangulated and incompletely obstructive. Therefore, both the symptoms and underlying causes of 'chronic' intussusception can differ from those of 'acute' cases. The triad of symptoms seen in acute intussusception: colicky intermittent abdominal pain, vomiting, and bloody stool is rare in CI. In this report, we present the case of an 11-month-old boy who developed idiopathic intussusception over a span of 2 months. We also provide a concise review of the literature [1-5].

### 1.1 Case Presentation

An 11-month-old boy was admitted to the Pediatric Surgery Department of Amirou Boubacar Diallo National Hospital of Niamey. He presented with a three-week history of anorexia, intermittent central abdominal pain and weight loss of 2kg was reported. Physical examination revealed a moderately malnourished boy, conjunctival pallor and fever. His abdomen was soft and not distended no abnormalities or organomegaly. Rectal examination showed yellow stools without

blood. Abdominal ultrasound concluded to acute intussusception although there was discrepancy between clinical and imaging findings. Hemogram results showed elevated white blood cell count ( $10.2 \times 10^9/\mu\text{l}$ ) and hypochromic and microcytic anemia ( $\text{Hb}=8,3\text{g/dl}$ ). Microscopic paludism blood test was positive at 80 Plasmodium falciparum parasites  $/\mu\text{l}$ , while electrolytes, and renal function tests were normal. The patient received blood infusion, treatment for malaria and empirical antibiotherapy and was discharged after a week. However, he was readmitted over the next three weeks for further investigations. Clinical examination still revealed persistent abdominal pain, occasional vomiting and ongoing weight loss. There were no signs of blood, but mucus presence in the stools. Physical examination revealed a soft and mildly distended abdomen, emptiness of the right iliac fossa, which was easily depressible, no palpable mass or organomegaly. Hemogram results indicated leucocytosis ( $25.1 \times 10^9/\mu\text{l}$  with neutrophile predominance), anemia ( $\text{Hb}=9\text{g/dl}$  (microcytaire hypochrom and microcytosis), thrombocytosis ( $1166 \times 10^9/\mu\text{l}$ ); hypokalemia ( $1.87 \text{ mmol/l}$ ) and hyponatremia ( $131 \text{ mmol/l}$ ). Urea was measured at  $0.19 \text{ mmol/l}$  and creatinine at  $9.61 \mu\text{mol/l}$ . Urine culture were sterile, while stools culture were positive to Escherichia Coli. Abdominal X-ray revealed poor digestive aeration, and abdominal asymmetry with the right fossa iliac projection deformity (Figure 1). The abdominal ultrasound performed revealed a rosette-shaped image, indicative of acute intussusception (Figure 2).



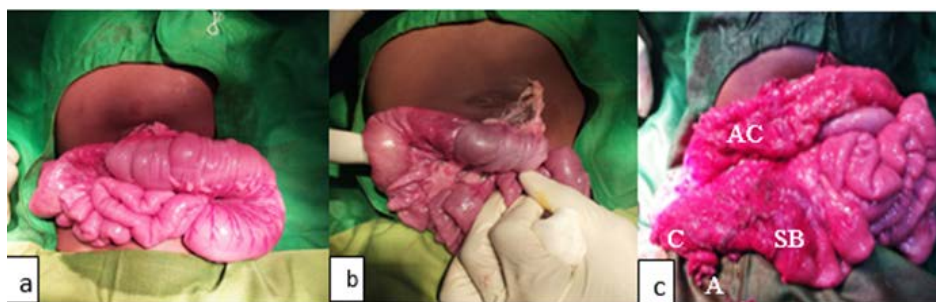
**Figure 1:** An abdominal x-ray highlighting a poor distribution of digestive aeration, opacity of the left flank and the peri-umbilical region and abdominal asymmetry with the right iliac fossa which seems uninhabited.



**Figure 2:** cross-sectional ultrasound image showing a sandwich-like tubular mass (intussusception).

Exploratory laparotomy was performed through a right flank quadrant transverse incision, revealing an ileocolic intussusception that extended up to the transverse colon. An invagination resembling a sausage shape (Figure 3.a) was located in the left hypochondrium, accompanied by multiple mesenteric lymph nodes. The presence of multiple adhesions, including grelo-grelic, grelo-colic, and ileo-ileal adhesions, was observed. The process of manual partial desinvagination by Hutchinson reduction maneuver was intricate and required considerable effort (Figure 3b). This was followed

by a meticulous adhesiolysis procedure involving the removal of fibrous tissue between the last 10 cm of invaginated ileal loops and the cecum, along with 5 cm of the invaginating ascending colon. The loops exhibited a healthy pink coloration and a thick, deperitonized intestinal wall (Figure 3c). Grossly, there were no necrosis or perforation. The appendix appeared normal and was not removed. Both the ileocecal junction and the ascending colon were found to be mobile without restriction (Figure 3.a and 3.b).



