ARTICOLI ORIGINALI - ORIGINAL CONTRIBUTIONS

Chronic inguinal pain in children



Ann. Ital. Chir., 2018 89, 6: 479-484 pii: \$0003469X18028142

Francesca Calzolari^{*}, Carmine Del Rossi^{**}, Valentina Maffini^{***}, Alberto Attilio Scarpa^{**}, Laura Lombardi^{**}, Enrico Vaienti[°], Paolo Bocchi^{°°}

*Specialty Training Program in Paediatrics, University of Parma, Nuovo Ospedale dei Bambini, Parma, Italy

**Paediatric Surgery Unit, Nuovo Ospedale dei Bambini, Parma, Italy

***Paediatric Unit, Nuovo Ospedale dei Bambini, Parma, Italy

°Orthopaedic Surgery Unit, Azienda Ospedaliero-Universitaria di Parma, Italy, Italy

°°Valparma Hospital, Surgery department, Langhirano, Parma, Italy

Chronic inguinal pain in children

PURPOSE: To present our experience in the management of chronic groin pain in children.

METHODS: We report 4 patients (age range 0-18 years old) who presented with history of chronic groin pain (April 2010 January 2017). After failure of all conservative treatments they underwent surgical management as ultima ratio. RESULTS: Currently, there is no consensus on treatment of 'Sportsman's Hernia' and literature on paediatric population is still poor. There are no appropriate randomised controlled trials supporting a standardized management of chronic groin pain. Initial approach should be conservative [Physical Medicine and Rehabilitation (PM&R) follow-up for 3-6 months] and surgical treatment proposed after failure of conservative therapies. It is advisable to consider surgery at least months after clinical onset. More recently, some studies suggested surgery as a first line treatment. Most of surgical cases manage to recover to full activity without pain, as reported in our experience. Surgical approach to the groin can be anterior (open) or posterior (laparoscopic), both these procedures have shown good results. Surgeons having different opinions on the aetiology of 'Sportsman's Hernia' may sustain the suitability of different surgical techniques. In our experience, pain was localized at the pubic tubercle therefore we preferred the 'open' approach ensuring a successful release of ilioinguinal and genitofemoral nerves.

CONCLUSIONS: Children with chronic groin pain can benefit of surgical treatment. In our experience, surgery always proved to be successful for treating chronic groin pain in paediatric age. Further multicentric studies are needed to support these results.

KEY WORDS: Bernhardt-Roth syndrome, Children, Chronic groin pain, Chronic inguinal pain, Meralgia Paraesthetic, Sportsman's Hernia

Introduction

The inguinal canal is a complex anatomic structure that should be understood in the context of its three-dimensional anatomy. It extends inferiorly and medially, from depth to surface. The nerves that run in the region of the inguinal canal are as follows: the lateral femoral cutaneous nerve, the genitofemoral nerve, the ilioinguinal and iliohypogastric nerves. The last three run into the inguinal canal (Fig. 1), whereas the lateral femoral cutaneous nerve runs externally and laterally.

Chronic groin pain is a common cause of disability in athletes from a variety of sports ¹. It can be the result of an identifiable pathology of the hip joint and pubic region or related tendons. Many of these cases are given the final diagnosis of 'Sportsman's Hernia': a condition that has no real definition in literature, nor a suggested diagnostic exam and recommended treatment, but results to be a debilitating and common condition, rep-

Pervenuto in Redazione Marzo 2018. Accettato per la pubblicazione Aprile 2018

Correspondence to: Francwesca Calzolary, MD, Speciality Training Program of Paediatrics, Nuovo Ospedale dei Bambini, University of Parma, Via Gramsci 14, 43126 Parma, Italy (e-mail: francesca.calzolari@studenti.unipr.it)



Fig. 1: Handmade drawing depicting normal anatomy of the inguinal canal with Ilioinguinal Nerve and the genital branch of the Genitofemoral Nerve running parallel to the spermatic cord.

