Sphincter denervation in anorectal incontinence and rectal prolapse

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SUMMARY Biopsies of the external anal sphincter, puborectalis, and levator ani muscles have been examined in 24 women and one man with long-standing anorectal incontinence, 18 of whom also had rectal prolapse, and in two men with rectal prolapse alone. In 16 of the women anorectal incontinence was of unknown cause, but in eight there was a history of difficult labour. Similar biopsies were examined in six control subjects. In all the incontinent patients there was histological evidence of denervation, which was most prominent in the external anal sphincter muscle biopsies, and least prominent in the levator ani muscles. Myopathic features, which were thought to be secondary, were present in the more abnormal biopsies. There were severe histological abnormalities in small nerves supplying the external anal sphincter muscle in the three cases in which material was available for study. We suggest that idiopathic anorectal incontinence may be the result of denervation of the muscles of the anorectal sling, and of the anal sphincter mechanism. This could result from entrapment or stretch injury of the pudendal or perineal nerves occurring as a consequence of rectal descent induced during repeated defaecation straining, or from injuries to these nerves associated with childbirth.

Faecal incontinence occurs in patients with various neurological disorders, particularly with spinal cord and lumbosacral root lesions, after trauma to the muscles of the pelvic floor (Parks and McPartlin, 1971), in association with rectal prolapse (Porter, 1962), and as an idiopathic disorder (Parks, 1975). Anorectal incontinence, a term used to denote faecal incontinence not caused by neurological disorders, occurs in about two-thirds of patients with rectal prolapse referred for operation, but a third of these remain incontinent after rectopexy, and these patients are then similar to those with idiopathic anorectal incontinence. The latter disorder occurs almost exclusively in women. These patients usually complain of a disturbance of anorectal sensation so that they are unable to differentiate flatus from faeces, though formal sensory examination of the skin of the anal margin reveals no abnormality, and the tendon reflexes in the legs are preserved. The rectal sphincter is commonly patulous, there is little if any voluntary sphincter contraction, and the anal reflex is usually absent. The normal anorectal angle is lost (see Parks, 1975) and the pelvic floor is usually dropped in relation to surrounding structures. During coughing or straining the puborectalis muscles become passively stretched so that the pelvic floor fails to rise, or may even descend (Parks et al., 1966; Parks, 1975). In many of these patients there may also be minor degrees of rectal prolapse, but this follows the development of faecal incontinence; in patients with primary rectal prolapse faecal incontinence, if it occurs at all, usually develops some months or years after prolapse has occurred (Porter, 1962). However, both idiopathic anorectal incontinence and rectal prolapse, with or without incontinence, are often associated with a long history of excessive straining during defaecation and there are thus many points of similarity between the two disorders (Porter, 1962; Parks, 1975).

During the past 15 years surgical reconstruction of the anorectal angulation, restoring the flap-valve mechanism responsible for continence (Parks et al., 1966), has proved an effective treatment of anorectal incontinence (Parks, 1975). The operative approach is postanal so that the anus and rectum are approached from their visceral aspect, thus exposing the internal and external anal sphincters, and the puborectalis and levator ani muscles. This surgical

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Table Details of 27 patients

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*Cases in which muscle biopsies were examined with enzyme histochemical stains and with electron microscopy.

Exposure of the muscles responsible for anorectal continence has provided an opportunity for their histological study, and in this paper we shall describe our observations of abnormalities found in 81 biopsies of these muscles, obtained from 27 patients. We have also made preliminary observations on the innervation of these muscles.

Methods

Clinical Material

Muscle biopsies, taken from the external anal sphincter, puborectalis, and levator ani muscles during operations for correction of idiopathic anorectal incontinence, or of rectal prolapse, have been examined in 27 cases (see Table). In three of the 27 patients, small nerves supplying the external anal sphincter and puborectalis muscles were also biopsied. The patients' ages ranged from 24 to 80 years (mean 58 years). Twenty-five of the 27 patients suffered from anorectal incontinence of from nine months to 40 years' duration (mean eight years). Two patients (cases 26 and 27) had rectal prolapse without incontinence: these were both men, but all the other patients, except one (case 19), were women. One patient (case 2) was found to have sensory impairment in the anus and in a small area of skin between the anus and coccyx, a distribution consistent with bilateral pudendal nerve lesions. In this case there was a history of anorectal incontinence with loss of anorectal and perianal sensation as a result of obstetric trauma; faecal incontinence began after a difficult or precipitate labour in seven other cases (see Table). Seven women (cases 5, 16, 17, 21, 22, 24, and 25), however, had never been pregnant. In case 16 faecal incontinence followed a haemorrhoidectomy. In most cases anorectal incontinence was of gradual onset and it was accompanied by rectal prolapse in 18 patients. There was a history of excessive straining...
during defaecation, for many years, in 16 patients (see Table). Only three (12%) patients (cases 6, 18, and 25) also suffered from urinary incontinence. There was no evidence of neurological disease in any patient, except case 2.

