

A pediatric surgeon's 35-year experience with pilonidal disease in a Canadian children's hospital

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Accepted for publication
Nov. 13, 2009

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DOI: 10.1503/cjs.028509

Background: There is an ongoing debate regarding the optimal surgical management for pilonidal disease in the pediatric population. The purpose of this study was to evaluate a pediatric surgeon's experience at a Canadian children's hospital over 35 years.

Methods: We performed a retrospective review of the charts of patients seen and treated from July 1969 to December 2003, inclusive. All patients were evaluated for age, sex, clinical diagnosis, infection, treatment, healing time, complications and results.

Results: In all, 121 adolescents with pilonidal disease (64 boys, 57 girls) with a mean age of 15 (range 12–19) years were evaluated at the same children's hospital. The 107 (88%) patients with infection (46% acute) underwent surgery. At operation, all 107 pilonidal cysts were either excised and packed open, marsupialized or excised and closed primarily without drainage under general anesthesia; the operation performed was arbitrarily chosen. Vacuum-assisted closure was not used. All patients received antibiotics. The time for healing after the initial operation in the group whose cysts were excised and packed open was at least twice as long (75 d) as in the other 2 groups ($p = 0.031$). Disease recurred in 24 (22%) patients, 6 (25%) of whom experienced 2 recurrences. Among the 90 patients in the excised and packed open group, 20 (22%) experienced recurrences and 5 (25%) experienced 2 recurrences. Among the 13 patients in the marsupialized group, 3 (23%) experienced recurrences and 1 (33%) experienced 2 recurrences. Among the 4 patients in the excised and closed primarily without drainage group, 1 (25%) experienced a recurrence and none experienced 2 recurrences ($p = 0.12$). Each recurrence was smaller than the original. All wounds eventually healed. There were no other complications and no deaths. A multivariable logistic regression analysis revealed that the type of surgical approach was not predictive of recurrence after controlling for age and sex.

Conclusion: Age, sex and surgical approach were not predictive of recurrence. From our experience, excision and packing open the wound produced a longer morbidity but offered the same results compared with marsupialization or excision and primary closure without drainage.

Contexte : Le débat se poursuit au sujet de la prise en charge chirurgicale optimale de la maladie pilonidale dans la population pédiatrique. Cette étude visait à évaluer l'expérience qu'un chirurgien pédiatrique a acquise en 35 ans dans un hôpital canadien pour enfants.

Méthodes : Nous avons procédé à une étude rétrospective des dossiers de patients vus et traités de juillet 1969 à décembre 2003 inclusivement. Nous avons évalué tous les patients selon l'âge, le sexe, le diagnostic clinique, l'infection, le traitement, la durée de la guérison, les complications et les résultats.

Résultats : Au total, 121 adolescents qui avaient une maladie pilonidale (64 garçons, 57 filles) et qui étaient âgés en moyenne de 15 (de 12 à 19) ans ont été évalués au même hôpital pour enfants. Les 107 (88 %) patients qui avaient une infection (aiguë dans 46 % des cas) ont subi une intervention chirurgicale. Au moment de l'intervention, les 107 kystes pilonidaux ont été soit excisés et bourrés d'une mèche, soit marsupialisés, soit excisés et fermés immédiatement sans drainage, sous anesthésie générale. L'intervention pratiquée a été choisie arbitrairement. On n'a pas utilisé la fermeture assistée par le vide. Tous les patients ont reçu des antibiotiques. La guérison après l'intervention initiale chez les patients dont les kystes ont été excisés et bourrés d'une mèche a pris au moins 2 fois plus de temps (75 j) que chez les 2 autres groupes ($p = 0,031$). La maladie est réapparue chez 24 (22 %) des patients, dont 6 (25 %) ont eu 2 récurrences. Sur les 90 patients qui ont subi l'excision et dont la plaie a été bourrée

d'une mèche, 20 (22 %) ont eu une récurrence et 5 (25 %) en ont eu 2. Chez les 13 patients dont la plaie a été marsupialisée, 3 (23 %) ont eu une récurrence et 1 (33 %) en a eu 2. Chez les 4 patients dont le kyste a été excisé et fermé immédiatement sans drainage, 1 (25 %) a eu une récurrence et aucun n'en a eu 2 ($p = 0,12$). Chaque récurrence a été plus petite que l'originale. Toutes les plaies ont fini par guérir. Il n'y a pas eu d'autres complications et il n'y a eu aucun décès. Une analyse de régression logistique à variables multiples a révélé que le type d'approche chirurgicale n'était pas un prédicteur de récurrence compte tenu de l'âge et du sexe.

