

INTUSSUSCEPTION AT THIRTEEN YEARS OLD BOY WITHOUT ANY LEAD POINT

Case Report

ONÜÇ YAŞINDAKİ ERKEK ÇOCUKTA SÜRÜKLEYİCİ NOKTASIZ İNVAJİNASYON: OLGU SUNUMU

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ABSTRACT

Intestinal intussusception is the most common cause of bowel obstruction in children. The peak incidence varies from the fifth to the tenth month of life, with only 10% to 15% of cases occurring after 2 years of age. It is well established, that in older children intussusception is frequently caused by a pathological lead point. Fluoroscopic images during reduction tend to miss pathological lead points. Therefore, when an older child presents with a clinic of intussusception, surgery is often performed without a radiologically guided reduction. A thirteen years old boy reported who emergency surgery was performed to him without any radiologically reduction because of ileocecal invagination.

Keywords: *Intestinal intussusception; older children; lead point.*

ÖZET

İntestinal invajinasyon, çocuklardaki barsak obstrüksiyonlarının en sık nedenlerinden birisidir. En sık görüldüğü dönem beş ile onuncu aylarken, iki yaştan sonraki olgular sadece %10 ile %15 oranındadır. Büyük çocuklarda invajinasyona sıklıkla bir sürükleyici nokta neden olur. Floroskopik redüksiyon sırasında patolojik sürükleyici noktalar gözden kaçırılır. Bu nedenle invajinasyon kliniğiyle gelen büyük çocuklarda radyolojik redüksiyon yerine cerrahi sıklıkla tercih edilir. İleoçekal invajinasyon nedeniyle radyolojik redüksiyon uygulanmadan acil cerrahi uygulanan onüç yaşındaki erkek olgu sunulmuştur.

Anahtar kelimeler: *İntestinal invajinasyon; büyük çocuk; sürükleyici nokta.*

INTRODUCTION

Intestinal intussusception is the most common cause of bowel obstruction in children. It occurs with invagination of the proximal intestinal segment into the distal. The peak incidence varies from the fifth to the tenth month of life, with only 10% to 15% of cases occurring after 2 years of age.

Intussusception in older children occurs due to a pathological lead point in up to 20–25% of the cases^{1,2}. In the adult population intussusception is uncommon, and very rarely idiopathic. In about 50% of adult cases it caused by a malignant lead point^{3,4}. Therefore, when an older child presents with a clinic of intussusception, surgery is often performed without a radiologically guided reduction.

CASE REPORT

Thirteen years old boy was referred to our clinic with only recurrent abdominal pain for six hours. Tenderness and mass were determined at right lower quadrant of abdomen on physical examination of the patient. Leucocyte count was 15800/mm³, plain abdominal radiography was normal. Our initial diagnosis was perforated appendicitis, but intussusception was determined at ultrasonographic evaluation. Emergency surgery was performed and, ileocecal invagination was determined (**Figure 1**).

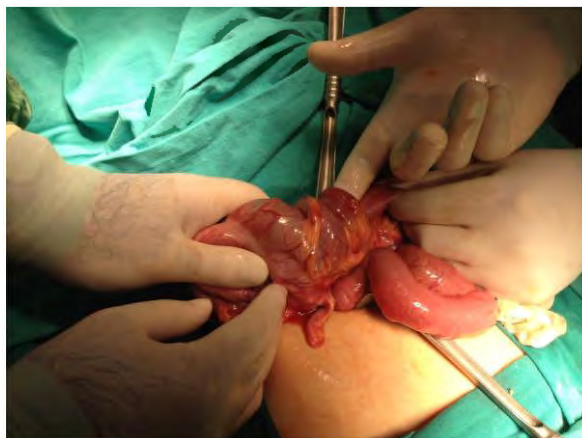


Figure 1: Ileocecal invagination was determined at surgery.

The invagination was reduced manually. The 40 cm length terminal ileum above cecum had invaginated. There were edema and hemorrhage at the ileal wall, but was not necrosis (**Figure 2**).



Figure 2: There were edema and hemorrhage at the invaginated terminal ileal segment wall, but was not necrosis.

The appendix had not invaginated and was normal. Also we excised the lymph nodes near the cecum and terminal ileum. The patient was fed orally at postoperatively third day, and was discharged at fifth day. The pathological examination of lymph nodes was normal.