resenting a large proportion of the cases of chronic groin pain, when alternative diagnoses have been excluded. According to the British Hernia Society at least 3 of the following clinical findings should be present to make a diagnosis of 'Sportsman's Hernia', i.e.:

1. Point sensitivity where the conjoint tendon adheres to the pubic tubercle;

2. Sensitivity to palpation in the deep inguinal ring;

3. Pain and/or dilatation in the outer inguinal ring without apparent hernia;

4. Pain at the origin of adductor longus muscle;

5. Diffuse inguinal pain extending to the perineum, inner surface of the femur and crossing the midline 2 .

The most common sports where this pathology is present are those that involve kicking and twisting movements while running, particularly in rugby, football and ice hockey ³. In other sports such as cycling and swimming this condition occurs less frequently, because the pelvic and torso movements predisposing to a painful groin are not so frequent.

However, the etiology remains still not entirely known, but is likely to be multifactorial, resulting from overload of the lower anterior abdominal wall and pelvis, with the impression that laxity of the posterior inguinal wall, conjoint tendonitis and/or compression of the ilioinguinal nerve play a role in the onset of symptoms. The control of rotation movements and pelvic stability are the most important factors in preventing the occurrence of initial or recurrent inguinal damage.

Patients often demonstrate non-specific physical examination findings and normal imaging studies. However, symptoms are often severe enough to have ended the career of professional athletes.

Materials and Methods

Here we report four cases of patients in the paediatric age with chronic groin pain, they presented between April 2010 and January 2017. After the failure of all conservative treatments previously carried out, they needed surgical exploration as ultima ratio.

C.L. 11-year-old boy

He practices competitive swimming. One day in the morning, just after waking up, he reported sudden pain in the left groin. He described this pain as a sort of "internal shocks". He had just come back home from one week of winter holiday during which he fell several times while skiing. He never reported the same pain in that region previously. As symptoms did not improve during the following days, L. was brought to a peripheral hospital Accident & Emergency (A&E) Department. On examination symptoms were associated to "pubalgia", thus he was discharged home with mild painkillers. Over the following weeks pain persisted, therefore the child was admitted at the Paediatric Haematology-Oncology Unit of our institute in order to undergo diagnostic investigations (MRI, bone scintigraphy, CT-scan, PETscan) all reporting normal results. No abnormalities were found on routine blood tests. Various analgesic therapies were administered (Ketorolac, Morphine) with partial and transitory relief. He was then discharged with a plan of outpatient follow-up in order to address anxiety and emotional components related with pain.

L. was said to suffer from a depressive disorder. He was prescribed with a combination of SSRIs (Fluoxetine) and Benzodiazepines (Lorazepam) but pain and paraesthesias continued and worsened causing severe functional limitation.

L. was no longer able to walk and started using a wheelchair to move around. He finally became bedridden for most of the day, with no possibilities of moving his legs as this movement was causing severe pain and internal shocks.

Following a specialty referral carried out by an Orthopaedic Surgeon, treatment with local anaesthetic injections in the left groin area was attempted with only transitory pain relief. This test is considered diagnostic for the syndrome of nervous compression. The case was discussed with a Pediatric Surgeon and an Adult General Surgeon who decided for surgical exploration. During the operation the inguinal canal was opened: there was no evidence of hernias, the ilioinguinal nerve, the genital branches of both the genitofemoral nerve and the iliohypogastric nerve were freed and preserved and a Ropivacaine injection was administered. Then the external oblique muscle aponeurosis was closed, avoiding nervous compression. From the following day, pain resolved completely. Patient underwent a two-month kynesitherapic rehabilitation in order to recover normal mobility and joint function that were highly compromised at the moment of operation due to antalgic reflex postures.

B.A. 11-year-old boy

Past medical history of appendectomy for acute appendicitis at the age of 10. Three years later he reported right groin pain. He plays competitive football, but was not reporting any recent injuries at presentation.

Initially, pain was described as irregular and occasional but later on became permanent and forced him to walk with an antalgic gait. Pain improved by lying down huddled or by offloading his body weight on the opposite side. Pain was described as a "pins and needles" sensation (paraesthesia) and sometimes as "electric shocks", its intensity causing nocturnal awakenings.

