Abstract: Duplicational anomalies of gastrointestinal tract have the reported incidence of 1 in 4500. Among them, rectal duplications seem the be the least common type of these anomalies with very few cases reported. We present two cases over the period od five years. Both children were presented with prolonged constipation, and one child had rectal bleeding. Both children were successfully treated by surgical excision.

Keywords: Duplication, children, rectum, constipation, surgery.

1. INTRODUCTION
Duplications of alimentary tract encompass a wide variety of mass lesions throughout the course of the gastrointestinal tract [1]. They can occur anywhere from mouth to the anus, and have a reported incidence of 1 in 4500 [2]. Duplications vary in size, can be either spherical or tubular, and may communicate with the gastrointestinal tract [3]. Rectum seems to be the least common site of the duplication anomalies [4,5], with fewer than 100 reported cases in the literature [6].

Constipation is one the most common complaints in childhood. Although most of the cases can be attributed to functional constipation, a small percentage of children have the organic cause of their symptoms. Mostly, retrorectal i.e. presacral mass may cause the problem. A very rare, but potentially serious cause of constipation may be rectal duplication; yet, the mass has to be differentiated from cystic sacrococcygeal teratoma and anterior meningoceles [7]. Delayed diagnosis increases the risk of complications, therefore, high index of suspicion is imperative in all the cases of constipation, unresponsive to conservative treatment.

2. CASES
We present two cases of rectal duplications for the period of five years (2011 – 2016). Both patients were boys (confirming the male predominance of the rectal duplication cysts), with prolonged constipation as the main symptom.

Case 1. A 4-year-old boy was presented to emergency department for constipation, that was treated with fiber supplements and laxatives over the last six months. Rectal examination revealed cystic mass adjacent to posterior rectal wall; no rectal bleeding was confirmed whatsoever.

Complete blood count and biochemical analyses showed no abnormalities. Initial imaging study included ultrasonographic examination that confirmed extensive, oval, homogenous and hypoechogenic mass (87x65x60 mm) behind rectum. Further magnetic resonance study (MRI) confirmed the presence of the cystic tumor (Figure 1). The patient was scheduled for the operative treatment after obtaining written consent from the parents.

We used posterior sagittal approach and revealed the cystic mass presacrally (Figure 2). The mass was completely excised, leaving the small part of the cystic wall in situ, just in the part that shared the wall with the posterior rectum. With proper mucosectomy and drainage placing, the intervention was finished. The postoperative course was uneventful; the drain was removed on the fourth operative day. Pathology result confirmed duplication cyst with columnar epithelium, mucosal muscularis layer and true muscularis, as seen in rectum.

Case 2. An 11-month-old male infant was admitted for further examination of the rectal bleeding. Patient history revealed chronic constipation over the period of last five months. Rectal examination showed cystic mass behind the rectum, that was the most probable cause of the obstruction.

Apart from significant anaemia (Er – 2.62, Hb – 8.2 g/l), all the other laboratory and biochemical analyses were within the reference values. Having performed plain abdominal radiography, the patient was scheduled for...
computed tomography scan (CT). Conclusion was well formed, unilocular cystic formation 33x33 mm in size in front of the urinary bladder, and extending into retrorectal space (Figure 3).
We used posterior sagittal approach for the exposition of the lesion; as no communication with the nearby structures was found, the cyst had been completely enucleated. Having reconstructed the parasagittal muscle complex as to preserve the normal sphincter function and continence, the operation was finished with excellent cosmetic result (Figure 4). The postoperative course was uneventful. Pathology result proved rectal mucosal lining within the cyst, as well as muscle coat of the wall.

Figure 1. MRI scan showing retrorectal mass

Figure 2. Operative finding of presacral cystic mass

Figure 3. CT scan of rectal duplication
3. DISCUSSION

Even though duplicational anomalies have been known for a long period of time, Ladd was the first to suggest the term duplication in 1937 [1]. They can be found anywhere along alimentary tract, causing variety of symptoms depending on their localisation. They vary in size, can be either tubular or spherical, and may communicate with the intestinal tract.

Although several theories have been proposed, the true etiology of the duplications remains obscure. Persistence of fetal gut diverticula, defects in fetal gut recanalisation, partial twinning and split notochord theory are some of many proposed [7]. All of them can be applied to some lesions, yet, no uniform theory has been published so far. Ladd's criteria for characterising the lesion as duplication are still in use. The lesion is to have well-developed coat of smooth muscle, inner mucosal membrane resembling any portion of the intestinal tract mucosa, and should have an intimate anatomic association with any part of the digestive tube.

Some rectal duplications remain asymptomatic throughout a person's life; others cause complications, primarily constipation, rectal bleeding and sepsis, but also more serious one as malignant transformation [6]. Our two patients were both presented with constipation (despite the dietary changes and laxative use), and the younger boy was presented with rectal bleeding.

Even though ultrasonography is widely in use as the initial imaging study and offers some information about the existence of the mass itself, further diagnostic imaging is imperative (MRI as the imaging modality of choice), in order to obtain exact dimensions, relation to other structures and MRI characteristics. As rectal duplications have bimodal presentation, and are mostly seen in perinatal period and during early childhood, clinical suspicion is possible. In both our cases, the patients were scheduled for operative treatment highly suspicious of duplicational anomalies.

Although benign, timely diagnosis is important, so to prevent complications. Malignancy is the most serious one, very rare in childhood, but observed in 7-18% of adult cases [6].

4. CONCLUSION

Every child with prolonged constipation, unresponsive to conservative treatment, is to be subjected to sonographic examination, in order to exclude the organic cause of the constipation. The widespread utilisation of ultrasonographic examination helps identifying the presence of abdominal and pelvic cystic and tubular lesions and yields for further diagnostic imaging modalities.

REFERENCES