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Congenital colonic stenosis after the neonatal period: A case report and review of the literature with criteria for safe primary anastomosis

Naser El-Mefleh[1], Muhammad Eyad Ba'Ath[2]

[1]Department. of Pediatric Surgery, National Hospital, Aleppo, Syria.

[1]Department of Pediatric Surgery, Bab Alhawa Relief Hospital, Edleb, Syria

[1]Department of Pediatric Surgery, Muhammad Wassim Maaz Hospital, Aleppo, Syria

[2]Danat Al Emarat Hospital, Dubai, United Arab Emirates.

[*]**Corresponding Author:**

NECMETTİN MAH., BEDIR SOK. NO. 33, 79000 KILIS MERKEZ / KILIS

TURKEY

Email ID: dr.naser1989@hotmail.com

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ABSTRACT

we present a congenital colonic stenosis (CCS) in a 9-month-old male. CCS have only been reported 8 times older than 6 months of age and 18 reports after the neonatal period in the English-language literature. Our infant had recurrent episodes of intestinal obstruction and failure to thrive and laparotomy shows colonic stenosis. Investigations are the abdominal X-ray, contrast enema, colonoscopy, and rectal biopsy to differentiate the CCS. The differential diagnosis (D.D) includes distal intestinal obstruction causes and its definitive treatment is surgical.

Keywords: *Colon, Differential, Intestinal Obstruction, Infant, stenosis*

1. BACKGROUND

CCS has been reported in 18 cases since 1966 after neonatal period and only 8 presented more than 6 months of age. D.D is any distal intestinal obstruction, such as Hirschsprung disease or acquired colonic stenosis (ACS)[1]. Although the exact cause is unknown, vascular insufficiency has been accepted[2]. The main investigations are the abdominal X-ray, contrast enema, colonoscopy, and rectal biopsy[3]. Associated abnormalities have an effect on the prognosis[3]. The definitive treatment is single- or multi-stage surgery[3].

2. CASE REPORT

A 9-month-old full-term male with a birth weight of 3 kg had recurrent episodes of abdominal distention and thrive failure. Physical examination revealed dehydration and severe abdominal distention, with a weight of 6 kg. There is no further data available on the histopathology of the stenosis area or the etiology of ACS, such as CMV. Contrast enema (Figure 1, a) revealed a severely dilated colon extending to the proximal sigmoid, followed by a stenosis involving the sigmoid and rectum with an acute caliber change at the recto-sigmoid junction. He was resuscitated and placed on colonic washouts but failed to respond. Laparotomy findings (operative appearance of acute calibre) (figure 1, b) was CCS and biopsy excluded Hirschsprung disease. A double barrel colostomy was preferred to us rather than an ileostomy (high morbidity), or primary anastomosis (the significant calibre of 9:1) (Figure 1, c). The child did well and was discharged on the 4th day. There was weight gain and the dilated colostomy regression on follow-up (Figure 2, a). Colostomy closure was done after 9 months. The discrepancy in diameters was 4:1, and side-to-end anastomosis was done with no complications. A subsequent contrast enema (Figure 2, b) confirmed a smooth barium passage. Follow-up continues for 2 years, and the boy is well.