He was admitted for routine evaluation and several instrumental investigations (MRI, Ultrasounds) were carried out. No abnormalities were found. The child underwent different treatments using nSAIDs and mild opioids with just a transitory relief. He started a PM&R follow-up for "severe osteitis of the pubis", though he became no longer able to walk and started using a wheelchair to move around. After failure of all conservative treatments previously carried out, surgical exploration was indicated. A multidisciplinary surgical team performed an exploration of the inguinal canal, which was opened and the genital branch of the ilioinguinal, iliohypogastric and genitofemoral nerves were preserved.

There was no evidence of hernias and compression on the nervous branches caused by the internal oblique muscle was removed. Finally, a troncular injection of the three nerves with Ropivacaine was performed. The external oblique muscle aponeurosis was closed, leaving a wide external inguinal ring. During operation no evidence of direct or indirect ipsilateral inguinal hernia was found. The day after, the child began walking normally and independently thanks to the disappearance of pain and discomfort.

N.G. 13-year-old girl

She had been playing competitive athletics (high jump, long jump, running) for the past 5 years. After the summer, while restarting the athletic training at the begin-ning of September, she began to complain pain at the right groin. Initially appendicitis was suspected: an abdominal ultrasound was carried out with no direct nor indirect signs of appendicitis. Collaterally, a right ovarian follicular cyst was pointed out but was no longer observed on a following ultrasound done as follow-up. Pain worsened over the following days and G. described it as a continuous pain, with an overlapping of intermittent stronger stabs and exacerbation on finger pressure. Symptoms caused severe and progressive limitation to physical activity inducing the child to definitively stop any physical activity since early October. An MRI was therefore carried out, but did not show relevant abnormalities. Patient underwent further admissions and investigations (all with negative results) within the following 2 months and intravenous analgesic treatment (NSAIDs) was prescribed with only temporary benefit. Once dis-

charged home, pain became so intense that G. was forced to be absent from school for several days. Consumption of analgesic drugs (paracetamol and codeine) was routinely required for symptoms control, with the onset of side effects. Finally, a surgical exploration was indicated, as for previous cases. At the opening of the external inguinal ring, there was no evidence of hernias and the ilioinguinal nerve in its cranial component (emergence from the abdominal wall) was clearly compressed from the muscular fibres of the internal oblique muscle.

An accurate decompression of the same nerve was performed, with stab incision of the muscular fibres. Finally, a troncular injection of the three inguinal nerves with Ropivacaine was administered and the aponeurosis of the external oblique muscle reconstructed, leaving a wide external inguinal ring.

After surgery, G. reported some pain limited within the area of the surgical wound, which promptly disappeared within a few days. One month after intervention patient was in good health and free from medications while reporting no residual pain. She went back to physical activity successfully and with no limitation.

S.A. 12-year-old girl She played competitive volleyball. She reports sudden groin pain in the right area, without history of recent local trauma. Pain was reported to trigger from the inguinal region, then irradiating towards the upper lateral region of the ipsilateral thigh. Within two months she was admitted 3 times in A&E, where routine blood tests (with normal results), surgical examination (that found no sign of appendicitis) and gynaecological examination with ultrasound (with normal results) were carried out. As pain did not improve she underwent an orthopaedic examination and X-ray of the right coxofemoral joint that excluded signs of fracture. She was finally admitted to the Paediatric Unit of our hospital for further investigation. The young girl presented with functional limitation during abduction and extra- rotation of the right lower limb. Abdominal examination showed a flat and soft abdomen with localized pain in the right iliac fossa. A. reported spontaneous pain in the right groin and the ipsilateral thigh, below the anterior superior iliac spine. Symptoms were described as a burning pain, with local paraesthesias. Pain was present also at rest and its intensity was causing nocturnal awakenings. Symptoms worsened while walking as well as by prolonged standing in an upright position and sitting cross-legged.

