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Clinical Vignette

Rapunzel Syndrome with a gastric ulcer

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CASE PRESENTATION

A 7-year-old girl presented with frequent episodes of abdominal pain and vomiting for the last 6 months. She had history of eating hair and threads for the last 4 years along with pulling out her own hair. Physical examination revealed pallor, and a palpable firm, non-tender mass in the epigastrium. Laboratory investigations were within normal ranges except for anemia (HB: 7.6g/dl).

Figure 1: Trichobezoar with long tail, Rapunzel syndrome.

Ultrasound abdomen suspected a gastric bezoar. CT scan (done outside) showed a large intra-gastric mixed density mass with internal areas of coarse calcification and air trapping causing massive luminal distortion of the stomach, suggestive of gastric bezoar. Psychiatric evaluation was unremarkable. At surgery, gastrotomy was performed and a huge gastric trichobezoar with 2-feet long tail (extended up to proximal jejunum) was removed (Fig.1). Before closure of gastrotomy, gastric mucosal inspection revealed a 2x3cm gastric mucosal ulcer on posterior gastric wall (Fig.2). Since it was not a bleeding ulcer, and outer gastric wall at the respective site of ulcer was not thin, therefore, it was left as such, and gastrotomy closed. Postoperative recovery was uneventful.

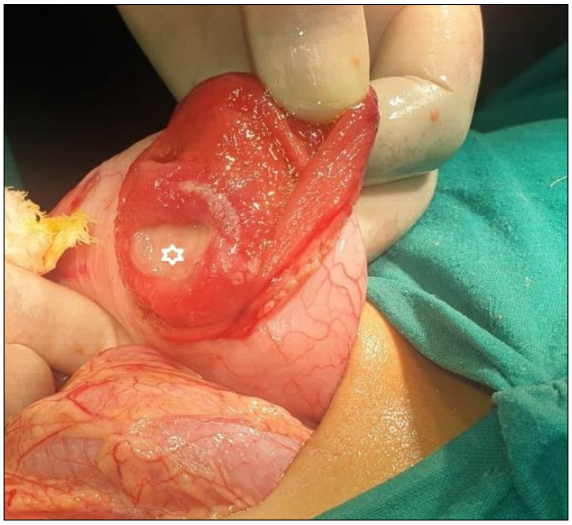

Figure 2: Mucosal ulcer is evident (Asterisk) on posterior gastric wall.

The patient was allowed orally on 5th postoperative day and discharged on oral antibiotic, multivitamin, analgesic and proton pump inhibitor. On first follow-up after a week, the patient was in a good clinical condition without any symptoms related to gastric ulcer. Psychiatric evaluation and counseling was again done in psychiatric department.

DISCUSSION

Rapunzel syndrome is trichobezoar with a long tail residing in gastrointestinal tract (GIT).[1] These bezoars may remain asymptomatic in GIT for a considerable period before developing any symptoms or complications.[2] The index case has had trichophagia for the last 4 years, but symptoms appeared 6 months before presenting to our hospital. It is quite possible that

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gastric ulcer developed 6 months earlier might have caused episodic abdominal pain in the index case. Sustained pressure of trichobezoar for a considerable period might result in local mucosal ischemia which could progress to mucosal ulceration.[2,3] Mucosal ulceration may produce various complications in addition to abdominal pain. Few cases of gastric trichobezoar with massive bleeding secondary to gastric ulcer or gastric perforation have been reported in literature.[2,4] A case of multiple small bowel perforations secondary to a trichobezoar has also been reported in literature.[5] Pressure induced ischemia at various points along the trichobezoar might explain these perforations.

Management of trichobezoar is its retrieval along with psychiatric evaluation and counseling to prevent recurrence. Regarding associated ulcer, if it is not bleeding, and serosa at respective site of mucosal ulcer is not paper thin, it can be left as such.[2] In case of bleeding ulcer or impending perforation, it has to be dealt surgically.[3] Similarly, in the index case, the ulcer was not bleeding and the serosal surface was not paper thin, therefore, we left it as such. Presumably, the ulcer would have healed after surgery as indicated by quite uneventful postoperative course. We have not applied any objective method to evaluate healing progress of ulcer such as endoscopy, because firstly the patient was asymptomatic and secondly due to covid-19 pandemic we were avoiding non-urgent procedures. The patient is doing fine on follow-up.

Conflict of Interest: MBM and NT are members of editorial team.

Consent to Publication: Author(s) declared taking informed written consent for the publication of clinical photographs /material (if any used), from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient, however it cannot be guaranteed.

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REFERENCES

1. Chauhan NS, Kumar S, Bhoil R. Rapunzel Syndrome: Rare 'tale' of a broken 'tail.' Polish J Radiol. 2016; 81:166–9.
2. Mirza B, Talat N, Saleem M. Gastrointestinal trichobezoar: an experience with 17 cases. J Pediatr Surg [Internet]. 2020 May [cited 2020 Jun 15]; Available from: https://www.jpedsurg.org/article/S0022-3468(20)30305-5/pdf
3. Athanasiou A, Michalinos A, Moris D, Spartalis E, Dimitrokallis N, Kaminiotis V, et al. Rapunzel Syndrome: A Rare Presentation with Giant Gastric Ulcer. Case Rep Med. 2014; 2014.
4. Ahmad Z, Sharma A, Ahmed M, Vatti V. Trichobezoar causing gastric perforation: A case report. Iran J Med Sci. 2016; 41:67–70.
5. Bux I. Multiple small bowel perforations in a patient with Rapunzel Syndrome. J Surg Pakistan. 2015; 20:117–8.