The normal appearances of the external anal sphincter, puborectalis, and levator ani muscles have been studied in six patients, aged 17 to 64 years. In one of these patients muscle biopsies were obtained during abdomino-perineal resection of the rectum for cancer, and in the remaining five cases muscle samples were obtained at necropsy. In all these patients there was no known history of rectal prolapse or of anorectal incontinence. In the cases coming to necropsy death had occurred as a result of road traffic accident, cardiac infarction, or cerebrovascular accident.

TECHNIQUES

During a preliminary survey, biopsies of the external anal sphincter, puborectalis, and levator ani muscles obtained from 20 patients undergoing surgical correction of anorectal incontinence (Parks, 1975) were fixed in formol-sublimate and embedded in paraffin block. Sections of these 60 blocks were stained for histological examination with haematoxylin and eosin and by van Gieson's method.

In seven later cases (cases 1, 2, 6, 13, 14, 18, and 25) small pieces of 21 biopsies, obtained from the same three muscles, were frozen in iso-pentane which had been cooled with liquid nitrogen. A series of nine consecutive transverse cryostat sections, each 8 μ thick, was cut from a block of each muscle, and these sections were prepared for light microscopy with haematoxylin and eosin, periodic acid Schiff, modified Gomori trichrome, and Sudan black B stains. A standard series of enzyme histochemical staining techniques, including nicotine adenine dinucleotide tetrazolium reductase (NADHtr), adenosine triphosphatase (ATPase) preincubated at pH 9.5, 5.4, and 4.3, and phosphorylase methods (see Dubowitz and Brooke, 1973), were also used, and longitudinal sections were also studied in most cases. The transverse sections of each biopsy, stained with haematoxylin and eosin, were rated as normal (0), mildly abnormal (1), moderately abnormal (2) or severely abnormal (3), in an attempt to compare the biopsies in the various cases (see Table).

Ultrastructural studies were also performed in these seven cases. Portions of each of the 21 biopsies were fixed in cold, 2.5% barbiturate-buffered glutaraldehyde, post-fixed in 2% phosphate-buffered osmium tetroxide, dehydrated in a series of graded alcohols, and embedded in Araldite. Semithin (1 μ) sections were stained with toluidine blue for light microscopy, and ultrathin sections of selected features from these blocks were mounted on Formvar-coated 1 mm open grids, stained with uranyl acetate and lead citrate using standard techniques, and examined in an AEI 6B electron microscope. The three nerve biopsies were prepared for light and electron microscopy in similar fashion.

Sections of the external anal sphincter, puborectalis, and levator ani muscles, obtained from the six control cases, were prepared using the same enzyme histochemical methods.

Results

PATIENTS WITH ANORECTAL INCONTINENCE OR RECTAL PROLAPSE

Almost all the 81 muscles examined were abnormal (Table). The changes found were similar in all the cases examined, though they varied in degree from case to case. The external anal sphincter was always the most abnormal of the three muscles and the levator ani was usually the least affected; in three cases the levator ani appeared normal. In the paraffin-embedded material a wide variety of abnormalities was present but it was difficult to be certain of their cause. Some of these problems have been clarified by study of the frozen material.

External anal sphincter muscle

The most abnormal of the external sphincter biopsies consisted of a few scattered striated muscle fibres embedded in fibrous and adipose tissue, situated adjacent to the smooth muscle fibres of the internal rectal sphincter muscle. In other, less abnormal biopsies the muscle fibres were arranged in groups of 10 to 60 fibres separated from each other by bands of fibrous or adipose tissue (Fig. 1). The fibres within each group were of approximately uniform size. Some of these fibres contained central sarcosomal nuclei (Fig. 1). In some cases myopathic changes were very prominent so that in haematoxylin and eosin preparations there was apparently random variation in fibre size, individual fibres being widely separated from each other by collagenous connective tissue. However, even in these cases, the ATPase and NADHtr stains (when frozen material was available) revealed fibre-type grouping consistent with reinnervation (Fig. 2).