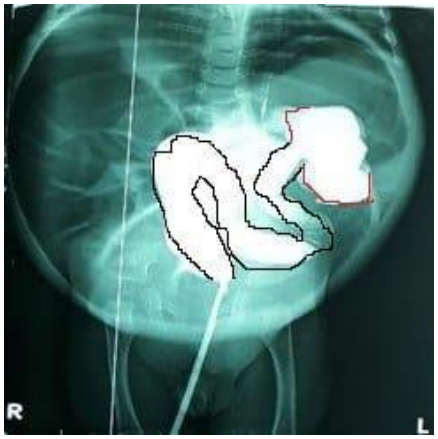

Figure 1, a

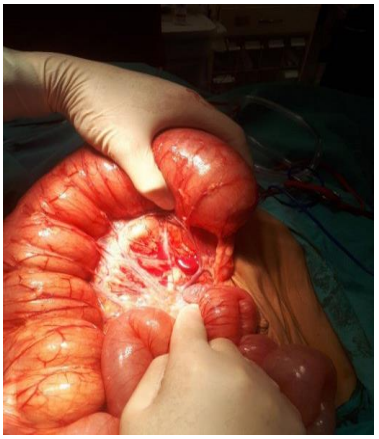

Figure 1, b

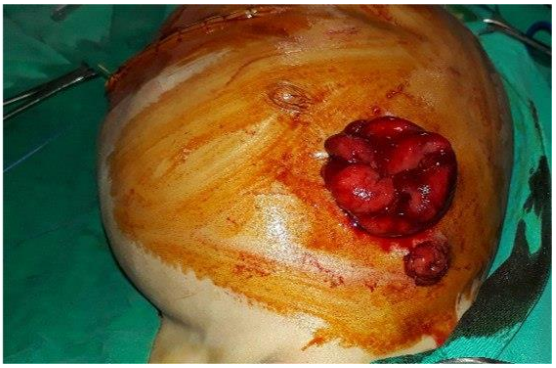

Figure 1, c

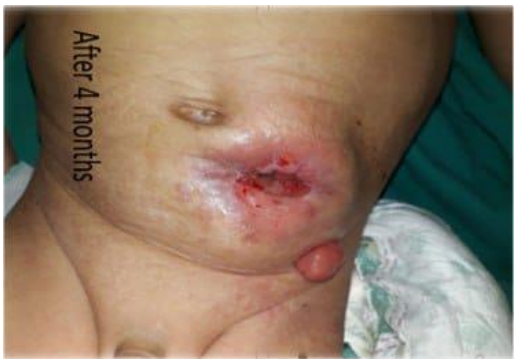

Figure 2, a

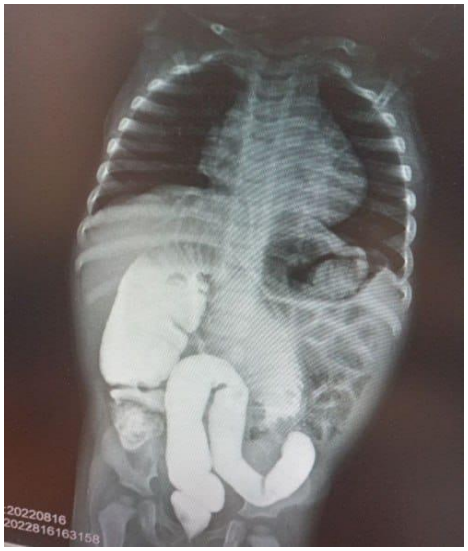

Figure 2, b

Fig. 1: Acute caliber change: a severely dilated colon extending to the proximal sigmoid, followed by a stenosis involving the sigmoid and rectum. A: pre-operative by contrast enema. B: Intraoperative. C: post-operative colostomy

Fig. 2: A: Appearance of colostomy after months from surgery with regression of the discrepancy of proximal colon and distal colon. B: Contrast enema after colostomy closure and a smooth barium passage and without abnormality.

3. DISCUSSION

Colonic atresia is a rare cause of distal intestinal obstruction, accounting for 1.8 to 15% of all intestinal atresia, and most tertiary-level centers in the world see an average of 1 to 2 cases per year[4]. CCS is even rarer, accounting for less than 1%[5]. The most accepted theory is that a vascular accident may involve intrinsic or extrinsic factors, and depending on its severity, stenosis or atresia may occur[6]. Preoperative diagnosis of CCS is difficult, and there are no management protocols. Any early case of vomiting, abdominal distention, or passing meconium failure is suspected. The later presenting case will have similar features in addition to constipation and failure of thrive[6]. Differential diagnoses include Hirschsprung disease, ACS, meconium plug syndrome, and small left colon syndrome[6]. ACS is more common than CCS, secondary to NEC[6], CMV, and norovirus[7]. An unusual presentation of recurrent episodes of melena substantiated by a drop in hemoglobin, constipation, and growth failure is described by Bharti et al[8]. Lengths of stenosis vary from 1–16.5 cm[9]. An abdominal X-ray is an initial investigation. If the child is stable, a detailed evaluation can be done with a contrast enema (a key to the diagnosis), a rectal biopsy, and a colonoscopy[3]. Contrast enema findings are usually a small distal colon followed by an abrupt distended segment proximally[3]. There are many classifications, such as Louws and Grosfeld classifications[3] and according to another classification, there are 5 anatomical types of colonic atresia; the rarest is V (CCS)[10].

Table 1: Review of the literature of CCS after neonatal period.