DISCUSSION

The classic triad of intussusception symptoms are abdominal pain, vomiting, and discharge mucus with blood "strawberry jam" through the rectum. The presence of abdominal mass is important finding for diagnosis, with an incidence of 48%. The most common first symptom in young children is vomiting, unlike abdominal pain is the first symptom in older children. There was only abdominal pain at our case. Also mass was determined at right lower quadrant of abdomen. Although of all these findings, our initial diagnosis was perforated

appendicitis because of the older age of our case.

Ultrasound has greatly facilitated the diagnosis of intussusception (5). Description of the classic ultrasonographic findings of the "target" sign and pseudokidney appearance. Fluoroscopic images during reduction tend to miss pathological lead points (6). We determined target sign at ultrasonography, too. We did not apply any further diagnostic and treatment procedure (barium enema, etc) because of the high incidence of malignant lead point at older children's intussusception.

It is well established, that in older children intussusception is frequently caused by a pathological lead point. The medical literature also states, that any child older than 6 years with colic abdominal pain, palpable abdominal mass and radiological evidence of intussusception should be considered as having an ileal lymphosarcoma, until proved otherwise (7,8). Partially due to this data, and partially due to the reports that adult intussusception is caused by malignant lead points in more than 50% of the cases (3,4). Pathological lead points in children may also include Meckel diverticulum, duplication cysts, lymphoma, hemangioma and appendix (9,10). We did not determine any tumoral mass at our case but we excised the lymph nodes near the cecum and terminal ileum.

Some authors have noted a recent increase in the absolute number of the cases of intussusception in older children (11). In several publications, prolonged and relatively mild clinical course was often present in the older age group of children, as well as a higher recurrence rate (1,2,11,12). So that the intussusception must be thought at older ages and abdominal exploration must be performed because of high rate of lead point and recurrence.

REFERENCES

- 1)Turner D, Rickwood AM, Brereton RJ. Intussusception in older children. *Arch Dis Child* 1980;55:554-6.
- 2)Shuh S, Wesson DE. Intussusception in children 2 years of age or older. *CMAJ* 1987;136:269-72.
- 3)Barussaud M, Regenet N, Briennon X, de Kerviler B, Pessaux P, Kohnen-Sharhi N, et al. Clinical spectrum and surgical approach of adult intussusceptions: a multicentric study. *Int J Colorectal Dis* 2006;21:834-9.
- 4)Azar T, Berger DL. Adult intussusception. *Ann Surg* 1997;226:134-8.
- 5)Burke LF, Clarke E. Ileocolic intussusception – a case report. *J Clin Ultrasound* 1977;5:346-7.
- 6)Miller SF, Landes AB, Dautenhahn LW, Pereira JK, Connolly BL, Babyn PS, et al. Intussusception: ability of fluoroscopic images obtained during air reduction to depict lead points and other abnormalities. *Radiology* 1995;197:493-6.
- 7)Lopez Alvarez-Buhilla P, Idigoras G, Torres C, Larrea F, Bezanilla JL. Non-Hodgkin lymphoma as a cause of intussusception in children. *An Esp Pediatr* 1987;27:467-70.
- 8)Wayne ER, Campbell JB, Kosloske AM, Burrington JD. Intussusception in older child-suspect lymphosarcoma. *J Pediatr Surg* 1976;11:789-94.
- 9)Navarro O, Dugougeat F, Kornecki A, Shuckett B, Alton DJ, Daneman A. The impact of imaging in the management of intussusception owing to pathologic lead points in children. *Pediatr Radiol* 2000;30:594-603.
- 10)Ein SH, Shandling B, Reilly BJ, Stringer DA. Hydrostatic reduction of intussusception caused by lead points. *J Pediatr Surg* 1986;21:883-6.
- 11)Belyaev MK. Features of intestinal invagination in children. *Khirurgiia (Mosk)* 2003;4:47-50.
- 12)Fallis JC. Intussusception in older child. *CMAJ* 1976;114:38-40.

NEPHROTIC SYNDROME ASSOCIATED WITH ISOTRETINOIN TREATMENT

Case Report

İSOTRETİNOİN TEDAVİSİ İLE İLİŞKİLİ NEFROTİK SENDROM VAKASI

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ABSTRACT

Isotretinoin is a synthetic retinoid used for the treatment of moderate to severe acne rosacea. Although various adverse effects during isotretinoin treatment have been reported, renal impairment is very rare. Herein we report a case of full-blown nephrotic syndrome associated with isotretinoin treatment.

