Coccygectomy for Chronic Refractory Coccygodynia in Pediatric and Adolescent Patients

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Aims: Coccygodynia (also known as coccydynia and coccyalgia) described for the first time by Simpson in 1859. It is a disabling pain in the coccyx usually provoked by sitting or changing position from a sitting to a standing position. Treatment started with adjustment of ergonomics as specialized cushion for sitting, application of local heat, and oral analgesics. The aim of this study was to assess the results of total or partial coccygectomy for patients suffering from chronic refractory coccygodynia and not improving on conservative treatment in pediatrics and adolescents.

Methods: The study included 22 patients, 8 of them with chronic refractory coccygodynia not responding to conservative management underwent partial or total coccygectomy using Key’s surgical exposure after good preoperative preparation.

Results: This study included 5 females (62.5%) and 3 males (37.5%), their ages ranged from 7 to 15 years. The results were excellent in 75% of patients and good in the remaining cases. No significant intraoperative or postoperative complications were reported. The parents were satisfied in almost all cases in the long term follow up.

Conclusions: Coccygodynia is a disabling problem, especially in pediatric patients. Coccygectomy is a feasible management option and has a favorable outcome when performed for an appropriate indication.

Keywords: Coccyalgia, coccydynia, coccygectomy, coccygodynia, pediatric

Introduction

The coccyx had important physiological and anatomical relations to the pelvic floor musculature, the fifth sacral and coccygeal nerve roots, and the terminal sympathetic plexus. Coccyx-related pathological conditions produce local pain and tenderness and also radiating and radiculopathic symptoms. Coccygodynia (also known as coccydynia and coccyalgia) was described by Simpson first time in 1859, is a disabling pain in the coccyx that is provoked by sitting or changing from seated to a standing position. Although it is a disabling problem, there is paucity in literature about coccygodynia in pediatrics and adolescents. This tail bone pain may radiate rostrally to the sacrum or lumbar spine or laterally to the buttocks. Patients may rarely present with associated rectal pain or radicular symptoms. One-third of the patients have associated back pain which may be the cause for misdiagnosis.

Maigne et al. described a condition of instability-related coccygodynia (dislocation or hypermobility);[3,4] it constitutes a very good indication for surgery and coccygectomy seems to produce the best results.[5]

Treatment started with adjustment of ergonomics as specialized cushion for sitting, application of local heat, and oral analgesics. Good results reported after using local injections of steroids and/or local sodium channel blocking anesthetic agents.[6] Added measures are repeated hyperextension and hyperflexion of the coccyx under general anesthesia. The most invasive therapy involves surgical removal of the coccyx.[6]
The aim of this study was to assess the results of total or partial coccygectomy for patients suffering from chronic refractory coccygodynia and not improving on conservative treatment in pediatrics and adolescents.

**METHODS**

This prospective study was done at the period between April 2017 to December 2019 at Pediatric Surgery Unit, Tanta University Hospitals, Egypt, and included 22 pediatric and adolescent patients presented with coccygodynia. Fourteen patients were improved on conservative treatment yielding 8 patients with chronic refractory coccygodynia who subjected to surgical treatment in the form of total or partial coccygectomy. Approval was obtained from the institute’s Research Ethics committee. Parents or the legal guardians of each patient were informed about all steps in this study including the operative idea and post-operative follow up. An informed consent was taken from parents of each patient included in the study. Privacy of the participants and confidentiality of the data were maintained.

For all patients, the following was done: analysis of demographics, duration of symptoms, clinical findings, and preoperative X-ray imaging ± magnetic resonance imaging [Figure 1].

During the period of the study, we had 22 patients presented with coccygodynia; patients selected for coccygectomy had a history of failed conservative treatment for at least 6 months (which includes ergonomic adjustments such as a specialized cushion for sitting, application of local heat, oral analgesics, local injections of steroids, and/or local sodium channel blocking anesthetic agents). Diagnosis was by clinical examination, based on the presence of pain at the tip of the coccyx with palpation of a painful bony excrescence under the skin. Patients may exhibit a guarding seated posture, in which one buttock is elevated to shift weight from the coccyx. The anatomical configuration was determined by radiographs of the sacrum and coccyx before surgery on anteroposterior and lateral views, and the coccyx type was evaluated following the classification of Postacchini and Massobrio. In 1983, they defined four types of the coccyx according to shape and size [Table 1].