CCS site	ascending	transverse	descending	sigmoid	descending-sigmoid junction	recto-sigmoid junction	multiple
Number.	6	2	1	3	1	1	4
%	33	11	5	16	5	5	22

13 of 18 cases with known sex (9 are male: 70%). Associated disorders are not common, unlike colonic atresia. Perinatal period was uneventful in all and no one has a history of delayed meconium. the Literature has estimated that coexistent Hirschsprung disease is present in 2% of cases, but it should be excluded in every case[20]. The stenosis site has been documented as Table 2:

The study	**Child age /sex**	**CCS site**	**CCS length**	**Calibre**	**Procedure**
Gupta [9] 2021	2 months /M	Ascending	1.2cm	>3-1	resection and ileo-colostomy then closed after 12 w
	4 months /F	Transverse	3 cm	<3-1	resection and primary anastomosis
	7 months /M	Multiple : Transverse	5.5cm	>3-1	resection and double barrel colostomy then closed after 10 w
	11 months /M	Descending-sigmoid junction	3 cm	>3-1	resection and double barrel stoma then closed after 10 w
	24 months /F	ascending	7 cm	>3-1	proximal ileostomy then resection with ileocolic anastomosis after 8 m
Ekenze [11] 2019	11 weeks /M/ preterm 35	Ascending	5cm		resection with ileo-transverse anastomosis
	9 weeks /M/ preterm 34	Multiple: cecum, ascending, transverse, descending			resection with ileo-sigmoid anastomosis
Khanna [12] 2018	7 months /F/ preterm 32	Ascending	2cm	significantly	Poor condition: double barrel ileo-colostomy, then closure after 9 m
Bharti[8] 2017	5 months /M	Sigmoid			Poor condition and failure of balloon dilation: primary anastomosis with a protective loop ileostomy

Zambait [1] 2016	2 months /M/ preterm 35	Multiple: ascending, transverse with appendix atresia		significantly	Resection, proximal cecostomy, distal ascending colostomy in order to preserve colonic length stage 2 later: colocolostomy and enteroplasty.
Saha[13] 2013	1.5 years	Descending	2cm		resection and primary anastomosis
Lim IIP[14] 2013	6 months /M	Multiple: Mid-Transverse/ descending		significantly	resection and coloplasty/ double barrel colostomy.
Mirza B[3] 2012	8 months	Sigmoid			resection and primary anastomosis
Galván-Montañ[15] 2010	3 years	Ascending			resection and primary anastomosis with removing foreign bodies.
Ruggeri[16] 2009	4 months	Ascending			
Garcia-Vázque[17] 2002	2 months /M	Sigmoid			video-assisted resection and primary anastomosis
Takahas[18] 1998	3 years	Transverse			resection and primary anastomosis
Pai G.K. 1990[19]	4 months /F	Recto-sigmoid Junction			

16 of the total cases were mentioned surgical procedures, as follows:

1. Primary anastomosis: 8 (50 %)
2. Primary anastomosis with protective stoma after balloon failure: 1 (6%)
3. Colostomy or ileo-colostomy and later closure: 6 (37.5%)
4. Proximal ileostomy and later resection and primary anastomosis: 1 (6%)

Suhaimi, M.S., et al[21] in a review of the literature in 2021 refers to the fact that most of the authors favored staged surgery. But in our literature review, approximately half of surgeons tend to perform a primary anastomosis and the rest to perform staged surgery. So this confirms the management is challenging because of its rarity.

Patency of the distal colon segment should be tested by retrograde or antegrade instillation of air and saline prior to the surgery or intraoperatively[22]. The treatment should be individualized and depends upon multiple factors, like the site of the anomaly to the splenic flexure[16], general condition, frozen suction biopsy availability or Hirschsprung disease exclusion, associated anomalies, and intraoperative findings (calibration, Multiple stenosis: close together or far apart). Advantages of primary anastomosis: no stoma complications, single surgery, and single anesthesia, but a risk of leakage, sepsis, and death[22]. Single-stage primary anastomosis has been described in the literature by a few methods, like distal Cheatle slit, plication or tapering for the proximal side, or end-to-oblique anastomosis[6]. Pratama et al[23] described a modified Santulli procedure to combine the advantage of primary anastomosis with preventing anastomotic failure with ileostomy. Cox et al. suggested that primary anastomosis could be safely performed with a diameter variance of 3:1[24].

4. CONCLUSION

CCS will be in mind with any distal intestinal obstruction with positive ganglion nerve in rectal biopsy, and it is investigated by contrast enema and colonoscopy. There is no clear management protocol, but with limited capabilities, the safest surgical option is staged. Decreasing lumen diameter may happen through a time factor. Primary anastomosis is safe with this criteria: normal distal colon, positive ganglions of rectum, normal anal position, no associated anomalies, and a diameter variance of 3:1.

5. DECLARATIONS

- Ethics approval and consent to participate: was obtained from the local committee.
- Consent for publication: was obtained from the patient's father

- Availability of data and material: is available within the article.
- Competing interests: None
- Funding: none
- Authors' contributions: both contribute equally.
- Acknowledgements: None

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