A previously healthy 29 year old lady, presented with periorbital and ankle oedeme. Based on the clinical and laboratory findings, she was diagnosed as nephrotic syndrome. A renal biopsy was taken and minimal change disease was diagnosed due to electromicroscopic examination which showed prominent effacement and microvillus formation in the pedicels of epithelial cells in the glomerulus. The only significant point in the history was the use of isotretinoin (40 mgr/day) which had been prescribed six months ago for acne rosacea. Isotretinoin was immediately stopped and she was treated with 1 mg/kg/day alfa-methyl prednisolone. The nephrotic syndrome responded to treatment in 3 weeks.

Nephrotic syndrome is a rare side effect of isotretinoin therapy and should be suspected in any patient with oedema formation during therapy.

Key words: nephrotic syndrome; isotretinoin; oedema.

ÖZET

Isotretinoin, orta ve ağır şiddetli akne tedavisinde kullanılan sentetik bir retinoiddir. Isotretinoin tedavisi ile farklı yan etkiler bilinmekte olup, tedavi ile ilişkili böbrek yan etkileri oldukça nadirdir. Bu vaka sunumunda isotretinoin tedavisi altında gelişen nefrotik sendrom vakası takdim edilmiştir.

Daha önce bilinen bir hastalığı olmayan 29 yaşındaki bayan hasta periorbital ve alt extremité ödemi ile nefroloji polikliniği'ne

başvurmuştur. Klinik ve laboratuvar bulguları sonucu nefrotik sendrom tanısı konulan hastanın hikayesinde tek göze çarpan nokta, 6 ay önce akne tedavisi için 40 mgr/gün dozunda isotretinoin tedavisi başlanmış olmasıydı. Isotretinoin tedavisi hemen kesilerek alf-methyl-prednisolone 1 mgr/kg/gün başlatılan hastanın 3 hafta içinde nefrotik sendrom klinik ve laboratuvar bulguları tamamen düzelmiştir. Sonuç olarak, isotretinoin tedavisinde çok nadir olarak nefrotik sendrom görülebilir ve bu tedavi sırasında ödem yakınması olan bütün hastalarda şüphe uyandırılmalıdır.

Anahtar kelimeler: nefrotik sendrom; isotretinoin; ödem.

INTRODUCTION

Isotretinoin, a vitamin A related molecule, has been widely used in the treatment of severe and antibiotic-resistant cystic acne vulgaris for more than 25 years. It is also rarely used in the treatment of certain malignancies such as certain skin cancers, brain and pancreatic tumours. Isotretinoin, normally found in small quantities in humans, is known to be a safe drug and most of the side effects are related to dry mouth, dry eyes, cheilitis, headache, nausea, elevated liver enzymes, a shift towards an atherogenic lipid profile and teratogenic birth defects (1,2). Despite this wide spectrum of side effects, little is known about its impact on the kidneys. Here we report a case of full-blown nephrotic syndrome following the use of isotretinoin.

CASE PRESENTATION

A previously healthy 29 year old lady was referred to our nephrology department due to periorbital and ankle oedema. She also complained of turbidity in urine and 10 kg weight gain in the last two months. No previous renal disease or familial renal disease history was found. There was no history of hypertension, diabetes or any clue for rheumatic and vasculitic diseases. The only significant

point in the history was the use of isotretinoin (40 mgr/day) which had been prescribed six months ago for acne rosacea. On the day of the admission, she was still on isotretinoin therapy. Physical examination revealed significant periorbital and pretibial oedema. Blood pressure was 110/80 mmHg with a pulse rate of 72 beats/min. Blood work showed normal blood urea nitrogen (11mg/dl), creatinine (0.57 mg/dl) and electrolyte levels with significant hypoalbuminemia (1.8 gr/dl). Total cholesterol (445mg/dl) and LDL cholesterol levels (347mg/dl) were significantly increased. Hematologic parameters were all within normal limits (Haemoglobin 14.3 gr/dl, Leukocytes 10,000/mm³ with normal differential count) except for ESR (64 mm/hr, normal <15 mm/hr). Serological tests including viral markers, ANA, Anti-dsDNA, RF, ANCA titers as well as the Direct Coombs test were all negative. Complement factor levels were also normal (C3:1.07 gr/l, C4: 0.227 gr/l). 24-hour urine collection revealed 4100 mg/day microalbuminuria. Urine microscopy was negative for cellular elements and casts. Ultrasonographic examination showed two normal-sized kidneys with normal echogenicity without any evidence of obstruction. Based on these findings, she was diagnosed with nephrotic syndrome and a percutaneous renal biopsy was undertaken to identify the underlying glomerular pathology. Histologic examination revealed 13 glomeruli without evidence of sclerosis. Thirty percent of glomeruli showed mild mesangial expansion without any cellular infiltration, necrosis or crescent formation. The remainders of the glomeruli as well as the tubulointerstitial and vascular areas were all normal under light microscopic examination. Immune-fluorescence revealed IgM and C3 deposition in only one glomerulus. Kongo-red staining was negative for amyloidosis. Electron microscopic examination showed prominent effacement and microvillus formation in the pedicels of epithelial cells in the glomerulus (**Figure 1**).