In Type I, 10 cases, the coccyx is slightly curved forward. In Type II, 7 cases, the coccyx has a more marked curve and points forward. In Type III, 3 cases, the coccyx characterized by a sharp forward angulation, and Type IV, 2 cases, it characterized by subluxation in the sacrococcygeal or intercoccygeal joint [Table 1]. All patients with type I coccyx and 4 of type II improved on conservative treatment, while 3 cases of Type II coccyx needed surgical intervention. None of Type III and IV patients respond to the conservative management and they needed surgical intervention [Table 1].

The shortest duration of pain with conservative treatment prior to surgery was 6 months. A preoperative bowel preparation had been done to all patients to minimize complications in the unlikely event of a rectal perforation and to prevent fecal contamination of the wound. Third-generation cephalosporin antibiotics were administered intravenously prior to skin incision. Under general anesthesia, coccygectomy (key’s surgical exposure with en bloc resection from a proximal to distal direction) was performed in the prone position.

To expose the gluteal cleft, the buttocks were retracted laterally with adhesive tape.

The operative technique is to reach the terminal coccygeal segment directly via a small vertical incision in the natal cleft followed by an exposure from the proximal to distal direction [Figure 2]. The coccyx is elevated and separated from the surrounding tissues circumferentially (the coccyx 1 is identified anatomically by the cornua at the articulation with the caudal sacral segment). In the case of a Fusion of 5th sacral segment and 1st coccyx disc, the resection proceeds distal to the first mobile segment [partial coccygectomy], unless the proximal portion is prominent and close to the skin [total coccygectomy], after removal of the sacrococcygeal disc, in a subperiosteal plane using monopolar electrocautery, proceeding with an en-bloc resection and removing of the terminal coccygeal segment [Figure 3]. The wound was irrigated with normal saline, closed in two layers without drain. All patients received postoperative dual antibiotic therapy for 3 days, and to avoid direct pressure on the surgical wound, the patient received nursing care in a lateral position.

**Follow-up**

The patients were followed up by the operating surgeon in the outpatient clinic 2 weeks, 1 month, 3 months and every 3 months after the operation. The median follow-
up period was 15 months (range between 7 and 25 months). Parents were advised to contact the department of pediatric surgery, if there were any concerns in the immediate postoperative period. According to the criteria described by Bayne et al.,[9] clinical results were assessed. Excellent referred to complete relief of pain with no change in lifestyle or occupation. A good result referred to occasional discomfort after prolonged sitting and minimal discomfort on digital examination, again no change in lifestyle or occupation, and analgesics were not required. A fair result referred to pain or discomfort on sitting for short period of time and considerable pain on digital examination, changes in lifestyle or occupation were fairly minimal, and analgesics were occasionally required. A poor rating implied no improvement following surgery with the continuous need for analgesics.

**RESULTS**

We performed eight consecutive coccygectomies in the Pediatric Surgery Unit at Tanta university hospitals, Egypt between 2017 and 2019. No cases in Type I coccyx needed surgery, Type II 3 of 7 cases, Type III all the 3 cases, and Type IV the 2 cases needed surgery. Total coccygectomy was done in five patients and partial coccygectomy was done in three patients. The study included 5 females (62.5%) and 3 males (37.5%), and their ages ranged from 7 to 15 years (mean 10.91 ± 1.87). The mean length of hospital stay was 1.43 ± 0.98 days. No significant surgical complications were encountered. One case of superficial infection was treated with oral antibiotics. The results were excellent in 75% of the patients and good in the remaining cases [Table 2]. All parents were satisfied that their children did not complaining of coccygeal pain any more on the long-term follow-up.

**DISCUSSION**

Coccygodynia is classified into idiopathic or posttraumatic (fall onto the buttocks or difficult childbirth). True coccygodynia must be distinguished from false in which pain is referred to the coccygeal region from the visceral organs, from a root, plexus, or peripheral nerve, or dural irritation. Coccygodynia also classified as acute or chronic (pain lasting more than 2 months).[10] In our study, the shortest duration of pain with conservative treatment prior to surgery was 6 months.

Coccygodynia is diagnosed by physical examination and radiological study. Patients may exhibit a guarding sitting posture, in which one buttock is elevated to shift weight from the coccyx. Palpation will elicit a tender point at or distal to the sacrococcygeal junction. The coccyx may be hypermobile and painful on rectal examination. In our study, 10 cases were Type I coccyx (45.5%), 7 cases were Type II (31.8%), 3 cases were Type III (13.6%), and 2 cases were