Scattered necrotic fibres, some undergoing phagocytosis (Fig. 3) were sometimes found, usually close to the larger fibres in the biopsy. Splitting or fragmentation of individual muscle fibres was also present in most of these biopsies (see Fig. 6) and rare basophilic regenerating fibres were seen.

The Gomori trichrome stain sometimes showed fibres with a granular red margin and with a densely stippled interior: these regions also stained intensely
Sphincter denervation in ano-rectal incontinence and rectal prolapse

Fig. 1 Case 6. External anal sphincter. 
Haematoxylin and eosin (paraffin), x 140. Groups of fibres of varying size are separated by fibrous tissue. The muscle fibres in the external anal sphincter were always smaller than those in the puborectalis (Fig. 6) and levator ani muscles (see Fig. 7). SM: smooth muscle.

Fig. 2 Case 6. External anal sphincter. ATPase (pH 4.3) (frozen), x 140. Fibre type grouping and grouped denervation atrophy.

Fig. 3 Case 18. Puborectalis. Gomori trichrome (frozen), x 350. Two vacuolated necrotic fibres: one contains a leucocyte.
with the NADHtr reaction, but they were unstained with the ATPase technique. These changes are characteristic of 'ragged-red' fibres (Engel, 1971). In the same sections other fibres contained scattered rod-like (nemaline) bodies (Fig. 4). Ultrastructural studies confirmed that these rod-bodies were derived from the Z band material (Fig. 5). They were located in clusters in regions of focal myofibrillar degeneration and were often associated with collections of lipid droplets.

**Puborectalis muscle**

The abnormalities found in the puborectalis muscle biopsies (Fig. 6) were similar to those found in the external sphincter muscles, but they were always less severe (see Table). In general, however, the degree of abnormality in this muscle was related to that found in the external sphincter muscle. Fibre-type grouping was clearly evident in ATPase preparations, but myopathic changes, such as fibre splitting or fragmentation, necrosis, phagocytosis, or regeneration of single fibres with distinct variation in fibre size and central nucleation, were usually far less evident. Accumulations of rod-bodies were found in several cases but they were less prominent than in the external anal sphincter muscles.

**Levator ani muscle**

The levator ani muscle biopsies usually showed only mild abnormalities such as disseminated neurogenic atrophy. Grouped denervation atrophy was found in 11 cases. Type 1 fibre preponderance was common in this muscle. Myopathic abnormalities were rarely found and accumulations of rod-bodies were rare.

**Other features**

Many of the muscle fibres in the external anal sphincter biopsies were smaller (range of least diameters: 10 to 80 μ) than in the puborectalis (50 to 80 μ) or in the levator ani muscles (50 to 80 μ). No consistent difference, apart from differences in degree of abnormality, were observed when the biopsies of patients with anorectal incontinence alone, anorectal incontinence with rectal prolapse, or rectal prolapse alone were compared. Similarly the abnormalities found in case 2, a 30 year old woman whose anorectal incontinence was associated with anal and perineal sensory loss, were similar to those found in the other cases (see Table). Muscle spindles were found occasionally in each of the three muscles examined. They usually appeared normal, but in the external anal sphincter muscles there was some fibrosis of their periaxial spaces and capsules (see Swash and Fox, 1974).

**Innervation**

Small intramuscular nerve bundles, examined both by light and electron microscopy in the external anal sphincter biopsies, contained few nerve fibres, and were fibroed. Intramuscular nerve bundles were less affected in the puborectalis muscles of these cases, and were normal in the levator ani biopsies. In the three biopsies of nerves supplying the external anal sphincter muscles (Fig. 7) there was a pronounced reduction in number of myelinated nerve fibres, with some proliferation of Schwann cells. Segmental or paranodal demyelination was not found, and the unmyelinated nerve fibres were normal. The interstitial collagen was unusually prominent.

**Control cases**

As in other human muscles, the normal external anal sphincter, puborectalis, and levator ani muscles consist of a mosaic of type 1 and type 2 fibres. In the six control cases there was a preponderance of type 1 fibres in each of these three muscles. The external anal sphincter muscles contained scattered smaller fibres (Fig. 8) which were histochemically type 1 fibres (Fig. 9). Some similar smaller fibres were also found in the puborectalis muscles (Fig. 9). In some of these normal muscles, particularly in the external anal sphincter muscles, rare necrotic fibres were found. A detailed account of the normal structure of these three muscles, and of their nerve supply, will be given in a subsequent paper (Beersiek and Swash, in preparation).
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Discussion

The external anal sphincter, puborectalis, and levator ani muscles form a continuous muscular tube around the anorectal junction, the levator ani muscles opening like a funnel superiorly. The external anal sphincter muscle itself consists of subcutaneous, superficial, and deep portions, but only the deep portion forms a complete sphincter around the anal canal. Our biopsies were all taken from the deepest portions of this muscle, and some of them included smooth muscle from the internal sphincter musculature. Since the operative approach was the same in all the cases, the biopsies were always taken from similar sites.