During admission, blood tests (with normal results), right hip ultrasound (negative for effusion and signs of joint inflammation), pelvis and dorso-lumbo-sacral spine MRI (negative for organic lesions, disk disease or other morphostructural abnormalities) were carried out.

A symptomatic treatment with Ketoprofen by intravenous injection was administered with partial benefit. Furthermore, patient underwent an orthopaedic exami-



Fig. 2: Intra-operative picture showing compression site (white arrow) of the lateral femoral cutaneous nerve at the level of the inguinal ligament once the sartorius muscle fascia was opened. Lateral femoral cutaneous nerve is lifted on a vessel loop.

nation. A compression syndrome to the lateral femoral cutaneous nerve (Meralgia Paraesthetica or Bernhardt-Roth syndrome) was suspected and a local injection with Lidocaine was performed in the region with rapid response and complete, although transitory, regression of pain (due to the duration of action of the local anaesthetic drug). The young girl was then discharged home with a program of regular painkillers and orthopaedic and PM&R follow-up. Over the following period Scrambler Therapy (interrupted after few sessions because of lack of improvement) and oral therapy with Gabapentin (soon suspended for little benefit and the onset of side effects such as drowsiness and hyporeactivity) were attempted.

Also in this case, surgical operation was finally indicated. Once the sartorius muscle fascia was opened, the lateral femoral cutaneous nerve was clearly compressed at the level of the inguinal ligament. Thus, an incision of 2-3 mm of the inguinal ligament was performed (Figs. 2, 3). The surgical exploration of the inguinal canal was carried out and demonstrated no evidence of hernias. The aponeurosis of the external oblique muscle was then closed, leaving a large external inguinal ring and an infiltration of the subcutaneous tissue with Ropivacaine was administered.

From the day after surgery the patient reports no pain. In the following period she gradually recovered to normal joint mobility and completed the PM&R program of rehabilitation successfully.

Discussion

Currently, there is no consensus on treatment of the 'Sportsman's Hernia' as it is based more on physicians'



Fig. 3: Intra-operative picture showing stab incision of the inguinal ligament at compression site with consequent releasing of the lateral femoral cutaneous nerve (lifted on a vessel loop).

experience rather than on scientific evidence. Randomised controlled trials on this subject are still few in number and often not conclusive.

The initial treatment should be conservative and a PM&R physician specialized in groin diseases should follow the patient for 3 to 6 months in order to address the dysfunctional pelvis stability. A randomised study carried out by Hoelmich examined the effects of using a structured exercise programme (six exercises including strengthening, coordination and core-stability exercises) versus a "normal" exercise regime in treating patients with adductor-related groin pain. This study proved a 31% reduction in the risk of groin injury with the use of the structured programme, but this result did not reach statistical significance ⁵. This study followed on from a previous one from the same group, which concluded that an active training programme was more effective than a non-active training programme, with kynesitherapy present in both arms ⁶. A Dutch group compared the effects of a multi-modal treatment approach versus a simple exercise therapy, confirming that a multi-modal approach was significantly effective in a quicker return to physical activity, but it also affirmed that neither treatment was completely effective 7.

More recently an Australian group supposed that inflammation and compression around the ilioinguinal nerve were the most relevant causes of chronic groin pain in athletes and carried out a study examining the effects of radiofrequency denervation of the inguinal ligament, compared to local anaesthesia and topical steroid injections in 36 patients. This study proved a significant benefit in terms of pain reduction with the use of radiofrequency denervation as compared to other local therapies. Surgical treatment should be considered after failure of conservative therapies. It is necessary to indicate surgery at least 3 months after the onset of symptoms. More recently, some studies propose surgery even as the first line treatment for groin pain in sportsmen ⁸. Most of surgical cases manage to return to full activity without pain ⁹.

Surgical treatment has been described using both the anterior (open) ¹⁰ and posterior (laparoscopic) techniques ¹¹.