Conclusion : L'âge, le sexe et la méthode chirurgicale n'étaient pas des prédicteurs de récurrence. Notre expérience indique que l'excision et le bourrage de la plaie ont produit une morbidité plus longue, mais les mêmes résultats que la marsupialisation ou l'excision et la fermeture immédiate sans drainage.

Pilonidal disease in pediatric patients is not a rare occurrence. However, only 7 papers have appeared in the pediatric surgery literature since 1972. There is an ongoing debate regarding the optimal surgical management for this disease. We present a series of patients treated by the same pediatric surgeon (S.H.E.) over a 35-year period.

METHODS

We conducted a retrospective chart study. Inclusion criteria were all children with infected, symptomatic pilonidal disease. All patients were evaluated for age, sex, clinical diagnosis,¹ infection, treatment, healing time, complications and results. All patients were amenable to 3 surgical treatments: excision and packed open, marsupialization or excision and primary closure without drainage. Vacuum-assisted closure was not used. We performed a χ^2 test for categorical variables and analysis of variance (ANOVA) for continuous variables, and we performed a multivariable logistic regression analysis. The Research Ethics Board of SickKids in Toronto, Ontario, approved our study protocol.

RESULTS

From July 1969 to 2003 inclusive, 121 adolescents with pilonidal disease were evaluated: 64 boys and 57 girls with a mean age of 15 (range 12–19) years. From this group, 14 (12%) patients were treated nonoperatively because their pilonidal disease was not infected and was asymptomatic; the 107 (88%) patients with infection (46% acute), including 1 with hemophilia and 4 with diabetes,

underwent surgery. All 107 operations were performed under general anesthesia, with the patient usually in the lateral or occasionally prone position. All patients in this series were amenable to 3 surgical approaches, with the procedure chosen arbitrarily by the surgeon. Ninety pilonidal cysts were excised and packed open, 13 marsupialized and 4 excised and closed primarily without drainage; vacuum-assisted closure was not used (Table 1). All patients received antibiotics. Follow-up was continued until their wounds were healed; the average time for healing after all operations was 39 days (75 d for those whose cysts were excised and packed open, 34 d for those whose cysts were marsupialized, and 8 d for those whose cysts were excised and closed primarily without drainage ($p = 0.031$).

Disease recurred in 24 (22%) patients, 6 (25%) of whom experienced 2 recurrences (Table 1). Among the 90 patients in the excised and packed open group, 20 (22%) experienced recurrences and 5 (25%) experienced 2 recurrences. Among the 13 patients in the marsupialized group, 3 (23%) experienced recurrences and 1 (33%) experienced 2 recurrences. Among the 4 patients in the excised and closed primarily without drainage group, 1 (25%) experienced a recurrence and none experienced 2 recurrences ($p = 0.12$). The mean time to recurrence was 195 (range 30–390) days. Each recurrence was smaller than the original. We performed a multivariable logistic regression analysis (with the outcome being recurrence), and the possible predictive variables were age, sex and surgical approach. None of these variables were predictive of recurrence (odds ratio 0.24–4.24, $p = 0.12$). There were no other complications and no deaths.

Table 1. Surgical results of a surgeon's 35-year experience with pilonidal disease in children

Result	Total no. (%) patients, n = 207	Treatment; no. (%)*		
		Excision and packed open, n = 90	Marsupialized, n = 13	Excision and closed primarily without drainage, n = 4
Mean healing time, d†	39	75	34	8
First recurrence‡	24 (22)	20 (22)	3 (23)	1 (25)
Second recurrence‡	6 (25)	5 (25)	1 (33)	0

*Unless otherwise indicated.
 † $p = 0.031$.
 ‡Odds ratio 0.24–4.24, $p = 0.12$, on multivariable logistic regression.

DISCUSSION

Although pilonidal disease is common, very little has been written about its occurrence in pediatric patients. There is no mention of it in the indexes of recent textbooks on pediatric surgery,²⁻⁴ and only 7 papers have appeared on this topic in the pediatric surgery literature since 1972 (all since 1990).⁵⁻¹¹ Moreover, the *Journal of Pediatric Surgery* has only 3 references to pediatric pilonidal disease in its indexes from 2000 to 2008, inclusive.⁹⁻¹¹ One must therefore surmise that this problem almost always occurs in adolescents, that its treatment is virtually the same as for adults and that many of these patients are treated by general surgeons.^{7-10,12} Interestingly enough, in 2005 there were 3 random prospective studies from general surgeons that showed the following.