The changes found in the muscle biopsies of these 27 cases were similar in all the three muscles examined, but always far more prominent in the external anal sphincter muscles than in the puborectalis or levator ani muscles. Many of the biopsies contained necrotic fibres, some of which were undergoing phagocytosis. Ragged-red fibres and fibres containing prominent accumulations of rod-bodies were also seen. Basophilic regenerating fibres were less evident, but fibre splitting was prominent in some of the biopsies. Most of the external anal sphincter biopsies...
One x enzyme histochemical of atrophy, contained this in and, NADHtr normal loads and such abnormalities (Schwartz et al., 1969), but these changes are usually mild. In our patients the abnormalities were severe, and they were not equally distributed in the three muscles studied. Further, similar abnormalities were found in the youngest and oldest patients (see Table). None the less, in some of our control subjects in whom biopsies of the same three muscles were taken at necropsy or during abdominoperineal resection of the rectum for cancer, some necrotic fibres were found in the external sphincter and puborectalis muscles (Fig. 9). The levator ani muscle was normal. Some of the type 1 fibres in the external sphincter muscles in these control subjects were smaller than those in the puborectalis and levator ani muscles, a finding that may account for the presence of small fibres in the biopsies of patients with anorectal incontinence and rectal prolapse. Further work on the normal structure and on the effect of age in these muscles is in progress (Beersiek and Swash, in preparation).

The pathogenesis of denervation in the three muscles studied is uncertain. In intramuscular nerve fascicles, and in small nerve twigs innervating the external anal sphincter muscle, there was loss of myelinated axons, with obvious endoneurial fibrosis. These abnormalities suggest a more proximally situated lesion of some chronicity. Further, the observation of accumulations of rod-bodies in some muscle fibres is consistent with long-standing denervation, though rod-bodies have been reported in a variety of other neuromuscular disorders (Dubowitz and Brooke, 1973), including a rod-body myopathy (Engel, 1966), and they do not therefore represent a specific disorder. Rod-bodies were usually most prominent in the external anal sphincter, the most severely affected of the three muscles examined.

The posterior part of the external anal sphincter muscle is innervated by branches of the inferior rectal nerve, and the anterior part receives innervation from the perineal nerve. Both these nerves are branches of the pudendal nerve (see Williams and Warwick, 1975). The inferior rectal nerve also contains cutaneous afferent fibres which are distributed to the lower part of the anal canal, and to the skin between the anus and the coccyx. This area of skin was anaesthetic in case 2. The perineal nerve supplies motor innervation to other perineal and pelvic muscles, including the puborectalis and levator ani muscles. All the various parts of the ano-rectal sling and of the anal sphincter mechanism are innervated, therefore, by branches of the pudendal nerve.

Fig. 6 Case 18. Puborectalis. Haematoxylin and eosin (frozen), x 140. Groups of fibres of differing size. Fibrous tissue is more prominent in the group of smaller fibres. One large fibre contains a separate central core and, in an adjacent fibre, splitting has occurred.

contained excessive interfascicular fibrous tissue, and this was also noted in many of the puborectalis biopsies. In the paraffin-embedded material it was often difficult to come to firm conclusions about the cause of the severe abnormalities found but, in the seven cases in which frozen tissue was available, the enzyme histochemical stains, particularly the ATPase and NADHtr stains, showed grouped denervation atrophy, a characteristic finding that suggested that denervation with subsequent reinnervation was the primary pathological process. There was no evidence of denervation in the control material. Myopathic changes, such as those found in these biopsies, are well recognised in long-standing denervation (Drachman et al., 1967) and there is evidence that these changes may be the result of mechanical trauma and of attempts at regeneration induced by normal loads imposed on a weakened muscle (Schwartz et al., 1976).