**Figure 3:** The sausage image (a) the intussusception could not be reduced even with the Hutchinson maneuver (b). After fibrous dissection/resection, the small bowel (SB) appendix (A) caecum (C) and ascending colon (AC) were deprotonated.

The child experienced an uneventful recovery. He was discharged on the seventh day after the surgery, with normal bowel movements. Three months following the surgery, he remained in good health without any recurring abdominal symptoms and had gained weight. The patient weighed 8 kg three weeks after the symptom's onset and 11 kg three months after the surgery.

#### 4. Discussion

More than 90% of the intussusception cases in children are idiopathic. It is most common in infants and children aged 3 months to 3 years. The incidence of chronic intussusception (CI) is about 3% of all cases of intussusception in children aged under one year. Although the incidence of chronicity is greater in older children (10 % of children over one year). Underlying pathological causes of intussusception can be identified in 1.5-12.0% of cases. The case described here involves an 11-month-old with CI. The classical triad of colicky abdominal pain, vomiting, and passage of blood through the rectum are mostly absent in CI. Sometimes, CI present with episodes of non-specific abdominal pain of prolonged duration lasting over 14 days (Table1). As reported in our case, because of non-specific symptoms of intussusception, CI may be misdiagnosed primarily. Recurrent abdominal pain is occasionally considered a functional disorder in the absence

of alarming symptoms such as significant vomiting, fever, diarrhea, and weight loss or delayed growth. A conspicuous feature of CI is a loss of weight attributed to protracted episodes of vomiting. In our study, patient had a 2-month history of abdominal pain, constipation with transient vomiting, weight loss; he had also fever due to malaria. An unusually high incidence of chronic and sub-acute intussusception was described in a study of 62 children from Nigeria. A high incidence of painless intussusception (41%) was reported. Although intussusception signs were not clear, ultrasound imaging likely contributed to clinical symptoms. As CI is non-strangulated, it may reduce intermittently or advance to strangulation [1-12].

CI is frequently associated with a high rate of unsuccessful hydrostatic reduction, as well as a high incidence of organic lesions as a leading point [8-9]. After the failure of air reduction, laparoscopic exploration of the abdomen and reduction of the intussuscepted bowel may attempt, all instances of chronic intussusception should be treated surgically. Laparoscopic reduction maybe challenges due to strictures and need laparotomy conversion to complete surgery. In the present case, there was clinico-imaging discordance laparotomy was performed for the CI [2-13].

**Table 1: Y= Year, M=Month, W=Week. The literature on chronic intussusception (CI) in children.**

Studies	No of case	Age	Sex	Duration	Abdominal	Vomiting	Weight. loss	Blood in	Mass	Type /diagnostic	Etiology	Surgery	Recovery uneventful
Zavras Al.2016(14)	1	4.5 Y	M	6W	+	+	+	-	+	Ileo colic	Mal rotation	Manual	+
Saka Al. 2015(4)	1	14	M	5W	+	+	+	-	+	Ileo colic	B cell lymphoma	Resection-anastomosis	+
Sutherland Al. 1932(10)	1	3Y	M	2M	+	-	-	-	-	Ileo colic	Mobile caecum	Manual	+
Ramesh Al.2021(9)	1	9Y	F	1M	+	+	+	-	+	Ileo colic	Duplication cyst	R hemi colectomy	+
Choi Al. 2016(2)	1	6Y	F	2M	+	+	-	+	-	Ileo colic	B cell lymphoma	Ileocolic resection	+
Page Al.1990(11)	1	16M	M	9W	+	+	+	-	+	Ileo colic	Peyers hyperplasia	Ileocolic resection	+
Sadd Al. 2018(5)	1	3Y	M	6W	+	+	-	-	-	Ileocolic	Burkitt's lymphoma	Ileocolic resection	+
Present case 2023	1	11M	M	2M	+	+	+	+	-	Ileocolic	Mobile caecum	Manual +strictures resection	+

Goyal and Al. 2022 funds coeliac disease as cause of ileo ileal CI, the child was managed conservatively and there was gradual resolution of obstruction. For Jawad and Al. Saad and Al. and Saka and Al. and others, lymphomas were the etiology (Table 1). patients got resection and end to end anastomosis. Ramesh and Al. the child had an enteric duplication cyst as the leading point of CI. Literature in adult confirm that malignancy accounts for nearly 65% of CI of all cases malignancy remains the leader etiology of CI in children (Table 1). Zavras and Al present that malrotation was responsible of CI. The present case, it was no possibility to complete manual reduction due to the strictures but reduction had been completed by strictures resection and there was no intestinal perforation the freely mobile ileocecal junction was incriminated as a principal factor of chronic intussusception. In literature (Table 1). the presentation form of ileocolic intussusception is predominant, the etiology of idiopathic CI is often linked to factors like malrotation, freely mobile cecum, hyperplasia of Peyer's patches, duplication cyst and lymphoma. Surgical resection is commonly performed due to the presence of intestinal fibrous adhesions. All of patients presenting CI had an uneventful post-operative recovery [4-15].

