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Interventional Radiology

Nutcracker syndrome in adolescent with perineal pain

An interesting case of an adolescent with perineal pain due to pelvic congestion from nutcracker syndrome with relief after balloon venoplasty and sclerotherapy

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ABSTRACT

Nutcracker phenomenon is the descriptor for a patient's anatomy whenever the left renal vein becomes compressed between the abdominal aorta and the superior mesenteric artery. Nutcracker syndrome is the terminology used when the nutcracker phenomenon is accompanied by symptoms including pain (abdominal, flank, pelvic), hematuria, and orthostatic proteinuria. Diagnosis can be made with Doppler ultrasound, venography, computed tomography, or magnetic resonance imaging. This case demonstrates some of the typical findings of nutcracker syndrome. The limited clinical features and interesting imaging findings, in addition to the young age of the patient, make this a notable case.

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Case report

A 13-year-old girl presented with a chief complaint of perineal pain of 5 weeks duration. Two weeks before, the patient

had complained of coccygeal pain, thought to be secondary to a fall during cheerleading practice, although no incident was reported. The patient's pain at baseline rated at 2 of 10, which increased when bearing down while stooling or sneezing. Initial workup included a sacrococcygeal radiograph to rule out

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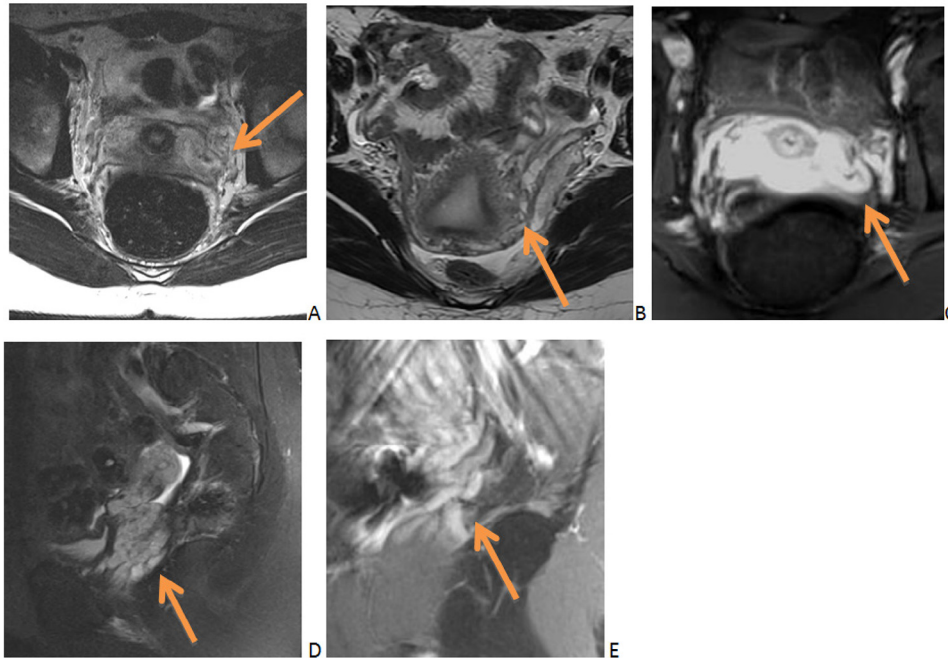


Fig. 1 – (A-E) Axial T2 fast spin echo (FSE) demonstrating dilated paracervical and parauterine veins (A); axial T2 FSE with fat suppression normal study for comparison; (B) paracervical and parauterine venous enhancement following gadolinium (C); sagittal T2 FSE with fat suppression demonstrating the dilated paracervical and parauterine veins (D); sagittal T1 FSE with fat suppression demonstrating enhancement following gadolinium, although artifact distorts the superior aspect of the image (E).

fracture, which only demonstrated a moderate stool burden. The patient was subsequently started on Mirilax therapy twice daily, resulting in regular bowel habits. Her pain persisted, becoming so severe that she stayed at home from school for 2 weeks.

The patient's medical history was remarkable for *Helicobacter pylori* gastritis, chronic constipation, exercise-induced asthma, multiple allergies, and anaphylaxis to nuts. Surgical history included tonsillectomy, adenoidectomy, and colonoscopy. The patient was adopted and lived with her adoptive mother, father, and younger sister. The patient was active in cheerleading and soccer. She denied alcohol, drugs, tobacco, and sexual activity. She did well in school, although had missed multiple days due to pain, which included the inability to sit at her desk for more than a few minutes secondary to perineal pain.

Review of systems revealed no saddle paresthesia, numbness of the extremities, or pain radiation. She denied lower limb weakness, changes in gait, loss of balance, or bladder and bowel incontinence. Lastly, she denied fever, decreased appetite, emesis, and diarrhea. The patient reported pain localized at her perineal and sacral regions, which worsened with any action that involved bearing-down (sneezing), sitting on hard surfaces, and activities such as running. Finally, the patient endorsed a moderate weight gain, which she attributed to the considerable decrease in physical activity from the pain.

On physical examination, the patient's vitals were normal. She had mild lower abdominal tenderness to palpation. There was no edema. She also had perineal tenderness at the right and left lower regions between the vagina and anus. A speculum examination was performed under anesthesia, and no visible perineal varicosities were seen at the vaginal intro-

itus. The patient's laboratory values at the time (including urinalysis and culture) demonstrated normal values. Following this examination, the patient left the office without clear etiology for her symptoms. Over the next 2 weeks, the patient's pain gradually became more severe with radiation up her back and around her hips. The patient's symptoms were not relieved by positional changes and became recalcitrant to ibuprofen (initially dosed at 400 mg as needed and then increased to 800 mg as needed), which led her to visit the emergency room.

Because of the unclear etiology of the patient's pain, multiple imaging studies were ordered including pelvic magnetic resonance imaging (MRI) and ultrasound. MRI with and without contrast of the patient's pelvis demonstrated dilated enhancing serpentine vessels extending to the left lateral aspect of the uterus (Figs. 1A-E).

These findings were most consistent with pelvic varicosities or plexiform neurofibroma. Ultrasound evaluation was then performed. Dynamic transperineal ultrasound, with valsalva maneuvers and Doppler, demonstrated multiple dilated perivaginal, left adnexal, and uterine veins (Figs. 2A-D). These sonographic findings further supported pelvic venous congestion vs vascular malformation.

Computed tomography (CT) of the abdomen and pelvis with contrast was performed next and it demonstrated a compressed left renal vein (LRV) at the level of the superior mesenteric artery (SMA) with a "beak sign" (rapid tapering of the vessel to a point) [1] (Figs. 3A and B). Reconstructed sagittal CT imaging demonstrates the angle of the SMA from the abdominal aorta (AA) acute at 24 degrees (Fig. 3B), less than

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