Both have shown good results and surgeons having different opinions on the aetiology of 'Sportsman's Hernia' may sustain the suitability of different surgical techniques. Furthermore, literature lacks of randomised controlled trials comparing different surgical strategies in order to demonstrate superiority of one technique over the others. As a result, based on their own expertise and practice, different surgeons adopt various techniques from "open" to laparoscopic surgery, especially when professional athletes are involved. In our experience, we preferred the 'open' approach to the laparoscopic one, because the pain was localized at the origin of the rectus abdominis muscle, at the pubic tubercle (insertion site for the conjoint tendon). Namely, the anterior approach is the only one that can ensure a successful release of the compression on ilioinguinal and genitofemoral nerves.

The fourth case reported is peculiar because of presenting two co-existing components: besides a chronic groin pain, the patient reports a characteristic irradiation to the upper lateral region of the thigh. This kind of pain is called Meralgia Paraesthetica (or Bernhardt-Roth syndrome). Meralgia Paraesthetica consists of an entrapment syndrome of the lateral femoral cutaneous nerve and it presents with sensory loss, pain and paraesthesias in the distribution of the nerve ¹². A lesion can occur at any point along the course of the nerve, but entrapment at the inguinal ligament is the most frequent and wellestablished cause ¹³. Such entrapment is often the result of anatomic variations in the course of the nerve.

The damage can also occur as a result of local trauma or during surgery. In any case, knee and medial thigh are never involved by symptoms and there is never any motor weakness of the quadriceps or decrease of patellar reflex. Walking and strenuous effort are often aggravating factors. Palpation of the nerve near the lateral end of the inguinal ligament can reproduce symptoms, while a lidocaine injection in the same area can relieve symptomatology, thus confirming the diagnosis. Meralgia Paraesthetica in children is more frequent than suggested from the literature, but this disease is often unrecognized or misdiagnosed and many patients are examined by different specialists (neurologists, orthopedic surgeons, general surgeons, gynecologists and general practitioners) before the diagnosis of Meralgia Paraesthetica is established. In children symptoms are more frequently bilateral, they have usually a chronic course and present often in thin and muscular subjects. Operative decompression is advisable when the pain due to

Meralgia Paraesthetica is severe and persistent. After open decompression, excellent results are reported in literature, also in children ¹². Less severe symptoms such as primarily sensory loss or mild pain do not need to be treated surgically, unless they progress.

As there is no universally accepted criteria or gold standard for its diagnosis, chronic groin pain due to nerve compression can remain unrecognised and can cause patients to be set within the context of psychiatric and mood disorders (e.g. major depressive episode). However, paediatric reports in literature are still few and there are no appropriate randomised controlled trials on chronic groin pain and its treatment. As in the four cases reported from our experience, it is often a longstanding, disabling pain, causing severe limitations in patients' lifestyle.

After surgical decompression, in all four cases there has been a complete regression of painful symptomatology, with progressive improvement of joint motility and functionality.

Conclusions

Therefore, also in children chronic groin pain with suggestive symptoms, normal laboratory investigations and instrumental examinations, unresponsive to conservative therapies, indicates surgical treatment.

In our experience, indeed, surgery always proved to be effective as successful treatment for chronic groin pain in the paediatric age. Further multicentric studies are needed to confirm these results.

Riassunto

Viene riportata la nostra esperienza sul tema del trattamento del dolore inguinale cronico in età infantile a proposito di quattro pazienti di età compresa tra 11 e 13 anni nell'intervallo di tempo dall'aprile 2010 al gennaio 2017. Dopo il fallimento delle cure conservative, come ultima ratio si è fatto ricorso alla chirurgia.

Di fatto non c'è accordo sul trattamento dell'ernia dello sportivo, e la letteratura sulla popolazione pediatrica è ancora limitata. Non vi sono adeguati trials che diano un indirizzo standard sul trattamento del dolore inguinale cronico.