## 5. Conclusion

Idiopathic chronic intussusception is rare in childhood, it should be the causes of weight loss in young children. prolonged vague abdominal symptoms and vomiting. Clinical examination contrasts with imaging results. Surgery remains the primary treatment. We found the freely mobile cecum as factor of chronic intussusception. Similar to findings in adults the literature underscores that malignancy is a leading etiological factor in chronic intussusception in children.

## Author's Contributions

NL designed, drafted the study and wrote the manuscript. MO and MH helper for design and draft of study and revised the manuscript. ER and SM collected data. YI AD and AH designed critically revised the manuscript. The authors received no specific funding for this study.

## Availability of Data and Materials

All data generated or analyzed during this study are included in this published article.

## Declarations

Consent: Written informed consent was obtained for publication of this case report and accompanying images.

## Competing Interests

The authors declare that they have no competing interests.

## References

1. Rees, B. I., & Lari, J. (1976). Chronic intussusception in children. *British Journal of Surgery*, 63(1), 33-35.
2. Choi, S. H., Han, S. A., & Won, K. Y. (2016). Chronic Intussusception caused by diffuse large B-Cell Lymphoma in a 6-year-old girl presenting with abdominal pain and constipation for 2 Months. *Journal of Korean medical science*, 31(2), 321-325.
3. Ali, A., Lee, Y. J., Gosse, M., Meade, C., Labath, H., & Murali, A. R. (2023). Ileocolic intussusception presenting as chronic diarrhea in an elderly woman. *VideoGIE*, 8(2), 89-91.
4. Saka, R., Sasaki, T., Matsuda, I., Nose, S., Onishi, M., Fujino, T., ... & Oue, T. (2015). Chronic ileocolic intussusception due to transmural infiltration of diffuse large B cell lymphoma in a 14-year-old boy: a case report. *Springerplus*, 4(1), 1-5.
5. Saad, A. A. M., Kkalid, T., Abbas, M., & Salih, K. M. A. (2018). Rare presentation of chronic ileocecal intussusception secondary to Burkitt's lymphoma in three years Sudanese boy: a case report and literature review. *Pan African Medical Journal*, 31(1).
6. Li, Y., Zhou, Q., Liu, C., Sun, C., Sun, H., Li, X., & Zhang, L. (2023). Epidemiology, clinical characteristics, and treatment of children with acute intussusception: a case series. *BMC pediatrics*, 23(1), 143.
7. Macaulay, D., & Moore, T. (1955). Subacute and chronic intussusception in infants and children. *Archives of Disease in Childhood*, 30(150), 180.
8. Jawad, A. J., Shibli, S. Y., Sahni, P. S., & Malabarey, T. (1997). Chronic intussusception. *Annals of Saudi medicine*, 17(5), 545-547.
9. Ramesh, R., Gurugopinath, S., & Muhaidat, S. (2021). Outage performance of relay-assisted single-and dual-stage NOMA over power line communications. *IEEE Access*, 9, 86358-86368.
10. SUTHERLAND DM: Chronic intussusception in children. *Arch Dis Child*: first published as 10.1136/adc.7.40.191 on 1 August 1932.
11. Page AC, Price JF, Salisbury JR, Howard ER, Karani J: Chronic intussusception. *Arch Dis Child*: first published as 10.1136/adc.65.1.134 on 1 January 1990.
12. Momoh JT and Lawrie JH: Tropical paediatric intussusception. Is it a different disease entity? *Annals of Tropical Paediatrics*. déc 1981;1(4):237-40.
13. Léonidas, N., Aurore, H., Marret, J. B., & Julien, R. (2022). Acute intussusception of Meckel's Diverticulum in a 15-year-old boy: Clinical, imaging and laparoscopy aspects.
14. Zavras N, Tsilikas K, Vaos G: Case Report Chronic Intussusception Associated with Malrotation in a Child: A Variation of Waugh's Syndrome? *Case Reports in Surgery*. Volume 2016, 5638451, 5 pages. Doi. org/10.1155/2016/5638451.
15. Goyal, P., Nohria, S., Grewal, C. S., Sehgal, R., & Goyal, O. (2022). Celiac disease and intussusception-a rare but important association. *Acta gastro-entomologica Belgica*, 85(1).