- Rhomboid excision and Limberg flap had a faster healing time and lower recurrence than excision and primary closure.¹³
- Limited excision of the fistulous tract and wound left open had a faster healing time than either excision with primary closure and drainage or excision with packing open. Apart from the minimal nonsignificant elevation of the rate of recurrence in the closed wound group, there was no difference in recurrence among the 3 groups.¹⁴
- Rhomboid excision and Limberg flap had fewer postoperative complications than excision and primary closure.¹⁵

It has been reported that the incidence of pilonidal disease in children is 2.5 in 1000;⁶ the incidence of this problem in the senior author's (S.H.E.) practice was 1.2 in 1000. The girl:boy ratio of 1:1 in this series is not the same as that reported elsewhere,^{5,6,8} but the mean age of 15 (range 12-19) years was similar.

There is nothing written about this disease in the Gray and Skandalakis textbook *Embryology for surgeons*.¹⁶ It has been assumed that this disease is caused by hair in the coccygeal region being "in-driven"¹² into the subcutaneous tissues, subsequently becoming infected in many cases. It is unknown if or when all pilonidal cysts eventually become infected; Lee and colleagues¹¹ reported that about 50% of their patients had initial drainage of an acute abscess before definitive treatment. In our series, 88% of patients had infected cysts; the remaining 12% were small, not infected and asymptomatic, and surgery was not suggested. It is assumed that, in many children, asymptomatic pilonidal disease remains undiagnosed and, similarly, many of these children never experience infection. General anesthesia was used in all surgeries. Initially, the operative position used was prone; however, the lateral position was soon favored because of the safer maintenance of the endotracheal tube and the ease of moving large teenage patients. All patients received routine antibiotics in therapeutic doses. The organisms cultured and treated were similar to those reported by Rowe and Fallat.¹²

In this series, 94 patients had their pilonidal disease

grossly excised, even if the presentation was an acute infection/abscess; the wound was packed open (90) or closed primarily without drainage (4). Marsupialization was used in 13 patients;¹² vacuum-assisted closure was not used. The most common procedure used during the 35 years of this series both at SickKids and throughout the pediatric surgical literature was excision and packing open; the procedure chosen in this series was not fixed by any rules and was chosen arbitrarily by the surgeon. In actual fact, there were really no significant changes in surgical or medical management of pilonidal disease throughout the pediatric surgical world during this study period. Both marsupialization and excision with primary closure without drainage were chosen during the earlier part of this series (between 1969 and 1994, inclusive), and all of the patients had the same baseline characteristics (e.g., age, sex, chronicity of disease).

The time for healing in the excised and packed open group after the initial operation was at least twice as long (75 d) as in the other 2 groups, whereas the recurrence rate was similar in all 3 groups. The reason for this equally high recurrence rate with all 3 operative procedures is unclear.¹⁴ One area of confusion in the literature has been the differentiation of a postoperative infection and a recurrence. We considered all postoperative infections as recurrences, because we observed that these infections never healed until treated as a recurrence.¹⁷ Other authors reported an equally high recurrence rate, causing a search for alternative methods of closure (vacuum-assisted primary closure,^{9,10} primary closure with suction drainage,^{7,12} Karydakias asymmetric skin flap^{8,12,18} and the Bascom cleft lift midline shift closure¹⁹), which lowered their recurrence rates to 14%, 12%, 4% and 0%, respectively. The latter 2 procedures seem to offer the best results but have not been universally embraced by pediatric surgeons.

The percentage of patients followed until the operative area returned to normal (range 2 wk to 1 yr; mean 6 mo) was virtually 100%. No attempt at a longer follow-up was made once healing occurred; however, it was expected that if there was a new long-term problem, the patient would return to the original surgeon. This seems to be the best that can be achieved under the circumstances. In Toronto (population 3 million) and surrounding communities (population 10 million), the referral patterns and patient base are mostly related to SickKids. Moreover, if 1 of the original patients underwent surgery elsewhere, more often than not, that information would be transmitted to the SickKids surgeon.

Limitations

The major focus of this paper was to evaluate the results and outcome (as defined by recurrence) of the surgical treatment of pediatric pilonidal disease in a single surgeon's practice; a minor focus was the healing time after each of the 3 operations. This study, however, has several limitations. It is a retrospective series, and there are a limited

number of patients who received 2 of the 3 operations available according to the purely arbitrary choice of the surgeon. The fact that most of the patients were surgically treated with excision and packing open, which was the most common and popular procedure both at SickKids and throughout the pediatric surgical literature at the time precludes any statistical conclusions because the number of patients receiving the 2 other operations was too small. As such, age, sex and surgical approach were not predictive of recurrence.

CONCLUSION

Considering the above limitations with this series, excision and packing open produced a longer morbidity (average time for healing after the initial operation was at least twice as long at 75 d), but the same results (recurrences), when compared with both marsupialization or excision and primary closure without drainage.

Competing interests: None declared.

Contributors: Dr. Ein designed the study and acquired the data. Drs. Nasr and Ein analyzed the data, wrote and reviewed the article and together approved its publication.

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