**Table 1: Radiographic Postacchini and Massobrio Classification of the coccyx**

| Radiographic Classification of the Coccyx | Total Number (n=22) | Improvement on Conservative Treatment | Needs Coccygectomy |
|-----------------------------------------|---------------------|---------------------------------------|--------------------|
| Type I: Curved slightly forward         | 10                  | 10                                    | 0                  |
| Type II: More marked curve, straight forward | 7                  | 4                                     | 3                  |
| Type III: Sharply angled anteriorly     | 3                   | 0                                     | 3                  |
| Type IV: Subluxation of the sacrococcygeal or intercoccygeal joints | 2                   | 0                                     | 2                  |
Table 2: Postoperative results according to the criteria described by Bayne et al.\textsuperscript{[9]}

| Results | Number of patients (n=8), n (%) |
|---------|--------------------------------|
| Excellent | 6 (75) |
| Good    | 2 (25) |
| Fair    | 0     |
| Poor    | 0     |

Type IV (9%), and this is almost similar to Karadimas et al.,\textsuperscript{[11]} who analyzed 153 radiographs and according to them, 57 cases were Type I (37.3%), Type II were 48 cases (31.4%), Type III had 29 cases (18.9%), and 19 cases in Type IV (12.4%).

This study included 22 patients (8 males and 14 females), and this distribution is in agreement with Karadimas et al.,\textsuperscript{[11]} who reviewed the literature and found the gender characteristics were provided on 617 patients, the vast majority were females bringing the male/female ratio to 1/4.4.\textsuperscript{[6,7,9,12,13]} In three studies, the demographics were presented only on the initial recruited population.\textsuperscript{[6,14,15]}

The age distribution found in our study was ranged from 7 to 15 years and is different with the literature where the mean age of those series was from 26.4–52.8 years.\textsuperscript{[12,16]} and the extremes of the age were 11 and 76 years.\textsuperscript{[6]}

In this study, the probable etiology of coccygodynia was idiopathic in 18 cases (82%) and traumatic in 4 cases (18%). In Karadimas et al.,\textsuperscript{[11]} review on 457 patients, direct trauma was the most common cause of coccygodynia, recorded in 270 patients (59.1%). Idiopathic coccygodynia (no identifiable causes) was present in 141 cases (30.9%). Childbirth in 37 females (8.1%). Recent lumbar spinal or rectal surgery or epidural injection in 9 (1.9%).

Chronic coccygodynia is defined as failure of conservative treatment for six months. Prior to coccygectomy, all cases had failed non-operative treatment for at least 6 months and this period of non-surgical treatment was noticed also in Hellberg et al., and Hodges et al. series.\textsuperscript{[14,17]}

In our study, low residual diet and fleet enema was used to prevent any contamination of the wound during the operation. For the same purpose, intravenous administration of the third generation cephalosporin was used. Perioperative use of antibiotics was recorded in all series\textsuperscript{[11]} except Bayne et al. study.\textsuperscript{[9]}

In our study, we used key’s surgical exposure and this was in accordance with the literature\textsuperscript{[12,18,19]} dissection from a proximal to distal direction limits the risk of rectal injury, especially in the case of an anteverted coccyx, whereas in only one study, the Gardner’s exposure was used (resection from distal to proximal direction).\textsuperscript{[9]} Statistical analysis of the incidence of complications between the two different techniques revealed a higher complication rate in patients treated with the Gardner method, but this difference did not reach significance.\textsuperscript{[11]}

Wound infection is the most frequently reported complication in patients treated with coccygectomy. To reduce the infection rate after coccygectomy, the surgeons have proposed various strategies, such as antibiotic prophylaxis before and after surgery, making a longitudinal incision,\textsuperscript{[17]} drainage, periosteal preservation, and closure.\textsuperscript{[12]}

Pennekamp et al.\textsuperscript{[20]} observed wound infection in 19% of the patients in their study. The present study suggests that prophylactic antibiotics over a period of 72 h postoperatively would be reasonable measure to decrease wound infection. No significant surgical complications were encountered in this study and the results were excellent in 75% of the patients and good in the remaining cases, and the parents were satisfied in almost all cases in the long-term follow-up. It is in accordance with the systematic analysis in other series in which 504 over 596 patients treated with coccygectomy had excellent or good outcome (84.6%), 46 patients fair (7.7%), and 46 poor outcomes (7.7%).\textsuperscript{[6,7,9,12,13]}

**CONCLUSIONS**

Coccygodynia is a discomforting and disabling problem, especially in pediatric and adolescent patients. Coccygectomy for coccygodynia was not commonly practiced by pediatric surgeons. Although the number of cases in this study is small, coccygectomy is a feasible management option and has a favorable outcome when performed for an appropriate indication. Strict measures must be taken to prevent surgical wound infection.

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**Conflicts of interest**

There are no conflicts of interest.

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