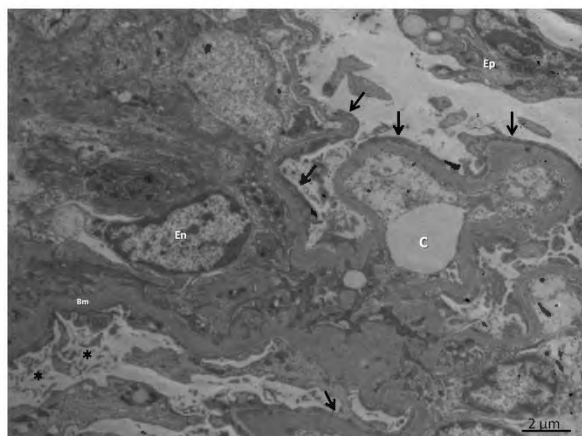


Figure 1: Electron microscopic examination; Prominent effacement (arrows) and microvillous transformation (asterix) in the pedicels of epithelial cells (Ep) in the glomerulus. (Bm): Basement membrane; (En): Nucleus of an endothelial cell; (C): Capillary lumen. Bar=2 μ m .

On the basis of data mentioned, diagnosis of minimal change disease due to isotretinoin use was made. Isotretinoin was immediately stopped and she was treated with 1 mg/kg/day alfa-methyl prednisolone and 80 mg/day furosemide. The nephrotic syndrome responded to treatment in 3 weeks. During that time, serum albumin level increased to 4 gr/dl, microalbuminuria decreased to 30mg/day while the patient lost 8 kg with complete resolution of oedema. Corticosteroids were tapered off every 2 weeks without any sign of relapse. On her last follow -up two months after the start of therapy, the patient was on 4 mg of alfa-methyl prednisolone without any evidence of relapse.

DISCUSSION

Drugs are well known causes of minimal change disease. ACE inhibitors, 5 aminosalicylic acid, sulphasalazine, NSAII and gold are the most known agents . To date, only a single case of nephrotic syndrome development after isotretinoin use has been reported before. In that report, the nephrotic syndrome was clinically manifest 2 months after the start

of therapy and clinical symptoms had resolved after discontinuation of the drug and a short course of corticosteroids (3). Our patient 's clinical, histologic and outcome features are similar. The only difference is that our patient developed nephrotic syndrome after 4 months of therapy. An interesting case report by Horber et al described renal impairment, hypercalcemia and proteinuria in a patient treated with etretinate (a synthetic retinoid) for superficial bladder tumour (4). Pavesse et al also reported mild proteinuria and renal impairment in a patient treated with isotretinoin for acne after 2 months of therapy. After discontinuation of therapy renal functions and proteinuria returned to normal [5]. However in both case reports, proteinuria was in the non-nephrotic range. Although it may be argued that the development of nephrotic syndrome was coincidental in our patient, rapid resolution of symptoms and proteinuria after withdrawal of isotretinoin and the quick response to a short course of corticosteroid therapy imply a causal relationship.

CONCLUSION

Nephrotic syndrome is a rare side effect of isotretinoin therapy and should be suspected in any patient with oedema formation during therapy. The drug should be stopped immediately with a short course of corticosteroid therapy started in patents with the full nephrotic syndrome.

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REFERENCES

- 1)Strauss JS, Krowchuck DP, Leyda JJ et al. Guidelines of care of acne vulgaris management. *J Am Acad Dermatol* 2007; 56 : 651-663.
- 2)Berard A, Azaoulay L, Koren G, Blais L, Perrault S, Oraichi D. Isoretinoin, pregnancies, abortions and birth defects; a population based perspective .*Br J Clin Pharmacol* 2007; 63 :190-205.

3) Oers van AJ, Leeuw DJ, Bommel van HE. Nephrotic syndrome associated with isotretinoin. *Nephrol Dial Transplant* 2000;15: 924.

4) Horber FF, Zimmerman A, Frey FJ. Impaired renal function and hypercalcaemia associated with etretinate. *Lancet* 1984; 10: 1093.

5) Pavese P, Kuentz F, Belleville C, Rouge P, Elsener M. Renal impairment induced by isotretinoin. *Nephrol Dial Transplant* 1997;12:129.