L'approccio iniziale dovrebbe essere conservativo: terapia medica ed esercizio fisico all'interno di un programma riabilitativo; e il trattamento chirurgico andrebbe proposto dopo il fallimento delle terapie conservative. È consigliabile prendere in considerazione la chirurgia non prima dei 3 mesi dall'esordio della sintomatologia clinica. Più recentemente alcuni studi suggeriscono il trattamento chirurgico come prima scelta. La chirurgia mira al recupero completo dell'attività senza dolore, come nella nostra esperienza. L'accesso chirurgico alla regione inguinale può essere anteriore o posteriore, per via laparoscopica, ed entrambi gli approcci hanno dimostrato buoni risultati.

I chirurghi che hanno opinioni diverse riguardo la causa dell'ernia dello sportivo possono essere assertori di tecniche chirurgiche differenti.

Nella nostra esperienza il dolore era localizzato a livello del tubercolo pubico e pertanto la nostra preferenza è andata all'accesso anteriore di chirurgia open con efficace sbrigliamento dei nervi ilio-inguinale e genitofemorale.

Possiamo concludere che i ragazzi con dolore inguinale cronico possono beneficiare del trattamento chirurgico e che nella nostra esperienza in età pediatrica la chirurgia è sempre stata coronata da successo: naturalmente sono necessarie ulteriori esperienze per confermare questi risultati.

References

1. Comin J, Obaid H, Lammers G, Moore J, Wotherspoon M, Connell D: *Radiofrequency denervation of the inguinal ligament for the treatment of 'Sportsman's Hernia': A pilot study.* Br J Sports Med, 2013; 47(6):380-86.

2. Sheen AJ, Stephenson BM, Lloyd DM, Robinson P, et al.: 'Treatment of the sportsman's groin': British Hernia Society's 2014 position statement based on the Manchester Consensus Conference. Br J Sports Med, 2014; 48(14):1079-87.

3. Kingston JA, Jegatheeswaran S, Macutkiewicz C, Campanelli G, Lloyd DM, Sheen AJ: *A European survey on the aetiology, investigation and management of the "sportsman's groin"*. Hernia, 2014; 18(6):803-10.

4. Garvey JF, Read JW, Turner A: Sportsman hernia: What can we do? Hernia, 2010; 14(1):17-25.

5. Hölmich P, Larsen K, Krogsgaard K, Gluud C: *Exercise program for prevention of groin pain in football players: A cluster-randomized trial.* Scand J Med Sci Sports, 2010; 20(6):814-21.

6. Hölmich P, Uhrskou P, Ulnits L, Kanstrup IL, Nielsen MB, Bjerg AM, Krogsgaard K: *Effectiveness of active physical training as treatment for long-standing adductor-related groin pain in athletes: randomised trial.* Lancet, 1999; 353(9151):439-43.

7. Weir A, Jansen JA, van de Port IG, Van de Sande HB, Tol JL, Backx FJ: *Manual or exercise therapy for long-standing adductorrelated groin pain: A randomised controlled clinical trial.* Man Ther, 2011; 16(2):148-54.

8. Siddiqui MR, Kovzel M, Brennan S, Priest OH, Preston SR, Soon Y: *A literature review on the role of totally extraperitoneal repairs for groin pain in athletes.* Int Surg, 2012; 97(4):327-34.

9. Morales-Conde S: Sportsman's hernia: an entity to be defined, diagnosed and treated properly? Videosurg Other Miniinvasive Techn 2009; 4:32-41.

10. Muschaweck U, Berger L: Minimal Repair technique of sportsmen's groin: An innovative open-suture repair to treat chronic inguinal pain. Hernia, 2010; 14(1):27-33.

11. Paajanen H, Brinck T, Hermunen H, Airo I: Laparoscopic surgery for chronic groin pain in athletes is more effective than non-operative treatment: a randomized clinical trial with magnetic resonance imaging of 60 patients with sportsman's hernia (athletic pubalgia). Surgery, 2011; 150(1):99-107.

12. Edelson R, Stevens P: Meralgia paresthetica in children. J Bone Joint Surg Am, 1994; 76(7):993-99.

13. Richer LP, Shevell MI, Stewart J, Poulin C: *Pediatric meralgia paresthetica*. Pediatr Neurol, 2002; 26(4):321-23.