Before considering the pathogenesis of these abnormalities the possible effect of ageing on the perineal musculature must be considered. The mean age of our patients was 58 years, and 13 of them were older than 60 years. During ageing, without evident neuromuscular disease, both neurogenic and myopathic changes can be found in apparently normal muscles (Tomlinson et al., 1969), but these changes are usually mild. In our patients the abnormalities were severe, and they were not equally distributed in the three muscles studied. Further, similar abnormalities were found in the youngest and oldest patients (see Table). None the less, in some of our control subjects in whom biopsies of the same three muscles were taken at necropsy or during abdominoperineal resection of the rectum for cancer, some necrotic fibres were found in the external sphincter and puborectalis muscles (Fig. 9). The levator ani muscle was normal. Some of the type 1 fibres in the external sphincter muscles in these control subjects were smaller than those in the puborectalis and levator ani muscles, a finding that may account for the presence of small fibres in the biopsies of patients with anorectal incontinence and rectal prolapse. Further work on the normal structure and on the effect of age in these muscles is in progress (Beersiek and Swash, in preparation).

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We suggest that anorectal incontinence may be the result of damage to the pudendal nerves, or to one of their major branches. This could itself be caused, firstly, by trauma sustained during childbirth, as in our case 2, in whom there was an area of perineal sensory loss; trauma during childbirth was probably also a factor in cases 1, 3, 6, 8, 9 and 10 in whom ano-rectal incontinence, not associated with sensory loss, began after delivery. Secondly, repeated straining during defaecation, which was prominent in 16 (64%) of our patients might be an important factor. If continued during many years this may lead to perineal descent and rectal prolapse (Porter, 1962), thus intermittently stretching the nerve supply to the external anal sphincter muscles and, also, to a lesser extent, to the nerve supply to the puborectalis and to the lower part of the levator ani muscles. Sunderland (1968) found that a whole nerve could be stretched only about 7%, rarely as much as 20%, before sustaining damage. All our patients showed perineal descent during straining and 18 (72%) of the 25 patients with anorectal incontinence also had rectal prolapse (see Table). Thirdly, entrapment neuropathy, perhaps itself exacerbated by stretch-induced
of the mechanical changes associated with perineal descent and loss of the ano-rectal angle. Injury to the pudendal or inferior rectal nerves must be sustained, therefore, at a site such that the sensory fibres are relatively spared.

In addition to supplying the muscles of the ano-rectal sling and the external anal sphincter the pudendal nerve also supplies part of the external urethral sphincter and ischiocavernosus muscle (Williams and Warwick, 1975). This may account for the urinary incontinence which was present in 12% of our patients. In a wider context, therefore, the observation of sphincter denervation in ano-rectal incontinence implies that denervation of the urinary

Fig. 8  Control case (necropsy). ATPase pH 4.5 × 140. External anal sphincter. There is a preponderance of dark type 1 fibres, and these are generally smaller than the pale type 2 fibres.

movement (McLellan and Swash, 1976) of the pudendal or inferior rectal nerves, might also be a factor. The pudendal nerve lies between the coccygeus and piriformis muscles before passing through the greater, and then through the lesser, sciatic notches to enter the pudendal canal and entrapment might, perhaps, occur at any of these four sites. This suggestion needs further anatomical and histological study. Further, the details of the innervation of the anal sphincter mechanism also need clarification (see Winckler, 1957).

Idiopathic ano-rectal incontinence occurs predominantly in women. This cannot be explained entirely by the stresses of pregnancy and childbirth, however, since seven of our patients were nulliparous. All but one of these seven patients, and the single man in our series (case 19), gave a history of excessive straining at defaecation during many years and we, therefore, attach particular importance to this symptom (see Porter, 1962). In considering the possible sites and mechanism of nerve injury in our patients it must be noted that in most cases the only sensory abnormality was difficulty distinguishing faeces from flatus. The latter sensation is probably more related to pressure changes than to stimuli arising in cutaneous receptors in the anal canal so that this sensory symptom may be the result simply

Fig. 9 Control case. Haematoxylin and eosin (frozen) × 140. (a) External anal sphincter; (b) puborectalis; (c) levator ani. There is slight variability in fibre size, most pronounced in the external sphincter and puborectalis muscles. The fibres of the external anal sphincter muscle are generally smaller than those in the other two muscles.
Sphincter denervation in anorectal incontinence and rectal prolapse

Sphincter mechanism may be a factor in some patients with idiopathic urinary incontinence particularly, perhaps, in women in whom urinary incontinence of gradual onset in middle life is associated with perineal descent, and in those who have experienced a difficult labour. This suggestion is consistent with the clinical findings in idiopathic urinary incontinence (Edwards, 1976) and warrants further work.

Finally, it is unlikely that attempts to treat anorectal incontinence by electrical stimulation of the anal sphincter musculature would be successful in the presence of such severe damage to these muscles, and to their innervation, and our observations are, therefore, relevant to current attempts to develop such appliances.

References


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